Development of social functioning in children with congenital visual impairment

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Declaration

I, Valerija Tadić, confirm that the work presented in this thesis is my own. Where information has been derived from other sources, I confirm that this has been indicated in the thesis.

Signed:

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Dedication

I dedicate this thesis to my family - my parents, Sofija and Božo, and my sister Isidora.

Mami, tati i šeni, s ljubavlju.

Abstract

Effects of significant visual impairment (VI) in childhood are profound and far-reaching, impacting on most developmental areas. In recent years, there has been a particular emphasis on the effects of VI on social communication and social cognition, with a focus on a potential link with autism. However, the mechanisms underlying specific socio-developmental difficulties and the 'autistic-like' presentation shown by some children with VI, as well as the mechanism by which many children with VI are able to overcome such developmental vulnerabilities, remain poorly understood and require further clarification. The goal of the research reported in the present thesis was to elicit further understanding of the developmental patterns of social functioning in children with VI, and gain a better appreciation of the role that language may play in these processes. The thesis focused on children with severe and profound degrees of congenital vision loss without additional impairments, as learning difficulties have been identified as a confounding factor. Their developmental outcomes were compared to those of a group of typically developing sighted children of similar age and ability. The children were assessed using parental/teacher questionnaires and a battery of developmental and experimental tasks targeting language, social communication, mental state understanding and discourse, and executive functioning. An important finding was a discrepancy between the structural language skills and pragmatic language use in children with VI. Additionally, a substantial proportion of children with VI showed socio-communicative profiles that were consistent with a broader autism phenotype. An investigation of the children's mental state language use, which was also reported, provided a useful context within which socio-pragmatic difficulties seen in children with VI in this research could be considered. A similar contribution was provided by a study of mother-child mentalistic language exchange, which emphasised specific strengths of socio-interactive environment of children with VI that future interventions can capitalise on. Furthermore, the developmental vulnerabilities imposed by VI were found to extend to a broader behavioural presentation in children with VI, including weaknesses in specific executive function domains. Such weaknesses, notably in cognitive shifting, were considered in the context of attentional mechanisms that may be particularly affected by vision loss in early development. A retrospective examination of attentional behaviours in pre-school development of children with VI helped to clarify these issues further, by shedding light on the potential precursors to the vulnerabilities in achieving social competence and adjustment in children with VI at school age. The findings are believed to offer original contribution to understanding the development of social functioning in children with congenital VI, and are hoped to contribute towards the diagnostic considerations and intervention strategies aiming to boost such children's developmental potential.

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Chapter 1

General Introduction

VISION, VISUAL IMPAIRMENT (VI) AND DEVELOPMENT: GENERAL OVERVIEW

Vision is involved in the development and functioning of most human processes. It is easy for any sighted person to take vision for granted; however it only takes closing one's eyes to fully appreciate the potency of its provision. It is a powerful sensory modality which integrates and co-ordinates the information provided by other senses, allowing the features of the external world to be consolidated as a unified experience (Rock, 1985). It is therefore easy to see how a dominant role of vision in human perception is beneficial for young children who are learning about the world and themselves in relation to that world, particularly in pre-lingual stages of development. What is perhaps more difficult to appreciate are the experiences of children who are born with a severe or profound vision loss and how they come to learn about the world that they share with others.

Children with congenital[1] visual impairment (VI)[2] provide a natural experiment for studying the role of vision in human development; however, understanding visual impairment itself and its impact on a child's life is not a straightforward issue, either psychologically or medically. Vision loss that has clinical and educational significance is a largely heterogeneous phenomenon in the aetiological and phenotypical sense, and even the terminology to describe individuals with this impairment both in the literature and practice has varied over the years (Cullinan, 1987). There is not a single cause or diagnosis of congenital VI, making children who are born with severely impaired or absent functional vision a highly heterogeneous population (Baird & Moore, 1993; Rahi & Dezateux, 1998). Furthermore, severe and profound congenital VI is thought to be relatively rare in childhood. There is a great variation in the incidence of severe VI in different

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[1] Congenital VI implies that some part(s) of the visual system fail to develop normally or are damaged at some point during gestation or the perinatal period (i.e., the period around birth) (Sonksen & Dale, 2002).

[2] Both in the literature and everyday language, 'blind' is the most commonly used term to describe an individual with sight loss. While such term implies a total lack of sight, most individuals who are labelled 'blind' have some, although profoundly or severely degraded, levels of vision. The term 'visual impairment' represents the heterogeneous nature of visual disorder more accurately and, for the purpose of the present thesis, it is used to refer to sight loss which has significant clinical and educational implications (see Chapter 2 for a further discussion).

parts of the world, with a greater prevalence occurring in developing countries compared to the developed world (Baird & Moore, 1993). However, in the UK it has been estimated that 4 to 5 in every 10 000 children are diagnosed with serious VI of significant clinical and educational concern in the first year of life, with the incidence increasing to nearly 6 per 10 000 by 16 years of age (Rahi & Cable, 2003). This relative rarity as well as diverse aetiology of congenital VI makes children with this impairment a particularly challenging model for psychological research, and in Chapter 2 some of the relating methodological issues are considered in further detail. Here it is important to mention that, in the study of children with congenital vision loss, theorists and researchers have over the years attempted to establish different levels of experimental rigour with an aim to paint an authentic picture of what the human brain provides to the developmental process, in the absence of visually-driven knowledge. However, even though methodological rigour seems like a reasonable scientific goal, the diverse presentation of children with congenital VI is powerful evidence that real life rarely provides the means for achieving such a goal. Nevertheless, children who are born with sight loss (both as a heterogeneous population and individually) provide a fascinating insight into human development when it takes a differential course. Despite many methodological obstacles, understanding such a course is of crucial importance, not only for illuminating the mechanisms that underlie typical vision-guided development, but also for understanding the impact that this specific disability may have on the lives of the children concerned and their families.

What is therefore the extent of such impact? Extensive clinical and research evidence shows that developmental constraints imposed by severely impaired or absent functional vision in childhood are significant and complex. Poor developmental outcomes as a result of VI in childhood have been reported in most areas, including personal, emotional, social, motor and cognitive development (e.g., Bigelow, 2003; Cass, Sonksen, & McConachie, 1994; Fraiberg, 1977; McConachie & Moore, 1994; Sonksen & Dale, 2002). Importantly, in recent years, there has been a particular emphasis on the effects of visual impairment on social communication and social cognition, and given these particular effects, a number of researchers and clinicians have highlighted striking behavioural resemblances between children with congenital VI and sighted children with autism (see Pring, 2005 for a general overview). Such research has not only brought under the spotlight specific developmental challenges and needs of children with VI; it has also offered novel insights into the development of social functioning in typical circumstances and into the mechanisms that underlie the social impairments in sighted children with autism. Importantly however, the mechanisms underlying specific social difficulties and the autistic-like presentation shown by some children with VI, as well as the mechanism by which many children with VI are able to overcome such developmental challenges and vulnerabilities, still remain poorly understood. The general rarity and heterogeneity of the population certainly contributes to the relative dearth of research with such children, maintaining the scarcity of knowledge of their developmental processes. Yet, gaining further insight and appreciation into the developmental trajectory of social communication and social cognition of children with VI is of great significance, from both psychological and clinical perspectives. Crucially, the goal of the research reported in this thesis is to provide a unique insight into the understanding of developmental patterns of social functioning in children with VI and to contribute to diagnostic considerations and intervention strategies aiming to boost developmental outcomes for such children.

Before specific developmental patterns of children with VI are considered empirically in later chapters, the aim of this chapter is to provide a review of the literature concerning the research and theory of social functioning in child development. Starting with the origins of such functioning and leading to its later manifestations, this literature review will also consider the role of other specific factors that are thought to be involved in the trajectory of social functioning in childhood. More specifically, in the first and main part of the review, the developmental trajectory of social functioning in sighted children will be considered, before addressing the characteristics of such trajectory in children with VI. The latter part of the literature review will specifically address language and its relationship with important social milestones in child development, with a particular focus on its role in the development of children with VI.

DEVELOPMENT OF SOCIAL FUNCTIONING IN CHILDREN WHO ARE SIGHTED

While vision has been given a central role in most developmental processes, its significance has been particularly emphasised in the development of socio-communicative skills and socio-cognitive understanding. Therefore, the aim of the following section is to consider the theory and empirical evidence that highlights this particular role of vision in the development of pivotal social milestones in childhood.

Early social functioning

Joint attention

Infants enjoy rich and stimulating social lives from the earliest stages of development. Early social experiences are dyadic in nature, with an infant taking part largely in face-to-face interactions only with one social partner at a time. These early social interactions are characterised by what Trevarthen described as *primary intersubjectivity* (Trevarthen, 1979), where infants demonstrate responsive conscious appreciation of the adult's communicative intentions and signalling by engaging in mutual eye-gaze, vocalization and rhythmic turn-taking patterns of behaviour (e.g., such as in social games like 'peek-a-boo'). From around six months of age, the new patterns of communication emerge, as the child moves from the purely dyadic interactions with one social partner into the world of objects. The main characteristic of these novel experiences, which have been conceptualised as *secondary intersubjectivity* (Trevarthen & Hubley, 1978), is the infant's awareness that their experiences of objects, people and events can be shared with others. This transition from the infant's dyadic (child-caregiver) engagement to triadically shared (child-object-caregiver) experiences marks one of the major milestones in socio-communicative development. The central component of this milestone is what is now commonly known as *joint attention*, referring to co-ordinated sharing of attention between the child, an adult and objects in space (Bakeman & Adamson, 1984; Corkum & Moore, 1998; C. Moore & Dunham, 1995; Mundy, Kasari, & Sigman, 1992). Its behavioural manifestation encompasses a complex set of actions, such as eye-gaze directing and following, point following, showing and pointing, the purpose of which is to negotiate and share the mutual focus of interest with a social partner. Research evidence suggests that, in typical development, such behaviours emerge between six and 12 months, and are consolidated by 18 months of age (Bakeman & Adamson, 1984; Markus, Mundy, Morales, Delgado, & Yale, 2000).

Joint attention experiences that infants and their caregivers mutually share are largely driven by the visual modality, so they are often referred to as 'joint visual attention' (Corkum & Moore, 1995; Hobson, 1993; Tomasello, 1995). The dominance of eye-gaze behaviours in this process has largely been reflected in the empirical studies of joint attention in children (both in naturalistic and laboratory settings), where the child's ability to make eye-contact and monitor the direction of another's eye-gaze, with the purpose of initiating or responding to adults' bids of attention, have been taken as measures of joint attention competence (e.g., Bakeman & Adamson, 1984; Scaife & Bruner, 1975).

Theoretical accounts of joint attention

The visual nature of joint attention has also been emphasised in theoretical accounts of this process. As will be described in this section, while different accounts of joint attention postulate the involvement of different mechanisms that underlie the development of the ability to share the mutual focus of attention with others, the common theme that these accounts share is an emphasis on vision in both the emergence and manifestation of these experiences.

The cognitive-modular account of joint attention highlights the involvement of cognitive mechanisms that are responsible for meta-representational functioning involved in the process of attention sharing (Baron-Cohen, 1995a). According to Baron-Cohen, these mechanisms (i.e., modules) enable the child to construct triadic representations that specify self and others attending to the same objects. This account highlights the importance of the 'intentionality detector' module (ID), which enables young children to appreciate other people as intentional agents who have their own representations of objects and events in the world. Further emphasis in this account is on the role of an 'eye direction detector' (EDD) module, the function of which is to interpret visually mediated information (e.g., looking where someone else is looking). The child's representations of intentionality and their ability to follow the visual focus of others combine within the 'shared attention mechanism' module (SAM), which is responsible for the triadic sharing of attention between a child, a caregiver and a mutual focus of attention (i.e., joint attention) (Baron-Cohen, 1995a).

In contrast to the cognitive-modular account, Hobson (1993; 2002) argues that joint attention has an affective origin, with affective sharing of experiences through smiles, facial expressions and gaze monitoring providing a basis for both dyadic and triadic interactions in which a young child takes part. The starting point in Hobson's interpersonal-affective account is the child's ability to apprehend other people as persons with attitudes. Central to this account is the concept of interpersonal engagement within the 'relatedness triangle' involving the child, the other and the referent (e.g., object) of mutual interest. Through experiences of many of these triangular relationships, the child learns that different people can have different attitudes toward the same referent and that the same person may produce different attitudes towards the same referent. Crucially, according to Hobson, the perception of the other's attitude, the affective component of that attitude, and the fact that the attitude is directed at a specific referent in the world, is dependent on the visual modality. Vision enables children to triangulate their own attitudes as well as the attitudes of others towards the visually-specified objects (e.g., through gaze-directing, gaze-following, pointing and showing), and provides them with the means for achieving psychological co-orientation and co-reference with other people.

Others have argued that joint attention has a more basic perceptual precursor, related to development of visual attention and the changes that attentional systems undergo in the first year of life (Corkum & Moore, 1995, 1998; Scaife & Bruner, 1975). For instance, while reflexive attentional orienting to sensory stimuli is typically present from birth, voluntary attentional control is generally poor until around the third month, when infants become able follow the cue of another person's eye-gaze, although this is only if the target stimulus is in the infant's visual field (Atkinson, Hood, Wattam-Bell, & Braddick, 1992; Hood, Willen, & Driver, 1998). Between nine and 12 months, an important change in attentional capacity occurs as infants begin to use another person's head-turn and eye-gaze as an attentional cue, even when the target stimulus is not visible (Butterworth & Cochran, 1980; Butterworth & Jarrett, 1991; Corkum & Moore, 1998). However, it is not until 18 months of age that they are able to rely on the adult's eye-gaze alone to establish a joint reference with another person (Butterworth & Jarrett, 1991; Corkum & Moore, 1995). This evidence emphasises the importance of visual attention orienting to sensory stimuli and attentional control in the development of joint attention.

Developmental implications of joint attention

It is generally agreed that early joint attention experiences are of great developmental significance, as they provide optimal conditions for the general learning of young pre-lingual children. For instance, the socio-interactive context within which joint attention occurs is thought to provide a rich framework for the child's emerging symbolic understanding and word learning, thus acting as an important precursor to symbolic play and language development (Charman et al., 2000; Mundy & Gomes, 1998; Tomasello & Farrar, 1986; Tomasello & Todd, 1983). The mutual exchange of interests, intentions and attitudes between the child and the interacting adult, towards each other and objects in their shared environment, is seen as providing a *scaffolding* mechanism upon the child's language learning takes place (Tomasello & Farrar, 1986). Indeed, the empirical evidence shows that the capacity of the child to respond to and follow the adult's focus of attention, as well as the time spent in joint engagement involving a caregiver, in early infancy is predictive of children's earliest gestural communication and linguistic competence and their later lexical acquisition (Carpenter, Nagell, & Tomasello, 1998; Markus et al., 2000; Tomasello & Farrar, 1986). Importantly, joint attention is seen as providing a framework within which young children learn clues to different mental states and expressions of emotions, through observing the facial expressions, gestures and bodily postures that caregivers direct towards them and other agents in their environment. As such, joint attention is believed to act as a stepping stone for the development of more sophisticated forms of social functioning later on (Hobson, 2002; Tomasello, 1995). A specific social and cognitive achievement, thought to be a direct consequence of joint attention ability, is now commonly known as *theory of mind*. This particular milestone in child development, which has been given a widespread attention over the past 25 years both in theory and empirical research, will be considered in the following section.

Later social functioning

Theory of mind

Theory of mind, 'mind reading', 'mentalising' and 'understanding of others' minds' have synonymously been used in psychology to refer to the child's ability to understand and attribute a range of mental states to self and others in order to explain and predict their actions and behaviours (Meltzoff & Gopnik, 1993; Perner & Wimmer, 1985; Wellman, 1990). In other words, to make sense of the sophisticated social environment that surrounds them, children must be able to understand that other people have intentions, desires, thoughts, beliefs and feelings which are different from their own and that such states of mind will influence people to act and behave accordingly.

Theory of mind has been extensively studied in children using false-belief tasks, which developmentally have been categorised as those of first and second order. First-order false-belief tasks typically employ a character-based scenario (using puppets or pictures) in which a child is required to predict behavioural outcomes of a story character, based on that character's belief, which may be false. For example in the well-known Sally-Anne task (an adaptation of a task originally designed by Wimmer and Perner, 1983), a child who has first-order false-belief understanding knows that Sally will look for her marble in the basket where she initially left it because she is unaware that Anne has moved it to another basket in her absence. That is, the child understands that Sally has a false belief about where the marble really is (Baron-Cohen, Leslie, & Frith, 1985). This is known as an 'unexpected transfer' false belief task. Another example of a first-order false belief task is the 'unexpected contents' type of task. In this task, where a child is presented with a familiar object that would usually contain familiar contents (e.g., a Smarties tube), but which now contains something unexpected (e.g., a pencil), a child who has false-belief understanding is able to correctly predict that the person who is unaware of the unexpected contents of the Smartie tube would think that the tube contains sweets (Perner, Leekam, & Wimmer, 1987).

Based on children's performance on variations of such tasks, it is now generally accepted that first-order theory of mind ability is typically acquired by the age of five (Ruffman, 2004; Tager-Flusberg, 2001; Wellman, 1990). Second-order theory of mind however, is thought to develop between the ages of six and eight, when the child begins to develop understanding of more complex and embedded mental representations (Baron-Cohen, 1989; Perner & Wimmer, 1985). To pass a second-order false-belief task, a child needs an awareness not just that people have beliefs about the world (which may be different from child's own beliefs), but also that they have beliefs about the content of others' minds (i.e., about others' beliefs), and similarly, that these too may be different or false. An example of such a task is Perner and Wimmer's (1985) story of John, Mary, and the ice-cream man in the park. Here, the child is told that, while Mary goes home to get money to buy ice-cream, the ice-cream man tells John that he is going over to the church to sell ice-cream there. On his way to the church, Mary happens to see him and the ice-cream man tells her the same thing he told John. However, John does not know this, so later, when he goes to Mary's house and her mother tells him that Mary has gone out to buy ice-cream, John runs off to look for Mary. Perner and Wimmer found that, by the age of seven, children are able to predict correctly where John thinks Mary has gone (i.e., the park) and they are able to justify their answer.

Theoretical accounts of theory of mind

A number of theories that have been proposed to explain a child's theory of mind have linked its development to specific visual precursors, such as joint attention and the processes occurring within this context. For instance, following the previously described cognitive-modular account of joint attention by Baron-Cohen (1995a), children's theory of mind ability has been viewed as a result of the functioning of an innate, specific neuro-cognitive module (Baron-Cohen, 1995a; Baron-Cohen & Swettenham, 1996; Leslie, 1987, 1994). According to Baron-Cohen, *the theory of mind module* (ToMM) is preceded by activation of the three previously mentioned modules that are involved in joint attention development, namely the intentionality detector (ID), the eye direction detector (EDD) and the shared attention mechanism (SAM). Importantly, all of these modules become available to the child at different developmental stages, following a specific developmental sequence. The first module that becomes available and which allows interpretation of intentionality (ID), feeds into the module that allows following the visual focus of others (EDD). The functioning of the EDD module in turn leads to activation of the SAM module, which enables the child to grasp the skill of sharing attentional focus with others and gives rise to joint attention behaviours and interactions. Activation of the SAM ultimately feeds to the ToMM, which is responsible for the child's developing appreciation of others as intentional agents with attitudes, thoughts and beliefs of their own.

Leslie (1987) argued that earliest representations of ToMM can be seen between 18 and 24 months of age when the child begins to engage in *pretend play*. Pretend play involves the capacity to comprehend simultaneously multiple representations of an object (i.e., understanding that one object can stand for another, that pretend properties can be attributed to real objects and that pretend interaction can be carried out with a non-real object) (Leslie, 1987, 1994). To understand pretence, children need to understand something about other people's minds. For example, in order to play along when their mother pretends a banana is a telephone, a child must have an understanding that the mother is projecting the thought of a telephone onto a banana. Therefore, pretend play is evidence of child's metarepresentational capacity and understanding that thoughts, and not reality, guide people's behaviours.

Like Leslie, Harris (1992; 2000) emphasised the role of pretence in children's theory of mind. According to Harris, children's understanding of the mind involves introspective awareness of their own mental states and they can use this awareness to infer the mental states of other people. According to Harris, children develop theory of mind understanding by *simulation* or role taking, and pretend play provides the context within which this ability develops. Through practice in role taking and the increasing ability to use pretence *by observing* the behaviour of others, children improve their simulation skills and learn to draw complex analogies between themselves and other people. This in turn facilitates the acquisition of sophisticated socio-cognitive knowledge and the understanding of other's subjective minds.

A different theory of mind account postulates that in their attributing of mental states children need to resort to a *theory* to predict and explain behaviour (e.g., Gopnik & Meltzoff, 1997; Gopnik & Wellman, 1994; Perner, 1997). This is known as the *'theory-theory'* account. According to this account, throughout the course of development, and in addition to a powerful innate representational system, children also develop, change and qualitatively reorganize their theories of the world, and they do so on the basis of the input they receive (Meltzoff, 1999). Through the developmental progression of theories (i.e., a developmental sequence by which different mental states are understood), the child develops a fully-fledged representational theory of mind at around the age of four (Gopnik & Wellman, 1994). Central to the theory-theory account provided by Meltzoff and Gopnik is *imitation* (Gopnik & Meltzoff, 1997; Meltzoff & Gopnik, 1993). Children are born with an innate predisposition to imitate others who they perceive are like them, and the evidence for this comes from the studies of infants being able to imitate adults' facial gestures days and weeks after birth (e.g., Meltzoff & Moore, 1977; Meltzoff & Moore, 1983). However, in order to imitate, the child is required to *see* the adult's actions (e.g., facial expressions and gestures), use this *visual perception* as a basis for an action plan, and execute the motor output. Through visual imitation, children model not only another person's motor acts but also their intentions, emotional attitudes and reactions towards specific objects or events. It is therefore clear how vision provides a premise for the development of joint attention and understanding of others' minds.

Atypical social functioning

Autism[3]

Theoretical and empirical advances in the understanding of the development of social milestones such as joint attention and theory of mind in the past 20 years have largely been a consequence of the clinical emergence of autism. Autism is a pervasive neuro-developmental disorder characterized by disturbances in social and communicative development and imagination (Wing & Gould, 1979). Although biologically based, with a strong genetic component, the disorder is defined and diagnosed on the basis of the triad of behavioural difficulties, namely in social interaction, communication, and repetitive behaviours and restricted interests (DSM-IV-TR, 2000).

Autism has been extensively researched, which resulted in a number of prominent accounts, the aim of which was to predict and explain the triad of impairment that is characteristic of the disorder. These accounts are beyond the scope of the current thesis and their detailed descriptions can be found elsewhere (Frith, 1996, 2003). However, one account of autism that is pertinent here, postulates that individuals with autism have a deficit in theory of mind. That is, the cognitive mechanisms responsible for understanding and attributing mental states to others are impaired in autism, giving a rise to the specific socio-interactive and communicative impairments that characterise the disorder (Baron-Cohen, 1995b, 2000). The evidence for the theory of mind deficit in autism comes from the numerous studies assessing false-belief understanding in such children, who have been consistently found to fail these tasks when compared to matched typically developing controls (Baron-Cohen et al., 1985; Happé, 1995; Perner, Frith, Leslie, & Leekam, 1989). The poor false-belief comprehension of children with autism is particularly striking, considering that it is generally independent from other skills that involve similar types of reasoning, but which exclude mental state concepts (e.g., comprehension of false photographs, Leekam & Perner, 1991; Leslie & Thaiss, 1992). Additionally, many high-functioning children and adults with autism have difficulties at the level of theory of mind (Baron-Cohen, Jolliffe, Mortimore, & Robertson, 1997; Ozonoff, Pennington, & Rogers, 1991) and non-autistic children with learning

[3] It is generally agreed that autism is a spectrum disorder, with a clinical picture that varies in severity across diagnosed individuals. In this thesis, the term 'autism' will be used throughout to refer to the spectrum of the condition.

difficulties (e.g., Down Syndrome) generally do better by comparison (Baron-Cohen et al., 1985). It therefore appears that the poor theory of mind performance of children with autism cannot be explained in terms of their general intellectual abilities, although general linguistic competence can make a significant positive impact (Bowler, 1992; Happé, 1995).

Importantly, the core theory of mind deficit in autism has been related to the disruptions in joint attention in early childhood and the lack of behaviours such as gaze and point following, showing and declarative pointing (Charman, 2003; Dawson et al., 2004; Hobson, 1993; Leekam, López, & Moore, 2000; Mundy, Sigman, & Kasari, 1994). Absence of these behaviours in children with autism forms one of the criteria for diagnosing the disorder (DSM-IV-TR, 2000). One argument is that poor joint attention in children with autism is indicative of the damage to the cognitive mechanism/ module that is responsible for developing triadic representations of self and other attending to the same object, and subsequently impairing the ability to form mentalistic interpretations of other's actions (Baron-Cohen, 1995a). As they are unable to represent another person's attention or interest towards the external world, children with autism are unable to participate in joint attention and the relevant behaviours. Consequently, this leads to the impairments in socio-communicative and socio-cognitive skills for which joint attention provides a stepping stone. As a result, children with autism have marked difficulties in symbolic understanding and pretend play (Baron-Cohen, 1987; Charman et al., 1997), language (Tager-Flusberg, 2000), and theory of mind (Baron-Cohen, 2000). Another explanation for the failure to engage in acts of joint attention in autism is the breakdown in interpersonal engagement and the affective, intersubjective experiences with others (Hobson, 1993, 2002). Thus, joint attention impairment in autism is not only confined to the triadic interactions (child-person-object), but is also seen at a more basic dyadic face-to-face level of interaction with another person. Without this innate predisposition, a child with autism is unable to comprehend attitudes of other people and relate to external events as shared. Similar to the previous argument, this lack of affect-driven joint attention experience in autism is also detrimental for the child's development of social interaction, symbolic understanding and concept of the mind (Hobson, 2002).

According to Hobson (1993) the failure to relate to other people's relatedness to the world is a defining feature of children with autism. Descriptively, such children generally seem socially aloof and distant. Thus, in many ways they appear *as if* they are *unable to see* the social world that surrounds them and within which different mental states and feelings arise (Cass, 1998). Keeping this insight in mind, it is now important to consider those children who are unable to see their social surroundings due to a significant loss of vision at birth, while autism will be considered again later in this chapter in relation to these children.

DEVELOPMENT OF SOCIAL FUNCTIONING IN CHILDREN WITH CONGENITAL VI

Following the underlying principles of the aforementioned developmental theories of joint attention and theory of mind, which emphasise the involvement of vision, it is reasonable to assume that any significant and long-term disruptions to visual processes will have a detrimental effect on the development of such functions. This certainly has implications for the development of children who are born with severely or profoundly impaired functional vision for whom, in absence of the shared attention mechanisms (Baron-Cohen, 1995a), visual imitation (Meltzoff & Gopnik, 1993) and visually-driven, triangular interpersonal engagement (Hobson, 1990) the social communication and social cognition are likely to be vulnerable developmental areas. The theory and empirical evidence illustrating the related developmental patterns in children with VI will therefore be reviewed in the following section.

Early social functioning

Joint attention

It has been suggested that, despite the dominance of the visual modality in development of joint attention, its acquisition is possible in children with VI. Baron-Cohen (1995a) argued that even though lack of vision may hinder the development of the SAM, this mechanism is amodal, and is able to build triadic representation in other non-visual modalities. In children who are visually impaired, for instance, SAM can function through auditory and tactile modalities and a child without functional vision may construe a representation of another person's attention on an object of interest by feeling that person's hand on that object. Similarly, Hobson (1990; 1993; 2002) hypothesised that joint attention is possible in children with severe and profound VI, emphasising the role of social experience in facilitating the child's joint engagement with other people and the possibility of an alternative non-visual route for achieving co-reference with others in such children. However, both theoretical accounts imply that, although joint attention can be attained via non-visual means in children with serious VI, its emergence and manifestation is likely to be delayed.

Empirically, however, very little is known about the development of joint attention in children with VI. The empirical study of joint attention in children in general has been so heavily influenced by the dominance of eye-gaze behaviours, facial expressions and visual gestures in this process that our knowledge of its non-visual aspects is extremely limited. Consequently, we cannot be certain of the full extent to which joint attention experiences may (or may not) be disrupted in children with VI. In fact, given the scarcity of research with such children, the way in which they develop joint attention is virtually unknown.

In typically developing sighted children, joint attention ability is believed to develop spontaneously, evolving out of a natural context of routine child-caregiver interactions, and the caregiver's sensitivity and responsiveness to the child are the key ingredients to the child's developing interpersonal engagement (Recchia, 1997; Sandler & Hobson, 2001). Vision is likely to facilitate the caregiver's involvement, the manifestation of which is likely to be different for children whose attention cannot be directed through eye-contact and visual gestures. However, the poor theoretical and empirical understanding of this aspect of development of children with VI does not automatically imply that the interactions between children with VI and their parents are devoid of affective sharing and social enjoyment. In fact, some evidence suggests that joint attention between children with VI and their parents can be established through alternative (i.e. tactile, auditory, kinaesthetic) modalities (Bigelow, 2003; Preisler, 1991; Urwin, 1978). However, the nature and the function of these non-visual behaviours in children with VI are still poorly understood and there is generally conflicting evidence regarding parental responsiveness to their visually impaired child's attentional cues.

In a longitudinal study, Preisler (1991) observed the patterns of social interaction between 10 infants with profound visual impairment and their mothers, across the children's first year of life (i.e., 3-12 months of age). Preisler noted that the early communicative patterns between the children with VI and their mothers very much resembled the dyadic experiences of primary intersubjectivity in sighted children, as described by Trevarthen (1979). More specifically, these interactions were characterised by the dyads concentrating on establishing an affective bond; while being largely unaware of the external world, the infants were responsive to their mothers and engaged in proto-conversations by exchanging smiles, showing lip movement in an articulation-like manner and imitating speech. Preisler also observed instances of infant hand movement that were co-ordinated with the mother's speech, a behaviour which, in sighted children, normally occurs in combination with mutual eye-contact between mother and infant (Trevarthen, 1979). Additionally, these infants with VI showed signs of intentionality, by exhibiting repeated body movements and hand gestures in play and body-touching games with the parent. Based on these observations, Preisler noted that vision may not be a necessary condition for certain aspects of social interaction to develop. Importantly however, around the children's first birthday, they had notable difficulty with establishing secondary intersubjectivity, the experience of which marks the ability to engage in triadic joint attention (Trevarthen & Hubley, 1978). Although they could share themselves with their mother, aided by the mother's affect attunement, the children were unable to co-ordinate their attention at the same time towards an object in the external world. Interestingly, Preisler noted that the infants with VI were attentive to the sounds in the environment and reacted to these by establishing frozen bodily and facial postures. However, while these subtle signs, in addition to distinctive body pointing towards the sound, may provide the means of attention directing from the visually impaired child's perspective, such behaviours may be too subtle and ambiguous for parents to interpret or notice (Preisler, 1991).

In relation to this, drawing from their study on social characteristics of play in children with VI, Rogers and Puchalski (1984) commented that both partners in the child-mother interaction, where the child is visually impaired, are disadvantaged. While the child is deprived of visual information and the lack of effective communication by the mother, who cannot interpret the child's signals, the mother is deprived of the positive and responsive cues from her child that would let her know that she is doing the right thing. This study strongly highlighted the poverty of responsive social exchanges and initiations in mother-child dyads in the case of children who are visually impaired, in contrast to the interactions of sighted children and their mothers. Presumably, this 'vicious circle' of impoverished parent-child responsiveness is likely to be both a cause and a result of the impoverished joint attention capacity seen in children with VI.

However, in a study of two infants with congenital profound VI, Urwin (1978) showed that the nature of caregiver-child responsiveness was largely adaptive. Once the mothers discovered particular cues that elicited the response of their child with VI, they were able to use these cues repeatedly: *"(They) used phased touching routines to alert the babies' attention; they would trace their fingers around the babies' mouths, blow on their faces, and encourage them to explore their own body parts. (They) would mock-imitate the babies' fusses, coughs, splutters and sneezes to 'dramatize' the babies' actions"* (Urwin, 1978, p. 88). However, despite the effective socio-interactive routines that facilitated the dyadic relationships between the children with VI and their mothers, both infants studied by Urwin showed difficulties and delays in their triadic interactions that required them to incorporate objects into their interactions with the adult and to establish reversible exchanges of actions on objects. Neither child exhibited spontaneous 'showing' behaviours to initiate joint interaction with the mother. If any reverse actions of 'giving and taking' emerged, they were largely the result of specific training provided by the mother. Therefore it seems that, while affective sensitivity and non-visual resourcefulness provided by the primary caregiver plays an invaluable role, severe lack of vision is still likely to affect the child's intersubjective engagement and joint attention capacity.

In a more recent study, Bigelow (2003) focussed specifically on non-visual actions that may be suggestive of joint attention in children with VI, differentiating three types of behaviours. The first type of behaviours were those that are preliminary to joint attention, but may not necessarily lead to it, such as using the adult as a social tool, and instrumental and self-stimulating behaviours that may be interpreted as communicative gestures. The second type included behaviours that may be liberally construed as joint attention, but may be somewhat questionable as true joint attention behaviours. These are involved in comprehension and production of language, such as spontaneous labelling of objects and actions, and using adult's verbal instructions to engage with objects. Finally, the third cluster of behaviours investigated by Bigelow were those that might be conservatively suggestive of joint attention and may be less questionable indicators, in that the child's actions are more clearly indicative of their awareness of the adult's role in their mutual interaction with objects (e.g., repeated giving and taking of an object, labelling of an object at the request of an adult, and joint manipulation of an object). While the interpretation of these behaviours (particularly the first two types) may often be ambiguous, Bigelow argued that they may serve a different function in children with VI compared to sighted children, and should be considered in the context of their development. For instance, while using adults as social tools may not necessarily be indicative of intention to share focus of attention in sighted children (e.g., children with autism, who typically do not engage in joint attention, often show such behaviour), this behaviour may serve a function of locating desired objects for a child who is visually impaired, hence providing an adult with a valuable attention-directing cue. Bigelow assessed the occurrence of these behaviours over a period of time in two infants with profound congenital visual impairment, using a series of object search tasks that involved presenting the infants with silent and continually sounding toys and observing the infants interacting with the toys and the familiar adult.

The theoretical background underlying this investigation was that in sighted children, joint attention development is closely tied with the development of self-knowledge, particularly the knowledge of self in relation to environment (Neisser, 1991). This knowledge in children with VI is shown to be largely dependent upon their understanding of space and objects (Bigelow, 1992, 1995). Children with VI need to develop the understanding that objects that they attempt to reach do exist and have a permanence of their own and which is independent of self. They have to learn where they are in relation to such objects and where the objects are in relation to one another as well as to understand that they can use their bodies to explore these objects. Typically, vision guides the co-ordination and understanding of relations between objects in the environment and self-position in space (Rieser & Rider, 1991). However, such co-ordination and understanding in children with VI is only likely to be possible after they have mastered the concept of object permanence and the ecological self (Bigelow, 1995, 2003). Bigelow (2003) observed that the two children she studied supported this developmental pattern; the three types of joint attention behaviours only began to occur in the two children with VI once they developed their sense of ecological self and began to reach for objects in their environment. Furthermore, there was a developmental sequence of joint attention behaviours. The preliminary joint attention behaviours, such as the infant's use of the adult's bodies to find desired objects, emerged before the more liberally construed (e.g., object labelling) and conservatively construed (e.g., giving and taking game) joint attention behaviours, which occurred in a close parallel with one another. Importantly however, the children examined by Bigelow achieved these milestones later (i.e., 13-19 months in one child and 17-29 months in the other) than sighted children, whose self-knowledge and joint attention are established before the first year of life. Importantly, the achieved joint attention episodes between the children with VI and their parents were always initiated by the caregiver. Even though they actively participated in the maintenance of joint attention episodes initiated by the adult, the children studied by Bigelow did not instigate triadic interactions themselves.

The findings so far (Bigelow, 2003; Preisler, 1991; Urwin, 1978) support the hypothetical stance (e.g., Hobson, 1990; 1993, 2002) that joint attention experience in the absence of visual stimulation, although delayed, is possible for children with VI, implying that the role of vision in these processes is not absolute. Children with severely impaired or absent functional vision depend developmentally on tactile information and memory, as well as auditory input such as sound changes, air currents, echolocation (Millar, 1988), and verbal guidance by others (Urwin, 1978). Such experiences must, at least to an extent, allow them to learn to co-ordinate the spatial placement of objects and to establish a shared focus on such objects with others. However, despite the evidence of joint attentional engagement in children with VI, it generally appears that the nature of such engagement is qualitatively different from what is known about the joint attention capacity of sighted children, and this is particularly evident at the level of triadic interactions of secondary intersubjectivity. Additional studies, with larger samples, are certainly necessary to further our understanding of joint attention and the processes underlying its development in children with VI. However, delayed attainment of specific joint attention behaviours in children with VI is suggestive of such children's developmental vulnerability which is likely to extend to further social developments. More specifically, given the importance of joint attention for the subsequent social achievements such as theory of mind, these achievements in children with VI are likely to be compromised (Hobson, 1990; 1993, 2002). The aim of the following section is to consider these developments in more detail.

Later social functioning

Theory of mind

In line with Hobson (1990; 1993; 2002), several studies to date have shown that children with VI indeed have a specific difficulty in theory of mind understanding. The first such study was carried out by McAlpine and Moore (1995). Here, the first-order false-belief understanding was assessed in a group of 16 children with varying degrees of VI using two tactile versions of the unexpected contents tasks (Perner et al., 1987). The study was descriptive in nature as the wide range of ages and levels of verbal impairment in the sample prevented the use of standard statistical procedures. Additionally, it did not include a sighted control group to which to compare the performance of the children with VI. However, descriptions at the level of individual children showed that some children with VI, and in particular those whose VI was of greater severity, were unable to pass false-belief tasks which are typically expected to be mastered by sighted children of similar mental ability (Perner et al., 1987). However, McAlpine and Moore noted that some children with VI were able to complete the two tasks correctly, although such performance was confined solely to those children whose verbal mental ages were greater than 11 years and whose VI was of lesser severity. Despite its methodological drawbacks, the study by McAlpine and Moore provided the first suggestive evidence that children with congenital and profound VI may be subject to a delay in theory of mind development, in line with the role of related visual precursors proposed by Hobson. Importantly, the study paved the way for more controlled studies, which were consistent in showing that a subset of children with congenital VI, who do not have any additional impairments, experience difficulty with standard false-belief tasks at mental ages older than four.

For instance, Minter et al. (1998) addressed some of the methodological problems of McAlpine and Moore's study by testing first-order false belief understanding in a larger and more homogeneous group of children in terms of the severity of their VI (i.e., those who had light perception or less) and by employing a sighted control group matched to their VI sample on chronological and verbal mental age. The 21 children with VI in this study all had verbal mental ages above four years. The VI and the sighted control groups were presented with a variation of the unexpected contents task (Perner et al., 1987) and a variation of the unexpected transfer task (Wimmer & Perner, 1983), both using the tactile presentation to suit the VI group. The results of the study confirmed the previous findings by McAlpine and Moore that a substantial number of children with congenital VI may indeed have a difficulty with first-order false-belief understanding at mental ages older than four years. However, there was a discrepancy in the VI groups' performance across the two tasks, with particularly notable lower performance on the unexpected contents task. It has been argued that the version of the unexpected contents task adapted by Minter et al. for this purpose may have underestimated the VI group's false-belief awareness, as it introduced an object (i.e., warmed tea-pot) which could be perceived as dangerous by children with VI, who have to explore such stimuli manually (Green, Pring, & Swettenham, 2004). In everyday circumstances children with VI may be more likely to identify materials such as hamburger wrappers and a teapot on the basis of aroma and heat, rather than their shape and this is likely to affect their judgement of what someone else would think is inside an odourless container. For this reason, the failure on the unexpected contents task involving a teapot or a hamburger wrapper may reflect a modality-specific perceptual problem in children with VI, confounding their theory of mind performance on this specific false-belief task (Peterson, Peterson, & Webb, 2000).

Following from this, Peterson et al. (2000) attempted to test more sensitively the discrepancy between unexpected contents and unexpected transfer task performance in children with VI, by using two variations of each type of task used in the study by Minter et al. (1998). Rather than carrying out a comparison study with a control group, Peterson et al. (2000) were interested in systematically investigating developmental change in false-belief understanding in children with VI, over the period of five to 12 years. They assessed two groups of children with differing levels of VI and across differing ages (averaging at six, eight and 12 years). The findings of the study showed that, while the majority of the six year olds failed all four false-belief tasks, false-belief performance improved with age, although significant difficulties could be seen in some eight year olds and to a lesser extent in the 12 year olds. However, while age was found to be a significant and unique contributor in false-belief performance in this VI sample, the level of visual impairment was not so, and the pattern of theory of mind delay was seen across the range of severity of VI. Peterson and colleagues suggested that in children with VI, the limited access to non-social cues (e.g., facial expressions and eye-contact), which are important for discerning the conversational information on conversational partner's attentional focus and emotional attitude, restricts the child's opportunity to appreciate what is in other's minds, impacting on their theory of mind. They argued that the theory of mind difficulties seen in children with VI, as a consequence of a restricted access to early socio-conversational experience, are a direct result of vision loss, rather than a delayed language or a lack of conversation partners, although this link was not specifically addressed by the study.

More recently, Green et al. (2004) assessed false-belief task performance of a group of 18 primary-school-aged children with congenital and total sight loss, and no other impairments. In order to control for intellectual level, lacking from the previous studies, the children with VI were individually matched to a group of sighted controls by age, verbal IQ and verbal mental age. All the children were assessed on three first-order false-belief task adapted from the previous studies on theory of mind in children with VI. The findings by Green et al. supported the previous studies' findings that children with congenital and profound vision loss have a difficulty with first-order false-belief task, when compared to developmentally-matched sighted controls. Interestingly, however, the verbal ability level significantly distinguished children with better and poorer false-belief understanding in this study. Importantly, further investigation by Green in her PhD (Green/née Cupples, 2001) revealed that many of the congenitally blind children she studied were able to catch up with their sighted peers on more advanced theory of mind tasks, once they mastered basic false-belief. However, as well as lending support to the general pattern of delay in theory of mind development observed in children with severely restricted vision, Green also identified a subset of children with VI who have-long term difficulties in the area.

Such findings were most recently supported by Roch-Levecq (2006) who also demonstrated that primary school aged children with congenital and profound vision loss who have normal intelligence have a significantly poorer false-belief understanding than developmentally matched sighted controls. Interestingly however, in addition to poorer false-belief task performance, the children with VI in this study were found to be less accurate in conveying emotions facially to adult observers than were sighted participants. Similar findings were reported by Galati and colleagues (Galati, Miceli, & Sini, 2001), who found that voluntary expressions of emotion distinguished children with VI from sighted children, although their spontaneous expressions did not. Such findings can be explained in terms of the breakdown in primary intersubjectivity (Trevarthen, 1979) and what Hobson (2002) called affectively-charged interpersonal engagement (discussed previously in this section, see p. 17). Interpersonal engagement is perceptually (i.e., visually) and affectively driven, with an innate predisposition towards the bodily appearances and behaviour of others (Hobson, 1991), and visual imitation is a manifestation of such predisposition

(Meltzoff & Gopnik, 1993). Accordingly, lack of vision hinders a child in developing representations of other people's mental states, resulting in both ambiguous facial emotional expression and delayed false-belief understanding (Roch-Levecq, 2006).

While the majority of the studies on theory of mind in children with VI assessed first-order false belief understanding, Pring, Dewart and Brockbank (1998) used the Strange Stories paradigm (designed by Happé, 1994) to assess their advanced theory of mind understanding. The task consists of presenting children with a number of stories about everyday situations where the story protagonists say things that they do not literally mean (i.e., tapping advanced mental state elements, such as sarcasm, misunderstanding, persuasion, pretence and deceit). Pring et al. (1998) found that the children with congenital VI were poorer than age-matched sighted controls in predicting whether the protagonist's statements were true and in giving contextually-appropriate mental state justifications for these statements. This suggested that the previously observed socio-cognitive difficulties, based on the children's false-belief performance, persist into later childhood in children with congenital VI (i.e., age 9-12). The authors also reported a significant relationship between the children's general intellectual levels and the frequency of their appropriate mental state justifications, suggesting that children with VI who are intellectually more able may also be more able to compensate for difficulties in their social cognition than children with lower intellectual levels (the finding supported by Green et al., 2004). However, as the children in this study were not matched on cognitive ability, it remains unclear whether the difference between the VI and the sighted children may have been confined to the lower ability group. Nevertheless, the study by Pring et al. is the only one to date that has shown that children with congenital VI have a difficulty with advanced theory of mind understanding.

Connections with autism

From the theoretical and empirical evidence given thus far, it is clear that children with congenital VI are vulnerable to impoverished socio-communicative and socio-cognitive outcomes. What is particularly striking about the presentation of difficulties in social functioning in children with congenital VI is that many of these features are shared by sighted children with autism. The similarities between the two populations are both fascinating and alarming, and have been an intriguing topic of interest both in research and clinical and educational practice for over 15 years (Pring, 2005).

The seminal research that has highlighted the resemblance between the two populations was carried out by Hobson and colleagues (R. Brown, Hobson, Lee, & Stevenson, 1997; Hobson, Lee, & Brown, 1999). In a systematic observation of children with VI without a diagnosis of autism, using autism screening checklists, R. Brown et al. (1997) found that the children displayed a range of 'autistic-like' clinical features, including poor sociability and communicative competence, repetitive and restricted patterns of play, unusual sensory preoccupations, unusual mannerisms, stereotypies and echolalia. Interestingly, while a broad range and severity of such features were observed in a group of children with VI and higher intellectual ability relative to a group of sighted non-autistic controls, the lower ability group of children with VI was found to be virtually indistinguishable from a group of sighted children with a diagnosis of autism (see also Hobson et al., 1999). The spectrum of autistic-like features in children with VI has also been reported elsewhere (Hobson & Bishop, 2003; Pring & Tadić, 2005).

A number of other studies have also reported autistic-like patterns in specific behavioural and cognitive domains. More specifically, difficulties have been found in the area of social interaction and communicative competence (Preisler, 1991; Rowland, 1983; Tröster & Brambring, 1992; Urwin, 1983); emotional expressiveness and emotional recognition (Dyck, Farrugia, Shochet, & Holmes-Brown, 2004; Minter, Hobson, & Pring, 1991; Tröster & Brambring, 1992); symbolic (i.e., pretend) and functional play (M. Bishop, Hobson, & Lee, 2005; Fraiberg & Adelson, 1977; M. Hughes, Dote-Kwan, & Dolendo, 1998; V. Lewis, Norgate, Collis, & Reynolds, 2000; Tröster & Brambring, 1994); behavioural mannerisms, rituals and stereotypes (Brambring & Tröster, 1992; Chess, 1971; Tröster, Brambring, & Beelmann, 1991); uneven profile of cognitive abilities, including difficulties with abstract thinking (Tillman & Osborne, 1969); autistic-like developmental regression (Cass et al., 1994; Dale & Sonksen, 2002) and repetitive and unusual patterns of language use (i.e., echolalia and pronoun reversal) (Andersen, Dunlea, & Kekelis, 1984; Dunlea, 1989; Fraiberg & Adelson, 1977; Wills, 1979).

It is important to mention that autistic-like clinical features in children with congenital VI were initially observed in small groups of children with specific diagnoses such as congenital rubella, Leber's Amaurosis and retinopathy of prematurity (Chase, 1972; Chess, 1971; Ek, Fernell, Jacobson, & Gillberg, 1998; Keeler, 1956; Rogers & Newhart-Larson, 1989). However, the prevalence found across different aetiologies implies that such psychopathology in children with congenital VI is not confined to any specific ophthalmologic disease (Fraiberg, 1977; Keeler, 1956; Mukaddes, Kilincaslan, Kucukyazici, Sevketoglu, & Tuncer, 2007). Additionally, the studies investigating the association between autistic-like features and specific VI aetiologies have shown that such associations are often mediated by brain damage, severity of VI and associated intellectual impairment, implying that they are not likely to be a result of a specific diagnosis per se (Ek et al., 1998; Mukaddes et al., 2007). Others have also advocated general caution in interpreting autistic features in children with VI, as it is uncertain to what extent these can be put down to lack of sight per se or instead are a result of brain damaging events, social-emotional deprivation or other environmental factors that may coincide with visual problems (Cass, 1998; Fraiberg, 1977; Keeler, 1956; Warren, 1994). However, even though the autism-like picture may often seem more prominent in those children with VI and intellectual impairment (Cass, 1998; Hobson et al., 1999) and those children with VI in whom there is a known brain damage (Ek et al., 1998; Mukaddes et al., 2007), crucially the occurrence of such a phenotype has also been seen in children with VI without any known additional disabilities (R. Brown et al., 1997). More specifically, R. Brown et al. observed that, even though there was a tendency of low ability children with VI to score higher on autism checklists, there was a considerable overlap in the clinical presentation of low and high IQ children with VI. However, even though 37% (nine out of 24) of their VI sample (including both low and high IQ children) met the DSM criteria for a diagnosis of autism, only 8% of children (two out of 24) were clinically judged as being classically autistic. Thus, it generally appears that the autism presentation in children with VI is a broad constellation of relevant clinical features that vary in severity, rather than the case of a clear distinction between those children who have autism and those who do not.

Interestingly, despite the striking resemblance in socio-developmental outcomes between the two populations of children, researchers and clinicians further speculate that the quality of social impairment and of affective engagement and communication with others in the two groups of children may not be fully comparable. For instance, despite a considerable overlap in the clinical presentation of autistic-like children with VI and sighted children with autism, R. Brown et al. (1997) noted that the socio-affective impairment in most of the autistic-like children with VI that they studied was less profound relative to sighted children with autism. In a fine grain investigation of the same children with VI studied by R. Brown et al. (i.e., those children with VI who were judged as autistic-like using the DSM criteria for autism), Hobson et al. (1999) observed that a minority of children with VI who were autistic-like, compared to the majority of sighted controls with autism, showed abnormalities in the way they related to people, their emotional expression, and the quality of affect. Conversely, a majority of the children with VI were found to engage in instances of pretend play, compared to a minority of children with autism. More recently, it has been shown that even socially impaired children with VI were able to show levels of social relating that are generally absent in sighted children with autism (Hobson &

Bishop, 2003). Therefore, while the clinical presentation of some children with VI may indeed be similar, even identical, to autism in sighted children, the nature of autism in children with and without sight is potentially qualitatively different (Cass, 1998).[4]

Hobson was the first to provide a comprehensive theoretical account for the clinical similarities between autism and congenital sight loss, by proposing that the impairments in social relations, communication and symbolic play seen in the two groups may be traced to a shared psychopathology (Hobson, 1990, 1993, 2005). Both groups of children experience problems with perceptions of other people's attitudes which are outwardly directed to a shared visually-specified world. However, children with congenital VI are unable to see visual evidence of their social partner's attitudes (e.g., facial expression manifesting fear in response to a fear-evoking event), whilst children with autism seem to be unable to process this kind of information, despite a functioning visual channel. Although the causal mechanisms in terms of sensory and perceptual processes are clearly distinctive in the two populations, both are equally challenged in their ability to understand the relationships between attitudes, and the objects and events in the shared world. Therefore, whilst particular genetic and neurological factors may be necessary for autism to occur (Bauman, 1999), visual deprivation of this specific kind of social perspective-taking in children who are visually impaired is likely to contribute to related social and cognitive difficulties and delays (including difficulties with joint attention and theory of mind), predisposing them to autistic-like psychopathology (Hobson, 2005; Hobson et al., 1999).

The mechanisms underlying the pathogenesis of autism in children with VI still require further empirical consideration and further research is necessary to clarify the related mechanisms. However, Hobson presents a convincing case that specific socio-developmental factors may be responsible for the autism-like picture in children with VI, and that VI per se is likely to predispose a child to socio-developmental impairments that lead to the autistic-like syndrome (Hobson, 1990, 1993, 2005). It is likely that social impairments seen in some children with VI who meet the full criteria for diagnosis of autism may indeed be a reflection of actual autism coinciding with vision loss, without an inherent connection with the lack of visual input. However, the wide spectrum of severity of autistic-like features and the quality of social impairment, observed even in those

[4] It is estimated that up to a third of all children with significant levels of VI present with a disorder that is very similar to autism in sighted children. However, there are major difficulties with defining diagnostic criteria for autism in children with VI, given the likely differences in the aetiology of the disorder in the two groups (Cass, 1998). Nevertheless, in clinical practice, some children with VI may also receive a diagnosis of autism, although the exact prevalence of dual diagnosis is unknown (N. Dale, personal communication).

children with VI who are not classically autistic, implies that there is something special about severe vision loss that plays a role in pathogenesis of autistic-like syndrome in children with VI (Hobson & Bishop, 2003). Finally, understanding of this role and the related mechanisms that underlie the autistic-like psychopathology in children with VI is of great importance, not only for understanding the developmental needs of such children, but also for understanding of autism in general, and for appreciating the mechanisms that underpin typical development.

Summary of social functioning in children with VI

Thus far, this chapter has dealt with the nature and potential causal mechanisms of socio-communicative and socio-cognitive impairments in children with VI, leading on to the clinical and empirical concern regarding the behavioural as well as cognitive resemblances with sighted children with autism. However, as implied in the section above, the quality of social impairment in children with VI and sighted children with autism does seem to differentiate the two groups. More specifically, although there may be a common final pathway that leads to similar difficulties which characterise both groups of children, it is likely that the actual causal mechanisms contributing to pathogenesis of the syndrome of autism in the two populations are distinct (Hobson, 2005). In line with this, Baron-Cohen (2002) recently commented that the resemblance between children with VI and sighted children with autism may '*be no more than a surface similarity*' and warned that '*we should be careful not to assume that just because two church bells are ringing simultaneosly they are causally connected by the same rope*' (p. 792). For this reason, it is also important to consider that, despite the generally vulnerable socio-developmental outcomes in children with VI, the developmental trajectory of social milestones such as joint attention and theory of mind in such children appears to be different from the trajectory seen in sighted children with autism. More specifically, the picture of joint attention and theory of mind development in children with VI depicts a delayed trajectory rather than a deficient one, which potentially contrasts the classic presentation of autism where disruptions to early joint attention behaviours and lack of theory of mind have been seen as core deficits. The trajectories of social functioning in the two populations are inherently complex and it is likely that along those trajectories various underlying processes and mediating mechanisms exert their influence on the children's developmental outcomes. However, while such mechanisms may indeed be involved in both groups, they may serve potentially different functions for the two populations of children. In children with VI in particular, compared to children with autism, such mechanisms may in fact play a compensatory, rather than a mediating role, stepping in the place of a missing information channel. The theory and research evidence exploring this hypothesis will be discussed in further detail in the following section.

LANGUAGE AND SOCIAL FUNCTIONING

One particular developmental mechanism that appears to distinguish children with VI and children with autism is language. More specifically, whilst impairments in language development and functioning are one of the defining features of autism (Rapin & Dunn, 2003; Tager-Flusberg, 1993), for children with VI language is generally believed to be an area of strength (Landau & Gleitman, 1985; Pérez-Pereira & Conti-Ramsden, 1999; Warren, 1994). The relationship between developments in language and social functioning in both groups of children is an important topic for both empirical research and clinical intervention. Importantly, however, while much is known about the aspects of this relationship in autism (Fisher, Happé, & Dunn, 2005; Happé, 1995; Tager-Flusberg, 2000), the interaction between the acquisition of language and social milestones remains largely unexplored in children with VI. For instance, impairments in early socio-communicative experiences of joint attention in autism may explain why language is delayed and, in fact, why some children with the disorder never acquire language (Tager-Flusberg, 2000). On the other hand, despite difficulties with early joint attention, and some reported idiosyncrasies in initial language acquisition, children with VI seem to develop language with relative ease (Landau & Gleitman, 1985; Pérez-Pereira & Conti-Ramsden, 1999). In fact, once it is developed, language appears to be the most salient channel through which children with VI acquire knowledge and experience the world (Landau & Gleitman, 1985; Peters, 1994). Thus, language seems an important topic for research and intervention with children with VI. The aim of the following section is to consider the evidence which relates to the role that language plays in the development of such children and, more specifically, to consider its relationship with their socio-communicative and socio-cognitive development. Before this role of language for children with VI can be addressed explicitly, the function of language in social milestones will be first considered with regards to sighted children.

Evidence from sighted children

The relationship between language and social functioning is likely to be complex and the nature and causality of this relationship has been the subject of ongoing debate amongst researchers (reviewed in Astington & Baird, 2005; de Villiers, 2000; Slade & Ruffman, 2005). While early social experiences and non-verbal communication in pre-lingual development are regarded as important predecessors to language acquisition, growing language repertoire and comprehension of various linguistic concepts is likely to have a facilitating effect on the child's development of social cognition. Additionally, it is possible that other cognitive and environmental factors play a significant role in mediating the relationship between the two processes. The intrinsic intertwining and causality between language, social milestones and many potentially mediating factors is an important topic for further research, but one which is beyond the scope of the current thesis. Nevertheless, it is important to consider some empirical evidence regarding this relationship.

Language and early social communication

As mentioned earlier in this chapter, joint attention has been given a central role in the development of language. This is because language is a socially learned skill and joint attention provides a crucial socio-communicative format within which such learning occurs (Adamson, Bakeman, & Deckner, 2004; Tomasello, 1995). Joint attention behaviours such as eye-gaze alternation and non-verbal gestures generally precede the acquisition of first words, thus it is easy to appreciate how they may be necessary for language acquisition (Butterworth & Grover, 1990). Indeed, a substantial body of research has linked early joint attention experiences with language development over time in typically developing sighted children (Carpenter et al., 1998; Charman et al., 2000; Mundy & Gomes, 1998; Tomasello & Farrar, 1986). For example, it has been shown that the capacity of the child to respond to and follow the adult's focus of attention early in infancy significantly predicts their later vocabulary growth (Markus et al., 2000; Mundy, Fox, & Card, 2003; Mundy, Kasari, Sigman, & Ruskin, 1995), while the time spent in joint attention early on is generally found to be related to receptive and expressive language over time (Mundy et al., 1995; Tomasello & Farrar, 1986; Tomasello & Todd, 1983). The reverse relationship also holds as receptive language at 12 months has been found to be predictive of the time spent in joint attention at 18 months (Markus et al., 2000; Tomasello & Todd, 1983). Importantly, the caregiver has been given the central role in the relationship between joint attention and language development (Bruner, 1983) and the social responsiveness and communicative style employed by the caregiver in the joint attention episode have been found to be related to increased vocabulary acquisition in the child (Bakeman & Adamson, 1984; Tomasello & Farrar, 1986).

Support for the longitudinal association of early joint attention and later language ability also comes from several studies with children with autism (Charman et al., 2003; Charman et al., 2005; Mundy, 1995; Mundy, Sigman, & Kasari, 1990; Sigman & Ruskin, 1999; Stone & Yoder, 2001). In fact, these studies have demonstrated the predictive value of joint attention capacity in later language development by consistently showing that in children with autism, greater ability to initiate and respond to joint attention in preschool years predicted language outcomes up to eight years later. For that reason, joint attention has been highlighted as an important target for intervention in autism (Charman et al., 2003) and recent evidence suggests that training joint attention skills in autism leads to significant improvements in language developments (Drew et al., 2002; Kasari, Paparella, Freeman, & Jahromi, 2008).

Language and social cognition

Given that language as a developmental process is borne explicitly out of a social context, it is easy to appreciate its facilitative effects on subsequent social development. Research with typically developing children has consistently supported a strong link between language and children's developing social cognition, the implicit understanding being that a child's developing knowledge of their own and others' mental states must be facilitated through language and communication, both verbal and non-verbal (Astington & Baird, 2005; Tager-Flusberg, 1993). With regards to theory of mind in particular, language has been seen as playing a fundamental role and numerous studies to date have demonstrated a significant correlation between standardized language measures and performance on theory of mind tasks in children (Astington & Jenkins, 1999; Cutting & Dunn, 1999; C. Hughes & Dunn, 1997). Although the causality of this relationship has been a matter of some disagreement, it seems likely that language plays a causal role in children's theory of mind development, rather than vice versa. More specifically, research evidence shows that linguistic abilities in children predict their theory of mind performance at different time points, whereas the reverse relationship generally does not hold (Astington & Jenkins, 1999; Ruffman, Slade, Rowlandson, Rumsey, & Garnham, 2003; Watson, Painter, & Bornstein, 1998). The causal role of language in theory of mind is also supported by research with children who are autistic and children born with hearing impairment (HI). For instance, children with autism with better linguistic competence show better theory of mind outcomes than children with autism with lower language ability and generally require a higher language level than typically developing children to pass false-belief tasks (Fisher et al., 2005; Happé, 1995; Tager-Flusberg & Sullivan, 1994b). Similarly, children with HI who are born of hearing impaired parents, and are therefore exposed to sign language from the earliest stages of development, do not show theory of mind delays and difficulties, which are typically observed in children with HI born of hearing parents and who acquire sign language somewhat later (Peterson & Siegal, 1995, 1999, 2000; P. A. Russell et al., 1998; Woolfe, Want, & Siegal, 2002).

Whereas researchers are in general agreement that the role of language is fundamental to children's developing social understanding, there has been an ongoing debate regarding what particular aspects of language play a greater role in children's theory of mind. With regards to understanding of false belief in particular, some researchers have argued that it is syntactic ability, which involves combining words into sentences, that enables children to attribute mental states to self and others, because understanding of the word sequences in a sentence is mirrored in the child's understanding of the sequence of events in the false-belief scenario (Astington & Jenkins, 1999; de Villiers & Pyers, 2002). Conversely, others have argued that semantic knowledge plays a more significant role in understanding false belief, as this is suggested to be dependent on understanding the terms used to refer to cognitive mental states such as 'think' and 'know' (Bartsch & Wellman, 1995; C. Moore, Pure, & Furrow, 1990; Olson, 1988). More recently, Ruffman and colleagues have demonstrated that general language ability, which incorporates both semantic and syntactic knowledge, in fact reflects on children's understanding of false belief (Ruffman et al., 2003; Slade & Ruffman, 2005). These authors have argued that the early knowledge of mental states (e.g., knowing, thinking, believing), which is implicit and often manifested through behaviours such as eye-gaze (Ruffman, Garnham, Import, & Connolly, 2001), provides children with the terminology for thinking explicitly about mental states. As both syntax and semantics develop in tandem, and are closely interlinked in normal language development, they may both assist in theory of mind development by allowing the child to refine their implicit intuitions about other people's beliefs and intentions into explicit theories about those states (Ruffman et al., 2003)

Language as a social function

Given that language is a skill learned essentially under social circumstances, language itself can be viewed as a social function. Therefore, to gain a better appreciation of its role in children's social understanding, it is essential to try and distinguish social aspects of language from general linguistic competence that is mirrored in children's understanding of syntax and semantics (i.e., from structural language).

An important social function of language, commonly referred to as *pragmatic language* ability, involves the ability to use and interpret language appropriately in social situations in order to achieve successful communication both by verbal and non-verbal means (D. V. M. Bishop, 2005; Rapin, 1996). In everyday social interaction non-verbal pragmatic skills such as facial expressions, gestures and body postures provide a useful context for conveying and interpreting language and the speaker's communicative intentions. Verbal pragmatics, on the other hand, underlie the ability to appropriately apply conversational rules such as initiating, responding, turn-taking, and maintaining meaningful conversation. Importantly, these skills are involved in keeping track of speaker's and listener's mental states; hence they are by definition related to theory of mind (Astington & Baird, 2005) and are believed to be at the heart of social functioning (Tager-Flusberg, 2000).

Another inherently social aspect of language that has been regarded as a key component of children' social cognition is *mental states discourse* (reviewed by de Rosnay & Hughes, 2006; Symons, 2004). It is now widely believed that the use of language that represents thoughts, desires and feelings in children's conversational discourse may be an important indicator of their theory of mind ability (Baron-Cohen, 2000; Bartsch & Wellman, 1995; Harris, de Rosnay, & Pons, 2005; Tager-Flusberg, 2000). In research with typically developing children, correlations between the comprehension and frequency of use of cognitive and emotion terms and the performance on false-belief tasks have been consistently demonstrated (J. R. Brown, Donelan-McCall, & Dunn, 1996; C. Hughes & Dunn, 1997, 1998). Moreover, it has been argued that use of mental state terms in children's language precedes their mastery of false-belief tasks (Bartsch & Wellman, 1995; Harris, 1996; Wellman, 1990), supported by the evidence that children's early references to desires and thoughts are in fact predictive of their subsequent false-belief performance (C. Hughes & Dunn, 1998).

Children's ability to engage in such discourse has been closely linked to their conversational interactions with family members and peers (Dunn, Brown, Slomkowski, Tesla, & Youngblade, 1991; C. Hughes & Dunn, 1997; C. Lewis, Freeman, Kyriakidou, Maridaki-Kassotaki, & Berridge, 1998). More recently, however, specific emphasis has been placed on the quality of mother-child discourse, and maternal mental state language input in particular. In relation to this, extensive research evidence now links such input to a child's level of social understanding (de Rosnay, Pons, Harris, & Morrell, 2004; Meins, Fernyhough, Russell, & Clark-Carter, 1998; Meins et al., 2003; Meins et al., 2002; Ruffman, Slade, & Crowe, 2002; Slaughter, Peterson, & Mackintosh, 2007; Taumoepeau & Ruffman, 2006). Importantly, a number of more recent studies have provided strong evidence that the role of maternal mental state input is in fact a causal mechanism in children's growing social knowledge of the mind (Meins et al., 2003; Meins et al., 2002; Ruffman et al., 2002; Taumoepeau & Ruffman, 2006). More specifically, such studies have shown that the mother's early mental state talk in their conversational interactions with their children is a unique predictor of the children's theory of mind task performance over time, as the direction of this relationship remains even after potential mediators (i.e., mothers' educational level, children's ages, language ability, children's own use of mentalistic language and their early theory of mind) are controlled for. Thus, the continuous exposure to mental state language provided by the primary caregiver provides a unique contribution to children's developing theory of mind and is likely to continuously promote their socio-communicative competence and understanding.

Social language in atypical circumstances: Insights from autism

Pragmatic communication impairment is a cardinal feature of autism (Lord & Paul, 1997; Norbury & Bishop, 2002). For example, sighted individuals with autism are impaired in making contextually appropriate inferences and inferences about social scripts (Dennis, Lazenby, & Lockyer, 2001) and show difficulties with comprehending figure of speech (Happé, 1994; Jolliffe & Baron-Cohen, 1999), narrative humour (Ozonoff & Miller, 1996) and contextual understanding of questions (Loukusa et al., 2007). Interestingly, such pragmatic language difficulties have been observed even in those individuals with autism who have good linguistic competence and structural language skills (Landa, 2000; Lord & Paul, 1997). Tager-Flusberg (1993; 1999; 2000) reviewed evidence which showed that individuals with autism at all ages have difficulty taking into account the listener's perspective, which affects their ability to engage in conversation in a sustained or meaningful way. They also have difficulty conforming to conversational rules, such as initiating and engaging in reciprocal conversations, and tend to talk about their own interests, without regard to their listener's interests or role in the conversation. Impaired pragmatic communication in autism echoes the poor theory of mind profile that characterises the disorder. In line with this profile, it is therefore not surprising to find reduced mental state discourse skills in autism. It has been shown that children with autism rarely use such language in their spontaneous conversational discourse or in their descriptions of pictures involving action and deception (Baron-Cohen et al., 1985; Tager-Flusberg, 1992, 1995). Lack of reference to mental states in conversational discourse in autism has also been observed in adulthood and those individuals with autism with good language competence (Dennis et al., 2001; Happé, 1994; Jolliffe & Baron-Cohen, 1999). On the other hand, subsequent studies have argued that mental state discourse is not necessarily impoverished in autism; the apparent lack of references to mental state terms may be a reflection of the restricted narrative techniques employed by individuals with autism, where such individuals are less likely to refer to mental states within a causal framework and to use pragmatic devices to engage the listener (e.g., supplying a conversational partner with contingent relevant new information, character speech, sound effects, repetition, empathic stress) (Capps, Kehres, & Sigman, 1998; Capps, Losh, & Thurber, 2000; Tager-Flusberg & Sullivan, 1994a). A significant correlation between reduced use of such pragmatic devices and performance on theory of mind tasks in autism has been empirically demonstrated (Capps et al., 1998; Capps et al., 2000).

There clearly is a link between social language abilities and social understanding in sighted children with autism. It is possible that such fundamental impairments in children with autism are at least to an extent mediated by specific linguistic mechanisms that may complement their social knowledge, given the better socio-cognitive outcomes in those children who are verbally able (Happé, 1995; Tager-Flusberg, 2000). However, while such children may rely on verbal scaffolding and the explicit teaching of others in order to deal with the demands of socio-cognitive tasks, the social understanding they may develop is likely to be superficial and insufficient for successfully dealing with the demands of the real-life social world and socio-interactive relationships with others (Dennis et al., 2001; Klin, 2000).

Implications for children with VI[5]

Following the research reviewed so far, a crucial question that forms the backbone of the present thesis arises: Is there a special role for language in the development of children with VI, and more specifically, what is the mechanism that language may provide for their social functioning in particular?

Language development in children with VI is likely to be a somewhat controversial topic. The evidence linking early socio-communicative experiences and language development in typically developing sighted children and children with autism implies that disruptions to joint attention experiences in VI may impede development of specific language skills in children with VI. Consequently, delayed theory of mind outcomes in children with VI may be seen as a direct effect of, not only impoverished and vision-deprived early social interaction, but also of subsequent difficulties in language development. Yet, whether language is an area of potential difficulty for children with VI has been widely disputed. Language has in fact been highlighted as an area of particular strength in development of such children. Pérez-Pereira and Conti-Ramsden (1999) in particular have argued that, in VI, the role of language is a compensatory one, providing a mechanism by which the visual information is transformed into verbal one. In a number of investigations that these authors have undertaken with individual children with VI and their families, these authors (and collaborators) have indeed demonstrated that the general characteristics of language acquisition and progression of linguistic competence in children with VI closely resemble the characteristics of language development in sighted children (e.g., Conti-Ramsden & Pérez-Pereira, 1998, 1999; Pérez-Pereira, 1994; Pérez-Pereira & Castro, 1997).

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[5] The issues raised in this chapter are a focus of the empirical investigations presented in this thesis. Thus, while in the present chapter only a brief exposition of the relevant literature may be provided, in-depth review will be given in the introductions of the subsequent chapters.

On the other hand, some contrasting evidence suggests that the initial language development of children with VI is characterized by specific delays and irregularities (Andersen et al., 1984; Dunlea, 1989; Fraiberg, 1977; McConachie & Moore, 1994). For instance, it has been shown that the emergence of first words and related development of word meaning is delayed in children with VI due to limited experiences compared to sighted children (Andersen et al., 1984; Dunlea, 1989). Similarly, referential language (e.g., use of personal nouns and spatial terms) has been found to be an area of difficulty for children with VI, as such language is thought to be particularly dependent on the utility of vision. More specifically, visual experience helps to resolve the ambiguity which occurs when words take on different meanings depending on the location and identity of the speaker (e.g., spatial terms like 'here' and 'there' and pronouns like 'I' 'he' and 'she') and young children with VI tend to confuse such terms (Fraiberg & Adelson, 1977; Mulford, 1983). However, it has been argued that, even though some language differences between children with VI and children who are sighted may indeed occur (particularly as a result of the child's unique perceptual experiences), the language of children with VI is not devoid of content or meaning, and there is in fact little evidence that it is impaired in any way (Landau & Gleitman, 1985). Such literature is explored in more detail in Chapter 3 of the thesis where the pertinent issues are of specific relevance. However, it is important to mention that despite possible idiosyncrasies in the early language development of children with VI, it is generally accepted that if any initial difficulties do exist early on they tend to be resolved by the school age when the language of such children appears to be indistinguishable from that of sighted children (Landau & Gleitman, 1985; Mills, 1993; Warren, 1994).

However, our knowledge about the language of children with VI is largely limited to the development and function of structural language, particularly in pre-school development, whilst very little is known about the social language use of children with VI and, more specifically, how such language may reflect their social competence and knowledge in later childhood. In fact, the existing studies on pragmatic language in children with VI, drawing upon investigations of very small numbers of children and often only individual cases, are largely inconclusive. While some evidence suggests that pragmatic language development in children with VI in fact is an area of vulnerability (James & Stojanovik, 2007; Mills, 1993), others have failed to find significantly different outcomes from those observed in sighted children (Pérez-Pereira, 1994; Pérez-Pereira & Castro, 1992). This area of language in children with VI certainly merits further investigations and is given detailed attention in Chapter 3, where the relevant literature is discussed further. Crucially, mental state discourse in children with VI remains largely unexplored and is given more detailed attention in Chapters 4 and 5.

To address the social language use of children with VI, it is important to consider the environment within which the use of such language is learned, because the social language characteristics of children with VI are likely to be a consequence of that environment rather than of the visual impairment per se (Warren, 1994). Within this environment, emphasis is generally given to the socio-interactive style of the parent-child relationship and on the parental language input in particular. While the general language input provided by a caregiver is important for any child, this specific contribution by the caregiver may be of particular importance to a child with VI, who may rely on this input as an essential source of information that is otherwise easily available to children who are sighted. In the case of children who are sighted, the exchange of communicative intentions between mother and child is not likely to be dependent on the verbal channel, given the abundance of non-verbal communicative means such as gestures, facial expressions and eye-gaze behaviours (i.e., joint attention) (Hobson, 2002; Stern, 1985). However, mothers and their children with VI may particularly rely on the verbal modality for sharing of thoughts, interests and feelings about the external world with each other (Pérez-Pereira & Conti-Ramsden, 1999; Urwin, 1978). However, this may not always be easy. Naturally, part of communicative competence involves the sensitivity to the needs and perspectives of the interactional partner. Sighted children can learn to respond contingently to their mother's previous turn in conversation and attend to their mother's interests by maintaining eye-contact, and this is likely to facilitate their mother's responsiveness. On the other hand, for children who are visually impaired, monitoring their mother's intentions and communicative approach may be more difficult, hence they may appear more passive than sighted children (Kekelis & Prinz, 1996). This passivity in turn may impact on the parent, who may indeed be at a disadvantage when trying to interpret the subtle social cues given by the child (Preisler, 1991; Rogers & Puchalski, 1984). However, the few studies that have explored the conversational interactions between children with VI and their mothers are inconclusive. For instance, some authors have argued that mothers' language input to their visually impaired children is restricted and impoverished compared to mothers with sighted children and may be a direct consequence of a different parent-child communication style, where attention in a child with a significant sight loss cannot be caught and directed by eye-contact (Andersen, Dunlea, & Kekelis, 1993; Kekelis & Andersen, 1984; Kekelis & Prinz, 1996; V. Moore & McConachie, 1994). However, others have shown that that the parents of children with VI are able to develop alternative strategies when conversing with their children (Behl, Akers, Boyce, & Taylor, 1996; Pérez-Pereira & Conti-Ramsden, 1999, 2001). The extent of mental state language input to children with VI, as well as their productive output, is yet to be addressed by research and is considered in further detail in Chapter 5.

AIMS AND OVERVIEW

It is clear that children with VI, both as a model experimental group or individually, have so much to offer to developmental science. Although the clinical existence of autism itself may have been an important catalyst for the emergence of the prominent theories of the mechanisms that are involved in the processes of social functioning, it is likely that children with VI may provide an equally useful, and potentially unique, theoretical and methodological model for investigating such mechanisms. Without a doubt, there is so much more that can be learned from autism in sighted children; however, it is possible to isolate a similar contribution to the theory and clinical practice which is exclusively provided by considering the development of children born with VI.

Thus far, in theoretical terms, the aim of the present thesis was to consider the empirical evidence of the developmental trajectory of social functioning in children with VI, while drawing upon the prominent theories and research evidence concerning such functioning in typically developing sighted children and children with autism. However, the present literature review has not delved into the methodological challenges, which are an integral part of research with children with VI and which contribute to its general scarcity. For this reason, Chapter 2 is solely devoted to this purpose.

In empirical terms however, and drawing upon the theoretical and methodological issues raised in Chapter 1 and Chapter 2, the present thesis has six general aims. The first such aim is to shed further light on the language abilities of children with VI, with a view to understand the potential role that these may play for their social functioning. Whilst previous studies have shown that good verbal ability is facilitative of developmental (including social) outcomes of children with VI, such studies have failed to isolate the true contribution of language from a child's general intelligence. This issue is therefore addressed by the research presented in Chapter 3. Another aim of the research presented within Chapter 3 is to provide further understanding of the quality of social and communicative impairments in children with VI by addressing the nature of the autistic-like socio-communicative phenotype in children with VI, and to gain an insight into the aspects of language that may explain the variation within such a phenotype in these children.

The third aim of the thesis is to provide an insight into the socio-cognitive competence of children with VI, which extends beyond the false-belief paradigm and places emphasis on their mental state language. No studies to date have examined the use of such language in children with VI. As an indicator of theory of mind in particular, the use of mental state terms in children with VI may be of particular importance, as the only measure of such knowledge in children so far has been their false-belief task performance. The dichotomous nature of false-belief tasks may not lead to an authentic representation of real-life social knowledge, and children are likely to show more variation in the degrees of theory of mind capacities than the false-belief paradigm allows. Thus, false-belief tasks may, to an extent, trivialise or even underestimate the social understanding of children with VI, whereas their use of mental state language when making judgements of social events may provide a more realistic assessment of their social knowledge. This hypothesis is explored in Chapter 4.

The fourth aim is to investigate the context within which social language learning occurs for children with VI and focuses particularly on maternal mental state language input to such children during a mother-child dialogue. Mother-child mental state discourse has not previously been investigated in children with VI. Although a small number of studies examining mother-child dialogue exist with such children, these are largely limited to very young, often pre-lingual children, providing a reduced context from which to sample the mother-child mental state discourse. The research presented in Chapter 5 explores this particular aspect of the socio-interactive learning environment in children with VI.

The fifth aim, which is addressed in Chapter 6, is to pinpoint the potential role of other underlying mechanisms that may account for developmental outcomes in children with VI, and which remain crucial in the socio-communicative and socio-cognitive development of sighted children. More specifically, while the cognitive mechanisms of attention and executive function are highlighted as crucial in socio-developmental outcomes in typical development (see Chapter 6 for a more detailed review of the literature), the contribution of such mechanisms remains unknown in children with VI.

The sixth, and last, aim of the thesis is to consider the relationships between the outcomes across the four experimental chapters, with a view to fuse different aspects of the developmental picture presented by the children with VI who participated in this research. The correlational analyses which address this aim are presented in Chapter 7.

While the aims of the thesis are presented here at a relatively general level, the theoretical and methodological underpinnings of these aims, and with specific questions pertinent to those aims, are given more detailed consideration within the relevant experimental chapters. Finally, a synthesis of the findings across these chapters, linking to the theoretical accounts that underlie those investigations, is presented in the general discussion in Chapter 8. The aim of this synthesis is to highlight the unique contribution of the present research to understanding of developmental needs of children with VI and the implications that they have for child development in general.

Chapter 2

Visual Impairment (VI): Methodological Considerations

SUMMARY

Rigorous inclusion criteria are crucial when investigating developmental outcomes of children with VI, as these outcomes must be considered in the context of the child's vision loss, rather than any other underlying, non-sensory impairment. However, the methodological difficulties of adopting such criteria impact directly on both the quantity and quality of research carried out with such children, and existing studies are notably rare and methodologically limited. The aim of this chapter is to consider the methodological challenges contributing to the scarcity of research with children with VI and with a view to understand the conflicting evidence that may have ensued as a result of these challenges.

THE IMPLICATIONS OF VI CHARACTERISTICS ON RESEARCH METHODS

Low prevalence

Given the relative rarity of the population (Chapter 1, p. 14), it is not surprising that one of the main methodological difficulties in research with children with congenital VI is obtaining an adequate sample size. For that reason, the majority of VI related developmental research is generally grounded in investigations of small numbers of children and often only individual cases. In addition to relatively low prevalence of congenital VI, modest study samples of children with VI are also a result of attempts to control for other potentially confounding effects that are inherently associated with presentation of visual impairment (see section below). Even though small samples are generally at risk of compromised experimental power and reduced generalisability of the findings, investigations of few or individual children with VI can provide a rich and more detailed insight into their developmental processes, as long as appropriate measures are taken to ensure a high quality of sampling. Such measures are considered throughout this chapter.

Population heterogeneity

Aetiology of VI

VI is associated with high heterogeneity, the main source of which is the diverse aetiology of the impairment. Congenital causes are now predominant, as the incidence of visual loss caused by acquired disease in childhood (e.g., corneal infections) has been reduced with economic development (Rahi & Dezateux, 1998). Congenital causes of VI in childhood are i) genetic disease (e.g., albinism), ii) intrauterine events (e.g., congenital rubella), and iii) perinatal disease (e.g., oxygen deprivation at birth and retinopathy of prematurity – growth of abnormal blood vessels in the retina associated with premature birth), with genetic and perinatal disease being the most common in the developed world (Baird & Moore, 1993; Rahi & Dezateux, 1998).

For the purposes of research investigating the impact of congenital VI on development, heterogeneity introduced by diverse aetiology (and associated confounding effects) can be dramatically reduced by applying an appropriate taxonomy for the classification of visual disorders. One such taxonomy has been developed by the clinical team at the Developmental Vision Clinic (DVC) at the University College London, Institute of Child Health and Great Ormond Street Hospital for Children NHS Trust, London, UK from which the participants for the research reported in this thesis were recruited (Dale & Sonksen, 2002; Sonksen & Dale, 2002).[6] According to the DVC taxonomy, the site of origin of the congenital visual disorder is classified as *cerebral* (posterior visual pathways and visual nervous system) or *peripheral* (the globe, the retina and the anterior optic nerve) and is established through ophthalmologic diagnostics. Based on the DVC records (in Dale & Sonksen, 2002; Sonksen & Dale, 2002), *cerebral* congenital disorders of the visual system (i.e., cortical VI) are the most common and are associated with additional disabilities, including learning difficulties and cerebral palsy (see also Good, Jan, Burden, Skoczenski, & Candy, 2001). Congenital disorders of the *peripheral* visual system (CDPVS) account for about 30% of children with VI. The CDPVS group is subdivided into two groups. The first group is referred to as *Potentially Complicated* CDPVS and it involves children in whom the peripheral eye disorder is a part of a diagnosed paediatric disorder including underlying damage to the central nervous system. Examples of Potentially Complicated CDPVS are cataracts in Down Syndrome and retinal dystrophy in peroxysomal disorders (i.e., a group of congenital diseases characterized by the absence of normal peroxisomes in the cells of the body, such as

[6] The DVC is a specialized tertiary-level clinic to which young children with severe visual disorders of heterogeneous aetiology are referred (by paediatricians or ophthalmologists) for specialist assessment and management of their vision and development.

Joubert Syndrome). The second group is referred to as *Potentially Simple* CDPVS and involves children in whom there is no known involvement of the central nervous system in the visual disorder diagnosis. Because of the lower expected incidence of confounding variables in this group, compared to the children with cortical VI and those with Potentially Complicated CDPVS, children with Potentially Simple CDPVS are proposed to be a model group for clinical and developmental research (Sonksen & Dale, 2002). This is thought to be a particularly rare subpopulation; the exact prevalence is unknown but estimated as 350 born annually in the UK (Sonksen & Dale, 2002). Example diagnoses falling under the Potentially Simple CDPVS classification are: glaucoma, micropthalmia, aniridia, coloboma, Norrie's Syndrome, cataracts, Leber's amaurosis, cone dystrophy, albinism, optic nerve aplasia and optic nerve hypoplasia.

Degree of vision loss

Another challenge in assessing the developmental impact of VI in childhood stems from the lack of consensus in definitions and measurement of severity of vision loss that have been used both in research and practice. Traditionally, categories such as 'blind' and 'partially sighted' have been used in educational and legal contexts and have been assigned to children and adults based on distance visual acuity measurements, such as the Snellen chart or, more recently, log MAR. The Snellen chart, for instance, estimates the power of the eye to distinguish fine detail (e.g., letters) at different distances. For example, a normal eye can read the top letter (which is approximately one centimetre high) on the 6-metre chart from a distance of 6 metres; thus in the UK normal vision is expressed as 6/6 (also known as 20/20 in the USA). According to the World Health Association (WHO, 1980) 'partially sighted' individuals (i.e., moderate to severe VI) have visual acuity ranging from less than 6/18 (i.e., they can see less at 6 metres than what a normal eye sees at 18) down to 3/60, whereas a 'blind' person (i.e., profound VI / total lack of sight) has a visual acuity ranging from 3/60 down to no light perception.

However, the visual acuity tests, such as Snellen and log MAR, do not provide criteria that grade the degrees of visual impairment which are below the lowest limits of such measures, and are not suitable for use with younger children. This has important implications for meeting the needs of individual children with VI. Few of these children are completely blind, although their existing vision levels cannot be measured by the traditional acuity methods (Hatton, Bailey, Burchinal, & Ferrell, 1997). For this reason, visual impairment may be viewed more appropriately as a continuum of decreasing visual function. Thus, while very broad categories like 'blind' and 'partially sighted' may be appropriate for grouping children with VI for legal and educational purpose, they may not be useful (nor accurate) when used in clinical and empirical context. In this respect, for considering the impact of differing levels of vision loss on the developmental process of a child with VI, more narrow grading of the levels of visual function may be more appropriate (Hatton et al., 1997).

Sonksen and colleagues (Sonksen, 1983, 1993) developed the Near Detection Vision scale (NDV), which allows for formal grading of vision levels that are below the standard acuity measures. The measure was specifically developed for the purpose of meeting the developmental needs of infants and young children with significant levels of VI and grouping them meaningfully for developmental research. According to the NDV, significant visual impairment in childhood can be categorised as profound (PVI) or severe (SVI), distinguishing children with and without 'form' vision (i.e., awareness of visual targets that do not reflect light). On the NDV scale, PVI status implies the absence of 'form vision' and is defined as having the ability to perceive light reflecting objects (e.g., a spinning silver tinsel ball) size 12.5 cm at the distance of 30 cm or less. Conversely, SVI status implies the presence of 'form' vision and is defined as having the ability to perceive a non-light reflecting objects (e.g., a spinning woolly ball) size 12.5cm from a distance of 30 cm or better. While the PVI population is relatively homogenous in terms of their vision loss, children with SVI vary with respect to their levels of form perception. However, the available level of form vision in this group is still severely degraded and below the standard acuity measures (i.e., below distance equivalents of Snellen 6/30), hence children with SVI are considered clinically to be a vulnerable group.

Previously, researchers have attempted to control for the potentially confounding effects of the varying levels of vision loss by specifying the inclusion criteria at the level of congenital and total sight loss (i.e., light perception or less from birth / PVI only) (e.g. Green et al., 2004; Hobson et al., 1999; Minter et al., 1998). Such rigorous selection criteria do seem vital in studies investigating the effects of vision on specific developmental outcomes. However, in such studies, systematic screening of the vision levels of children with VI was usually based upon teacher and parental reports and case notes. It can therefore never be certain whether the 'pure' samples of children with 'congenital and total blindness' reported in the previous research were in fact homogenous, or whether they were likely to have included also children with some useful levels of, however degraded, form vision (e.g., those with SVI).

Another reason for this uncertainty is the fact that VI in childhood tends to present as an unpredictable factor, owing to the maturational processes of the visual system (Atkinson, 1984; Day, 1997). As a result of visual maturation, some children with profound VI may obtain useful levels of form vision - giving them an SVI status - in the course of their development, as was the case with 8% of the preschool sample studied by Dale & Sonksen (2002). Similarly, visual levels of children with SVI may significantly improve, particularly if appropriate intervention strategies designed to promote such development are implemented in the first year of life (Sonksen, Petrie, & Drew, 1991). On the other hand, the vision levels of some children born with SVI remain suboptimal and can degrade to the profound level (Sonksen et al., 1991). Hence, unless the nature of the visual diagnosis implies that no visual perception is possible (e.g., anophthalmia - absence of eyes; or bilateral optic nerve aplasia - absence of optic nerve), a certain risk of changing presentation of vision level in the samples of children with VI remains.

Co-occurrence with intellectual impairment

Another dominant source of heterogeneity in the VI population is the high co-occurrence of intellectual impairment with VI, which may also co-exist with an additional disability (e.g., cerebral palsy, hearing impairment) (Cass et al., 1994; Reynell, 1978; Sonksen & Dale, 2002; Teplin, 1995). It has been estimated that between 30% and 70% of children with VI have a co-occurring disability, with intellectual impairment being the most frequent (C. A. Mervis, Boyle, & Yeargin-Allsopp, 2002; G. T. Scholl, 1986). Co-occurrence of additional disabilities, including cognitive abnormalities, is almost always present in children with cortical VI (Good et al., 2001). Hence, as argued earlier, the incidence of intellectual impairment can be significantly lessened by excluding such children from research designs (Dale & Sonksen, 2002; Sonksen & Dale, 2002). For instance, Dale and Sonksen (2002) reported an incidence of learning difficulties of 17 % in their group of children with Potentially Simple CDPVS, compared to previously reported incidence of 60-70% in samples with VI of more heterogeneous aetiology (Hirst, Poole, & Snelling, 1993; C. A. Mervis et al., 2002; Robinson, 1977). Naturally, for the purposes of research, another way of reducing sample heterogeneity imposed by the presence of children with intellectual impairment is excluding such children.

Issues involved in developmental matching

Another way of dealing with heterogeneity introduced by intellectual impairment in children with VI is to ensure that appropriate developmental matching to a suitable control and/or comparison group is carried out, although this is likely to be a challenging task. For instance, the concept of a control group in research with children with VI is likely to be both theoretically and practically problematic. In investigations of developmental processes of children with VI, including a suitably matched group of sighted children is generally seen as a useful method for obtaining a reference point to which to compare the outcomes of children with VI. However, in tests that require manual exploration, it has been proposed that utilising a blindfolded sighted group can act as a useful control condition, the method that has been successfully applied in research with blind adults (e.g., Röder, Rösler, & Spence, 2004) and in a few studies with school age children with VI (e.g., Hermelin, 1972; Landau, Spelke, & Gleitman, 1984). However, blindfolding sighted children (particularly at a younger age) may not only be practically difficult; this condition is likely to impose extra demands on sighted children and introduce an additional confounding factor. More specifically, blindfolding may place sighted children in a perceptual situation that is unnatural to them, potentially leading to incomplete exploration and underestimating their ability (also Pérez-Pereira & Conti-Ramsden, 1999). Indeed, there seems to be an inherent theoretical difficulty in attempting to control for lack of sight and the unique sensory experience of children with VI. Additionally, such sensory experiences are likely to differ between individual children with VI, depending on how little or how much residual vision they may have. For this reason, as an alternative to a blindfolded sighted control group, it may be more useful to compare children with differing levels of VI (e.g., PVI vs. SVI), as well as to a suitably matched comparison group of sighted children.

However, acquiring a developmentally matched sighted comparison group in research with children with VI can be challenging for two reasons. First, while the developmental levels of many children with VI, especially in the early years, reflect a lag in comparison to sighted norms, these levels may be seen as appropriate when the severity of the child's VI is taken into account (Reynell, 1978, 1979). What is especially interesting about this 'lag behind sighted norms' in children with VI is its potential to be overcome in later school years (Landau & Gleitman, 1985; Mills, 1993). More specifically, whereas the discrepancy with sighted norms may be particularly noticeable in the early development of children with VI (Reynell, 1978), the developmental profiles of the two groups appear to even out by school age as the children with VI 'catch up' with their sighted peers in a number of areas, most notably in language (Landau & Gleitman, 1985). The learning levels of some children with VI may always remain suboptimal, the reasons for which are beyond the scope of the current thesis. However, the 'catching up' phenomenon observed in many children with VI implies an inherently different developmental trajectory relative to children who are sighted (Warren & Hatton, 2003). This is generally problematic when utilizing a comparison group, particularly in the early, preschool years, when the slower rate of learning in the child with VI may be directly related to the severity of their visual impairment, rather than an underlying cognitive deficit (Reynell, 1978, 1979).

This links into the second challenge relating to developmental matching: the lack of appropriate measures for assessing developmental levels of children with VI. Most developmental tests and assessment materials used in research and clinical practice are not designed with children with VI in mind. Typically, these tests rely on visual stimuli which, while particularly useful with preschool and prelingual children, cannot be used with children with significant degrees of VI. Furthermore, reliable non-visual tests that are appropriate for use with both visually impaired and sighted children are exceptionally rare.

The most common way of matching children with VI and sighted children for the purpose of developmental research is by using tests of verbal intelligence. For children with developed linguistic skills, the verbal subtests from the Wechsler scales of intelligence for both pre-school and school children (i.e., the WPPSI and the WISC) (Wechsler, 1989, 1992) have been consistently shown to provide a reliable measure of the language-based intelligence in children with VI. However, using only verbal tests for assessing intelligence is not without its problems, as intelligence involves skills and abilities that cannot be tapped by purely verbal means (Warren, 1994). A small number of intelligence tests tapping haptic/performance abilities have been developed particularly for children with VI, such as the Blind Learning Aptitude Test, (Newland, 1979) and the Intelligence Test For Visually Impaired Children (Dekker, 1993), and these have been shown to provide a useful method in the educational context. However, the psychometric properties of these tests are relatively weak, and they may not be suitable for use with sighted children (i.e., they have not been tested or normalised using sighted samples); hence they may not be appropriate for research studies requiring comparison groups (Warren & Hatton, 2003).

Furthermore, tests of verbal intelligence such as the Wechsler scales are not suitable for young prelingual children with VI or those children whose language and cognitive levels are below the basal levels of such tests. The only test that has been designed specifically for this purpose is the Reynell-Zinkin Developmental Scales for Young Visually Handicapped Children (Reynell, 1979; Reynell & Zinkin, 1979). The Reynell-Zinkin scales (RSZ) are widely used in clinical and educational contexts for monitoring the developmental progress of young, pre-school children with differing levels of VI in the area of language, sensory-motor understanding and social adaptation. They rely on age equivalents, which are derived from comparison with other standardised scales for such children (e.g., Maxfield & Buchholtz, 1957), and as such provide semi-standardised norms. Importantly, separate norms are used for blind, partially sighted and sighted children, thereby providing a useful monitoring tool and a guide to developmental stages, and allowing for the consideration and implementation of appropriate teaching as early as possible. However, even though they are the most commonly used test of intellectual ability and progress of young children with VI, the RZS have never been psychometrically validated, thus they are generally lacking in reliability. Additionally, they have been found to overestimate the cognitive levels of children with VI, particularly in the area of verbal comprehension (Dale & Sonksen, 2002; Timer-Van de Vosse & Hamers, 1994), reflecting further the lack of appropriate psychometric properties of the RZS.

THE CURRENT METHODOLOGICAL CONSIDERATIONS

In the research reported in this thesis, a number of steps have been undertaken to ensure appropriate levels of experimental rigour, so the potential confounding effects of extraneous variables discussed above could be suitably addressed. Although more detailed accounts of the specific methods employed in individual aspects of the present research will be provided in the later chapters, a general overview of the current methodology will be given in the following section, so that these steps can be clarified.

An overview of the general method

For the purpose of the research presented here, the aim was to recruit a group of children with congenital PVI and SVI who met the Potentially Simple CDPVS criteria specified by Sonksen and Dale (2002). An additional inclusion criteria was that these children would be in their primary school years (approximately between ages 6 and 12) during the period in which the present thesis would be undertaken. Subsequently, in consultation with the DVC team, a group of 34 children meeting these criteria were identified and put forward for inclusion in the present research[7]. Most of these children attended the DVC for continuous clinical assessment and guidance throughout their pre-school years, while a minority visited the clinic only once. Out of the 34 children whose parents were approached for participation in this research, only 18 consented to participate at the time of the present research. To meet developmental matching criteria (see below), three of the 18 children were subsequently excluded from the part of the research that involved comparison with a typically developing group of sighted children at school age.

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[7] To avoid biased sampling, the children were identified consecutively through the DVC records, strictly following the inclusion criteria.

The main body of the thesis therefore comprises of research carried out with 15 children with VI in their school years. The design that is largely adopted throughout the thesis is independent-samples, whereby the developmental outcomes of the VI group (n = 15) have been compared to developmental outcomes of age and ability matched sighted children. In addition to the research carried out on concurrent developmental outcomes of this group, in a later part of the thesis (see Chapter 6, Part 2) a retrospective approach is undertaken, tracing back to most of the initially identified sample of 34[8], which also included the 15 children who have been seen at school age. The purpose of this was to explore the early (i.e., pre-school) developmental patterns in a wider presentation of children with Potentially Simple CDPVS by incorporating a cross-sectional design, whereby the outcomes of those with PVI and SVI could be compared to one another, as well as to a group of developmentally matched sighted children.

Reducing heterogeneity

In the present research, aetiology-related heterogeneity, which has been related to the high incidence of additional disabilities and intellectual impairment among children with congenital VI, has been decreased by excluding children with cerebral VI and children with Potentially Complicated CDPVS (Sonksen & Dale, 2002). However, in terms of the degree of VI severity, the current sample of 15 children remained a heterogeneous group. On the other hand, despite their varying levels of congenital VI, in clinical terms, all the children had a level of VI that was considered sufficiently significant to pose a developmental concern. Furthermore, in favour of the current methodology, each child's VI history had been clinically documented over their preschool years (including a formal functional vision assessment by the DVC paediatrician), so each individual child could be characterised very precisely with respect to the level of their VI. Thus, the group variation that may potentially be influenced by the children's differing levels of VI has been addressed by consulting individual children's clinical records and taking their varying levels of VI severity into account when interpreting the results. Importantly, the group was strongly homogeneous in terms of their cognitive levels (see below), a feature typically lacking from other similar studies.

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[8] During the children's clinic attendances spanning their preschool years, in addition to the database containing the information on children's vision levels and cognitive development, a database of video recordings of the clinical assessments in which children participated was also set up and archived - with parental consent - for the purposes of clinical research and training. Video data were available for 31 out of 34 children with VI who were identified as prospective participants.

Decision on developmental matching

Matching on verbal IQ and chronological age was seen as the most appropriate method for between-group comparisons with a typically developing group of sighted children in the school-age phase of the research. Three of the 18 children for whom consent was obtained for this part of the research were excluded due to the children's intellectual impairment. These children were assessed at the DVC where their intelligence was estimated to be below the normal range (VIQ ≤ 70). This lead to the rest of the group (n = 15) being strongly homogenous in terms of their verbal reasoning, with none below the average range.

However, the retrospective study of the VI children's early developmental outcomes included a more cognitively varied sample of children. As described earlier in this chapter, the rate or learning of a child with VI in the early years, although behind sighted norms, may be appropriate for that child's VI level (Reynell, 1979). Hence, it was decided that developmental matching at this early developmental stage to a sighted group would be most suitable if carried out on actual ability levels, that is matching on mental age equivalents using the raw scores, but not on chronological age (C. B. Mervis & Klein-Tasman, 2004). This is because finding a child with normally developing vision who matches the child with VI on both chronological and mental age in the pre-school stage is not only practically difficult, but may also introduce a different, yet potentially more challenging methodological problem, in that the reasons for developmental lag in the sighted child are likely to stem from inherently different, non-sensory origins (e.g., a genetic condition/disability such as Down syndrome).

Statistical considerations

In common with the previous research with children with VI, the present research design also reflects the methodological challenge imposed by a small sample size. Small samples are associated with increased and heterogeneous variances and the data distributions are more likely to deviate from normality (Tabachnik & Fidell, 2001). For this reason, on those occasions where the data distribution may require such consideration, non-parametric tests have been used in the present research as an appropriate alternative to parametric analyses.

Importantly, small sample size is the major contributor of reduced power in experimental design (Campbell, Julious, & Altman, 1995; Florey, 1993). Reduced power in empirical research is associated with an increased probability of Type I Error (i.e., false positive) (Howell, 2002). The most common method for controlling the Type I Error rate in multiple comparison designs is employing a mathematical correction such as Bonferroni, which adjusts the study-wide significance value to keep it constant at .05 and subsequently reduces the probability of a spurious (i.e., false positive) result (Howell, 2002). However, this method has been criticized as being too conservative and insensitive, reducing the likelihood of false positives at the cost of rejecting a truly significant result (i.e., Type II Error) (Benjamini & Hochberg, 1995; Bland & Altman, 1995; Perneger, 1998), the phenomenon to which a study with a small and rare clinical sample, such as the present one, is relatively susceptible (Perneger, 1998). For this reason, corrections for multiple comparisons or correlations have not been carried out in the present research. However, certain precautionary steps have been taken to acknowledge the likelihood of false discovery rates, while not capitalising on Type II Error. Results found to be significant at $p \leq .01$ were generally considered, whereas those found to be significant at $p \leq .05$ were interpreted only if they were pertinent to the specific research hypotheses or questions that the present research aimed to address (see individual chapters). In cases of multiple comparisons, the results significant at $p \leq .05$ have also been accompanied by the calculation of effect sizes, as were those found not to be significant ($p > .05$). The inclusion of effect size calculations is increasingly being advocated in empirical research (McCartney & Rosenthal, 2000). Use of these statistics is not only helpful in determining whether the phenomena of interest may truly be absent or due to lack of power (Cohen, 1992, 1994). It may also be more meaningful in assisting the making of an informed judgement about the practical importance of a given finding, rather than a binary choice between a significant and non-significant result (Folger, 1989; McCartney & Rosenthal, 2000). This is because the mechanical yes/no decisions, based on a clear-cut index of probability, although practical, are independent of the content and may oversimplify the practical importance of a given finding (Folger, 1989; McCartney & Rosenthal, 2000). To aid the interpretation of the given findings, a number of conventions for the estimates of effect sizes have been established. However, in the present research Cohen's conventions for the estimates of effect size '*d*' have been adopted as follows: Small effect - $d = .20$; Medium effect - $d = .50$; and Large effect - $d = .80$ (Cohen, 1992, 1994). The calculation of effect sizes has been carried out using G*Power (Faul, Erdfelder, Lang, & Buchner, 2007).

Ethical considerations

The recruitment of children with VI who took part in the research reported in this thesis and the relevant methodological procedures with these children and their families were carried out in strict guidance with the research protocol approved by the NHS Research Ethics Committee, University College London, Institute of Child Health and Great Ormond Street Hospital, London, UK, and the Research Ethics Committee at Goldsmiths, University of London, UK.

Chapter 3

Language and Social Communication in Children with Congenital VI

SUMMARY

The research presented in this chapter sheds light on language and social communication in children with congenital VI. This was done by examining language and socio-communicative profiles of a group of children with VI who have normal intelligence in the context of i) a structured language assessment, and ii) a parental report of everyday communicative behaviours, and by comparing these profiles with the profiles of a group of typically developing sighted children of similar age and verbal intelligence. The findings suggest a discrepancy in presentation of language ability in children with VI. That is, relative to their good and potentially superior structural language skills, children with VI showed poorer use of language for conversational and social purpose. The pragmatic language difficulties in the VI group were observed within a broader socio-communicative profile, which in a substantial proportion of children with VI in this study appears to be consistent with the pattern found in sighted children with autism.

INTRODUCTION

The developmental impact of visual impairment on social communication and social cognition has been well documented. As discussed in Chapter 1, empirical evidence suggests that children who are visually impaired from birth experience difficulties in early social relations (Preisler, 1991; Recchia, 1997; Rowland, 1983) and have delayed theory of mind understanding (Green et al., 2004; McAlpine & Moore, 1995; Minter et al., 1998; Peterson et al., 2000). It has also been highlighted that the pattern of these difficulties seen in children with VI often bears striking resemblance to the pattern of difficulties experienced by children with autism (R. Brown et al., 1997; Hobson & Bishop, 2003; Hobson et al., 1999; Pring, 2005).

Theoretically, such difficulties in both children with VI and sighted children with autism have been attributed to disruptions in interpersonal engagement in early childhood and a breakdown in understanding that one's experiences of the world can be shared with others (Hobson, 1990, 1993). Crucially, in both groups of children, poor social outcomes in early development involve interruption to processes (i.e., dyadic and triadic joint attention) and behaviours (i.e., eye-gaze directing and following) that are driven by the visual modality. However, according to Hobson (2002), whilst children with autism fail to understand how other people '*relate*' to the world for the reasons that extend beyond vision, the difficulty to understand how other people relate to the world in children with VI is likely to be a consequence of their inability to '*see*' these relationships (p. 192).

The important role of vision in social interaction has been implied by numerous research studies of joint attention development in young children (see Chapter 1). Consequently, it can be hypothesised that lack of visual input for children with congenital VI may ultimately have a detrimental effect on their socio-communicative competence and socio-cognitive outcomes and contribute to a presentation of autism-like characteristics. Importantly, however, such difficulties are not evident in all children who are visually impaired. The same studies that highlight the impact of VI on specific social milestones also identify some children with VI who show the level of social engagement (Urwin, 1978) and theory of mind competence (Green et al., 2004) that seems to be comparable with sighted children's achievements. This implies that children who are visually impaired can achieve relatively normal social development. Thus, it appears that, while vision may be the main means for social relating for children who are sighted, children with VI may be able to rely on alternative strategies to achieve social relations with other people.

With this in mind, language has generally been seen as playing a powerful role in the development of children with VI (Landau & Gleitman, 1985; Pérez-Pereira & Conti-Ramsden, 1999; Warren, 1994; Warren & Hatton, 2003). Pérez-Pereira and colleagues have maintained over the years that language provides an important tool for children with VI, who rely on it and benefit from it to a greater extent than do children who are sighted. According to these authors, language allows children who are visually impaired to participate in social interactions by providing them with a mechanism which compensates for the absence of visual input and transforms the visual world into a verbal one (Pérez-Pereira, 1994; Pérez-Pereira & Castro, 1997; Pérez-Pereira & Conti-Ramsden, 1999). The idea that language may be of crucial importance in promoting the ability to communicate and reason within a social context for children with VI has also been supported by others (e.g., Hobson, 1993).

Support for the role of language in social communication and social cognition in children with VI comes from the research described in more detail in Chapter 1. For instance, in her longitudinal examination of theory of mind in children with VI, Green (Green et al., 2004; Green/née Cupples, 2001) found that children with VI with an initial theory of mind delay were able to 'catch up' with their sighted peers when followed up approximately a year later, indicating that with an advancement of language, the social cognition of children with VI receives a positive boost. Crucially, research by Green et al. (2004) and also Minter et al. (1998) yielded the findings that the children with VI who passed a standard theory of mind task had significantly higher verbal IQs and verbal mental ages than did those who failed it. In a similar way, R. Brown et al. (1997) found that children with VI who had higher verbal ability (VIQ > 70) showed fewer autistic-like behaviours on the Childhood Autism Rating Scale (CARS) (Schopler, Reichler, & Renner, 1988) than did children with VI with lower verbal ability (VIQ < 70). Thus, it is argued that linguistic competence may be an important factor, which mediates social outcomes in children with VI (R. Brown et al., 1997; Hobson, 2005).

In children with VI, these studies clearly demonstrate that language, at least in the form of verbal IQ, distinguishes those children who show better from those who show poorer socio-communicative competence and socio-cognitive outcomes. However, the contribution of the mechanisms that language provides for such children requires further clarification. For children with VI, language-based measures are commonly used to assess general intellectual level, making it difficult to isolate the contribution of language irrespective of a child's general cognitive ability. The same issue arises from grouping children with VI with a wide range of intellectual abilities in research studies; thus, the better social outcomes of children with VI who have higher compared to those with lower verbal intelligence (as shown by Green et al., 2004; Minter et al., 1998; and R. Brown et al., 1997) may not be fully appreciated as a consequence of better language per se as much as a result of a higher intellectual level. It is likely that the developmental interaction between language, cognition and social outcomes in general is a complex one, and the overlap between these processes in a child with severe vision loss may be especially remarkable and adaptive (Peters, 1994). The aim of the current research is to attempt to understand the contribution of the mechanisms that language provides for children who have a congenital VI, by examining a wider presentation of language in a group of children with VI with normal intelligence.

Language is a complex system, consisting of a rich network of functions and skills that provide building blocks for communicative and social interaction. While *structural* language skills, such as articulation of speech, use of grammar, vocabulary level and conceptual understanding of the vocabulary in question, may enable a person to converse fluently, they are not sufficient for achieving successful socio-communicative interaction with another person. For this, one must also master *pragmatic* language skills, that is, the ability to use language appropriately in a given context. Vision is implicated in language development in general as visually-driven joint attention experiences in early childhood are seen as providing a framework within which language learning occurs (Charman et al., 2000; Tomasello & Farrar, 1986). For this reason, visual input may be of particular importance in the development of pragmatic language skills which are a cardinal feature of social communication. It is clear that important aspects of pragmatic language are carried out in the visual modality, with an emphasis on non-verbal behaviours that have a communicative purpose, such as the use of gestures, bodily postures and facial expressions to convey meaning and intentions. It is therefore important to consider long-term language outcomes for children with VI in order to ascertain whether visual impairment per se is likely to give rise to any lasting difficulties in either structural or pragmatic language development.

With regards to the structural language skills of children with congenital VI, research generally shows that these are developed with relative ease. A number of studies have demonstrated some specific delays and irregularities in early vocabulary acquisition and production, syntactic knowledge, and the acquisition of semantic concepts in children with VI (Andersen et al., 1984; Dunlea, 1989; Fraiberg, 1977; McConachie & Moore, 1994). But on the other hand, many researchers argue that the early presentation of structural language in children with VI is largely in line with that of sighted children (Landau & Gleitman, 1985; McGinnis, 1981; Mulford, 1988; Pérez-Pereira & Castro, 1997; Urwin, 1983), even though their expressive language generally tends to be ahead of their receptive language (Reynell, 1978; Reynell & Zinkin, 1979). Despite some conflicting evidence, the prevailing view amongst the researchers in the field is that the early language of children who are visually impaired from birth is not necessarily deviant, but simply different from the language of sighted children (Andersen et al., 1984; Landau & Gleitman, 1985; Mills, 1993; Mulford, 1988; Pérez-Pereira & Castro, 1997; Urwin, 1983); and whatever the delays and differences in early language structure, they generally seem to be overcome by school age (Landau & Gleitman, 1985; Reynell, 1978). Despite following what seems like an alternative route of language development, children who are visually impaired ultimately seem to arrive at the same point as do sighted children (Mills, 1993). Interestingly, the majority of evidence concerning language structure in children with VI concerns the early years whereas language ability at school age (and in adulthood) has been largely ignored.

The picture is also less clear regarding language use for social and pragmatic purpose in children with VI. It has been suggested that the pragmatic language of children with VI has features that are similar to those of children with pragmatic language impairment (PLI) (Mills, 1993). Such features involve the extensive, and sometimes inappropriate, use of questions (Erin, 1986; McGinnis, 1981; Mulford, 1983), an absence of communicative gestures (Preisler, 1991; Rowland, 1984; Urwin, 1978) and the extensive use of imitative speech, repetitions and verbal routines (Dunlea, 1989; Norgate, Collis, & Lewis, 1998; Pérez-Pereira, 1994; Pérez-Pereira & Castro, 1992). It has been argued that such features of the pragmatic language use of children with VI may have an important function in promoting their cognition and social interaction by providing an adaptive strategy by which to gather information (Erin, 1986), analyse speech (Kekelis & Andersen, 1984; Pérez-Pereira, 1994; Peters, 1994), reduce memory load (Spiedel, 1989), and avoid isolation (Peters, 1994). However, a number of researchers emphasise a concern revolving around these language features in children with VI (e.g., stereotypic speech and echolalia) which are largely seen in children with autism and thus contribute to the presentation of an autism-like syndrome in children VI (R. Brown et al., 1997; Fraiberg, 1977; Hobson, 1993; Wills, 1979).

Based on the evidence from aforementioned studies, involving mostly preschool children with congenital VI, it generally appears that structural language is an area of relative strength for children with VI, whereas their pragmatic skills may be challenged. More recently James and Stojanovik (2007) demonstrated irregularities in the language presentation of a group of eight children with congenital VI (age-range 12-17 years) based on parental ratings of language and communicative behaviours using the Children's Communication Checklist (D. V. M. Bishop, 2003). The checklist targets both structural and pragmatic language behaviours observable in an everyday context, as well as social interaction skills evident from everyday language use, the impairment of which (in addition to pragmatic abnormalities) is a cardinal feature of children with autism (Lord & Paul, 1997). When examined with reference to the developmental norms for typically developing sighted children, the pattern of language presentation in the group of children with VI in James and Stojanovik's study suggested a discrepancy between certain aspects of structural language and pragmatics, with the group's use of context and non-verbal behaviours for communicative purpose falling below normal range limits. Additionally, a substantial proportion of children appeared to show a communicative profile that warranted further clinical investigation.

However, the study was largely descriptive and of a preliminary nature, and even though an attempt had been made to exclude children with an intellectual impairment in this study, the children's intellectual capacity was not systematically assessed and their profiles were not examined in comparison to a developmentally matched sighted group. The mismatch between structural and pragmatic language ability in children with VI with normal intelligence, and who do not have an additional diagnosis of autism or pragmatic language impairment, needs to be further substantiated with research.

The primary objective of the current study was to provide further investigation of the language ability and communicative behaviours of children who are born with severely impaired vision. In order to examine variation in language skill, independent from overall intellectual level, the current study a) focused on children with a significant congenital vision loss who have normal intelligence, b) utilised a comparison group of typically developing sighted children matched on verbal IQ, c) used a standardised test designed specifically to assess language function in children in the context of a structured assessment, and d) utilized a parental report of language and communicative behaviours in an everyday context. Language function and behaviours of the children with VI in this study were also examined in the context of a broader socio-communicative profile, in order to provide further insight into the nature of autistic-like presentation of children with VI with normal verbal intelligence. The outcomes of children with VI were compared to the outcomes of children who are sighted in order to see the extent to which the two groups differ and to gain further appreciation of specific strengths and weaknesses that may characterise the VI group.

Research questions and hypotheses

In line with the theoretical framework outlined thus far, the objective of the research presented in this chapter was to test the following experimental hypotheses and address some specific questions relating to those hypotheses:

Hypothesis 1 (H1): Language competence, as measured by a standardised test of language function, will differentiate children with VI from children who are sighted.

Question 1 (Q1): Will this difference be confined to a particular language domain (i.e., receptive vs. expressive)?

Question 2 (Q2): Will this difference vary as a function of a specific language skill (i.e., standardised language subtest)?

Question 3 (Q3): Is the performance on the test of language function independent of the measure of verbal IQ in children with VI?

Hypothesis 2 (H2): Children with VI and children who are sighted will show different social and communicative profiles, as assessed by parental reports.

Question 4 (Q4): Will this difference vary as a function of a specific socio-communicative skill (i.e., communication checklist scale)?

Hypothesis 3 (H3): There will be a discrepancy between language structure and pragmatic language in children with VI, compared to children who are sighted on a parental communication checklist?

Hypothesis 4 (H4): Socio-communicative profiles of children with VI, compared to the profiles of their verbal ability matched sighted peers, will be reminiscent of profiles typically seen in sighted children with autism.

Question 5 (Q5): Can severity of VI and the level of language competence explain the variation in the severity of such profiles in the VI group?

Hypothesis 5 (H5): The pattern of specific socio-communicative weaknesses in the VI group will be consistent across different measures (i.e., cross-measure correlations).

METHOD

Participants

Fifteen children with congenital visual impairment and 26 children with normally developing vision took part in the current research. The children with VI were recruited with parental consent through the Developmental Vision Clinic at the Great Ormond Street Hospital, London, UK where they had been referred to for developmental and functional vision assessments in their early years. All but one child from the VI group attended a mainstream school in England. The sighted children were recruited with parental consent through primary schools in South East London and Kent, UK. For the purpose of developmental matching, the inclusion criteria for the sighted children were: age (6 - 12 years) and verbal IQ within the normal range (VIQ $\geq$ 80). The majority of children in both participant groups came from white British families (over 50%), although the VI group included a somewhat larger proportion of children from a variety of ethnic backgrounds (VI - 47 %; Sighted - 31%).

The children with VI all had a degree of vision loss which was present from birth and was of peripheral, rather than cerebral origin. The inclusion criteria based on the origin and the site of the VI, which was adopted from the taxonomy by Sonksen and Dale (2002), is described in more detail in Chapter 2 (i.e., Congenital Disorders of Peripheral Visual System / CDPVS). The group consisted of children with varying levels of congenital VI, deemed sufficiently clinically severe to pose a developmental concern (Dale & Sonksen, 2002; Sonksen & Dale, 2002). This included children whose VI was profound - PVI (light perception or worse / no form vision) and those children whose VI was severe - SVI (visual acuity of worse than 6/30; some limited form vision, but severely degraded). Even though the children with differing levels of VI were grouped together for a comparison with the control group in the current study, the heterogeneity of the sample in terms of the degree of vision loss has been taken into consideration in the interpretation of the subsequent results. None of the children with VI had a known additional diagnosis (e.g., severe hearing impairment or autism). Individual child characteristics, including level of vision loss in the early years and specific diagnoses are summarized in Table 3.1.

Table 3.1: *Individual characteristics of children with VI*

N	***ID***	***Age***	***Gender***	***Visual diagnosis***	***VI level***
1	02	6:08	M	Familial exudative vitreo-retinopathy and Norrie's Syndrome	SVI
2	04	7:00	F	Leber's amaurosis	PVI
3	07	9:02	M	Bilateral micropthalmia and optic nerve aplasia	PVI
4	09	7:00	M	Bilateral optic nerve hypoplasia	PVI
5	10	12:11	F	Bilateral optic nerve hypoplasia	PVI
6	16	8:03	F	Bilateral microphthalmia	PVI
7	17	11:04	M	Bilateral Aniridia and glaucoma with Peter's anomaly in one eye	SVI
8	18	6:06	F	Leber's amaurosis	SVI
9	19	9:11	M	Bilateral micropthalmia with multiple corneal opacities	PVI
10	20	8:05	F	Multiple opacities and sclerocornea	SVI
11	23	8:07	M	Leber's amaurosis	SVI
12	25	7:03	F	Persistent primary hyperplastic vitreous	SVI
13	26	8:01	F	Bilateral mycropthalmia and sclerocornea	SVI
14	31	10:11	F	Leber's amaurosis	SVI
15	32	6:06	F	Leber's amaurosis	SVI

The VI data reported in Table 3.1 were obtained from the archived clinical records, which contain each child's history of comprehensive formal functional vision assessments by a paediatrician across the preschool years. Based on these early assessments five children had a consistent PVI (no presence of form vision) and six had a consistent SVI (presence of form vision) through their early years. It is worth noting that four children obtained some useful form vision (SVI) after an initial PVI in the first year of life (Participant IDs: 02, 18, 19 and 20). Interestingly, one of these children (Participant ID: 19) experienced a total vision loss approximately two years after gaining a level of useful form vision (i.e., SVI) in the first year of life. Cases like these highlight the difficulty of fully accounting for a degree of vision level in research with children with congenital VI (the related methodological considerations were discussed in Chapter 2). No formal screening for the severity of VI was carried out at the time of the current research and the vision levels in the

Table 3.1 are those obtained from the latest preschool assessment of functional vision that children underwent before their participation in this study.[9]

The children with VI and the sighted children in the study were matched on verbal IQ (VIQ), age and gender (see Table 3.2). Independent-samples *t* tests showed that that the two groups did not differ significantly in terms of their VIQ ($t_{(39)} = -.105$; $p = .917$) or Age ($t_{(39)} = -.502$; $p = .618$). Chi-square tests revealed that the two groups were comparable in terms of gender ratio, with more girls than boys in each group ($\chi^2_{(1)} = .702$; $p = .754$).

Table 3.2: *Matching characteristics of the VI and Sighted groups*

Matching criteria	***VI*** *N = 15*	***Sighted*** *N = 26*	***p value***
VIQ / WISC-III			
Mean (SD)	105.9 (10.7)	106.3 (11.1)	.917
Range	84 - 128	80 - 130	
Age			
Mean/ months (SD/ months)	103.1 (23.0)	106.5 (20.3)	.618
Range/years	6:06 – 12:11	6:02 - 11:11	
Gender ratio (Female/Male)	9/6	14/12	.754

Materials

Wechsler Intelligence Scales for Children- III / WISC-III

Verbal scales from the WISC-III (Wechsler, 1992) were used for the purpose of developmental matching. The WISC is a widely used children's intellectual test that measures intellectual abilities in both Verbal and Performance areas. The Verbal subtests primarily measure the child's ability to solve verbal problems using verbal and auditory skills, whereas the Performance subtests primarily measure the child's ability to solve visual and constructional problems using non-verbal, or only partially verbal skills.

The WISC-III consists of 13 subtests which are divided into 2 scales: Verbal Scale and Performance Scale. The Verbal Scale consists of 6 subtests containing language based items,

[9] The greatest development to the visual system occurs across the early years of life and the visual level is usually stable by the early school years (Sonksen, 1993).

whereas the Performance Scale contains 7 subtests that use visuo-motor items that are less dependent on language. Due to their visual content, the items on the Performance Scale are not suitable for children with severe VI. The Verbal Scale items are commonly used with such children to obtain an index of the children's general ability and Verbal IQ (VIQ) respectively. Thus, in the current study the VIQ was derived from 5 verbal subtests presented in Table 3.3.

Table 3.3: *WISC-III verbal subtests*

Subtest	***Subtest description***	***Item example***
Information	A test of general factual knowledge that relies on long-term memory.	How many days are there in a week?
Similarities	A measure of abstract/logical reasoning requiring the ability to understand categories and relationships between category domains.	How are **guitar** and **piano** alike?
Vocabulary	A measure of expressive language/verbal fluency. An example question would be to ask the child to describe a concept in their own words	What does **brave** mean?
Comprehension	A measure of knowledge of appropriate social behaviour and judgement.	What would you do if you lost a ball that belongs to one of your friends?
Digit Span [a]	A test of verbal, short-term working memory. The examiner reads a sequence of single digits and the child is asked to repeat the numbers verbatim, forwards and backwards. The sequences start with 2 digits (2 trials) and increase to up to 8 digits.	N/A

[a] Digit Span is a supplementary subtest, used here instead of the core Arithmetic subtest.

The administration and scoring of the subtests was carried out in the accordance with the WISC-III manual. The raw scores on each subtest were given scaled values, the sum of which is standardised in order to obtain an index of child's verbally based intellectual level (i.e., VIQ). The standard scores on individual subtests are based on normalized standard scores with a mean of 10 and a SD of 3. The composite standard scores (e.g. VIQ) have a mean of 100 and SD of 15.

The Clinical Evaluation of Language Fundamentals-3 / CELF-3

The CELF-3 (Semel, Wiig, & Secord, 2000a) is a standardised clinical tool used for the identification, diagnosis and evaluation of language skill deficits in school age children, adolescents and young adults. It consists of 11 subtests (8 core and 3 supplementary) that provide measures of receptive and expressive language in areas of morphology, syntax, semantics and memory. However, most of the CELF-3 subtests require the visual presentation of stimuli, which are unsuitable for use with children with visual impairment and for that reason only the non-visual subtests (2 core and 2 supplementary ones) were used with the current sample. Six age-appropriate subtests must be completed by each child in order to obtain a Receptive (3 subtests), an Expressive (3 subtests) and a Total Language composite score (sum of Receptive and Expressive language score). However, as only four non-visual subtests were used with the current sample, the Receptive and Expressive Language scores were calculated as a pro-rata of two subtests for each language domain. Importantly, this significantly reduces the reliability and value of the CELF-3 as a clinical tool, and although the obtained composite scores are investigated here for research purposes, they would have to be treated with caution if they were to be interpreted clinically.

The details of the individual subtests are given below. The administration and scoring of each subtest was carried out in accordance to the CELF-3 manual. As for the WISC, the standardized CELF-3 scores are based on normalized standard scores with a mean of 10 and a SD of 3. The composite standard scores have a mean of 100 and SD of 15.

CELF – 3 Receptive language subtests

Word Classes (WC) is a core subtest on the Receptive language scale of the CELF-3, used to assess the ability to perceive relationships in the meaning of words and to form word associations. These relationships may be categorised by part-whole and semantic class features and by synonyms and antonyms. The stimuli consist of 10 three-word items and 24 four-word items. These are read out to children and the children are asked to judge which two of the three (e.g., *simple*, happy, *easy*) or four words (e.g., horse, plane, *ship*, *boat*) that they heard go together best. The children are given a score of 1 for each correct response and a score of 0 for each incorrect or unanswered item.

Listening to Paragraphs (LP) is a supplementary subtest on the Receptive language scale of the CELF-3, used to assess comprehension, recall and interpretation of factual, inferential, sequential and predictable information. The stimuli consist of 2 short paragraphs (different paragraphs depending on the age group) which are read out to the child, each paragraph followed by five questions pertinent to the content of the paragraph (see example paragraph in Table 3.4). A correct item scores 1 and incorrect or unanswered item scores 0.

Table 3.4: Example paragraph from Listening to Paragraphs subtest from the CELF-3

Trial A (ages 6 to 9.11 years):	***Questions:***
Lisa was excited because her sixth birthday had finally come. After her family sang 'Happy Birthday', Lisa blew out the candles on her cake. Then, Lisa's mother told her to look in the garage for her present. As Lisa opened the garage door, she heard a 'meow' and felt something furry rub against her leg. Lisa was happy that she got her birthday wish.	Why was Lisa excited? How old was Lisa on her birthday? What was Lisa's birthday present? What did Lisa do after her family sang 'Happy Birthday? What do you think Lisa will do now?

CELF – 3 Expressive language subtests

Recalling Sentences (RS) is a core subtest on the Expressive language scale of the CELF-3 and is used to assess immediate recall and reproduction of sentence surface structure as a function of syntactic complexity. There stimuli consist of 26 sentences that are read out to the child and which become successively longer and more complex. The scoring for each sentence ranges from 0- 3 depending on the number of errors, omissions, substitutions that the child makes.

Word associations (WA) is a supplementary subtest on the Expressive language scale of the CELF-3 used to asses semantic organisation and word association strategies to generate and name members of a semantic class rapidly and efficiently. During the test the child is asked to name as many items as possible within a given category and in one minute. There are 3 test trials corresponding to 3 different categories, namely: *Animals*, *Foods* that people eat and *Jobs and Occupations* that people do. An appropriate response scores 1 while and incorrect or repeated response scores 0. The scores are added up over the 3 trials.

The Children's Communication Checklist – 2 (CCC-2)

The CCC-2 (D. V. M. Bishop, 2003) is a parental report questionnaire used i) to screen for children who are likely to have a language impairment, ii) to identify pragmatic impairments in children with communication problems and iii) to help identify children who may need further assessment for an autistic spectrum disorder. The function of the test is to obtain an evaluation of communicative skills that are not easy to evaluate in a context of a traditional structured assessment, as this context is not sensitive to the pragmatic communicative problems in children. It is designed to be completed by an adult who has regular contact with the child, typically the parent.

The CCC-2 consists of 70 multiple choice items and is divided into 10 scales (Table 3.5), each with 7 items. For each scale, 5 items describe communication difficulties and two describe communication strengths. On each item, the ratings of frequency of occurrence of a given behaviour are made on a 4 point scale ranging from 0 (less than once a week /or never); 1 (at least once a week, but not every day); 2 (once or twice a day) and 3 (several times /more than twice a day /or always). The raw scores on the CCC-2 are standardized on a normative population and scaled with a mean of 10 and SD of 3.

Table 3.5: *CCC-2 scales*

Scale	***Item example***
a) Speech	Pronounces words in a babyish way, such as "chibley" for "chimney" or "bokkle" for "bottle".
b) Syntax	Leaves out "is" and so says "Daddy going to work" rather than "Daddy's going to work" or "Daddy is going to work". Or might say "the boy big" rather than "The boy is big".
c) Semantics	Forgets words s/he knows, e.g., instead of "rhinoceros" may say "you know, the animal with the horn on its nose".
d) Coherence	Can be hard to tell is s/he is talking about something real or make-believe.
e) Inappropriate Initiation	Talks repetitively about things that no one is interested in
f) Stereotyped language	Repeats back what others have just said. For instance if you ask "what did you eat?" might say, "what did I eat?"
g) Use of context	Misses the point of jokes and puns (though may be amused by nonverbal humour such as slapstick).
h) Non-verbal communication	Stands too close to other people when talking to them.
i) Social relations	Hurts or upsets other children without meaning to.
j) Interests	Talks about lists of things s/he has memorised e.g., the names of the capitals of the world or the names of varieties of dinosaurs.

The first four scales (i.e., A – D) assess aspects of language *structure*. These scales cover the language domains that are often impaired in children with specific language impairments (SLI). The scales E – H cover *pragmatic* aspects of communication that are not easily assessed by conventional language assessments. The remaining scales, I and J, assess social behaviours that are usually impaired in cases of ASD. Although low scores on these two scales, as well as low scores on the scales assessing the pragmatic aspects of communication, may suggest further clinical evaluation, they cannot be used to diagnose autism.

Two composite scores can be derived from the CCC-2. The *General Communication Composite (GCC)* based on the first eight scales can be used to identify children likely to have clinically significant communication problems (i.e., if their GCC score is below 55). It is also possible to derive a *Social Interaction Deviance Composite (SIDC),* which reflects the mismatch between the sums of scales E, H, I and J, and the sums of scales A, B, C and D. The SIDC composite can therefore help identify the children in whom pragmatic language skills and social interaction skills are disproportionately impaired relative to their structural language. However, the SIDC provides qualitative information about the pattern of impairment and would normally only be interpreted in combination with the GCC and, more specifically, if the child obtains a score on the GCC below 55. For example, a negative score on the SIDC in combination with the GCC score below 55 is common in children with ASD. A positive score on the SIDC (i.e., above 9), combined with the GCC score below 55, is common with children with specific language impairment (SLI). However, a low SIDC score of ≤ -15, even if the GCC is within normal limits, may have clinical significance. Such extreme scores are rare in a normative population and are frequently seen in Asperger Syndrome (AS).

All of the items on the CCC-2 were considered appropriate for use with children with VI except item 14 (i.e., '*does not look at the person s/he is talking* to'). In over 50% of the cases, the parents of children with VI omitted this item, which was subsequently removed from analyses for both VI and Sighted groups.

The Social Communication Questionnaire (SCQ)

The SCQ (Rutter, Bailey, & Lord, 2003) is a parental report questionnaire that screens for the socio-communicative difficulties associated with ASD in children who are 4 years of age or older. It consists of 40 items with a yes/no response format. There are two forms of the SCQ: the Lifetime Autoscore which is used to obtain a child's entire developmental history and assist a diagnostic workup; and the Current Autoscore, which is used to assess a child's behaviour over the most recent 3 month period with a purpose to understand everyday living experiences of a child and evaluate treatment and educational plans. For the current thesis the Lifetime Autoscore has been used for its screening function. In this context, the SCQ Total Score is interpreted with reference to the cut-off score of 15 or greater, which is indicative of a possible autism and requires further clinical evaluation. Unlike the CCC-2, elevated scores on the SCQ are indicative of higher prevalence of behavioural difficulties. The SCQ can be broken down into three sub-scores that match three behavioural domains on the Autism Diagnostic Interview-Revised (ADI-R) that form the basis for diagnosis of autism (Rutter et al., 2003): *Reciprocal Social Interaction Domain, Communication Domain and the Restricted, Repetitive and Stereotyped Patterns of Behaviour Domain*. Although formal scoring of the subscales is not supported in the SCQ manual, the subscales can be investigated for research purposes as has been done here.

Procedure

The overall procedure took on average an hour and a quarter. The majority of children were assessed in a single session at home. In minority of cases, due to practical reasons, an additional assessment was carried out at school. Typically, the children took part in the language tasks while their parents completed the questionnaires. All of the questionnaires were completed by the parents of the children except in the case of two children with VI where the questionnaires were completed by a teacher who knew the children well. These two children came from the families where English is not the main language spoken at home, although both children are bilingual and attend an English school. All the language tasks were presented in the same order for all the children, with the WISC-III subtests being presented first, followed by the CELF-3 subtests, and in the order specified in the test manuals.

RESULTS

Structured language assessment

CELF-3

First, it was of interest to examine whether the CELF-3 would discriminate between the two groups in terms of their language ability (H1). The means and standard deviations (SDs) of the two groups on the CELF-3 composites and individual subtests, as well as the WISC-III performance, are summarised in Table 3.6.

Table 3.6: *Means and SDs on WISC-III and CELF-3 for the VI and Sighted groups*

Measure *Mean (SD)*	***VI***	***Sighted***	***p value***
WISC – III			
Verbal IQ / VIQ	105.9 (10.7)	106.3 (11.1)	.917
Information	12.2 (3.1)	11.8 (2.7)	.674
Similarities	11.6 (2.7)	12.8 (3.0)	.214
Vocabulary	11 (3.6)	10.7 (3.3)	.810
Comprehension	8.9 (1.9)	9.7 (2.1)	.218
Digit Span	11.1 (2.7)	10.3 (2.5)	.347
CELF - 3			
Receptive Language Composite	104.5 (10.3)	96.7 (9.1)	.016
Word Classes	11.4 (2.4)	10.04 (2.4)	.089
Listening to Paragraphs	9.9 (1.5)	8.8 (2.6)	.097
Expressive Language Composite	113.8 (15.6)	102.5 (14.4)	.024
Recalling Sentences	12.6 (2.7)	9.8 (3.1)	.005
Word Associations	11.9 (3.5)	10.7 (2.6)	.217
Total Language Composite	109.6 (12.9)	99.3 (11.3)	.011

As predicted (H1), the two groups were found to differ significantly in terms of their language competence as assessed by the CELF-3 (Total Language). More specifically, independent-samples *t* tests revealed that, despite being comparable in verbal ability (as measured by the WISC-III VIQ – Table 3.1, as well as the individual WISC-III subtests[10]), the children with VI

[10] Information ($t_{(39)} = .423$, $p = .674$, $d = .14$), Similarities ($t_{(39)} = -1.262$, $p = .214$, $d = .40$), Vocabulary ($t_{(39)} = .243$, $p = .810$, $d = .09$), Comprehension ($t_{(39)} = -1.252$, $p = .218$, $d = .40$), and Digit Span ($t_{(39)} = .952$, $p = .347$, $d = .31$).

achieved significantly higher scaled composite scores than the Sighted group on the CELF-3 overall (Total Language: $t_{(39)} = 2.674$, $p = .011$). This language strength did not seem to be confined to a specific language sub-domain (Q1), as the VI group achieved higher performance in terms of both their receptive and expressive language (Receptive: $t_{(39)} = 2.528$, $p = .016$, $d = .80$; Expressive: $t_{(39)} = 2.352$, $p = .024$, $d = .75$). Figure 3.1 graphically illustrates this apparent language strength of the VI group across the three CELF- 3 composites.

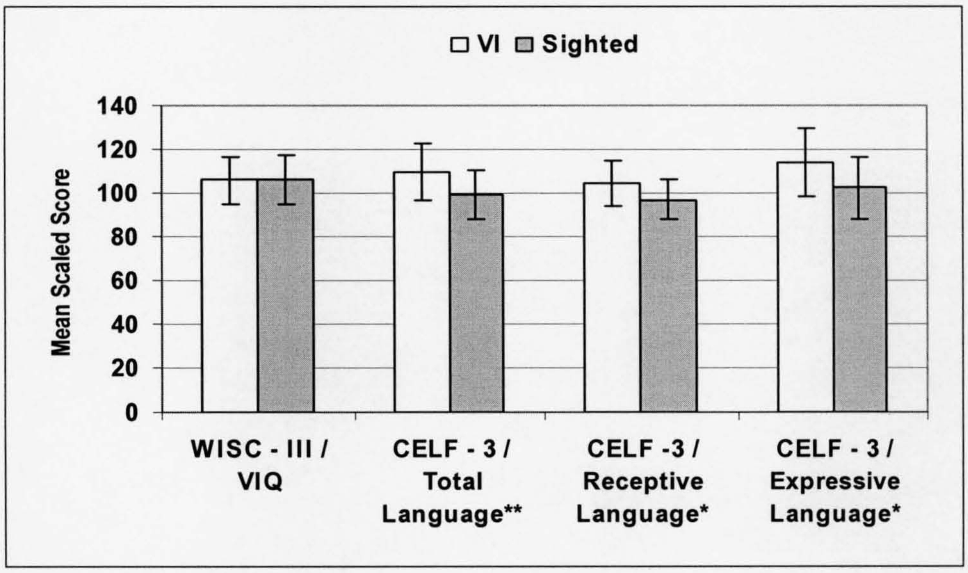

Figure 3.1: *Mean scaled scores on the verbal WISC-III and CELF-3 composites (error bars represent the SDs, ** $p \leq .01$ and *$p \leq .05$)*

However, the difference in language competence (as measured by the CELF-3) between the two groups did seem to vary as a function of a specific language skill (Q2), as the only CELF-3 subtest discriminating the children with VI as significantly better than the Sighted was Recalling Sentences ($t_{(39)} = 2.956$; $p = .005$). With regards to the other three CELF-3 subtests, there were trends towards a significant difference on Word Classes ($t_{(39)} = 1.742$; $p = .089$) and Listening to Paragraphs ($t_{(38.9)} = 1.702$; $p = .097$). The effect sizes for these trends were only medium ($d = .56$ and $d = .48$ respectively). The group difference on Word Association was not significant ($t_{(39)} = 1.256$; $p = .217$, $d = .40$). Figure 3.2 and Figure 3.3 graphically illustrate the performance of the children in the two groups on the individual subscales on both WISC-III and CELF-3.

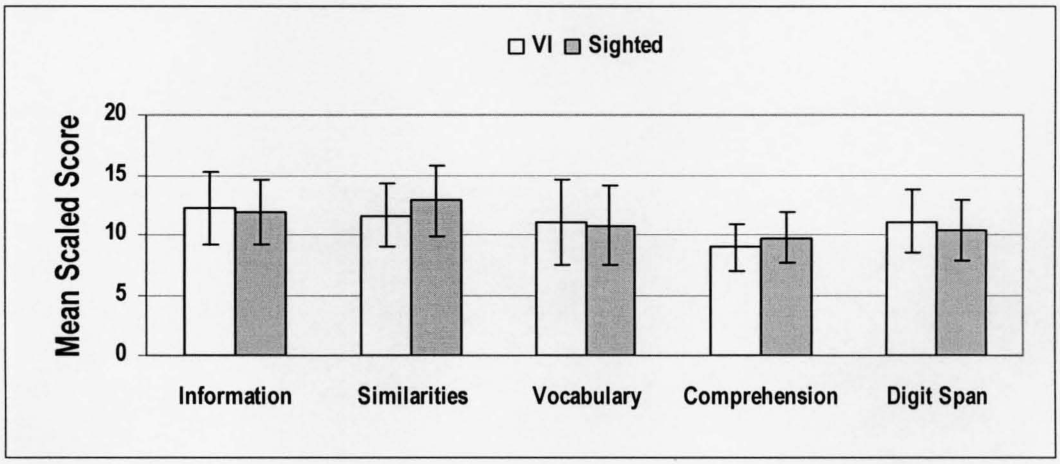

Figure 3.2: *Mean scaled scores on the individual WISC-III subscales (error bars represent the SDs)*

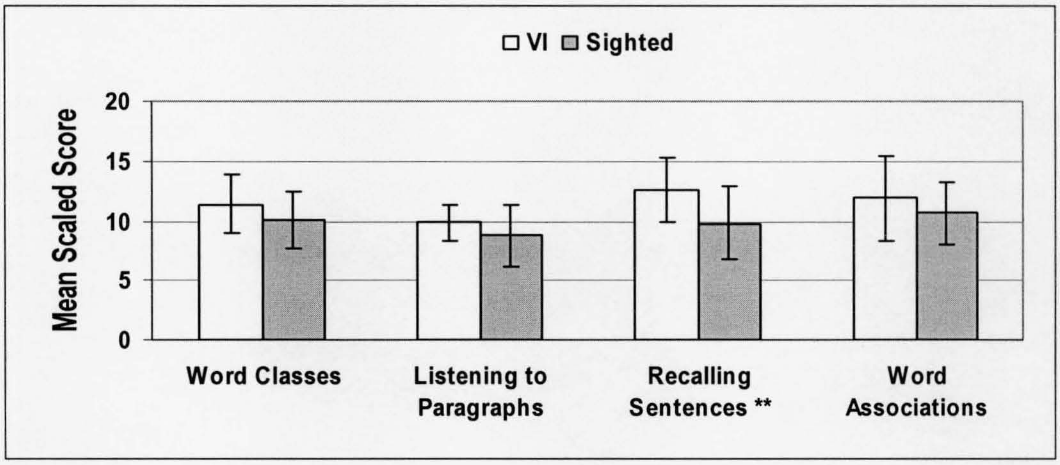

Figure 3.3: *Mean scaled scores on the individual CELF-3 subscales (error bars represent the SDs, ** $p \leq .01$)*

It may be worth highlighting that the performance of the Sighted group on the CELF-3 Total Language was significantly discrepant from their VIQ ($t_{(25)}$ = 4. 231; $p \leq .001$), whereas the scores of the VI group seemed comparable across the two composites ($t_{(14)}$ = -1. 262; $p = .228$, d = .33). The differing correlational pattern between individual WISC-III and CELF-3 subtests in both groups (Appendix A1) potentially supports this picture of differing language profiles manifested by the two groups (in line with H1). However, the overall performance on the two tests (CELF-3/Total Language and WISC- III / VIQ) was significantly correlated in both children with VI ($r = .559$; $p = .03$, n = 15) and sighted children ($r = .715$; $p \leq .001$, n = 26) signifying that the skills required for the two tests may not necessarily be independent (Q3).

Parental reports

CCC-2

With regards to the range of every day language and communicative behaviours based on parental reports on CCC-2, the profile of the VI group was examined in comparison to that of the sighted group. The means and SDs of the two groups on the individual CCC-2 scales are summarized in Table 3.7.

Table 3.7: *Means and SDs on the CCC-2 for the VI and Sighted groups*

Measure *Mean (SD)*	***VI*** *Missing N = 1*	***Sighted*** *Missing N = 1*	***p value***
CCC – 2			
Speech	9.9 (3.5)	10.3 (2.6)	.691
Syntax	9.6 (3.6)	10.9 (1.9)	.228
Semantics	8.6 (3.1)	11.1 (2.7)	.01
Coherence	8.7 (3.3)	11 (2.5)	.021
Inappropriate Initiation	7.5 (3.1)	11 (2.6)	.001
Stereotyped language	6.6 (3.6)	10.3 (3.3)	.003
Use of Context	6.5 (2.4)	11.2 (2.9)	.001
Non verbal	4.3 (2.6)	10.9 (2.7)	.001
Social	5.4 (2.8)	10.4 (2.9)	.001
Interests	6.5 (2.5)	9.5 (2.9)	.003
General Communication Composite (GCC)	61.8 (18.8)	86.8 (14)	.001
Social Interaction Deviance Composite (SIDC)	-13.14 (7.04)	-1.5 (7.7)	N/A

Profile Analysis was carried out to compare the profiles of the VI and Sighted groups across the CCC-2 scales (H2). Profile Analysis is a multivariate technique which examines three components of the profiles: 1) Flatness of the profiles, or whether performance, when collapsed across between-participant groupings, differs across measures (cf. 'main effect of Measure' in ANOVA), 2) Levels, or whether there is a between-group difference across measures (cf. 'main effect of Group' in ANOVA) and 3) Parallelism, or whether the profiles of different groups are parallel (cf. 'Interaction' in ANOVA).

The Profile Analysis comparing the profiles of the two groups revealed a significant test of Flatness, indicating that when averaged across the groups the children's performance differed across different CCC-2 subtests (Pillai's Trace criterion: $F_{(9, 29)} = 5.323$; $p \leq .001$). Furthermore, the test of Levels revealed a significant difference between groups when their scores were averaged across different CCC-2 scales ($F_{(1, 37)} = 26.6$; $p \leq .001$). However, these tests are qualified by a significant test of Parallelism, indicating distinguishable profiles between the two groups across different CCC-2 scales (Pillai's Trace criterion: $F_{(9, 29)} = 7.266$; $p \leq .001$). In summary, and in line with H2, the Profile Analysis results indicate that the two groups were significantly different in terms of their language and communication profiles as measured by the CCC-2 scales. However, this between-group difference varied as a function of a different CCC-2 scale on which specific communicative behaviours of the children were rated (Q4).

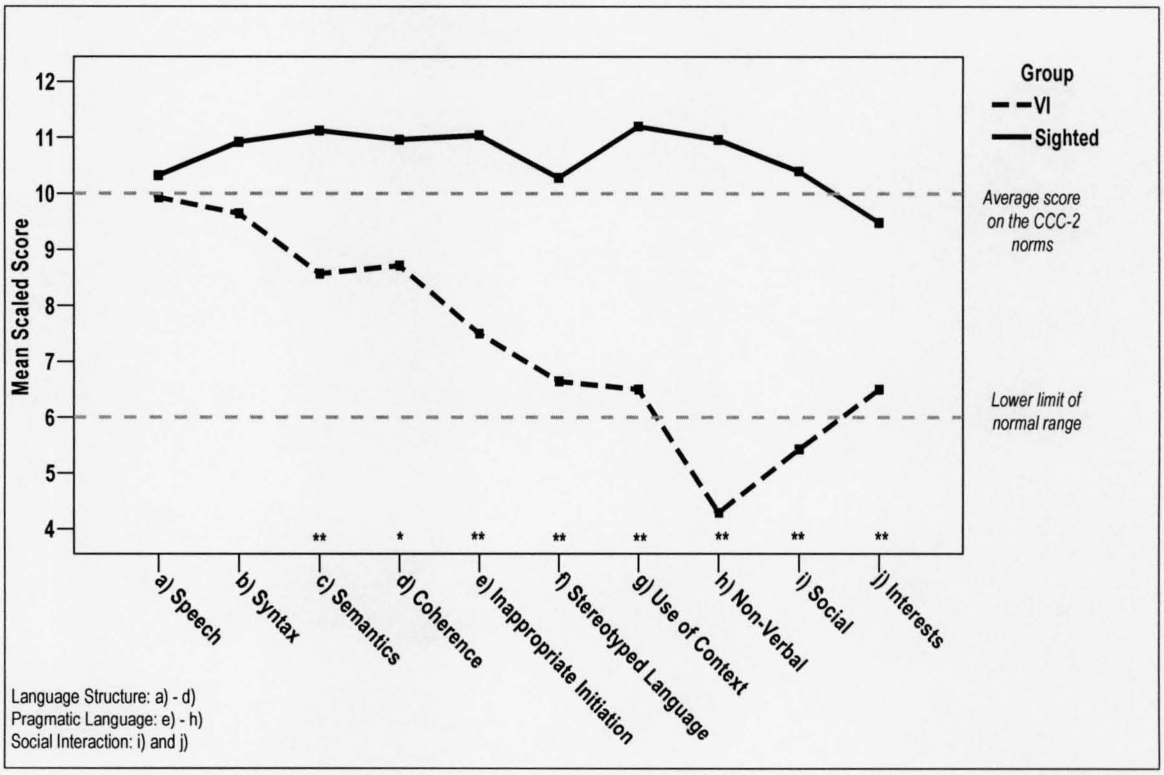

Figure 3.4: *Differing behavioural profiles between the two groups across the individual CCC-2 scales (** p ≤ .01; * p ≤ .05)*

Figure 3.4 graphically illustrates divergent CCC-2 profiles in the VI and Sighted groups. Here, in line with the significant deviation from profile parallelism, it is graphically evident that the pattern of parental rating for Sighted children's behaviours across the CCC-2 scales is relatively consistent across different scales, averaging at around the mean scaled score of 10 (top reference line) which is in line with the CCC-2 developmental norms (D. V. M. Bishop, 2003). In contrast, the profile of the VI group appears more uneven across the different scales. Despite the irregularity of their profile however, the mean scores of the VI group across the CCC-2 scales are largely within the normal range limits (i.e., scaled score $\geq$ 6) except for the Non-verbal and Social scales, where the mean scores of the VI group fall below normal limits (i.e., scaled score < 6).

In order to further explain the results of the Profile Analysis, post-hoc independent-samples *t* tests were conducted in order to examine the between-group difference on individual CCC-2 scales (Q4). With regards to the *structural language* aspects of the CCC-2, the *t* tests revealed that the children with VI were comparable to children who are Sighted with respect to their Speech ($t_{(37)}$ = - 0.401; *p* =.691, *d* = .13) and Syntax ability (equal variances not assumed - $t_{(16.99)}$ = -1.250; *p* =.228, *d* = .48). However, they were significantly poorer than their Sighted peers on Semantic ($t_{(37)}$ = - 2.717; *p* = .01) and Coherence ability ($t_{(37)}$ = - 2.404; *p* = .021, *d* = .80). With respect to the *pragmatic language* aspects of the CCC-2, the *t* tests showed significantly poorer performance of the VI group on all four scales tapping pragmatic language skills when compared to the Sighted group (Inappropriate Initiation: $t_{(37)}$ = - 3.838; $p \leq .001$; Stereotyped language: $t_{(37)}$ = - 3.18; $p \leq .003$; Use of Context: $t_{(37)}$ = - 5.105; $p \leq .001$; and Non-verbal: $t_{(37)}$ = - 7.49; $p \leq .001$). Similarly, with respect to the two CCC-2 scales tapping *social interaction*, the VI group performed significantly poorer than the Sighted group on both scales (Social: $t_{(37)}$ = - 5.17; $p \leq .001$; and Interests: $t_{(37)}$ = -3.15; *p* = .003). In addition, the children in the Sighted group obtained significantly higher General Communication Composite (GCC) scores on average, signifying their higher communicative competence, than the children in the VI group ($t_{(37)}$ = - 5.105; $p \leq .001$). Overall, these findings are in stark contrast to the VI group's superior language ability seen earlier with the CELF-3.

In the VI group, there appeared to be a notable disparity between children's scores on the scales assessing language structure, and their scores assessing pragmatic language skills. With a view to examine whether this apparent disparity in the VI group is statistically significant (H3), the scaled scores on the scales assessing structural and pragmatic language skills were summed in order to derive a Structural and Pragmatic Index for the VI and Sighted groups. A 2x2 mixed ANOVA was carried out to examine the difference between Structure and Pragmatics in each group. The results were as expected following from the Profile Analysis, with significant main effect of Language Index ($F_{(1, 37)}$ = 32.471, $p \leq .001$) and Group ($F_{(1, 37)}$ = 789.94, $p \leq .001$), qualified by a significant Language Index x Group Interaction ($F_{(1, 37)}$ = 34.261, $p \leq .001$). The result of interest concerned the within-group contrasts of Language Index. As the relatively flat profile suggested, there was no difference in structural and pragmatic language skills of the Sighted group ($t_{(24)}$ = -.125; *p* = .901, *d* = .023). However, in line with the experimental prediction

(H3), the structural language skills of the VI group were significantly better than their pragmatic language ($t_{(13)} = 7.716$; $p \leq .001$).

Qualitative examination of the CCC-2 profiles

As mentioned earlier, the VI and Sighted groups were found to be significantly different on the two scales assessing social interaction skills (Social and Interests). These two scales are used to calculate the Social Interaction Deviance Composite (SIDC), which can be considered in a screening context when identifying children with a potential socio-communicative impairment. A between-group comparison on the SIDC was not considered useful as this composite provides qualitative information about the pattern of impairment on an individual child's level and cannot be interpreted without the GCC. Instead the SIDC scores of each child have been examined qualitatively in relation to their GCC's in an attempt to subgroup the children from the two groups and identify the ones with potentially deviant socio-communicative profiles (H4).

Accordingly, Figure 3.5 shows the individual children's socio-communicative profiles, based on the relationship between their individual GCC and SIDC scores. Three reference lines plotted on the scatter-graph indicate the clinical cut-offs used to subgroup children with specific communication impairments corresponding to the broader autism phenotype. The green section of the scatter-plot marks the distribution of individual GCC/SIDC profiles that are considered to be typical (i.e., GCC > 55 and SIDC ≤ 15). The blue section of the scatter-plot marks a region of profiles where both the GCC (general communication) and the SIDC (social interaction skills) are considered to be below normal range (i.e., GCC < 55 and SIDC < 0), and such profiles are typical of a broader autism spectrum. Finally, the red section of the scatter-plot highlights the profiles of those children whose GCCs are within normal range (GCC > 50), but whose SIDC is considered to be deviant (SIDC < -15), and such profiles are frequently seen in Asperger Syndrome (AS).

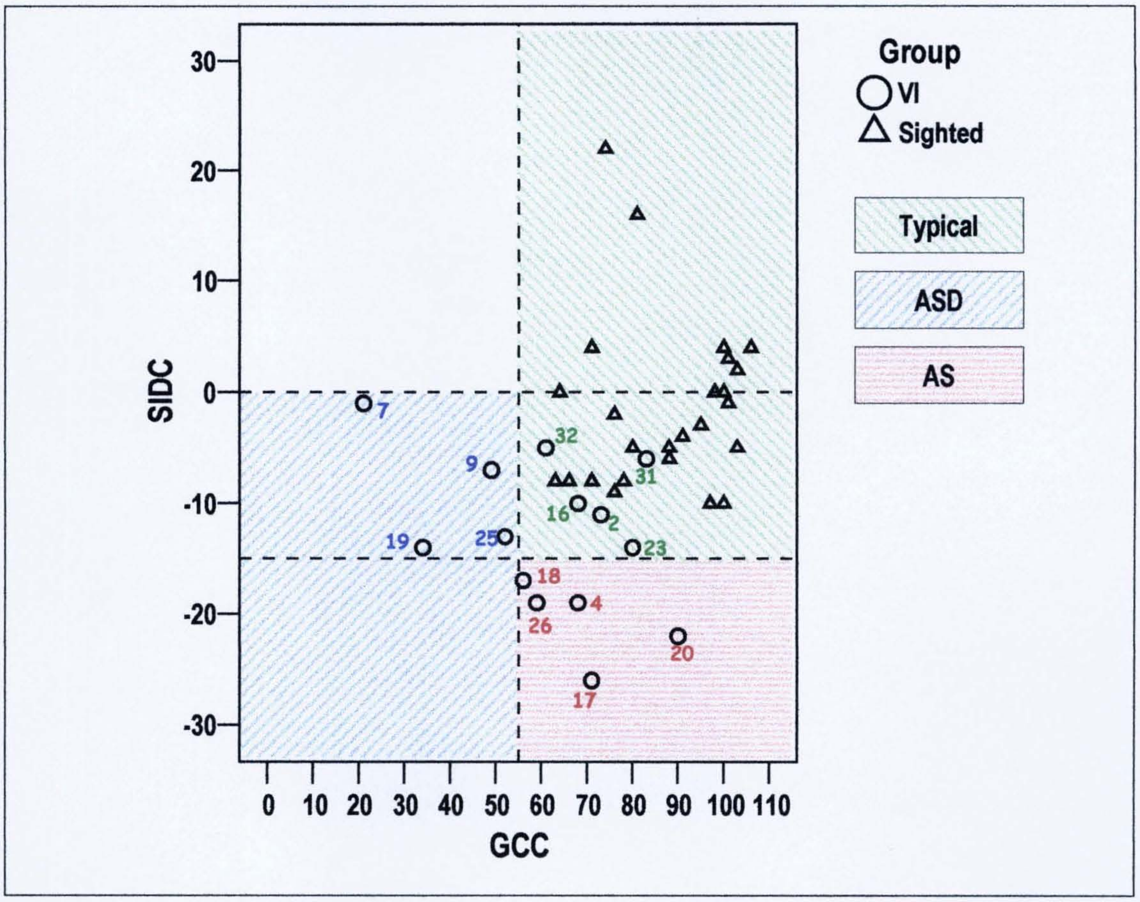

Figure 3.5: *Individual children's socio-communicative profiles*
(Note: participant ID numbers are given for the children with VI only)

Figure 3.5 illustrates that the socio-communicative profiles of all of the children in the sighted group were within a typical range. It is also important to highlight that the profiles of five children in the VI group were distributed within this section. However, it is crucial to emphasise further that four children with VI showed GCC/SIDC profiles that are typical of ASD and five that are associated with Asperger Syndrome. Overall, in support of H4, a substantial number (64 %) of the children with VI in this study (9/14, with the data of one child - Participant ID: 10 - missing) showed socio-communicative characteristics that are consistent with broader autism phenotype and which may warrant further clinical evaluation.

SCQ

To test H4 further, parental ratings of the children's socio-communicative behaviours on the SCQ were also examined. Table 3.8 summarizes the means and SDs for the prevalence of 'autistic-like' behaviours overall (SCQ total raw score) and across the three separate SCQ domains corresponding to the DSM-III criteria for autism diagnosis (Reciprocal Social Interaction Domain, Communication and Restricted, Repetitive and Stereotyped Patterns of Behaviour).

Table 3.8: *Means and SDs on the SCQ for the VI and Sighted groups*

Measure *Mean (SD)*	***VI***	***Sighted*** *missing N = 1*	***p value***
SCQ Lifetime Autoscore			
Total raw score	14.3 (3.9)	4.4 (3.9)	.001
Reciprocal Social Interaction Domain	4.1 (2.3)	0.9 (0.9)	.001
Communication Domain	5.3 (2.0)	2.1 (2.0)	.001
Restricted, Repetitive and Stereotyped Behaviours Domain	4.5 (2.0)	1.2 (2.0)	.001

In further support of H4, independent-samples *t* tests revealed a highly significant difference between the two groups on the SCQ total raw score, indicating higher prevalence of 'autism-like' socio communicative behaviours in the VI group ($t_{(38)} = 7.727$; $p \leq .001$). This difference between the two groups is also graphically illustrated in Figure 3.6. Further *t* tests revealed highly elevated scores in the VI group, relative to the sighted group, on all three individual SCQ domains that comprise the total SCQ score and form the basis for diagnosis of autism on ADI-R (Reciprocal Social Interaction: equal variances not assumed - $t_{(16.9)} = 5.306$; $p \leq .001$; Communication Domain: $t_{(38)} = 4.835$; $p \leq .001$; and Restricted, Repetitive and Stereotyped Patterns of Behaviour Domain: $t_{(38)} = 4.941$; $p \leq .001$) (see also Figure 3.7).

Figure 3.6 highlights the five children in the VI group (34% overall) whose scores may be of potential clinical concern (SCQ total raw score ≥ 15). It is worth noting that four of these children (i.e., Participant ID numbers: 7, 18, 25 and 26) were also within a domain for clinical concern of a potential communicative disorder on the CCC-2 (Figure 3.5). Additionally, a number of other children in the VI group (i.e., Participant ID numbers: 4, 17, 19 and 20) achieved overall SCQ scores that were just below the clinical cut-off of 15. Children obtaining such scores are frequently considered worthy of further clinical evaluation where there has been a raised concern of a potential autism spectrum disorder (Rutter et al., 2003). Thus, it is also worth noting that the profiles of three of these children fell within the red section of the CCC-2, signifying a concern of a socio-communicative disorder that is consistent with a broader autism presentation. Interestingly, one sighted child (Participant ID: 38) obtained an SCQ total score of potential clinical significance. However, the same child's socio-communicative profile on the CCC-2 was in the normal range and her parent did not report any concerns relating to her everyday behaviour or general development.

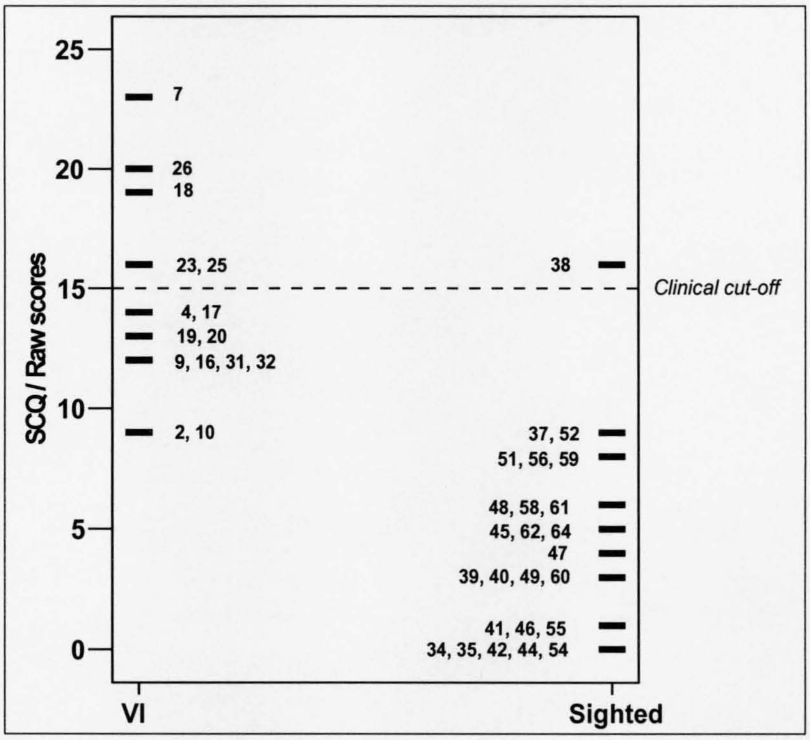

Figure 3.6: *Individual children's raw scores on the SCQ*

(Note: participant ID numbers for all the children are listed to clarify the overlapping data points)

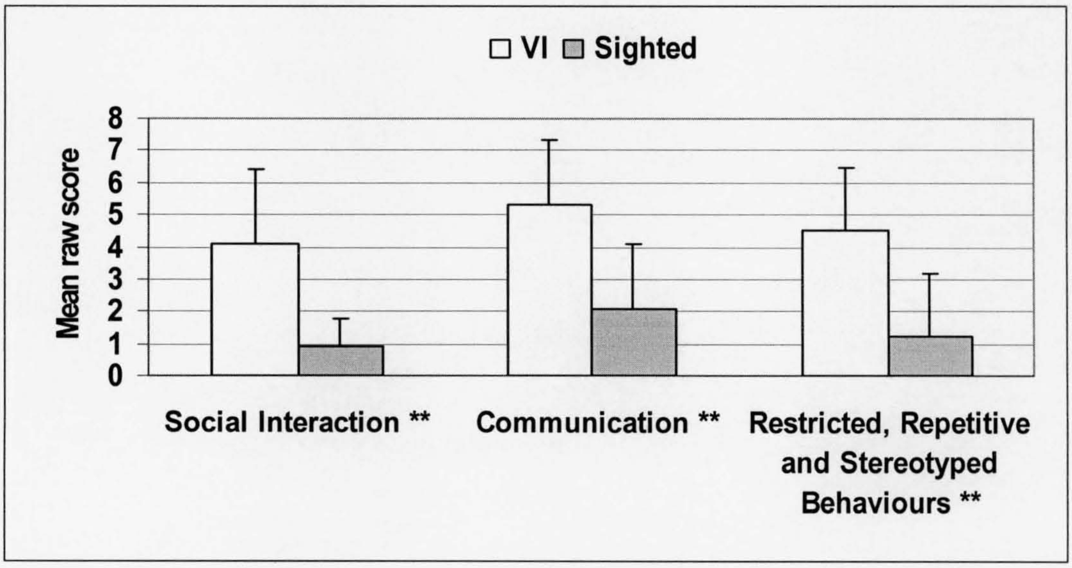

Figure 3.7: *Mean differences between the VI and Sighted groups across the individual SCQ domains (error bars represent the SDs, ** $p \leq .01$)*

Further data considerations

Association of the autistic-like profiles and severity of VI in the VI group

It is important to emphasise that clinically elevated scores on the CCC-2 and SCQ in the present study were seen in both the children with PVI and those with SVI. For a graphic illustration of this pattern of scores, the Figure 3.8 shows scores of individual children from the VI group on the CCC-2 and the SCQ, highlighting the degree of VI for each individual child. Individual ID codes appearing within the Venn circles indicate scores of clinical concern on the two parental checklists. For example, participant 23, who is a child with severe VI (SVI), had a clinically elevated score on the SCQ but not on the CCC-2. ID codes appearing outside the Venn circles indicate the profiles that fall within normal range. For instance, the profiles of participant 16, who is a child with profound VI (PVI), are in the normal range as this participant's scores fell outside the SCQ and CCC-2 domain for clinical concern. Chi square tests revealed that there was no significant association between the degree of VI in the early years (PVI and SVI) and the clinical cut-off for autism concern on the SCQ ($\chi^2_{(1)}$ = 1.250; p = .3) and the CCC-2 ($\chi^2_{(1)}$ = .837; p = .4) respectively. Therefore, the distinction between having and not having a degree of useful (i.e., form) vision in early childhood does not seem likely to explain the prevalence of autistic-like characteristics in this group of school-age children with congenital VI (Q5).

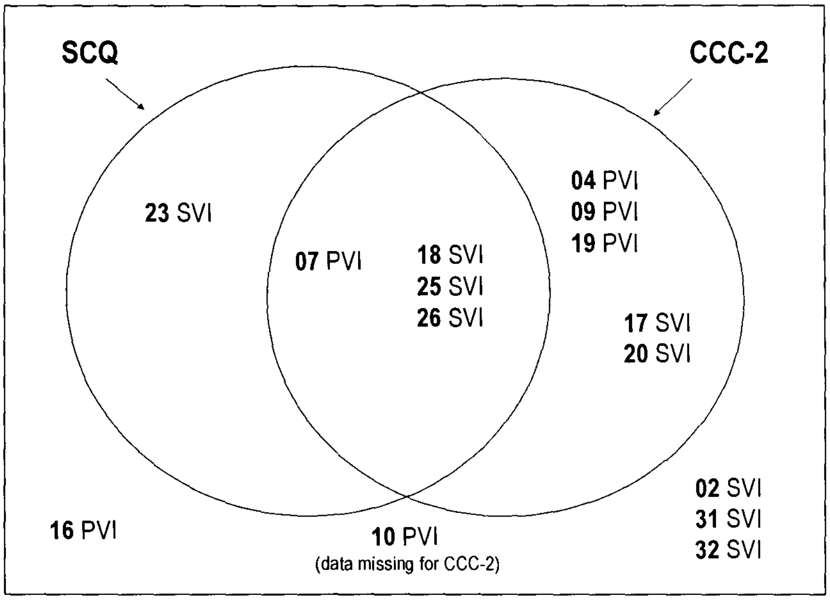

Figure 3.8: *Venn diagram illustrating the individual children with PVI and SVI with CCC-2 and/or SCQ profiles of potential clinical significance*

(Note: the participants outside the Venn circles are those who did not reach clinical cut-off criteria)

Further consideration of the autistic-like profiles in the VI group

Table 3.9: *Language and social communication scores of individual children with VI*

Child ID	*WISC – III Verbal IQ*	*CELF-3 Total Language*	*SCQ raw (atypical ≥ 15)*	*CCC-2 profile*
02	109	102	9	Typical
04	110	114	14	AS
07	100	91	23 (atypical)	ASD
09	101	95	12	ASD
10	99	108	9	Data missing
16	99	106	12	Typical
17	113	95	14	AS
18	128	126	19 (atypical)	AS
19	108	126	13	ASD
20	119	128	13	AS
23	100	97	16 (atypical)	Typical
25	113	121	16 (atypical)	ASD
26	95	120	20 (atypical)	AS
31	84	98	12	Typical
32	111	117	12	Typical

No significant correlations were found between the overall performance on the WISC-III and CELF-3, and socio-communicative behaviour rating on the SCQ and the CCC-2 (*p* values > .05, see Appendix A2). Thus, in it appears that the individual differences in verbal intelligence and language competence cannot explain the prevalence of autism-like socio-communicative profiles in the present sample of children with VI (Q5). Data examination at individual children's level (Table 3.9) did not throw any further light on this and it is difficult to get a clear grasp as to why some children showed atypical profiles while others did not. Moreover, it was not possible to ascertain what factors may have contributed to the better socio-communicative outcomes of those children with the lowest SCQ scores and normal range CCC-2 profiles (i.e., the participants shaded in grey in Table 3.9). None of these children's personal characteristics (e.g., age, gender, visual level or diagnosis), nor their VIQ and language characteristics, seemed to distinguish them from the rest of the VI group (see both Table 3.1 and Table 3.11). Interestingly however, even though these few children showed socio-communicative profiles that appeared within the normal range limits, it is questionable to what extent they can be considered 'comparable' to sighted children. For instance, even the two children with VI with some of the lowest (i.e., indicative of better outcome) SCQ scores in the VI group (Participant IDs: 02 and 10) were still in line with the 10 % of children who showed the highest prevalence of undesirable SCQ behaviours within the sighted group (Figure 3.6). Similarly, the four children with VI, whose CCC-2 profiles were considered to be in the typical domain, achieved scaled scores that were below the sighted group mean on Social, Non-Verbal and Context scales, and all but one on the Interests scale of the CCC-2. This has been given further attention in the discussion of this chapter.

Overlap between the CCC-2 and SCQ ratings

In line with H5, the pattern of specific socio-communicative weaknesses in the VI group appeared to be consistent across the CCC-2 and the SCQ, given the distribution of negative correlations between the two measures (Table 3.10). This pattern was similarly consistent also in the sighted group, suggesting that the CCC-2 and the SCQ are likely to tap similar underlying abilities. Most notably, the SCQ total scores correlated highly in both participant groups with the Social scale (VI: $r = -.653$, $p = .01$, n = 14; Sighted: $r = -.580$, $p = .002$, n = 25) and the Non-Verbal scale on the CCC-2 (VI: $r = -.631$, $p = .015$, n = 14; Sighted: $r = -.469$, $p = .018$, n = 25). Interestingly, both the scores of children with VI and sighted children on the restricted and stereotyped behaviour domain of the SCQ were significantly correlated with the CCC-2 scales assessing stereotyped language (VI: $r = -.703$, $p = .005$, n = 14; Sighted: $r = -.544$, $p = .005$, n = 25) and restricted interests (VI: $r = -.604$, $p = .022$, n = 14; Sighted: $r = -.396$, $p = .05$, n = 25). Therefore, based on the correlations between the two measures, it is clear that they are both sensitive to the same underlying socio-communicative strengths and weaknesses.

Table 3.10: *Pearson coefficients for correlations between SCQ and CCC-2*

	VI group - SCQ (raw score)				Sighted group - SCQ (raw score)			
	Total Score	Social Interaction	Communication	Repetitive, Restricted and Stereotyped Behaviours	Total Score	Social Interaction	Communication	Repetitive, Restricted and Stereotyped Behaviours
CCC-2								
Speech	-.589	-.331	-.084	-.610	-.021	.173	.058	-.132
Syntax	-.260	-.091	-.024	-.302	-.552**	-.371	-.275	-.409
Semantics	-.455	-.179	-.282	-.429	-.320	-.079	-.071	-.432
Coherence	-.335	.022	.017	-.653**	-.200	.088	-.188	-.139
Initiation	-.271	.055	.104	-.667**	-.454	-.086	-.408	-.292
Stereotype	-.278	.247	-.222	-.703**	-.461	-.192	-.158	-.544**
Context	-.328	-.111	-.117	-.405	-.297	.055	-.297	-.170
Non-verbal	-.631	-.413	-.051	-.585	-.469	-.051	-.216	-.542**
Social	-.653**	-.490	-.258	-.369	-.580**	-.490**	-.447	-.240
Interests	-.436	-.094	-.158	-.604	-.472	-.234	-.301	-.396
GCC	-.517	-.117	-.111	-.731**	-.519**	-.075	-.290	-.508**

** - significant at $p \leq .01$

DISCUSSION

Three main findings emerged from the research presented thus far. Firstly, despite being comparable on age and verbal intelligence, children with VI performed significantly better than developmentally matched sighted children on a standardised test of language function. Secondly, and in contrast to the first finding, the children with VI showed a significantly poorer range of skills than their sighted peers in terms of their language functioning in an everyday context (based on parental reports), with a particular weakness in use of language for pragmatic and social purpose. Thirdly, judging by the parental reports of children's everyday socio-communicative behaviours, a large proportion of children with VI showed a level of behavioural difficulties that is consistent with a broader autism phenotype in sighted children and may be of a potential clinical concern.

As discussed previously in the introduction of this chapter and also in Chapter 1, language has been generally seen as a strong point of the development of children with VI. However, what is especially remarkable about the present findings, unlike those of previous research, is that they differentiated children with VI as better than their sighted peers. These findings carry particular significance in light of the early language irregularities and delays in language development of children with VI reported by previous research (Andersen et al., 1984; Dunlea, 1989; McConachie & Moore, 1994; Reynell, 1978), and bear important implications for language-based interventions. Crucially however, how can we explain the language strength that children with VI have demonstrated in this study?

In line with Pérez-Pereira and Conti-Ramsden (1999), the language domain may be more salient to children with VI than children who are sighted and is therefore likely to serve a different function in the two groups. Importantly, the CELF-3 was shown in this study to be a successful tool in separating this language function from general intelligence, allowing us to illuminate the strength of children with VI that may be specific to their dominant domain. Being a test of language ability, a child's performance on the CELF-3 is likely to be related to their verbal IQ as assessed by the WISC-III (Semel, Wiig, & Secord, 2000b), a pattern which is also supported by the significant correlations between the two tests in the present research. However, unlike the majority of verbal WISC-III subtests, which essentially measure reasoning capacity through verbal means, the CELF-3 is less comprehension based, largely tapping language elements such as content and structure, which can be 'scrutinized independently and out of context' (Semel et al., 2000b, p.2). This potential of the CELF-3 to isolate language-specific strengths and difficulties in children has also been demonstrated in research with other clinical groups of children, such as those with specific language impairments (SLI) and those with autism (Lloyd, Paintin, & Botting, 2006; Nash, 2008).

However, we must remind ourselves that the CELF-3 assessment in the current study was based only on four out of required six subtests (two of which were supplementary) due to the visual content of most of the CELF-3 stimuli. For that reason the language profiles provided by the CELF-3 in this study are incomplete, so the composite language scores obtained in the present research need to be treated with some caution. For example, this incomplete CELF-3 assessment may explain why there was a significant discrepancy between the CELF-3 Total Language and the VIQs in the sighted group, who did not performed on the CELF-3 as it would be expected based on their verbal IQ. However, given that the CELF-3 assessment in this study was not presented in its required format, it is unlikely that the difference between the two groups of children was solely the effect of the sighted group.[11] In fact, the mean and standard deviation of the sighted group was still in line with the CELF-3 normative data which has a mean of 100 and standard deviation of 15. On the other hand, the CELF-3 Total Language scores in the VI group were higher than their VIQ score, even though this within-group difference was not statistically significant. Hence, it is possible that the CELF-3 subtests that were used here were especially favourable to children with VI, an advantage which was particularly obvious on the Recalling Sentences subtests.

Even though the CELF-3 subtests generally all have a strong memory component (Semel et al., 2000b), the Recalling Sentences subtest is particularly verbal short-term memory (STM) based. It is plausible to suggest that, given adequate intelligence, verbal STM may play a particularly important role for children with VI. The VI group's superior performance on Recalling Sentences accentuates their STM advantage, which traditionally has been demonstrated using Digit Span subtest from the WISC (Hull & Mason, 1995; Smits & Mommers, 1976; Tillman & Bashaw, 1968; Tillman & Osborne, 1969). The Digit Span superiority of children with VI has not been replicated in the present research, although direction of the means suggested this pattern. It is likely that Recalling Sentences places slightly different demands on the child than does the Digit Span, in that the words to be recalled need to be recognised within a language context (i.e., syntax and semantics). For this reason, Recalling Sentences may better capture a language-specific STM advantage than would a traditional Digit Span test.

Importantly, STM advantage may not only be obvious at the level of STM tests, but may also feed into all the other skills required for successful language function.[12] Hence, for a child with ample vocabulary, grammar and semantics, good verbal STM may especially boost an overall language outcome. This may explain why the overall CELF-3 performance (i.e., composite scores) of the VI group was more successful than for the sighted children, even though at the level of an individual subtest, only Recalling Sentences had the power to differentiate the two groups (i.e., Word

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[11] For instance, within the sighted group the lowest scores were obtained on the Listening to Paragraphs subtest from the Receptive Language domain. However this is a supplementary subtest, which was used here as a replacement for the required subtest that involves visual stimuli.

[12] In relation to this, it is important to note that the Recalling Sentences subtest has been demonstrated in previous research as a highly sensitive measure for discriminating between children with and without language impairment, including those with SLI (Botting & Conti-Ramsden, 2003; Conti-Ramsden, Botting, & Faragher, 2001).

Classes and Listening to Paragraphs were trends). Interestingly, STM strength may also provide an explanation for certain characteristics of the language of children with VI that are considered to be a disadvantage, such as modelled and imitative speech. Some researchers have argued that imitative speech, to an extent, may serve a purposeful, self-scaffolding function for children with VI (e.g., Pérez-Pereira, 1994), although this has been disputed by others (Norgate et al., 1998). Nevertheless, imitative speech is likely to be driven by STM (i.e., rehearsal). While this implicit rehearsal-like strategy may be beneficial in a STM-based context like Recalling Sentences for children with VI, the demands of such a task may be more challenging for sighted children. Sighted children may get distracted by the semantic context of the task and they may attempt to chunk meaningfully the semantic content of the sentence as a remembering strategy. Thus, future studies may need to consider the types of errors that sighted children make within such tasks, in order to unpack the processes that may underlie their poorer performance, in comparison to children with VI.

Furthermore, it is likely that the observed language competence in children with VI is dependent on the context within which it is assessed. For instance, while the children with VI performed better than sighted children on the CELF-3, their parental ratings of specific structural language skills revealed a potentially contrasting picture. More specifically, the VI group's Semantics and Coherence, as assessed by the CCC-2, although largely within normal range limits, were significantly lower than those of sighted children. It is possible that the children with VI benefited from the context of a traditional one-to-one assessment in which both the WISC-III and the CELF-3 are carried out. Such a context is structured and therefore rigid and scripted. This context, in addition to the provision of clear instructions by the assessor, may provide a useful scaffolding tool for achieving successful performance in a child with VI and may better capture the strength that is not necessarily apparent in an everyday conversation with such a child. On the other hand, an everyday context (within which language is generally used) is less rigid, largely spontaneous, and inherently social. For this reason, parental reports may be more likely to reveal a VI related disadvantage in Coherence and Semantics than would be evident in the context of a structured assessment. However, more rudimentary language elements, such as Speech and Syntax, may be less susceptible to contextual influences than are Semantics and Coherence, explaining why the two groups did not differ on these two CCC-2 components.

A particularly striking language weakness of children with VI that was brought into spotlight by the parental reports in the present study was the children's use of language for social and communicative purposes. More specifically, despite scores that generally appear to be within the normal range limits, the children with VI were found to be significantly poorer that their sighted peers at pragmatic language skills as measured by the CCC-2 (Inappropriate Initiation, Stereotyped Language, Use of Context and Non-Verbal Communication). Concerns about poor pragmatic skills in children with congenital VI have been raised previously in research studies looking primarily at young preschool children (Dunlea, 1989; Fraiberg, 1977; Preisler, 1991; Urwin, 1978), although research attempts to address this issue systematically have been methodologically limited (e.g., James & Stojanovik, 2007). The current research therefore provides unique evidence that pragmatic language concerns in children with VI are present at school age and in those children who are cognitively able, highlighting the non-verbal aspects of pragmatics as a particular challenge.

It could be argued that the parental ratings on the CCC-2 were negatively biased towards the children with VI, as this questionnaire has not been developed with such children in mind and is therefore less sensitive to their strengths. However, this is possibly a circular argument, because pragmatic language skills are likely to be particularly vision-driven. For instance, the non-verbal aspects of pragmatics, such as use of facial expressions and gestures, may be the most potent communicative tools in maintaining a conversational partner's focus of attention. Similarly, the ability to initiate conversations appropriately, understand irony and sarcasm, and adjust conversational topics based on others' levels of interest may be easier to achieve through monitoring of the conversational partner's facial expressions and bodily gestures. Hence, considering the use of visual behaviours for pragmatic purpose, it is not surprising that these skills in children with VI are not as good as those of their sighted peers. Interestingly, the visual nature of pragmatics may be the reason for why pragmatic language my not benefit from scaffolding in the same way as structural language does (as discussed previously). More specifically, pragmatic language is possibly more dependent on successful development of joint attention in early childhood than are structural language skills, which seem to develop without much difficulty in children with VI. On the other hand, early joint attention is a recognised area of developmental vulnerability in such children, and poor communicative pragmatics of verbally able children with VI at school age may be a consequence of this vulnerability. This is certainly in line with the developmental patterns observed in autism, as even high-functioning children with autism who show better language outcomes show poor use of language for pragmatic and social purpose (Dennis et al., 2001; Klin, 2000).

Following from this, it may not be surprising that the VI group obtained notably impoverished ratings on the CCC-2 scales targeting autism-related social difficulties (Social Relations and

Interests). Furthermore, the overall CCC-2 profiles of children with VI were significantly uneven in comparison to the typically flat profiles of the sighted group, and revealed a disproportional weakness of social and pragmatic skills relative to the presentation of structural language skills. This pattern is largely reminiscent of the presentation of autism in sighted children and is accentuated by the fact that a substantial proportion of the children with VI reached or exceeded the clinical cut-off for autism concern on both the CCC-2 and the SCQ.

These findings support the seminal research by Hobson and colleagues who were amongst the first to raise a concern about the alarming prevalence of autism-like features in children with congenital VI (R. Brown et al., 1997; Hobson et al., 1999). In this previous research, the autism-like clinical picture was found across a cognitively heterogeneous population of children with congenital and total sight loss, although it was significantly more prominent in those children who had learning difficulties. Crucially, the present research provides an important additional insight in that autism-like socio-communicative impairments have been observed in an intellectually homogeneous group of children VI with advanced language skills, some of whom have above average verbal IQs (e.g., Participant ID: 18, VIQ = 128; Participant ID: 25, VIQ = 113). In relation to this, it can be argued that the behavioural characteristics observed in the present VI sample overall bear strong resemblance to the presentation of Asperger Syndrome, which is a form of high-functioning autism that is marked by a good language outcome (Wing, 1981). Interestingly, sighted children with an Asperger diagnosis may provide a useful comparison group in further research, to help illuminate some of the subtleties that underlie autistic-like presentation in verbally proficient children with congenital VI.

Furthermore, while the previous research investigated autistic-like characteristics exclusively in children with total sight loss (i.e., PVI) (R. Brown et al., 1997; Hobson & Bishop, 2003; James & Stojanovik, 2007), the present research provides unique evidence that such characteristics are also prevalent amongst the children with some limited levels of functional although severely degraded vision in their early years (SVI). It is worth noting that some previous studies with infants and young children with congenital VI demonstrated poorer developmental outcomes in a range of areas in children with PVI compared to those with SVI. However, these studies were of an epidemiological nature and included a more representative sample of children with VI, including children with low IQ, the prevalence of which is higher in the PVI group (Cass et al., 1994; Dale & Sonksen, 2002). The present results suggest that it is unlikely that the distinction between children with total sight loss and those with some limited form perception in early childhood can explain the autistic-like features in the current sample of verbally-able children in their primary school years. With this in mind, the reader should be reminded that all of the children with VI in this study had a very significant vision loss and the severely degraded form vision in the children in SVI is still likely to interfere with the processes that are particularly at risk from reduced vision.

In relation to this, it is important to reiterate the previous point about the developmental significance of early joint attention. Vision loss may impose seriously limited opportunity to engage in that very special form of relatedness, affective sharing and perspective taking that the context of joint attention provides to the child and their interactional partner, creating a developmental vulnerability with possible long-term consequences for a child with VI (Hobson, 1990, 1993, 2002). Hence, the presentation of a broader autism phenotype in later childhood in children with VI is potentially a consequence of this early vulnerability rather than presentation of core autism per se. Similarly, this vulnerability may account also for why even those children with VI whose socio-communicative profiles were within normal range limits did not reach the levels of social and communicative competence that is typical of the majority of sighted children. However, it still remains unclear why some children with VI, despite their good language and verbal intelligence, present with autistic-like behaviours more strongly than others, and to the extent that may warrant further clinical evaluation. Similarly, individual characteristics of those children whose socio-communicative profiles scores are in the normal range do not provide any clues with respect to the potential factors that may contribute to their seemingly better socio-communicative outcomes. The reasons for this possibly extend beyond linguistic competence and, the research presented in the following chapters of this thesis may help to clarify some of these issues.

Finally, we must remind ourselves that the measures of socio-communicative competence used in this study were based on parental reports. The subjective nature of parental reports may not only underestimate children's behavioural difficulties that may require professional attention; it may also exaggerate the severity of a child's behaviours that parents may find particularly difficult to deal with. This may provide an explanation for why some children with VI in this study presented with more prominent socio-communicative difficulties than others. Thus, utilizing more structured measures and direct clinical assessments may be a useful addition to the parental questionnaires in future research. Nevertheless, the existing clinical measures that target autism-related socio-communicative problems may not be as revealing on their own, as they are not developed with children with VI in mind, and are likely to be difficult to adapt to their needs (i.e., they are largely dependent on the utility of visual stimuli)[13]. This further emphasises the lack of measures that are suitable for use with children with VI and which are sensitive to their developmental strengths. With this in mind, parents are a valuable source of knowledge about their children and are likely to provide a window into their children's characteristics that may not be easy to evaluate otherwise. Therefore, in the context of the present research, we can conclude that parental reports provided a valuable insight into the everyday socio-communicative competencies and difficulties of their children with congenital VI. From these reports we have learned that there is a wide spectrum of socio-communicative difficulties in cognitively able children with congenital VI of varying severity. However, while these difficulties (particularly in some children with VI) may resemble socio-communicative difficulties in sighted children with autism, we cannot generalise from these findings that there is a distinction between visually impaired children with and without autistic features. More accurately, these findings can be seen as providing useful baseline norms for cognitively able children with VI, from which future diagnostic criteria for autism (or autism-like socio-communicative disorder) in such children may potentially be developed.

[13] For instance, the Autism Diagnostic Observational Schedule (ADOS) (Lord, Rutter, DiLavore, & Risi, 1999).

Chapter 4

Understanding Mental States as Causes of Emotions in Children with Congenital VI

SUMMARY

The aim of the research presented in this chapter was to investigate social cognition in the same children with congenital VI who took part in the research presented in Chapter 3. Here, the children's theory of mind capacity was examined in a way that differs from the standard false-belief task, with an emphasis on the children's spontaneous use of mental states terms when explaining the emotions of story characters. Considering the VI group's socio-pragmatic difficulties that were highlighted in the preceding chapter, it was of interest to examine whether such difficulties would also transpire in their mentalistic language use. The children with VI were compared to age and VIQ matched sighted children on the extent to which they spontaneously referred to mental states rather than to situational factors as the causes of emotions. The results showed that the two groups did not differ in the frequency with which they used mental state language to explain basic emotions, and that their use of mental state terms (both for beliefs and desires) was as frequent as was their use of situational terms in their explanation of both typical and atypical emotional scenarios. These results are discussed in relation to i) methodological limitations of the task used in the current study, and ii) in comparison to the previous research evidence that highlighted social cognition as an area of vulnerability for children with VI.

INTRODUCTION

Numerous studies are in agreement that children with congenital VI are delayed in developing theory of mind understanding compared to sighted children of the same developmental level (McAlpine & Moore, 1995; Minter et al., 1998; Peterson et al., 2000; Pring et al., 1998; Roch-Levecq, 2006). As discussed in the previous chapters, theory of mind delay in children with VI is consistent with contemporary theories of social cognition that emphasise the importance of visual precursors to theory of mind. To reiterate, in typically developing sighted children, learning about subjective mental states is thought to be facilitated by the experiences of visually-driven shared attention (Baron-Cohen, 1995b), visual imitation (Meltzoff & Gopnik, 1993), and affectively charged triadic interactions (Hobson, 1993), which take place in early childhood and precede the development of theory of mind. On the other hand, children who are visually impaired are denied the chance to benefit from the rich cues provided within this visually-driven social context of imitation and affective sharing of attention (e.g. eye-gaze directing, gestures and facial expressions), leading to an impoverished framework for learning about the mental states of others, thus resulting in a delayed theory of mind (Hobson, 2002).

Methodologically, the majority of the aforementioned studies, which demonstrated a theory of mind delay in children with congenital VI, used variations of the false-belief task as a measure of theory of mind understanding. There is general consensus among researchers that in typical development, mastery of false-belief tasks is acquired around the age of four years (Ruffman, 2004; Wellman, Cross, & Watson, 2001; Wimmer & Perner, 1983). However, children with congenital VI have repeatedly been found to have difficulties with false-belief tasks at chronological and verbal mental ages older than four years (Green et al., 2004; McAlpine & Moore, 1995; Minter et al., 1998; Peterson et al., 2000; Roch-Levecq, 2006).

In most theory of mind studies that employ a false-belief paradigm children are asked explicitly to predict a story character's mental or emotional state. However, it has been suggested that theory of mind development should be studied in relation to real world consequences (Astington, 2001; Rieffe & Meerum Terwogt, 2000). For instance, even when children demonstrate that they can make an adequate false-belief prediction it is still uncertain whether they can use this ability spontaneously and whether they can formulate an explanation for the cognitive or emotional states they observe in other people. Related to this, it has been argued that the use of the language representing mental states (i.e., thoughts, desires and emotions) is fundamental to social cognition and may be an important indicator of a child's theory of mind (Baron-Cohen, 2000; de Rosnay & Hughes, 2006; Symons, 2004). In fact, it has been suggested that children's discourse about inner mental states in real life precedes their mastery of false-belief tasks (Bartsch & Wellman, 1995). To the contrary, children with autism, for whom the theory of mind deficit is seen as a defining characteristic (Baron-Cohen, 1995b; Happé, 1995), have been found to produce fewer words representing mental states when describing pictures involving action and deception and in their spontaneous conversational discourse compared to their typically developing peers (Baron-Cohen et al., 1985; Dennis et al., 2001; Tager- Flusberg, 1992).

However, little is known about the extent to which children with congenital VI spontaneously refer to mental states (particularly those of other people) in their everyday language use. The only investigation to date that has addressed this specific aspect of social cognition in children with congenital VI is the study by Pring et al. (1998), that used the 'Strange Stories' task (designed by Happé, 1994) for this purpose. In this study, sixteen children with congenital VI and no other impairments, with IQs reported to be in the average range, were compared to a group of chronological age matched sighted controls on the frequency of mental state justifications when explaining the story characters' behaviours. There were twenty four stories tapping sophisticated theory of mind and involving a range of advanced mental state elements such as sarcasm, misunderstanding, persuasion, pretence and deceit. In their study, Pring et al. found that the children with congenital VI produced significantly fewer mental state justifications for the story protagonists' behaviours than did the sighted children overall, concluding that that there is a difference between children with VI and sighted children in understanding the uses of language that requires insight into the beliefs and desires of others. The study further highlighted that this subtle difference in the way social understanding manifests in children who are visually impaired, compared to children who are sighted, is seen at an age (i.e., 9 - 12) by which it had previously been assumed that earlier developmental problems would have been overcome (Hobson, 1993). Finally, the authors reported a significant relationship between the children's general intellectual levels and the frequency of their mental state justifications. This supported the previous research, which suggested that children with VI who are intellectually more able may also be more able to compensate for difficulties in social cognition than children with lower intellectual levels (R. Brown et al., 1997; Green et al., 2004). For this reason, however, as the children in this study were not matched on cognitive ability, it remains unclear whether the difference between the VI and the sighted children was confined to the lower ability group.

Following Pring et al. (1998), the purpose of the current research was to investigate social cognition in children with congenital VI by focussing on children's spontaneous use of conversational means as indicators of their understanding of mental states. Unlike Pring et al.'s study, the aim of the current investigation was to compare children with VI to sighted controls matched on verbal ability (VIQ) as well as on chronological age. Furthermore, whilst Pring et al. noted the children's mental state language use in terms of justifications for the story characters' behaviours, in the current research the focus was on the children's insight into the story characters' mental states as causes of their emotions.

The design for the current investigation was built upon the series of experiments carried out by Rieffe and colleagues with children who are typically developing (Rieffe, Meerum Terwogt, & Cowan, 2005), children with autism (Rieffe, Meerum Terwogt, & Stockmann, 2000) and children with hearing impairment (Rieffe & Meerum Terwogt, 2000). More specifically, Rieffe et al. designed a task which involves presenting six stories, initially requiring children to predict emotional states of the story characters and subsequently to provide subjective explanations for the factors that caused them. The authors argued that understanding the causes of emotions is more sophisticated than just recognizing and reacting to them as it places the emotional experience in a socio-cultural context. In this context, emotions can also be a consequence of a person's interpretation of a situation and not just a mechanical product of the situation itself, the understanding of which requires theory of mind (Rieffe et al., 2005, p. 260).

Despite difficulties with theory of mind, it has been reported that children with congenital VI have an understanding of cause-effect relationships that evoke basic emotions (i.e., happiness, sadness, fear and anger). More specifically, it has been found that children with VI are as able as sighted children to identify such emotions as they occur typically in specific situations, from their own perspective (e.g., *How do you feel when you receive a new gift?*) (Roch-Levecq, 2006) and from the perspective of others (e.g., *Susan is given a new bicycle for her birthday? What will Susan feel?*) (Dyck et al., 2004). Moreover, Dyck et al. (2004) reported that, when asked explicitly to explain the meaning of emotions (e.g., W*hat does the word 'angry' mean?*), the semantic knowledge of children with VI even exceeded the knowledge of the sighted controls. However, both studies found that, in the task which required children to represent mental states more implicitly, children with VI were not as proficient. For example, the false-belief task performance of children in Roch-Levecq's study was significantly poorer than the performance of sighted controls (Roch-Levecq, 2006). Similarly, whilst able to explain the meaning of basic emotions, the children with VI studied by Dyck et al. (2004) were less able than their sighted peers at recognising vocal intonations specific to different categories of emotion. It is worth noting that a similar difficulty with recognising vocally expressed emotions in children with congenital VI has been reported by others (Minter et al., 1991). Thus, in tandem with these studies, it is expected that the children with VI in the current study will be able to predict the four basic emotions (i.e., happiness, sadness, fear and anger) without much difficulty. However, it is their explanations of the causes of emotions that may differentiate the children with VI from sighted children, as this process is more likely to call upon a child's theory of mind (Rieffe et al., 2005).

The paradigm designed by Rieffe and colleagues, which was adopted for the current study, also included another important element. They suggested that, within a socio-cultural context, within which emotional states occur, it is important to consider how typical certain emotions are. For example, a typical emotional reaction to receiving a present would be happiness and excitement, whereas an atypical reaction would be anger or sadness. By the age of five children understand the causal link between these typical emotions and subjective desires in that they are able to accurately predict a happy emotion when the protagonist receives what s/he desires and an unhappy emotion when the protagonist is frustrated in fulfilling his/her desire (Wellman, 1990). It has been argued that, when explaining emotions, typical emotional scenarios may not necessarily require a mental state inference and can easily be explained in terms of situational factors, which tend to be self-evident and usually represent a shared view of the world (Gnepp, 1983; Rieffe et al., 2005). For example, children can easily attribute a story protagonist's happiness at receiving a present to the possibility that it is the protagonist's birthday. However, such situational explanations may not be sufficient in the case of an unexpected emotional reaction, such as anger at receiving a present. In order to provide a justification for the strangeness (i.e., atypicality) of this situation, insight into the protagonist's mental state may be required (e.g., '*he is angry because he thinks it will be something he doesn't like*'). Therefore, if children have a theory of mind, atypical emotional scenarios should most strongly encourage them to make references to story protagonist's mental states (Gnepp, 1983; Rieffe et al., 2005).

Rieffe et al. (2000) found that the atypical emotion scenarios did in fact induce more mental state language in both typically developing children and children on the autism spectrum. However, whilst typical emotions differentiated children with autism from typically developing children, the atypical emotion did not. More specifically, when explaining typical emotion scenarios the autistic group referred to fewer desire and belief terms as causes of story protagonist's emotions than even a younger control group. However, when explaining atypical emotions, the autism group referred to mental states as frequently as did the typical controls of the same age. Based on the children's performance on this task, the authors argued that children with autism may have an insight into the mental states of others. However, they do not use this capacity in every-day circumstances and in the same way as do typically developing children. This is consistent with other findings that some high functioning individuals with autism who pass false-belief tasks still show poorer levels of social adaptation in everyday context (Klin, 2000).

In the current study it was of interest to examine whether children with congenital VI would perform like the children with autism studied by Rieffe et al. (2000). Given the theory of mind delay and the reduced propensity for mental state language reported previously in children with VI (Green et al., 2004; Pring et al., 1998), it would not be unexpected to find a difference between the children with VI and their sighted counterparts in the frequency with which they use mental state attributions rather than situational attributions in their explanations of emotions. Moreover, in keeping with Rieffe et al. (2000), while atypical emotions are more likely to induce mental state inferences in both groups, the typical emotion scenarios may be more likely to differentiate children with VI from children who are sighted.

Finally, it was of interest to investigate whether the two groups would differ in the extent to which they referred to different categories of mental states. Previous research has established that in the course of theory of mind development there is a clear developmental progression in the understanding of various mental state concepts (Wellman & Liu, 2004). More, specifically it has been found that children's understanding of desires precedes their understanding of beliefs and furthermore, that the causal relationship between desire and emotion is understood before the causal relationship between belief and emotion (Bartsch & Wellman, 1995; Harris, 1989; Wellman & Liu, 2004). One explanation for this is that separate modules, dedicated to each distinctive mental state concept, become available at different points in development (Fodor, 1992; B. J. Scholl & Leslie, 1999). Another explanation is that, in order to understand belief, children need to develop a concept of mental representation, which might not be necessary for understanding desire (Bartsch & Wellman, 1995). The theories of developmental lag between the understanding of desire and belief do not fall withing the scope of this thesis and will not be discussed in further detail here (see Flavell, 1999; Harris, 1996 for the reviews of literature). Importantly, there seems to be general agreement amongst researchers that there is a clear developmental sequence by which the understanding of different mental state concepts occurs. Thus, a similar pattern of understanding of desire before understanding of belief should also be observable in children with congenital VI. However, following the theory of mind delay hypothesis for children who are visually impaired, it is of interest to examine this pattern in comparison to children who are sighted. Finally, as children in general are more likely to make mental state inferences overall at an older age (Harris, 1989), an overall developmental trend in the frequency of references to mental states should also be observable in the two groups of children.

Research questions and hypotheses

In summary, in order to assess the frequency of mental state language use as an indicator of theory of mind in children with congenital VI, the current research used the paradigm designed by Rieffe et al. (Rieffe & Meerum Terwogt, 2000; Rieffe et al., 2005; Rieffe et al., 2000) with an aim to address the following questions:

Question 1 (Q1): Do children with congenital VI differ from sighted children of the same age and verbal ability in the extent to which they refer to mental states rather than situational factors in their explanations of emotions of story characters?

Question 2 (Q2): Does the extent to which the two groups may differ in their spontaneous use of mental state terms depend on whether the emotional scenarios are typical or atypical?

Question 3 (Q3): Does the extent to which the two groups may differ in their use of mental state terms depend on the specific type of mental state (i.e., desire terms relative to belief terms)?

Question 4 (Q4): Can severity of VI, chronological age and verbal IQ explain the variation in the extent to which children with VI refer to mental states?

METHOD

Participants

The participants were the same children described in Chapter 3. There were 15 children with congenital VI of differing degrees (i.e., severe and profound) and 26 children with normally developed vision, matched on verbal IQ, chronological age and gender. Average VIQ across the two groups was 106.2 (SD = 10.8) and the mean chronological age was 8 years and 3 months (SD = 1.7, range = 6 years and 3 months - 12 years 11 months) (see also Table 3.2). In both groups there were marginally more girls than boys (VI group gender ratio: 9/6; Sighted group gender ratio: 14/12).

Design

The study used a mixed 2 (Group) x 2 (Emotion typicality) factorial design. The first Independent Variable (IV) Group was between-subjects with two levels: VI and Sighted. The second IV, Emotion typicality, was within-subjects, and the two conditions were Typical and Atypical. There were two Dependent Variables (DVs). The first was frequency of mental state explanations, relative to situational explanations, when explaining the emotions of story protagonists. The second DV focused upon mental state explanations only and was measured in terms of the frequency of desire explanations, relative to belief explanations, when explaining the emotions of story protagonists.

Materials

The Emotion task was adopted from Rieffe and colleagues (Rieffe & Meerum Terwogt, 2000; Rieffe et al., 2005; Rieffe et al., 2000). It consisted of six short stories that described emotion eliciting situations (Table 4.1 and Appendix B1). The stories were designed to elicit typical emotions, with two stories provoking a positive emotion (e.g., happy, excited, surprised), two stories provoking sadness or anger and two stories provoking fear (Table 4.1, Typical Emotion column).

All but one story (*Story 5*) were the same as those used previously by Reiffe et al. In the original stimuli *Story 5* depicts a girl who *sees* a person in a dark living room who she cannot identify. The visual aspect of the story was modified as it was felt that this would be more suitable for the experiences of the children with VI (i.e., a girl hears a person walking, in what she thinks is an empty house).

Anger and sadness are placed in one emotion cluster. Rieffe et al. argued that they are both seen as plausible reactions to one and the same scenario because a situation can arouse either depending on whether the person concentrates on the cause of the negative outcome or on the negative outcome itself.

Table 4.1: *Content of the six stories with typical and atypical emotions*

Story	*Story content*	Typical	Atypical
1	Boy receives a present from his mother	Happy	Angry
2	Girl goes outside to play hide-and-seek with other children	Happy	Afraid
3	Girl cannot go to the Zoo, but has to stay at home	Angry / Sad	Happy
4	Boy has a dog that is ill	Sad	Afraid
5	Girl hears a person walking, in what she thinks is an empty house	Afraid	Happy
6	Girl lies in bed and hears a strange noise	Afraid	Angry

Procedure

The task administration for each child lasted approximately 10 minutes and was tape-recorded for subsequent transcription. All of the children in the study completed all six stories. The order of presentation of the six stories was randomized. The experimenter read the stories to the children in a neutral tone of voice.

The task had two elements: *Emotion prediction* and *Emotion explanation*. First, after hearing each story, the children were asked how the protagonist in the story would feel (*Emotion prediction*). If a child failed to identify an emotion the experimenter prompted the child, for example "*Do you think Linda feels happy, sad, angry or afraid?*" The order of the suggested emotions was randomized to avoid biased responses. Second, if the child predicted the typical emotion (Table 4.1, Typical Emotion column), they were asked to explain it (*Emotion explanation - Typical*). For instance "*Why does Linda feel sad?"* After the child had explained the typical emotion, the experimenter informed the child that the protagonist felt differently and named an atypical emotion. For example, "*Yes, that's what I thought as well. But Linda doesn't feel sad, she feels happy now that she is not going to the Zoo. Why is Linda happy?*" The atypical emotions were fixed for each story (Table 4.1, Atypical Emotion column). The children were then asked to provide an explanation for the atypical emotion (*Emotion explanation – Atypical)*.

If in the emotion prediction stage the child predicted an unexpected (i.e., atypical) emotion, the child was again asked to explain this emotion. After this explanation was given, the experimenter continued the task in the normal way by providing the emotion that was opposite to the one given by the child. The child was again asked to provide an explanation for the emotion given by the experimenter. However, any explanations given for emotions on stories where an atypical emotion had been predicted were not included in the analyses involving emotion explanations (see below). The purpose of continuing with emotion explanations on stories where the child had predicted an atypical emotion was to facilitate the general procedure and in order for the children not to become discouraged.

Scoring[14]

Emotion Prediction

On each story each child could predict either a typical or atypical emotion. All of the children in the study were able to make an emotion prediction for each story, except one child with VI who failed to predict an emotion for Story 4 even after a prompt. The frequency of predictions in each category (typical and atypical) was calculated for each individual story and each vision group (the single instance where a child with VI failed to predict an emotion for Story 4 was treated as missing). In the VI group (N = 15), one child made atypical emotion predictions on two stories while five children made atypical emotion predictions on one story. In the Sighted group (N = 26), two children made atypical emotion predictions on two stories while five children made atypical emotion predictions on one story. The two groups did not differ significantly in terms of the mean number of atypical responses ($t_{(18)} = .859$; $p = .396$).

Emotion explanation

As mentioned above, emotion explanations were only included for further analysis from those stories in which typical emotions had been predicted (valid stories). Explanations from stories on which an atypical emotion was predicted (along with the single instance in which no emotion was predicted) were not examined further. Therefore, for six children in the VI group and seven children in the sighted group, the subsequent examination of mental state references had to be carried out for fewer than six stories. The minimum number of stories on which the frequency of children's mental state references could be calculated was four. Table 4.2 below shows the proportion of children in each group based on the number of stories included in the subsequent analyses.

Table 4.2: *Proportion (%) of children in each participant group based on the number of valid stories*

	Number of stories		
	6	5	4
VI N = 15	60% (N = 9)	27% (N = 4)	13% (N = 2)
Sighted N = 26	73% (N = 19)	19% (N = 5)	8% (N = 2)

[14] Two examples of scoring procedure, including the data of one child from each group is given in Appendix B2.

For all valid stories, the emotion explanations were categorised as either 1) *Mental State* or 2) *Situational* as described below. If no explanation was offered, the response was classified as '*Don't know*'. Classification of explanations was performed separately for the typical and atypical emotions on each valid story.

1) *Mental State* explanations were further classified as *Beliefs* or *Desires*. *Belief* explanations were those in which children referred to a broader range of protagonist's cognitive states about the situation (e.g., know, think, imagine, wonder, guilty, guess). For example, Maggie is scared to hear someone moving in the living room '*because she doesn't know who it is*' or '*because she thinks it's a monster*'. *Desires* were those explanations in which children referred to the protagonist's states of desires and preferences (e.g., want, wish, like, look forward to, prefer, keen on). For instance, Linda is happy that she is not going to the Zoo because '*she didn't want to go in the first place*' or '*because she doesn't like animals*'. Importantly, these two mental state categories (*Beliefs* and *Desires*) were not exclusive. If a child used both to explain a protagonist's emotion (e.g., '*She wanted to go to the Zoo and she doesn't know why she can't go*') then the responses were assigned to both categories.

2) The responses were categorised as *Situational* when children elaborated on the situation or referred to another situation without reference to the protagonist's mental state. For example, Linda is happy that she is not going to the Zoo '*because it's raining*'.

When the children failed to provide an explanation for an emotion the response was classified as '*Don't Know*'. Across the Typical and Atypical emotion explanations the mean percentage of *Don't Know* responses in the VI group was 8% compared to 3% in the sighted group. However this difference between the two groups was not statistically significant (equal variances not assumed - $t_{(17.04)} = 1.291$; $p = .214$). It is worth noting that in both groups the proportion of *Don't Know* responses appeared higher when children were asked to provide explanations for atypical emotions. Consequently, related-samples *t* tests (collapsed across the two groups) revealed that the difference in the proportion of *Don't Know* responses between Typical and Atypical emotion explanations was statistically significant ($t_{(40)} = -2.918$; $p = .006$). The *Don't Know* responses were excluded from the subsequent analyses.

Inter-rater reliability was calculated for each explanation category (sum of explanations for typical and atypical emotions), using the Pearson correlation coefficients (*r*). An independent judge who was unaware of the children's individual characteristics or the hypotheses of the study coded approximately 50% of randomly selected transcripts from each vision group. The reliability ratings overall were high (*Beliefs*: $r = .943$; *Desires*: $r = .880$; *Situations*: $r = .985$; *Don't Know*: $r = .978$).

RESULTS

Typical emotion predictions

Predictions of the typical emotions were frequent in both groups. For most stories, typical emotion predictions were over 90% (Table 4.3). However, *Story 5*, which was initially modified to be suitable for experiences of children with VI, produced more atypical emotion predictions than any other story. *Story 5* evoked 27% atypical emotion predictions in the VI group and 15% of atypical emotion predictions in the sighted group. However, Chi square tests revealed that there was no significant association between Group (VI and sighted) and type of emotion prediction (Typical and Atypical) for any of the 6 stories (*Story 1*: N/A; *Story 2*: $\chi^2_{(1)} = .015$; $p = .903$; *Story 3*: N/A; *Story 4*: $\chi^2_{(1)} = .004$; $p = .950$; *Story 5*: $\chi^2_{(1)} = .771$; $p = .380$; *Story 6*: $\chi^2_{(1)} = .163$; $p = .686$).

However, it is worth noting that initially, when asked to predict an emotion, 20% (3/15) of the children in the VI group needed a prompt by the experimenter to identify an emotion. More specifically, two children needed a prompt on two stories each (Child 1: *Story 2 and 4*, and Child 2: *Story 3 and 6*) and one child required a prompt on one story (*Story 6*). By comparison only 4% (1/26) of the children in the sighted group needed a prompt to predict an emotion (*Story 5*). There was no obvious association between the story and the prompt requirement. Only prompted emotion predictions that were typical were included in the subsequent analyses.

Table 4.3: *Proportion (%) of typical emotion predictions in VI and Sighted groups across the six stories*

	Happiness		*Anger/Sadness*		*Fear*	
	Story 1	Story 2	Story 3	Story 4	Story 5	Story 6
VI	100%	93%	100%	87%	73%	93%
Sighted	100%	92%	100%	92%	85%	96%

Typical and atypical emotion explanations

First, it was of interest to examine whether the two groups differed in the extent to which they referred to mental states, rather than situations (Q1), as causes of the story protagonists' emotions, and if this was dependent on whether the emotion was typical or atypical (Q2). Thus, for each participant, the proportion of mental state emotion explanations was calculated as a proportion of all emotion explanations across all valid stories (i.e., number of *Mental State* explanations / number of all explanations [*Mental states* + *Situations*]). Each explanation was classified as either a mental state or situational. For mental states it was irrelevant which sub-type of mental state (i.e., *Belief* or *Desires*, or both) was given. This was repeated for typical and atypical emotion explanations. Table 4.4 shows the mean proportion of Mental State explanations, in the two groups of children across the typical and atypical emotion conditions. Notably, data variability was considerably large in both groups of children.[15]

Table 4.4: *Mean proportions of mental state references in the two groups for typical and atypical emotion conditions*

	Typical	*Atypical*
VI n = 15		
Mean (SD)	.49 (.33)	.48 (.32)
Sighted n = 26		
Mean (SD)	.45 (.22)	.44 (.22)

Looking at the proportions in Table 4.4, it is clear that mental state references were made approximately half the time for both typical and atypical emotions (i.e., the other 50% of the explanations were *Situational*, see Figures 4.1 and 4.2). Furthermore, it can be seen that the proportions of mental state references were similar across the two groups of children, and the proportions of mental state references were comparable for both typical and atypical emotions. Not surprisingly, a mixed 2 (Group) x 2 (Emotion typicality) ANOVA revealed no significant effects of Group ($F_{(1, 39)} = .284$; $p = .597$), Emotion typicality ($F_{(1, 39)} = .001$; $p = .841$) or interaction ($F_{(1, 39)} = .041$; $p = .979$). These results suggest that the children in the VI and the sighted groups were

[15] It is worth noting that Log transformation of the data was carried out, but did not improve the data distribution. Importantly, the subsequent analyses on transformed data as well as on raw scores did not yield different results from those presented here (i.e., proportion scores).

comparable in terms of the frequency with which they used mental state attributions to explain emotional states of the story characters (Q1), and the frequency with which the mental state references were made in both groups was the same for typical and atypical emotions (Q2).

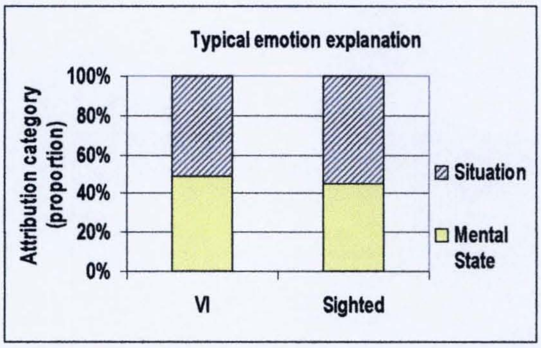

Figure 4.1: *Proportion of mental state and situational explanations for typical emotions*

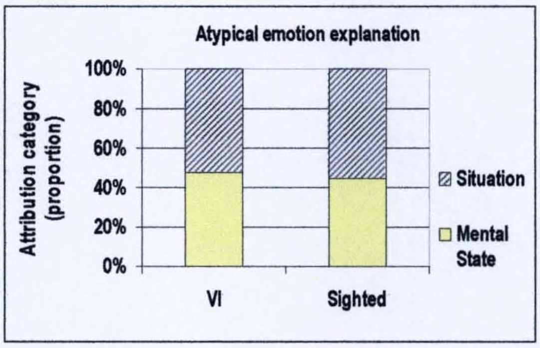

Figure 4.2: *Proportion of mental state and situational explanations for atypical emotions*

Mental states explanations: Desires vs. Beliefs

A further analysis of interest was to examine whether there was a difference between the two groups in the type of mental states explanations offered, namely *Desires* and *Beliefs* (Q3). Table 4.5 below shows the mean proportion of *Desire* explanations in the two vision groups, calculated as proportions of all the mental state references (i.e., number of *Desire* explanations / [number of *Desire* and *Belief* explanations]), for typical and atypical emotions. Naturally, children who did not refer to mental states in their emotion explanations on either typicality condition were excluded from these calculations. This was the case for 3 children in the VI group (i.e., one child on both typical and atypical emotion; one child on typical; and one child on atypical emotion condition) and 2 children in the sighted group (one child per typicality condition).

Table 4.5: *Mean proportion of references to Desires in the two participant groups for typical and atypical emotions*

	Typical	*Atypical*
VI (n = 12)		
Mean (SD)	.39 (.31)	.62 (.30)
Sighted (n = 24)		
Mean (SD)	.61 (.35)	.64 (.34)

Table 4.5 shows that, overall, children in the sighted group referred more frequently to *Desires* than *Beliefs*, as the mean proportions of *Desire* attributions, for both typical and atypical emotion explanations, were over 60%. However, in the VI group, *Desires* seemed to be referred to more frequently than *Beliefs* only for atypical emotion explanations; in the typical emotion condition the references to *Desires* made up fewer than 40% of all the references to mental states. Figures 4.3 and 4.4 graphically illustrate this pattern.

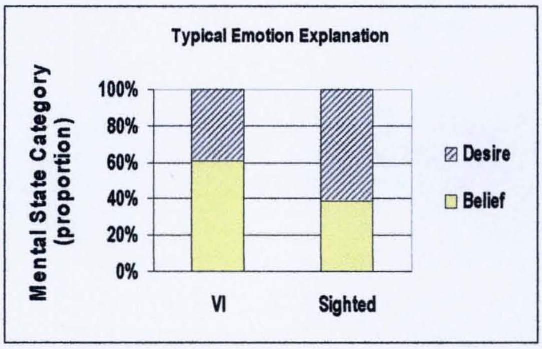

Figure 4.3: *Proportion of Belief and Desire explanations for typical emotions*

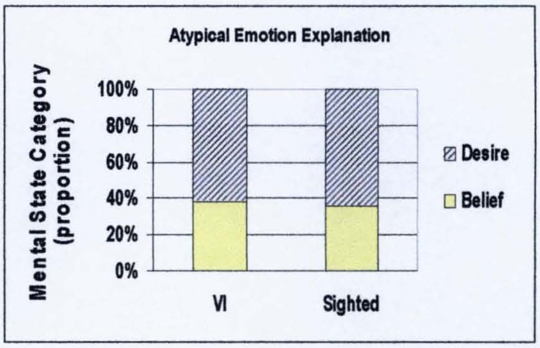

Figure 4.4: *Proportion of Belief and Desire explanations for atypical emotions*

A mixed 2 (Group) x 2 (Typicality) ANOVA, however, did not show a significant effect of Group ($F_{(1, 34)} = 1.612$; $p = .213$) and the effect of Typicality of emotion just failed to reach significance ($F_{(1, 34)} = 3.937$; $p = .055$). Furthermore, the interaction of Group x Typicality was not significant ($F_{(1, 34)} = 2.213$; $p = .146$). Given these findings, any further analysis and interpretations regarding Q3 would have been speculative.

Accounting for chronological age, intellectual level and VI level

The data were potentially too variable to carry out correlational analyses that would address Q4. Not surprisingly, using either raw or proportion scores, no significant correlations were detected in either participant group between the frequency of explanations in the *Mental States* category (overall and individual *Belief* and *Desire* categories) and the children's chronological ages and VIQs (i.e., based on the non-parametric Spearman's test; p values > .05). Additionally, Mann-Whitney tests did not significantly differentiate the PVI and SVI groups on the frequency of mental state explanations (overall, or for individual categories), using either proportion or raw scores, suggesting that the distinction between the PVI and SVI levels in the early years in children with VI could not account for the current task performance (p values > .05).

Qualitative considerations

The lack of power in the current experiment to detect results of statistical significance may have obscured the true richness of the children's language produced in a conversational context. For that reason it was felt that examinations at a level of individual children may throw further light on their use of mentalistic discourse to explain causes of emotions.

Judging by the qualitative data (examples of which can be seen in Appendix B.2), even the youngest children in the study, and in both participant groups, were able to express themselves very well using a range of mentalistic language terms across the typical and atypical conditions. However, closer inspection of such data may reveal some underlying subtleties in individual childrens' discourse, which could not otherwise be detected by a systematic coding method or a statistical test. The testing session with an 8 year old girl with SVI was chosen to illustrate such subtleties and the transcript from this session is presented in Table 4.6.

This child's data was chosen partly as she produced possibly the richest repertoire of mentalistic language in the whole sample. Her explanations were somewhat challenging to score using the coding method suggested by Rieffe et al. as she tended to predict a range of potential emotional outcomes for the story characters (e.g., Story 2, Table 4.6), unlike the majority of children who generally provided only one prediction. This made it clear that the task itself, including the coding system, was still relatively rigid, and did not necessarily account for the natural fluency of a child's language. Nevertheless, considering that this child's verbal IQ was above the average range (VIQ = 118) her verbal expression in the context of the present task may not be surprising. Interestingly, the majority of her explanations were justified using mentalistic discourse, rather than drawing on situational causes, which revealed a rich insight into the inner mental states held by this child. For instance, where many of her VI and sighted peers produced explanations that were relatively simple and had meaningful representations in real life (e.g., Walter is afraid that his dog is not very well because *'he might not know anyone around the area to play with'*; *'he doesn't want his dog to die'* or *'he won't have much fun without his dog'*), her explanations seemed quite intricate for a child of her age (e.g., *'he feels sorry for his dog, he feels guilty, 'cause he blames the dog'*, Story 1, Table 4.6). However, despite her extensive knowledge of mental states and her clear ability to attribute them to other people, her cause-consequence reasoning within the emotional scenarios sometimes appeared to lack pragmatic quality (e.g., Nadia feels angry when she hears the strange noise at night *'because she's too scared and too frightened of her mother to tell her about the strange noises because she's heard them before'*, Story 6, Table 4.6). This occurred throughout the testing session and she sometimes needed to be re-directed to the topic (e.g., Story 1, Table 4.6). Additionally, it appeared as if she had learned the right thing to say in a given context, even though her own reasoning for that answer did not always seem pragmatically meaningful (e.g., Maggie is happy that someone is in the living room '*because she knows someone's there even though it's her arch enemy*', Story 5, Table 4.6). While it may be speculative to generalise from this pattern of findings, it may be interesting to point out that, in Chapter 3, this child was within the domain for Asperger syndrome on the CCC-2 and just below the clinical cut-off for autism spectrum on the SCQ (participant ID: 20).

Table 4.6: *Emotion task transcript for a child with VI*

Story 1

Assessor: How did Max feel when he got the package?
Child: Very excited and apprehensive
Assessor: Why did Max feel like that?
Child: 'Cause, when I got a parcel this morning and it was a t-shirt and a pair of track suit bottoms, and I didn't know, and I was so excited because I thought it was like a big toy or something.
Assessor: That is interesting. But how about Max, why did he feel excited and apprehensive?
Child: Because he got a present and he doesn't know what it is.
Assessor: Actually Max feels angry when he gets the package. Why does he feel angry?
Child: 'Cause his mum won't tell him what's inside.

Story 2

Assessor: How does Anna feel when she goes out to play hide-and-seek with her friends?
Child: Nervous, in case they don't want her and happy, 'cause she likes the game, and maybe a bit apprehensive in case they start fighting with her.
Assessor: That's what I thought as well but, actually she feels afraid when she goes out to play hide-and-seek. Why does she feel afraid?
Child: Because she may have her arch enemy among them.

Story 3

Assessor: How does Linda feel when she is told that she can't go to the Zoo anymore?
Child: Upset and Angry
Assessor: Why does she feel upset and angry?
Child: Because they never told her on the day, because she really wanted, she was really looking forward to it.
Assessor: Hmm, I see. But Linda is happy not to go to the Zoo. Why is Linda happy?
Child: Because she doesn't want to lose them, her parents, doesn't want them to go away.

Story 4

Assessor: How does Walter feel now that his dog is not very well?
Child: Upset and angry
Assessor: Why does Walter feel that way?
Child: Angry, because his dog is not very well and he'll blame the dog, upset because his dog is not very well and he'll feel sorry for him.
Assessor: But Walter is actually afraid now that his dog is not very well. Why is Walter afraid?
Child: He feels sorry for his dog, he feels guilty, 'cause he blames the dog.

Story 5

Assessor: How does Maggie feel when she hears that someone is moving in the living room?
Child: Scared and determined.
Assessor: Why does Maggie feel like that?
Child: Scared because she doesn't like the fact that no one is there, and determined because she's heard them before, and she knows who they are, and she can defend her room.
Assessor: Yes, I would have thought that as well, but Maggie is actually happy when she hears that someone is in the living room, why does she feel happy?
Child: Because she knows someone's there, even though it's her arch enemy.

Story 6

Assessor: How does Nadia feel when she hears the strange noise?
Child: Scared
Assessor: Why is she scared?
Child: She doesn't know who it is ... but also she may feel a bit comforted, because she might think she knows who it is, whilst she doesn't.
Assessor: Nadia actually feels angry when she hears the strange noise? Why would she be angry?
Child: Because she's too scared and too frightened of her mother to tell her about the strange noises, because she's heard them before.

DISCUSSION

The present findings suggest that children with congenital VI do not differ from their age and verbal ability matched sighted peers in the extent to which they refer to mental states in their explanations of emotions of story characters. This lack of difference is a finding that was consistent across typical and atypical emotional scenarios and the differing types of mental states (i.e., beliefs and desires) in the present design. Importantly, in the interpretation of these results, two main issues need to be considered. First, the result did not support the methodology by Rieffe et al., who designed and validated the Emotion task used in the present study. Second, they contrast with the previous research evidence highlighting theory of mind development as an area of relative difficulty for children with congenital VI, in comparison to developmentally matched sighted children.

First, we must consider the present results in comparison to those of Rieffe and colleagues and take into account the methodological limitations of the task. In their studies with sighted typically developing children and sighted children with autism (Rieffe et al., 2005; Rieffe et al., 2000), Rieffe et al. supported their hypothesis that atypical emotions would produce more mental state justifications than typical emotions. It is possible that their findings may have been a result of having a fixed order of 'typical first, atypical second' explanations, whereby probing children for an additional explanation on the same story may evoke more thinking in general. The authors themselves suggested that the first emotion explanation may have caused a 'priming effect', resulting in an increased incidence of mentalistic language in the atypical emotion condition, even in the autism group (Rieffe et al., 2000). This cuing strategy that increased mentalising capacity of children who are known for their impoverished mental state insight is striking, particularly as the typical emotional scenario significantly distinguished the autism group as poorer than the control children.

Following from this, it is possible that the sample-size related lack of power had contributed to the current results, given that the studies by Rieffe et al.'s included larger groups of children. However, considering the distribution of the means in the present experiment (whereby the proportions of the mentalistic explanations for typical and atypical emotions were virtually indistinguishable), low power does not seem to provide a sufficient explanation. Here it is important to consider again the fact that the Emotion task required the children to provide an explanation for the novel (atypical) emotion after their initial (typical) emotion prediction had been challenged by the assessor (e.g., *'yes, that's what I thought as well, but Linda doesn't feel sad, she is actually happy now that she is not going to the Zoo. Why is Linda happy?'*). Theoretically, it makes sense that the atypical emotions should be more likely to provoke children to think about mental states, as the cause for the strangeness of an atypical emotional scenario (e.g., seeing someone angry at receiving a present) may not be self-evident (Gnepp, 1983; Rieffe et al., 2005). However, methodologically, the task design may not necessarily facilitate this process, as the children's responses may be affected by their learning that their prediction in the first place is not 'correct'. For instance, this may account for significantly more *Don't Know* responses for atypical emotion explanations in this study. Additionally, learning that their prediction may not be what the assessor expects, is likely to affect the children's prediction of typical emotions after the first story. Anecdotally, as a result of the assessor suggesting the emotions that were opposite to the ones provided by children, it was observed that a number of children developed a strategy of spontaneously predicting and explaining both typical and atypical emotions for a given story, before giving the chance for the assessor to suggest an alternative emotion. This would at least to an extent cancel out the priming effect that Rieffe et al. suggested of the atypical emotion scenarios, explaining the lack of difference of mental state use across typical and atypical emotion conditions in this research.

The findings comparing children with VI and sighted children in their use of specific mental state terms were not clear-cut in the present study. Overall, it may be speculative to discuss these results in any detail as the group variances with regards to this data were too heterogeneous. However, in line with Rieffe et al., it may be worth noting that the mentalistic language of typically developing sighted children appeared to contain more desire terms than belief terms overall, although Rieffe et al. found this effect to be particularly pronounced with younger children[16] (owing to the small sample, it was not possible to capture the age trend in this research). On the other hand, mentalistic language of children with VI appeared to follow a somewhat different pattern as they seemed to use more belief terms to explain typical emotion scenarios and more desire terms to explain atypical emotion scenarios. Even though it was not possible to affirm such findings statistically, the fact that the VI group appeared to rely on belief terms more than on the desire terms in certain conditions is indicative of existing mentalistic insight in these children, as

[16] It may be worth noting at this point that, when analysed further, Reiffe et al.'s findings regarding the emotion typicality and the type of mental state were found to vary as a function of type of emotion (i.e., sadness, happiness, fear). However, the present sample (particularly the VI group) was deemed too small to investigate potential effects of this variable. Having said that, it is possible that different types of emotion may interact with task performance in the current study. However, larger groups of children with VI would be required to address this systematically.

belief terms are developmentally more sophisticated and acquired later than desire terms (Bartsch & Wellman, 1995).

Following from this, the methodological limitations (including the lack of power) discussed thus far do not provide sufficient explanation for why the Emotion task failed to differentiate children with VI and children who are sighted in this study, despite its relative sensitivity with typically developing children and children with autism in previous research. For this reason, it is possible that the present null results reflect a real effect. More specifically, if performance on the Emotion task is a true index of mentalising capacity, then based on the present findings, such capacity in children with VI at school age indeed may be as proficient as that of sighted children of similar age and intelligence. In support of this, the qualitative data from individual children confirms that children who are visually impaired use a wide repertoire of mental state terms in their explanations of emotions. This, in turn, contrasts with previous research which demonstrated poor theory of mind outcomes, including mentalistic language use, in children with VI relative to sighted children (e.g., Green et al., 2004; Pring et al., 1998).

However, this interpretation requires the consideration of a number of issues. For example, the previous studies, which demonstrated a delay in theory of mind development in children with congenital VI, have shown that such children often require a mental age of at least four years in order to pass a standard false-belief task (Green et al., 2004; Minter et al., 1998; Peterson et al., 2000). Furthermore, an association between intellectual level (i.e., verbal IQ) and theory of mind performance in children with VI had also been demonstrated previously, with better socio-cognitive outcomes being observable in children with VI who were more able (Green et al., 2004; Pring et al., 1998). The children with VI in the present study, who were age-range 6 to 12 years, all had verbal mental ages that were in line with, or exceeding, their chronological ages. Even though the association between verbal IQ and the frequency of mentalistic discourse could not be established statistically, the children with VI in this research all had verbal IQs that were at least within the normal range limits, with a number of children with above average verbal intelligence levels. Therefore, it is likely that these children have already reached the verbal mental ages by which any potential theory of mind difficulties at an earlier age would have been resolved.

In relation to this, however, a number of research studies have reported that theory of mind difficulties in samples of children with VI often persist at considerably older ages (e.g., 9-12 years) (McAlpine & Moore, 1995; Peterson et al., 2000) and in cases of children with normal IQ (Green et al., 2004; Pring et al., 1998; Roch-Levecq, 2006). Hence, it is possible that the variations of the standard false-belief tasks used in such studies were generally more complex than the Emotion task used here, providing perhaps a more sensitive measure for distinguishing children with good and poor theory of mind. Similarly, it is clear that the 'Strange Stories' task, used by Pring et al. (1998), calls upon higher-order mental state understanding as it involves knowledge of more advanced mental state concepts, such as sarcasm, misunderstanding and figure of speech, compared to the basic emotions presented to children in the current study. This, in turn, may be more likely to capture the difficulties of those children who have poorer theory of mind ability.

On the contrary, it can be argued that the Emotion task had specific advantage in comparison to the standard false-belief tasks and 'Strange Stories', as these may be too complex and difficult for children with congenital VI for reasons other than poor theory of mind. For example, it has been suggested that children with VI fail false-belief tasks because lack of visual input affects their information-processing and representation (Roch-Levecq, 2006). It is likely that the false-belief task may place an additional information-processing load on children with VI, who may have to rely on attention and memory to deal with the demands of the task. However, this use of information-processing resources may not be as efficient as for children who are sighted (Roch-Levecq, 2006). Similar argument was given by Marschark, Geen, Hindmarsh and Walker (2000) with regards to children with hearing impairment (HI). They found that, contrary to the difficulties with understanding false-belief reported by others (Peterson & Siegal, 1995, 1998), children with HI were able to produce a rich repertoire of mental state terms, which was comparable to the use of mental state language in hearing controls. They argued that the false-belief paradigm tests more than just theory of mind understanding and that this may be the reason why children with HI fail false-belief tasks, despite having a reasonable insight into the mental states.

Following from this, the strength of the current paradigm may be in the similar information-processing demands that it places on both VI and sighted children. Additionally, in comparison to the 'Strange Stories', the Emotion task may be more suitable for use with younger children. The children who completed the 'Strange Stories' in the study by Pring et al. (1998) were above nine years of age. Moreover, they were also matched to a group of sighted children on chronological age only. Considering the lack of intellectually matched sighted controls (as well as the relationship between the intellectual levels of the children with VI and the frequency of their mental state justifications), there is a possibility that the poorer 'Strange Stories' performance of the VI group studied by Pring et al. may have been confounded by variables extraneous to theory of mind. Hence, it can be argued that the Emotion task provided a simple, but adequate measure that was sensitive to the understanding of mental states in children with congenital VI and at different chronological ages. Similarly, this measure is likely to have a greater ecological validity than the false-belief task or the 'Strange Stories'. This is because in real life, children may be frequently required to make judgements for why someone may be feeling happy or sad, than they are to be involved in scenarios such as those depicted in the other types of tasks. However, without a doubt, use of a range of measures of mental state reasoning of different complexity, along side the current task, would have provided a more comprehensive picture of socio-cognitive skills in the present sample of children with VI.

At this point it may be worth pointing out that, despite the ability of the children with VI to predict typical emotions without much difficulty (as expected, in line with Roch-Levecq, 2006 and Dyck et al., 2004), a somewhat larger percentage of these children needed a prompt by the experimenter to predict an emotion than did the sighted group, although this difference was not statistically significant (the prompt requirement was not an effect of any particular story). What may be particularly interesting about this pattern of finding is that the children in the VI group (N = 3) who required a prompt to predict an emotion were those children whose levels of vision were poorer (i.e., PVI). Statistically, the task did not differentiate children with PVI and SVI in the frequency with which they referred to mental states. Indeed, such small numbers of children within the two VI subgroups preclude us from drawing any firm conclusion with regards to the PVI/SVI distinction in this group of children. Additionally, eyeballing the qualitative outputs of the individual children did not reveal any obvious disadvantage for children with PVI in their use of mental state discourse. This may not be surprising, given that, despite some prompt requirements, the emotion prediction abilities of these children were near ceiling. However, even the minimal need for prompts by these children may reveal the subtle difficulties in the way that children who have no functional vision in their early years learn about emotions, compared to children with SVI whose limited form perception may enable them to discern some limited patterns of emotional expression on human faces very close by. Although speculative in this context, these issues are likely to be of relevance for future research with such children, particularly at younger ages.

Thus far, it cannot be disputed that the Emotion task called upon the implicit knowledge of mental states in children with VI in this study, as their references to beliefs and desires of the story characters were produced spontaneously, frequently and in a meaningful way, even when situational explanations were available to them. Without a doubt, such ability must be indicative of an active theory of mind. Therefore, it may be presumptuous to attribute the lack of difference between the two groups in the present study solely to a methodological limitation of the current task to distinguish the children with VI as poorer than their sighted counterparts. It is not a trivial achievement for children with congenital VI to recognise the emotions of others and to consider the subjective beliefs and desires that underlie them. For this, they have to assimilate, coherently and meaningfully, different mental state concepts and different perspectives from which they arise into their repertoire of experiences. Hence, the success in showing contextually appropriate mental state awareness by children with VI in this study is undoubtedly an important finding. However, we still cannot be certain whether this ability is indicative of the fully fledged, sophisticated theory of mind that is required to deal with the complex day-to-day demands of the social world. This is a particularly important question to ask, as the same children with VI who completed the Emotion task were found to show poorer social communication and pragmatic language use relative to the sighted children in the research presented in the previous chapter. This pattern is also somewhat illuminated through the qualitative consideration of an individual child in the current result section, as this child showed an unusual pattern of responses in pragmatic terms, despite her rich knowledge and use of mental state language that distinguished her even from the majority of sighted children.

In relation to this point, it has been proposed that *having* a theory of mind is different from *using* theory of mind abilities spontaneously to describe, explain and interpret other people's behaviours (Meins, Fernyhough, Johnson, & Lidstone, 2006). For this reason, mental state discourse in children may not necessarily be an indicator of the child's theory of mind ability per se, but rather of their tendency to use it. In line with this, several studies have failed to find an association between theory of mind competence (i.e., false-belief task performance) and spontaneous use of such capacities (e.g., using mental state narrative to account for the behaviours of storybook characters) in school-age typically developing children (Charman & Shmueli-Goetz, 1998; Meins et al., 2006; Tager-Flusberg & Sullivan, 1995), even though such association is present in young pre-school children (Dunn et al., 1991). The presence of this association in young children may be down to the fact that they may still be in the process of acquiring the theory of mind ability, which is then likely to constrain their mental state language use. However, once the baseline stage of representational theory of mind is complete, the fact that a child possesses this capacity does not necessarily entail that it will be applied on-line, particularly in a non-interactional context.

This reasoning may throw some light on the present findings. While the Emotion task may have provided evidence that primary school-age children with congenital VI and normal VIQ have an active theory of mind (based on the fact that they use mentalistic language), we are still uncertain if there may be subtle differences between children with VI and sighted children in the way they use this ability to deal with real-life social demands[17]. This uncertainty is particularly qualified by the findings in Chapter 3. For this reason, future research may need to consider (and potentially develop) a theory of mind measure that is sensitive to such demands, as well as to the individual differences in socio-cultural variables that interact with these processes in children.

The Emotion task itself may provide a more useful measure in future research studies, if appropriate modifications are adopted. For instance, the task may benefit from removing the emotion prediction factor. Children could be provided with a greater number of stories, where typical and atypical emotional scenarios (with a wider repertoire of emotions, ranging in complexity) would be presented by the experimenter in a counter-balanced order, requiring children only to provide explanations for the given emotions. Importantly, this would remove the methodological problem of having to treat a certain number of responses as invalid (i.e., if the child predicted an atypical emotion to begin with). Furthermore, a larger number of stories would provide a basis for a greater number of explanations, increasing the experimental power to potentially distinguish between the frequencies of desire and belief terms in children's explanations. For the purposes of future research, such modified version of the Emotion task, in addition to a range of theory of mind measures of different complexity, would help to chart the socio-cognitive abilities of children with VI and, more comprehensively, at different ages. However, these abilities in children are likely to be mediated by certain socio-interactive factors that are involved in theory of mind development and would also need to be considered in more detail. Interestingly, the role of one of these factors (i.e., maternal mental state language input) is addressed in the following chapter.

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[17] In its current form, the Emotion task may still lack the naturalistic aspect of everyday social reasoning, because of the structured way in which the children's responses are cued.

Chapter 5

The Role of Language Input to Children with Congenital VI: Mother-Child Mental State Discourse

SUMMARY

The research presented in the previous chapter investigated children's social understanding by examining the extent to which they used mental state discourse in their explanations of emotions. Despite its methodological limitations, it is implied from this research that school-aged children with congenital VI do have an implicit understanding of subjective mental states, judged by their ability to use the language that represents those states. The purpose of the research presented in this chapter was to consider the context within which learning of such language occurs in children with VI. An impressive body of evidence now shows the developmental link between mothers' mentalistic language input and children's growing understanding of the mind in typically developing children (reviewed by de Rosnay & Hughes, 2006). Thus far, the role of such input for children who are born with VI has not been considered empirically. Here, the quality of mother-child discourse was examined in the context of a joint book-reading interaction. Mothers of children with VI and mothers of children who are sighted were compared in the extent to which they used mentalistic language to elaborate on the story-book content, and the frequency with which such language occurred in the mothers' language was examined in relation to the children's own mentalistic discourse. The results suggest that the extent to which the mothers in this study used mental state language to elaborate on story-book contents was comparable across the two vision groups, although the mothers of children with VI referred to the story characters' mental states (as opposed to their own or their children's mental states) to a greater extent than did the mothers of sighted children. Additionally, the mothers of children with VI provided significantly more descriptions in their overall discourse than mothers of sighted children, providing more detailed information of physical and behavioural aspects of the story which are not easily accessible to their children with VI. Both groups of children used surprisingly little mentalistic language overall. However, the language of sighted children included more descriptions than did the language of children with VI. Although further research is needed to understand the unique contribution of maternal mentalistic language input in the socio-cognitive outcomes of children with VI, the current study is the first to date that has investigated such input

to these children, offering a unique insight into the nature of verbal scaffolding provided by mothers to their children with VI and highlighting a potential area for future interventions.

INTRODUCTION

A growing body of evidence highlights the influence of socio-interactive input within a family context on children's social understanding and theory of mind development (reviewed by Carpendale & Lewis, 2004; de Rosnay & Hughes, 2006; Symons, 2004). Among the factors believed to promote development of these processes in children is the presence of siblings (especially older ones), presence of older relatives, higher socio-economic status and secure attachment (J. R. Brown et al., 1996; Cutting & Dunn, 1999; Dunn et al., 1991; C. Lewis et al., 1998; Meins et al., 1998). While there is no doubt that different kinds of social and conversational environments may have discernible effects on children's developing social cognition, some researchers have placed special emphasis on the role of mothers' conversational input in this process (Meins et al., 2003; Meins et al., 2002; Ruffman et al., 2002).

In the context of maternal language input, mental states discourse has been given specific emphasis and several studies have now demonstrated a direct link between maternal mental state language input and children's social understanding (de Rosnay et al., 2004; Meins et al., 1998; Meins et al., 2003; Meins et al., 2002; Ruffman et al., 2002; Taumoepeau & Ruffman, 2006). A number of these studies have provided strong evidence, which suggests that the role of maternal mentalistic input to their children's social cognition is causal, providing a scaffolding framework upon which children learn their own verbal expression and understanding of mental states. For example, Ruffman et al. (2002) found a consistent correlation between mothers' early mental state talk and children's false-belief task performance at three different time points (i.e., between children's ages of two and four years), and this relationship remained even after controlling for other potential mediators (i.e., mothers' educational levels and frequency of other types of utterances, and children's ages, language ability levels, own use of mentalistic language, and early theory of mind). In a similar vein, Meins and collaborators (Meins et al., 2003; Meins et al., 2002) found that mothers' tendency to make appropriate comments on their children's own mental states (or what they called mothers' *mind-mindedness*) in the first year of life had unique value in predicting the children's performance of theory mind tasks at the age of four, even when other socio-environmental correlates of children's social understanding (e.g., the mothers' education, number of siblings, exposure to general mental state language and attachment style) were controlled for.

Whilst Meins and collaborators (Meins et al., 2003; Meins et al., 2002) emphasised the value of mothers' proclivity to comment appropriately on their children's mental states (and therefore treat their children as independent persons with their own thoughts and feelings), Ruffman et al. (2002) suggested that it is the mothers' general discussion about mental states, rather than one specific type of utterance, that facilitates children's learning about the mind, although it is directed at a level that is appropriate to that of the child (Taumoepeau & Ruffman, 2006). Just like children's understanding of desire precedes their understanding of belief (Bartsch & Wellman, 1995; Wellman & Liu, 2004), mothers also tend to talk more frequently about desires when children are younger, with talk about thoughts and beliefs increasing with children's age. Thus, it appears that specific types of mental state language *at critical points* in the child's development scaffold children's mental state language and bootstrap their understanding of the mind (Taumoepeau & Ruffman, 2006).

The positive relationship between mothers' mentalistic input and children's socio-cognitive outcomes has also been demonstrated with atypical groups of children with known theory of mind problems, namely children with autism and children with hearing impairment. For instance, Slaughter, Peterson and Mackintosh (2007) reported that, like in the case of typically developing children, children with autism whose theory of mind understanding is most advanced appear to have mothers who frequently talk abut mental states. However, rather than general talk about mental states, it was the mothers' mentalistic discourse that was explanatory, causal and contrastive that was found to have a unique contribution to the theory of mind task performance of children with autism. Similarly, in the case of children with hearing impairment (of hearing mothers), Moeller and Schick (2006) found a significant correlation between maternal talk about mental states and children's performance on verbal theory of mind tasks. Finally, as the overall amount of maternal talk was unrelated to the children's false-belief performance, the study highlighted the quality, rather than quantity of input as an important contributor to the social understanding of children with hearing impairment

Given the known difficulties in theory of mind development in children with autism (Happé, 1994) and late-signing children with hearing impairment (Peterson & Siegal, 1995), the studies described so far illuminate the language input provided by mothers as an important candidate for early intervention. This has important implications for children who are born with significantly impaired vision, whose vulnerable theory of mind outcomes have been well documented (see Chapter 1 and Chapter 4). Crucially however, no studies to date have examined mental state input to children who are visually impaired. Although some attempts have been made to address mother-child dialogue characteristics and interaction styles in young children with VI (e.g. Andersen et al., 1993; V. Moore & McConachie, 1994), the specific role of mentalistic language input provided by the mothers to their children with VI has not been addressed by research. The research presented in this chapter is the first to date to examine such input to children with VI and it is hoped that the findings will provide a unique insight into the quality of mother-child discourse in this population.

Nevertheless, it is first important to consider what is generally known about language input to children who are visually impaired. Andersen et al. (1993) argued that, in a conversational interaction involving a child with VI, where the child's attention cannot be caught and directed by eye-contact, parental input is likely be restricted and impoverished in a number of ways. For instance, it has been shown that mothers' language input to their children with VI tends to be highly directive and controlling, involving relatively few descriptions (e.g. on the functions and attributes of objects, events and people) (Andersen et al., 1993; Kekelis & Andersen, 1984; V. Moore & McConachie, 1994), although these patterns may be exacerbated in children whose VI is of greater severity (V. Moore & McConachie, 1994). Furthermore, it has been shown that mother-child conversational interactions involving children with VI tend to be asymmetrical. This means that the mothers tend to initiate a greater proportion of topics (which are almost exclusively child-centred) than do their children with VI who, in turn, when compared to sighted children, rarely initiate conversations with their mothers and are unable to sustain conversation around a particular topic (Kekelis & Andersen, 1984; Kekelis & Prinz, 1996; V. Moore & McConachie, 1994). Others have reported that mothers of children with VI tend to be generally less responsive vocally (Rogers & Puchalski, 1988; Rowland, 1983, 1984).

However, Pérez-Pereira and Conti-Ramsden (1999) argued that the existing research has underestimated and misinterpreted the quality of maternal language input to children with VI and that characterising mothers of children with VI as a homogenous 'non-responsive' group may be too simplistic and misleading. They argued that maternal directiveness and conversational asymmetry is a common feature of mother-child interaction in general, particularly in young children (Ninio & Snow, 1996), and provides scaffolding for a young language learner. In fact, Dote-Kwan (1995) found mothers' responsive behaviours to be positively related to the development of children with VI, with a high percentage of responses and compliances, and a low percentage of ignoring or refusing their children's initiations and requests for help. Furthermore, Pérez-Pereira and Conti-Ramsden (2001) observed that mothers of children with VI often speak more to their children than do mothers of sighted children (also Behl et al., 1996), and use significantly more descriptions when directing the child than do the mothers of sighted children. This lends support to the idea that parents of children with VI are able to develop alternative strategies when conversing with their children (Urwin, 1978) and exploit the use of language as a way to share the world with their child, who depends on language for social interaction and learning.

The studies investigating mother-child conversational interactions in children with VI have been scarce and, as seen from the discussion above, the available evidence is inconclusive. However, the general consensus appears to be that maternal language input to children who are visually impaired is qualitatively different from that received by sighted children. In the present study, it is of interest to examine whether this difference in the quality of language input will be also reflected in the extent to which the mothers in of children with VI and mothers of sighted children use mental state discourse when interacting with their children.

All of the aforementioned studies on mother-child discourse and interaction were carried out with very young and often pre-lingual children with VI, sometimes including children with additional non-sensory impairments. Furthermore, most of these studies have been examinations of very small numbers of children, and often only individual cases, generally lacking control groups. While such studies have been invaluable in furthering our understanding of mechanisms involved in the language learning and interaction of young children with VI, further controlled studies, including also older (i.e., school-age) children, are needed to fully appreciate the quality of mother-child discourse involving children with VI and the role of maternal language input in this process. With this in mind and in the context of the present study, one advantage of examining mother-child discourse with school-age children, specifically in terms of mentalistic language, is that the vocabularies of older children contain a wider repertoire of mental state terms and the quality of maternal mental state talk is likely to reflect this, providing a richer context from which to sample this kind of data. This follows from the research findings mentioned earlier, which suggest that mothers' mentalistic language input tends to be directed at a level that is appropriate for their child (Taumoepeau & Ruffman, 2006). Thus, given that the children in this study were of school-age, it is expected that the mentalistic language provided by mothers would be relatively rich and varied.

However, it is important to consider a context within which such language can be suitably examined. Researchers have used different methods and contexts for assessing mothers' mentalistic discourse, although in cases of young children, this has most commonly been done during the play interactions, both in sighted children (e.g. Meins et al., 2003; Meins et al., 2002) and children with VI (e.g. Kekelis & Prinz, 1996; V. Moore & McConachie, 1994). In other studies with typically developing sighted children, mothers were asked to provide open-ended descriptions of their children, independent of the mother-child interactional context, and measures of maternal mentalistic language were sampled from such descriptions (de Rosnay et al., 2004; Meins et al., 1998). Alternatively, in the studies by Ruffman et al. (Ruffman et al., 2002) and Taumoepeau and Ruffman (Taumoepeau & Ruffman, 2006), mothers of sighted children were presented with pictures depicting scenarios involving different mental states, which they were asked to discuss with their children. A popular method for assessing mother-child dialogue has been the book narrative (Slaughter et al., 2007; Symons, Peterson, Slaughter, Roche, & Doyle, 2005). It has been argued that the joint book reading context is facilitative of mental states discourse, as it provides a naturalistic setting and an opportunity to elaborate and ask questions by both conversational partners, including questions and elaborations about the story characters' thoughts and feelings (Dyer, Shatz, & Wellman, 2000; Symons et al., 2005). Accordingly, it was felt that in the present study a joint-book reading session, especially when using an unfamiliar story-book, would provide a suitable context within which to examine the incidence and nature of language input to children who are visually impaired, in that it would resemble an every-day situation that involves processing of novel information and thus facilitate joint discourse. This context is also useful as a means for assessing the children's own mentalistic language, enabling us to provide a clearer picture of how the verbal scaffolding provided by the mother takes place.

In summary, the aim of the present study was to gain further insight into the language input provided by the mothers to their children who were born with visual impairment, by focussing on their mental state language input in particular. Accordingly, it was of interest to examine whether the mothers of children with VI and mothers of children who are sighted would differ in the extent to which they talked about thoughts, desires and feelings during a joint book-reading session with their children. It was also of interest to undertake a more exploratory analysis of such input, by making a distinction between mentalistic language that is sensitive to mental states of a) the story characters, b) the conversational partners and c) participants themselves; and to examine potential between-group difference accordingly. For instance, whilst Meins et al. emphasised the role of appropriate commentary on child's mental states in particular, the story-book context provides an opportunity to discuss mental states of the story characters, and this may be reflected in the findings of this study. Furthermore, following from the idea that mothers of children with VI develop alternative strategies to facilitate their child's language and learning, it was also of interest to explore non-mentalistic features of the mothers' language input that may have a particular value for children with VI and distinguish their mothers from those of children who are sighted. For this reason, emphasis was placed on the extent to which mothers elaborate on descriptive aspects of the story-book, given the limited, yet conflicting evidence regarding the use of descriptions in mothers' language input to young children with VI (Kekelis & Andersen, 1984; V. Moore & McConachie, 1994; Pérez-Pereira & Conti-Ramsden, 2001). Finally, it was of interest to examine the relationship between the language characteristics of mothers and children in the two groups of children and provide a clearer picture of the role of verbal scaffolding provided by mothers to children with VI.

Research questions and hypotheses

In line with the theoretical framework above, the following predictions and questions were addressed by the research presented in this chapter:

Hypothesis 1 (H1): Mothers of children with VI and mothers of children who are sighted will differ in the extent to which they talk about thoughts, desires and feelings during a joint book-reading session with their children.

Question 1 (Q1): Will this difference vary depending on whether the mental state refers to the self, child or character?

Hypothesis 2 (H2): Children with VI and sighted children will differ in the extent to which they talk about thoughts, desires and feelings during a joint book-reading session with their mothers.

Question 2 (Q2): Will this difference vary depending on whether the mental state refers to the self, child or character?

Hypothesis 3 (H3): Mothers of children with VI and mothers of children who are sighted will differ in the extent to which they use descriptive attributes during a joint book-reading session with their children.

Hypothesis 4 (H4): Children with VI and children who are sighted differ in the extent to which they use descriptive attributes during a joint book-reading session with their mothers.

Hypothesis 5 (H5): There will be an association between the language characteristics of mothers and the language characteristics of their children with VI, in terms of both mentalistic and descriptive elaborations.

Question 3 (Q3): Can the severity of VI explain the variation in the extent to which mothers of children with VI use mentalistic discourse during the joint book reading session?

METHOD

Participants

The participants were a subgroup of the children who took part in the research presented in the previous chapters, and their mothers. There were 12 children with congenital VI and 16 sighted children. The two groups of children were comparable in their verbal ability ($t_{(26)}$ = .881, p = .386), chronological age ($t_{(26)}$ = .187, p = .853), and distribution of gender ($\chi^2_{(1)}$ = .012, p = .912) (Table 5.1).

Table 5.1: *Matching characteristics of the sample*

Matching criteria	***VI*** *N = 12*	***Sighted*** *N = 16*	***p value***
VIQ / WISC-III			
Mean (SD)	109 (9.2)	105.2 (11.9)	.386
Range	95 - 128	80 - 126	
Age			
Mean / months (SD / months)	101 (24.4)	100 (18.7)	.853
Range / years	6:06 – 12:11	6:02 - 11:11	
Gender ratio			
(Female/Male)	7/5	9/7	.921

Additionally, there was no significant association between group membership (VI/Sighted) and the children's ethnic background ($\chi^2_{(3)}$ = .142, p = .986), birth order ($\chi^2_{(1)}$ = .324, p = .569), number of siblings ($\chi^2_{(3)}$ = 1.410, p = .703), or the educational background of the mothers (the distributions of mothers' educational backgrounds was identical across the two groups)[18]. These details are given below in Table 5.2.

Table 5.2: *Demographic characteristics of the sample*

Demographic data (percentages)	***VI***	***Sighted***
Ethnicity		
White British	67 %	73 %
Black British	8 %	7 %
Asian	8 %	7 %
Mixed	17 %	13 %
Mothers' educational background [a]	N = 10 (missing 2)	N = 15 (missing 1)
Basic (up to A levels)	40 %	40 %
Higher (post A levels)	60 %	60 %
Number of siblings		
0	17 %	6 %
1	67 %	62 %
2	8 %	19 %
3	8 %	13 %
Birth order		
First child	42 %	30 %
Not a first child	58 %	70 %

[a] Although initially, the mother's educational levels were assigned to eight different categories (ranging from some secondary school education to postgraduate qualifications), they were re-assigned to two categories (basic and higher) because of the small numbers.

Materials

Following Symons et al. (2005), an illustrated children's book 'First Day Jitters' (Dannenberg, 2000) was used for the joint book-reading session between the children and their mothers. The book depicts a character dealing with the anxiety about the first day of school. Given the age range of the children in this study, the topic was seen as relevant to their experiences. The book also permitted a discussion about the internal mental states as the main theme in the story involves a case of mistaken identity revealed at the end of the book (i.e., the main character, who is anxious about her first day of school, is in fact a teacher, and not a child, as the reader is initially lead to believe). Importantly, since the book is unavailable for purchase in the UK, it was expected that none of the dyads would be familiar with it (which they all later confirmed).

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[18] The small sample size provided a limited opportunity to statistically examine the impact of these variables, which empirically have been found to be related to the levels of mothers' and children's mentalistic language. For that reason, here it was ensured that the groups were relatively comparable with regards to those variables.

Procedure

The task was carried out in the participants' homes. The children and their mothers were asked to spend some time reading the book in the manner that was most typical for them. They were told that the experimenter wished to obtain an insight into the language used between children and parents in a real-life situation and were asked to read and/or discuss the book content as they would usually do so, for example before bedtime or when looking at magazines. The parents were reassured that they would not be judged on their reading skills and that the experimenter was only interested in the way the dyads responded to each other in a naturalistic setting. In most cases, the experimenter left the room in order not to discourage the parents and to reduce the audience effect. In a minority of the cases however, the experimenter was present during the session. The dyads were tape-recoded throughout their book-reading session, which took seven minutes on average (VI group: Mean = 7.1, SD = 2.8; Sighted group: Mean = 6.5, SD = 3.9). There was no significant between-group difference in the duration of the joint book-reading session ($t_{(18)} = .391$; $p = .7$).

While the book reading in the VI sample was carried out by the parents for obvious reasons, it is worth noting that in the sighted group, the book reading was shared between the children and their parents and, in a couple of cases, it was carried out by the children themselves. This is not surprising, given the children's chronological ages and the fact that they were asked to discuss the material in a manner that was typical of them. However, in both groups the discussion about the story events and characteristics was facilitated by the mothers.

Scoring

All the speech produced by children and their mothers was tape-recorded for subsequent transcription. Only the language that was not read directly from the book, but was added by the parent or child was considered for coding and the subsequent analyses. First, the number of utterances that were relevant to the book's content was derived for parents and children respectively. Off-task utterances (such as those where dyads talked about the experimenter) were excluded automatically. An utterance was defined as a word or string of words identified by a pause or grammatical completeness (Symons et al., 2005). Then, for each partner, their utterances were examined and coded for the type of elaboration they contained. Even though in most cases the number of utterances was equal the number of elaborations ($r = .998$), for the purpose of the current study a distinction was made between utterances and elaborations because it was sometimes possible for one utterance to contain more than one elaboration, as will be explained below.

The elaborations were first classified broadly as either *mentalistic* or *non-mentalistic*. The elaborations were coded as *Mentalistic* according to the criteria for mental state language used by Ruffman et al. (2002), and in line with Bartsch and Wellman (1995) (Table 5.3). This included references to desires (e.g., '*She doesn't want to get up.*'), emotions (e.g., '*She seems quite scared.*'), modulations of assertion (e.g., '*I wonder why she's hiding.*'), think and know[19] terms (e.g., '*They're thinking hard.*') and other mental states (e.g. '*Do you remember your first day at school?*'). In the current study, a distinction between different types of mental states was not made and they were grouped together into a generic mental state category for subsequent between-group comparisons.

Table 5.3: *Examples of mental state language criteria used by Ruffman et al. (2002)*

Category	***Examples***
Desires	Want, like, love, hope, wish, dream, prefer, keen on
Emotions	Happy, sad, feel, cross, angry, grumpy
Modulations of Assertion	Sure, guess, figure, reckon, certain, suppose, wonder, expect
Think and know	Know, think
Other mental states	Remember, understand, forget

Non-mentalistic elaborations were classified as *descriptive* and *general*, following the categories specified by Symons et al. (2004). *Descriptive* elaborations involved language referring to behavioural and physical aspects of the story and the book which added a descriptive quality to the dyad's language (e.g., '*the girl has brown hair*'; '*the doggy is barking*'). *General* elaborations were all the other utterances that did not add to the descriptive value of the book-reading discourse (e.g., '*What's that?*', '*Oh, no!*', '*Let's continue*', etc). *Mentalistic* and *descriptive* elaborations appeared not to be exclusive and utterances containing both types of elaboration were often produced (e.g., '*do you think her heart is beating fast or slow?*'). On those occasions, the elaborations were assigned to both *mentalistic* and *descriptive* elaboration category. On their own, *general* elaborations were not examined further in the present study, and only *mentalistic* and *descriptive* elaborations were analysed in the subsequent between-group comparisons.

Each child and mother received a score for *mentalistic* and *descriptive* elaborations, expressed as a proportion of all elaborations (e.g., proportion *mentalistic* = total number of *mentalistic* / [sum of all elaborations: *mentalistic* + *descriptive* + *general*]). The proportional data were seen as more

[19] In line with Ruffman et al. (2003), 'I don't know' responses (i.e., responses that consisted of only these three words and which did not elaborate on what was unknown) were not treated as mentalistic because of their possible use to mean simply 'I can't answer'.

appropriate than frequency data for the current independent samples design as they were not susceptible to the confounding effect of mothers' verbosity (i.e., this ensured that the dyads' mentalistic language was not simply picking up on mothers' involvement).

In the first stages of the study, it was of interest to look at the incidence of references to mental states overall, for children and parents respectively, and a distinction was not drawn between mental state language relating to characters in the story and participants themselves. However, in further analyses, within the *mentalistic* language produced by the participants, mental state elaborations were classified as those referring a) to *self* (e.g., '*I don't remember seeing that*'), b) to *partner* (e.g., '*What do you think about this book?*'), c) to the *character* (e.g., '*She thinks it's horrible*') and d) *other,* less specific, mental state references (e.g., '*It's a mind trick*'; '*It's an idea.*'). These different mental state references were not exclusive. If two mental state elaborations were produced in one utterance (e.g., '*I think she's scared.*'), the responses were then assigned to both categories (e.g., '*I think*' = *self mentalistic*; and '*She's scared*' = *character mentalistic*). In the main, the *mentalistic* language produced by the dyads consisted of the more specific references made to *character*, *self* and *partner*, whereas *other* mental state references, which were more general, were made rarely. Only the incidence of mental state references to the former three types of referent was of interest for subsequent analyses. The scores for each of the three types of mental states in question (for children and mothers respectively) were expressed as proportions of all mental state elaborations (e.g., proportion of *self mentalistic* = number of *self mentalistic* / [total number of *mentalistic*: *character* + *self* + *partner* + *other*]).

An independent rater, unaware of the children's characteristics or the hypotheses of the study, coded approximately 50% of randomly selected transcripts from each vision group (see Appendix C1 for coding instructions, and Appendix C2 for an example of a coded transcript for a child with VI). Overall, the reliability ratings were high (Table 5.4).

***Table 5.4:** Inter-rarer reliability coefficients for mother and child elaborations during the joint book-reading task*

Elaborations	*Pearson's correlation coefficient r*
Mother	
Mentalistic (all)	.990
Self	.988
Partner	.942
Character	.961
Other	.402 [a]
Non-mentalistic (all)	.949
Descriptive	.929
General	.880
Child	
Mentalistic (all)	.889
Self	.492[a]
Partner	1 [b]
Character	.912
Other	.645 [a]
Non-mentalistic (all)	.999
Descriptive	.821
General	.977

[a] Certain elaborations were made rarely (e.g. mentalistic *self* and *other* for children and mentalistic *other* for parents) resulting in less variation, potentially explaining low inter-rater reliability on these components

[b] None of the children for whom the reliability rating was carried out made references to the mothers' mental states (i.e., partner) resulting in an inter-rater correlation coefficient of 1.

RESULTS

Table 5.5 shows the total number of elaborations produced by the children and mothers in each vision group, and the proportion scores for *mentalistic* and *non-mentalistic* elaborations. Here, it can be seen that in both groups, unsurprisingly, mothers elaborated on the book content more than did their children overall (VI group: $t_{(11)} = -5.440$, $p \leq .001$; Sighted group: $t_{(15)} = -3.229$, $p = .006$). In both groups, individual children and parents varied greatly in the number of elaborations they produced during the joint book-reading discourse. However, the overall numbers of elaborations spoken by children were comparable between the two groups ($t_{(26)} = .632$, $p = .533$, $d = .23$). Interestingly, the direction of the means in Table 5.5 suggested that the mothers of children with VI produced more elaborations overall than mothers of children who are sighted, although statistically this was only a trend with a medium effect size ($t_{(26)} = 1.811$, $p = .08$, $d = .67$). It is worth noting that one mother in the sighted group made many more elaborations than did the other mothers (3.5 standard deviations above the group mean, Figure 5.1). The *t* tests revealed a highly significant difference between the two groups on the total number of mothers' elaborations, after this particular dyad was excluded (equal variances not assumed: $t_{(13.8)} = 2.981$, $p = .01$), implying that the mothers of children with VI typically elaborated more during the book-reading session than the mothers of sighted children. Because of the small sample size, it was decided to retain the data of this parent (and her child) in the subsequent analyses on proportional data. Importantly, the results from the subsequent between-group comparisons on proportional data were the same with or without retention of the data belonging to this dyad.

Table 5.5: *Number of all on-task elaborations and the proportions of mentalistic and non-mentalistic elaborations for mothers and children in each group*

Measure	***VI***	***Sighted***
Mother		
All on-task elaborations - raw number		
Mean (SD)	75.3 (48.4)	42.1 (47.7)
Range	13 - 159	1 - 207
Mentalistic / Non – mentalistic [a]		
Means - proportions of all on task elaborations	.27 / .73	.34 / .66
(SD)	(.11)	(.12)
Child		
All on-task elaborations - raw number		
Mean (SD)	24.8 (21.8)	19.6 (21.3)
Range	1 - 56	3 - 89
Mentalistic / Non - mentalistic		
Means - proportions of all on task elaborations	.15 / .85	.12 / .88
(SD)	(.28)	(.13)

[a] Mentalistic + Non-mentalistic = 1 (100 %)

From Table 5.5 it can also be seen that *non-mentalistic* elaborations made up a large proportion of the dyad's language (over 80% for children and approximately 70% for mothers). Approximately a third of all the elaborations spoken by the mothers were those referring to mental states, compared to 12-15% of elaborations spoken by the children. A similar proportion of mentalistic language within the overall discourse produced by mother-child dyads during joint book-reading was reported previously with 5 - 7 year olds (i.e., 10% for the children and 28% for the mothers) (Symons et al., 2005) Therefore, the incidence of mental state language spoken by mothers and children in this study may provide a relatively realistic picture of the frequency with which mental state language occurs in parent-child conversations (i.e., in a context such as joint book-reading) in the general population.

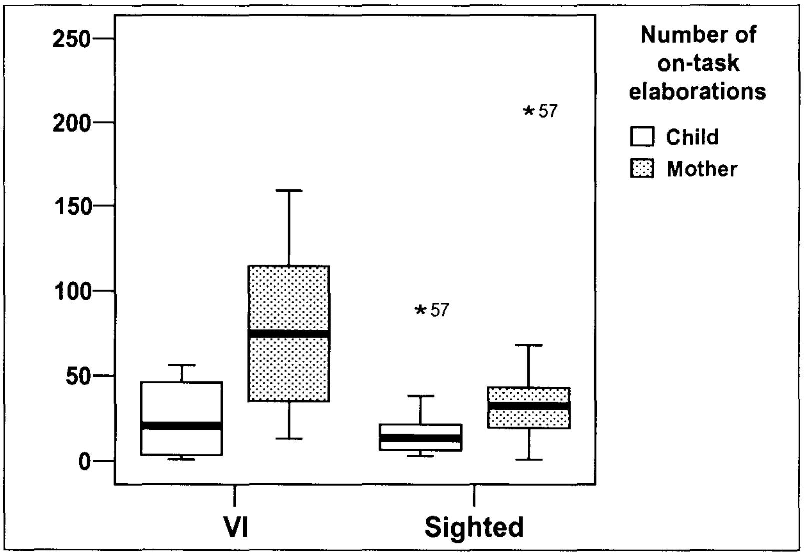

Figure 5.1: *Distribution of on-task elaborations for mothers and children in the two groups and an outlier dyad (57) in the sighted group*
(Note: black band represents the group median/ 50th percentile)

Between-group comparisons

A set of independent-samples *t* tests was carried out to examine the extent to which the two groups differed in the proportions of mentalistic and descriptive elaborations during the joint book-reading discourse, for mothers and their children respectively. The mean proportion scores of mental state and descriptive elaborations for children and mothers in each vision group are given in Table 5.6. It is interesting to note that there is a larger variation in the proportions of mentalistic language of children in the VI group, although this apparent difference in group variation was not found to be statistically significant (i.e., Levene's test for homogeneity of variances was not significant, $p = .395$).

Contrary to the experimental predictions (H1 and H2), there was no significant difference between the two groups in the proportions of mentalistic language spoken by mothers ($t_{(26)} = -1.619$, $p = .118$, $d = -.50$) or their children ($t_{(26)} = .455$, $p = .653$, $d = .14$). However, in line with H3 and H4, the two dyad groups differed with regards to their use of descriptive elaborations during the joint book-reading discourse. More specifically, while the sighted children's language contained significantly more descriptive elaborations than did the language of children with VI ($t_{(26)} = -2.617$, $p = .015$, $d = -.93$), the language of mothers of children with VI contained significantly more descriptive elaborations about the book than did the language of mothers of sighted children ($t_{(26)} = 2.855$, $p = .008$). These results are also graphically illustrated in Figure 5.2 and Figure 5.3.

***Table 5.6:** Proportions of mentalistic and descriptive elaborations for children and mothers in each group*

Proportion scores *Mean (SD)*	***VI***	***Sighted***	***p value***
Mother			
Mental States			
Mean (SD)	.27 (.11)	.33 (.12)	.118
Descriptions			
Mean (SD)	.49 (.20)	.29 (.17)	.008
Child			
Mental States			
Mean (SD)	.15 (.28)	.12 (.13)	.653
Descriptions			
Mean (SD)	.17 (.15)	.32 (.16)	.015

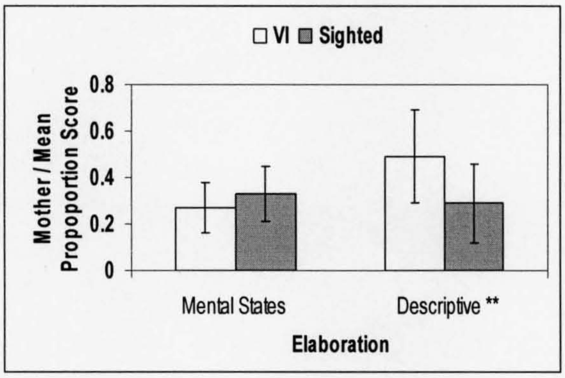

***Figure 5.2:** Mother elaborations*
*(error bars representing SDs, ** p ≤ .01)*

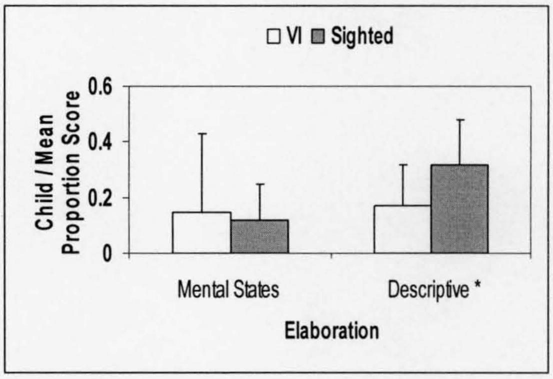

***Figure 5.3:** Child elaborations*
*(error bars representing SDs, * p ≤ .05)*

As an illustration of these findings, Table 5.7 shows an example of the mother-child discourse from each vision group about the same section in the book. More specifically, the two columns illustrate the differing quality of the discourse in the two vision groups, with an emphasis on the descriptive language produced by a mother of a child with VI in the left column, and the descriptive language spoken by a sighted child in the right column.

Table 5.7: *Examples of mother-child discourse*

VI group - boy, 9 years and 2 months	***Sighted group - girl, 7 years***
*I hate my new school, "Sarah said. She tunnelled down to the end of her bed.**	*I hate my new school, "Sarah said. She tunnelled down to the end of her bed.*
Mother: What does tunnelled down mean?	Mother: I still haven't seen Sarah, where is she?
Child: I don't know.	Child: There's her bed, and there's her pyjamas, and there's a big lump where she is...
Mother: She buried herself under the bed, hoping that her dad won't make her get out of bed. The cat's licking his paw on top of, I think, Sarah's bottom. 'Cause he's sitting on Sarah's duvet, and the father's got his hand on his head and he's thinking: 'Oh, my goodness'. And then he says:	Mother: I still don't know what she looks like, do you?
	Child: Just keep going so we can find out.
"How can you hate your new school, sweetheart?"	Mother: OK, let's see. The cat is so cute, isn't it?
	Child: It looks more like a bird to me.

*The text in *Italics* represents the text that is directly from the book

Mothers' mentalistic elaborations

To gain further insight into the nature of maternal mental state language input, a more detailed analysis of the mentalistic language produced by the mothers in the present study was carried out. It was of interest to examine whether the mothers in the two groups differed in the types of mental state references they made (i.e., difference in the proportions of references to the mental states of the story characters, mental states of the children and their own mental states) (Q1).

From Table 5.8 and Figure 5.4 it can been seen that in both groups the proportions of mothers' mentalistic language that is sensitive to the child's mental states (Mentalistic Partner, e.g., '*What do you think*?') were considerably larger than the proportions of mental state references made to self (Mentalistic Self, e.g. '*I'm wondering if...*') and the story characters (Mentalistic Character, e.g. '*She's frightened*'). More specifically, over 40 % of all mentalistic elaborations produced by mothers in both groups were those that referred to their child's mental state, implying that mothers generally may be sensitive towards their child's subjective beliefs, desires and emotions (in line with Meins et al.'s, 2003 concept of *mind-mindedness*). However, in relation to this, it may be worth noting that the variation in the VI group was substantially larger than in the sighted group (also Figure 5.4). Independent-samples *t* tests showed that the proportions of child-minded mentalistic language of the mothers (Mentalistic Partner) in the present study did not differ between the two vision groups, and the effect size was medium (equal variances not assumed: $t_{(16.2)}$ = - 1.519, p = .148, d = -.60). Additionally, the mothers in the two groups did not differ in the extent to which they referred to their own mental states (Mentalistic Self) ($t_{(25)}$ = .639, p = .529, d = .25). However, despite the heterogeneous variances in the two groups, the proportions of references to the mental states of the story characters were significantly higher in the mothers of children with VI, compared to the mothers of sighted children, and the size of this effect was substantial (equal variances not assumed: $t_{(14.3)}$ = 2.416, p = .03, d = .89).

Table 5.8: *Proportions of mothers' mentalistic elaborations*[a]

Proportion scores *Mean (SD)*	***VI***	***Sighted*** *N missing = 1*[b]	***p value***
Mentalistic Self	.19 (.11)	.16 (.12)	.529
Mentalistic Partner	.40 (.31)	.55 (.17)	.148
Mentalistic Character	.27 (.20)	.13 (.10)	.03

[a] Proportions ≠ 1, because the *other* mentalistic elaborations were not included in analyses (see Scoring section)
[b] One mother in the sighted group did not produce mentalistic language referring to self, partner or character, so her proportion scores for these elaborations could not be calculated.

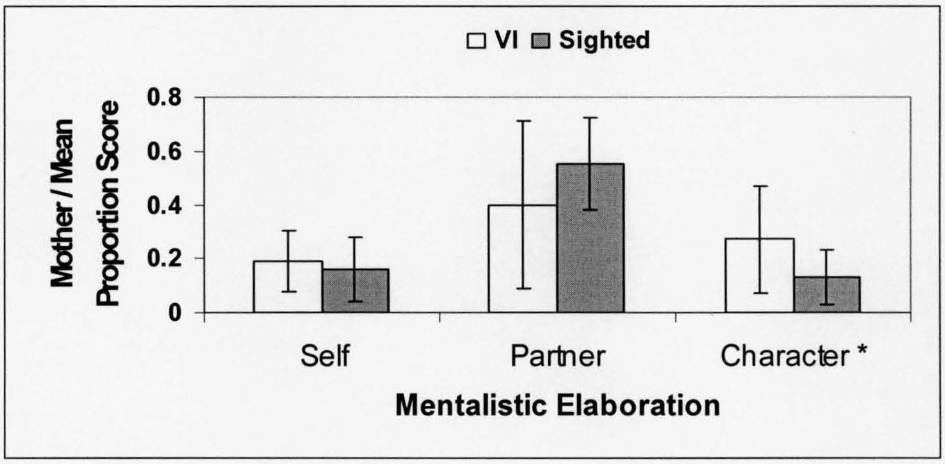

Figure 5.4: *Mean proportions of different mentalistic elaborations spoken by mothers (error bars representing SDs, * p ≤ .05)*

Children's mentalistic elaborations

As seen earlier from the proportion scores in Table 5.6, the proportions of mentalistic elaborations in the mothers' language were twice as large as the proportions of mentalistic elaborations in the overall language produced by the children. Children generally elaborated less on the book content than did their mothers, resulting in fewer data points, particularly in terms of their mentalistic elaborations. Furthermore, the proportion scores of mentalistic language referring to self, partner and character could only be calculated for 8 children in VI group and for 11 children in the sighted group (presented in Table 5.8 below), as some children did not produce any mentalistic language. However, while this was likely to be problematic in statistical terms, it was still of interest to explore whether the pattern of different mental state elaborations for children was similar to the pattern observed for their mothers (Q2).

Table 5.9: *Proportions of children's mentalistic elaborations*

Proportion scores *Mean (SD)*	***VI*** *N missing = 4*	***Sighted*** *N missing = 5*	***p value***
Mentalistic Self	.35 (.35)	.66 (.37)	.085
Mentalistic Partner	.06 (.13)	.02 (.05)	.323
Mentalistic Character	.43 (.36)	.19 (.29)	.119

Whereas the largest proportion of mothers' mentalistic language consisted of mental state references to partner (Table 5.8), from Table 5.9 above it can be seen that this type of elaboration made up the smallest proportion of children's mentalistic language. Here, it can also be seen that the mentalistic language of sighted children was largely about self, whereas the mentalistic language of children with VI was about the character and self. Independent-samples *t* tests showed that the groups did not differ with regards to the proportions of mentalistic language about partner (equal variances not assumed: $t_{(8.63)} = 1.047$, $p = .323$, $d = .34$) or character ($t_{(17)} = 1.643$, $p = .119$, $d = .71$), whereas there was a trend towards a significant between-groups difference in the proportions of mentalistic language to self, with a large effect size ($t_{(17)} = -1.829$, $p = .085$, $d = -.82$). It is possible that children who are sighted may reflect on their own mental states (e.g., '*I think that...*'), particularly as a result of the mother's child-minded mentalistic language (e.g., '*What do you think?*'), whereas children with VI may be more likely to rely on their mother's input, without explicitly reflecting on their own thoughts and feelings. However, considering the substantially small numbers of children on which these analyses were performed, and importantly, the scarcity of data points (particularly with regards to the mentalistic elaborations about partner), any further interpretations relating to these results would be speculative.

Relationship between mothers' and children's mentalistic discourse

Although the relationship between mothers' and children's mental state discourse was of particular interest in this study (H5), it was understandable that the small sample, as well as the general scarcity of language produced by the children, would pose a problem for conducting correlational analyses. The proportion scores in general, particularly those of the children, were not suitably distributed to allow correlational analysis. Unsurprisingly, when correlating the proportion scores for elaborations produced by children and mothers, no significant relationship between the mothers' and children's discourse was obtained for either mental or descriptive elaborations (r range = .195 - .308, p values ≥ .05). Hence, an attempt was made to illustrate the quality of mother-child language relationship by reverting to the absolute (raw) numbers of elaborations produced by the dyads.

Raw scores analysis

Firstly, there was a highly significant correlation between the mother's and children's overall numbers of on-task elaborations in both groups (VI: $r = .843$, $p \leq .001$; and Sighted: $r = .961$, $p \leq .001$), signifying that the quantity of children's discourse was directly related to the extent to which the mothers elaborated on the book content. This was not surprising, considering that the mothers consistently facilitated the discussion about the book. Using Fisher's Z transformation revealed that the correlations between mothers' and children's on-task elaborations were not of significantly different strength across the two groups (z-score = - 1.68, $p = .093$).

Mothers' and children's mentalistic (overall) and descriptive elaborations were then correlated in order to see whether these correlations would also reflect the same pattern of the mother-child language relationship. With regards to descriptive language during the mother-child discourse, there was a significant positive correlation between mothers' and children's total number of descriptive elaborations, both in the VI ($r = .624$, $p = .03$) and the sighted groups ($r = .940$, $p \leq .001$). This shows that the more descriptive input the mothers provided the more likely were the children to elaborate on the book content in the same manner. However, the correlation in the sighted group was significantly stronger than that in the VI group (z-score = - 2.3212, $p = .020$).

In terms of the mentalistic elaborations, in the VI group there was a highly significant positive correlation between the mothers' and children's mentalistic language ($r = .802$, $p = .002$) (Figure 5.5). Interestingly however, this correlation was not significant in the sighted group ($r = .202$, $p = .453$). From Figure 5.6 below it is clear that one mother in the sighted group was an outlier. As mentioned previously at the beginning of this section, the extent to which this particular mother spoke to her child in general distinguished her from the rest of the group (i.e., the overall number of elaborations she produced was 3.5 SDs higher than the mean of the sighted group), which would have naturally resulted in a distinctively high overall number of her mental state elaborations (Figure 5.1). Consequently, removing this dyad from the analyses resulted in a highly significant positive correlation between mothers' and children's mentalistic language in the sighted group (r = .648, p = .009), which was not of a significantly different strength to that observed in the VI group (z-score = 0.75, p = .451).

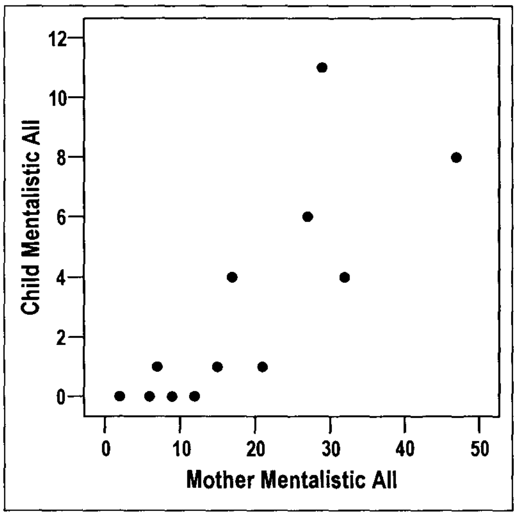

Figure 5.5: *Mother-child mentalistic language correlation in the VI group*

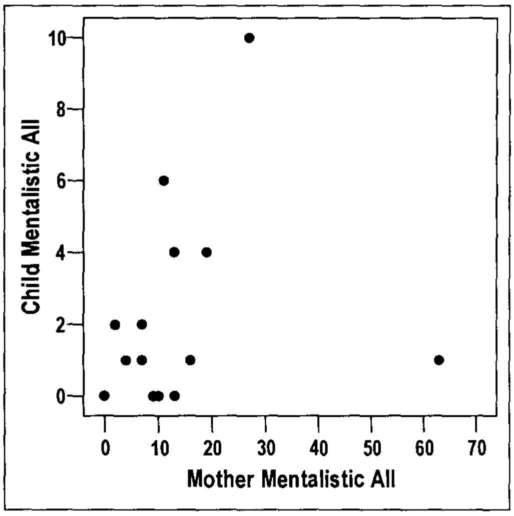

Figure 5.6: *Mother-child mentalistic language correlation in the sighted group*

It is clear from these correlations that the more mentalistic language the mothers produced, the more mentalistic language was also spoken by their children. However, given the highly significant relationship between the overall number of elaborations spoken by mothers and children (as well as the descriptive elaborations spoken by the dyads), it cannot be ascertained in this study whether the mothers' mentalistic language accounted for any unique variance (independent of their verbosity) in the relationship with the children's mentalistic language. The unsuitability of the current proportional data, particularly regarding the children, prevents us from answering this question at the present time. However, the current results confirm previous findings that the type of verbal scaffolding that is provided by mothers and which includes talk about thoughts, desires and feelings is an important feature of the child's learning environment in general and both children with and without sight equally benefit from it. A qualitative example illustrating how such scaffolding takes place can be seen in the two short extracts of mother-child discourse presented in Table 5.10. Given that this is the first study to date to investigate mentalistic language input to children with congenital VI, it was of particular interest to show examples of mentalistic language exchange between children with VI and their mothers.

Table 5.10: *Examples of mother-child mentalistic discourse in the VI group*

Girl with PVI, age 7 years
'They walked to the car. Sarah's hands were cold and clammy'
Mother: Why do you think that could be?
Child: I don't know.
Mother: Well, what makes your hands go cold and clammy? Can you think?
Child: When you're sick!
Mother: When you're sick, yeah. What else?
Child: I've no idea.
Mother: No idea? Do you ever get cold and clammy hands when you feel a bit nervous?
Child: Yeah!
Mother: Now, there you go.
Child: I felt well nervous...
Mother: When?
Child: When I went to that music thing, oh, my...
Girl with SVI, age 6 years and 6 months
Mother: Aaaah that might be what it is...jitters...
Child: What?
Mother: That means you're a bit nervous. First day at school. That might be why she's nervous.
Child: So she doesn't want to get up...
Mother: She doesn't want to get up. That's why she's got a pillow over her head, so she can't hear the alarm clock.

*The text in *Italics* represents the text that is directly from the book

Accounting for the VI severity

The numbers of children in each VI subgroup were very small (PVI n = 5; and SVI n = 7), preventing us from examining in more detail potential effects of the degree of VI on the mother-child discourse characteristics in this study (Q3). Thus, here non-parametric analyses were seen as more appropriate. The Mann-Whitney test revealed that the groups of children with PVI and

SVI did not differ significantly in terms of the overall number of elaborations produced by mothers (z = -.731, p = .465) or children (z = -.570, p = .568). Furthermore, the PVI and SVI groups did not differ significantly with regards to the proportions of mothers' mental state elaborations and descriptions (z = -.731, p = .465; and z = -.406, p = .685 respectively), or children's mental state elaborations and descriptions (z = -1.653, p = .1; and z = -.653, p = .1 respectively). These results imply that the extent to which mothers and children spoke and elaborated during the joint discourse was unrelated to the degree of the child's visual impairment. However, these analyses, with their small numbers, preclude us from drawing any firm conclusions.

DISCUSSION

The present study was the first to date to examine mothers' mentalistic language input to children who are visually impaired and the present findings provide evidence that the language input provided to children with VI is qualitatively different from that received by sighted children. However, the unique contribution of the current investigation is twofold. Firstly, it is shown that, in the context of a joint book-reading session, language input by mothers of children with VI consists of the same amount of mental state talk as does the language of mothers of sighted children. However, mothers of children with VI elaborate significantly more on the mental states of story characters than do mothers of children who are sighted. Furthermore, mothers of children with VI do not only seem to elaborate more than do mothers of sighted children in general, but these elaborations consist of significantly more descriptive information than the elaborations provided by mothers of sighted children.

The finding that the mothers in the two groups of children were comparable in the extent to which they elaborated on mental states is an important one. Approximately one third of all elaborations produced by mothers in both groups were about mental states. A similar proportion (i.e., 28%) of mentalistic language within the overall discourse produced by mothers during joint book-reading with their children (using the same story book as in the present study) was reported previously by Symons et al. (2005). Therefore, it appears that the extent to which mothers elaborate on mental states is a feature that is inherent in the language input that they direct to their children (at least in the context of joint book-reading behaviours), and the current findings imply that this important aspect of mothers' contribution to their child's learning may be unaffected by the child's sensory deficit. Furthermore, the finding that the mothers of children with VI refer to the mental states of the story characters more than do the mothers of sighted children may be of particular significance. It illustrates the sensitivity of mothers of children with VI in terms of their readiness to contribute to their child's understanding of important details of the social world (e.g., what other people are feeling or thinking), which for their children are otherwise difficult to access, and which sighted children may obtain spontaneously through vision (e.g., through observing facial expressions). The current findings therefore bear important implications for early intervention, where certain aspects of mother-child interaction should particularly be encouraged.

Interestingly, the finding regarding the mothers' mentalistic elaborations of story characters' mental states may throw some light on the findings presented in Chapter 4, where the children's own ability to refer to the mental states of story characters was investigated. To reiterate, the same children with VI who took part in the research presented in this chapter were also found to be comparable to the sighted controls in the extent to which they referred to mental states in their explanations of emotions of story characters. Perhaps it is not surprising to find that these children have good sensitivity towards other people's subjective mental states (at least in the context of the Emotion task, Chapter 4), because their mothers appear to be sensitive to the context in which they may need explicit encouragement. It is likely that the proclivity of the mothers of children with VI to elaborate on the mental states of others is beneficial to their children's understanding of those states. Perhaps they have learnt from their children that this is helpful to them. However, the causal nature of this relationship was not explicitly addressed in this research, and the data on children's own mentalistic language during the joint book-reading was not very revealing. Hence, we cannot be certain in this study to what extent the mothers' mentalistic language itself may be influenced by the children's level of socio-cognitive understanding. Nevertheless, the significant correlations that were found between the mothers' and children's overall language, including their mental state elaborations, are affirmative of an existing relationship that binds mother-child dialogue and interaction (for both children with VI and those who are sighted). Furthermore, while in this study it was not possible to isolate the unique contribution of the mothers' mentalistic language input (i.e., independent of mothers' verbosity) in the VI child's understanding of mental states, these correlations do offer an insight into the scaffolding mechanism that maternal language provides in this process. Alongside these correlations, the examples of mother-child dialogue in Table 5.10 provide further evidence for such scaffolding.

In addition to their mentalistic language input, the tendency of mothers who have a child with VI to provide a greater volume of descriptive language than do mothers of children who are sighted implies that maternal language input is largely adaptable to the child's needs, affirming its role in the child's learning processes. However, it was interesting to find that, in terms of the children's own discourse, sighted children elaborated on the descriptive aspects of the story-book to a greater extent than did the children with VI. These results are very meaningful, in that children with VI may produce fewer descriptive elaborations than their sighted peers because the information upon which to base such elaborations is not available to them. For this, they may instead rely heavily upon their parents' sensitivity to provide them with this information, resulting in mothers of children with VI describing people, objects and events to a greater extent than do mothers of children who are sighted. The descriptive nature of the language input provided by mothers of children with VI does seem to reinforce the notion that such mothers are able to adopt alternative strategies in order to bring the external events closer to the experiences of their child and this is likely to be facilitative of the child's development (Pérez-Pereira & Conti-Ramsden, 1999; Urwin, 1978). The current finding also may be particularly meaningful, given the evidence of impoverished descriptive language input to such children in early development (Kekelis & Andersen, 1984; V. Moore & McConachie, 1994).

The present findings have emphasised the strengths of the language input provided by the mothers of children with VI, which contrasts with some of the previous research which suggests that this input tends to be restricted (e.g., Andersen et al., 1993). However, the previous research studies, which addressed mother-child conversational characteristics involving children with VI, were carried out primarily with young and often pre-lingual children. Thus, we cannot be certain to what extent the level of mothers' language input in this research is determined by the children's own developmental level and the fact that these children are exceptionally verbal (as shown in Chapter 3). It is likely that the mothers' input would have been to an extent adapted to their child's level (in line with Taumoepeau & Ruffman, 2006). However, what remains uncertain is whether the same mothers would have experienced a certain level of difficulty when these children were much younger. This highlights the importance of longitudinal research in such processes (although this is a particular challenge with samples of children with VI). Another uncertainty stems from the fact that the aspects of maternal language input to children with VI that have previously been identified as an area of weakness (e.g., the level of maternal directiveness, control and responsiveness) were not assessed in this study.

Another factor that may have impacted on the current findings is the book-reading context itself. Although the strengths of this context had been identified previously (Dyer et al., 2000; Symons et al., 2005), the shared book narrative prevented us from drawing any firm conclusions regarding the children's own discourse (which was relatively scarce) or the mother's language input in other contexts. One reason for the reduced output by children in this study is that the children were too old to be stimulated by the task[20]. Another reason is that the nature of the task may have required the children, particularly those with VI, to be passive by default, despite their good verbal skills. Thus, the reduced verbal contribution by the child would have been likely to affect, at least to some extent, the level of parental involvement. Future studies therefore may benefit from a different context for examining the mother-child mentalistic discourse. For instance, while the method used by Ruffman et al. (2002) (whereby mothers were asked to discuss photographs depicting mental state scenarios with their children) may impose the same constraints on the mother-child dialogue as the book narrative, this method could be adapted to be more suitable for use with school-age children. For example, mothers could be provided with a set of topics (e.g., topics about friends, family members and familiar events, such as holidays) and encouraged to facilitate a discussion with their child, improving the ecological validity of the assessment context.

Finally, it is important to reflect on the current findings once more, from an intervention perspective. Despite the methodological limitations highlighted above, assessing the characteristics of mother-child discourse in a joint book-reading context has thrown a positive light on the mother-child relationship that involves children with VI. This is crucial, given that this relationship has been given a central role in the socio-cognitive development of typically developing children. Although the constraints that visual impairment may impose upon this relationship may indeed be present in the early years (Andersen et al., 1993; V. Moore & McConachie, 1994), the current findings suggest that, given a suitable context, maternal input to children with VI may receive a positive boost. For this reason the mothers' verbal involvement with their children with VI, while sharing book-reading time, should be highlighted as a strength that can be capitalised on, implementing it as an intervention strategy as early as possible.

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[20] In the sighted group, for instance, even the youngest child (approximately age 6) would have been a competent print reader, impacting on the dynamics of the joint book-reading context.

Chapter 6

Neuro-cognitive Processes in Children with Congenital VI: Implications for Executive Functioning and Attention

Thus far, this thesis has focused on socio-developmental outcomes in children with VI. In relation to this, it has been argued that poor social communication and autism-related characteristics in children with congenital VI may be a consequence of disruptions to joint visual attention in early childhood and related inability to participate in social attention sharing through eye-contact and gestures. Hence, despite normal intelligence, proficient language skills and adequate socio-conversational input provided by the caregiver (as it has been demonstrated by the present research), such children are likely to experience difficulties in social relating long term. Hobson's account of vision-driven interpersonal engagement in childhood has played a key part in explaining the nature of social impairments in children with VI in general, as well as in the current study. However, this account does not provide a sufficient explanation for why some children with VI, despite intact intelligence and regardless of their VI severity, have more difficulties than others in relating to other people, and why some children with VI do not seem to show marked difficulties with social engagement and/or autistic-like characteristics. It is therefore important to ask what other underlying mechanisms may play a part in their socio-developmental outcomes.

One such mechanism that has been linked to social functioning in sighted children, including the achievement of the milestones such as joint attention and theory of mind, is the executive function (Carlson & Moses, 2001; C. Hughes, 1998a; Pennington & Ozonoff, 1996). Executive function (EF) is a broad neuro-cognitive construct used in psychology to refer to goal-directed and problem-solving behaviours that are thought to be mediated by the frontal lobes and which are involved in the processes of planning, impulse control, inhibition, the deployment of attention, mental flexibility, working memory, the initiation of activity and monitoring of action (Rabbit, 1997; Stuss & Knight, 2002). Rather than a single neuro-cognitive function, EF is best described as a collection of neuro-anatomically related processes with a common need to regulate behaviour and organise cognitive activity (Duncan, Emslie, Williams, Johnson, & Freer, 1996; Stuss & Knight, 2002).

Importantly, executive functioning is not confined to cognitive processes and behavioural action, and has been implicated also in emotional control and social interaction. Evidence for the relationship between social and executive functioning comes from several studies with sighted children who are typically developing and also from children with autism (e.g., Carlson, Mandell, & Williams, 2004; Carlson, Moses, & Breton, 2002; Gilotty, Kenworthy, Sirian, Black, & Wagner, 2002; McEvoy, Rogers, & Pennington, 1993; Ozonoff & McEvoy, 1994). While this has important implications for children with VI, understanding how difficulties at the level of EF may contribute to impairments in social functioning in such children is a great challenge, particularly because very little is known about EF skills in this population. Although some inferences can be made from the measures that indirectly assess certain executive processes (e.g., verbal fluency and digit-span performance on intelligence tests), no research studies to date have explicitly addressed executive capacity and control in children with congenital VI. However, given the developmental vulnerabilities in social communication and social cognition in this population of children and their similarities with autism, it is plausible to suggest that such vulnerabilities may link in to some underlying neuro-cognitive factors that involve the executive processes of cognitive flexibility and attentional control. The aim of the research presented in this chapter was to provide a preliminary insight into this area of development in children with VI. Following from this, the data from two research investigations relating to EF processes in children with VI are presented. The first provides a broad outlook onto a range of EF skills in children with VI, in terms of the relevant everyday behaviours seen at school age (Part 1). The second investigation, which takes a retrospective approach, has a more specific focus on attentional capacity and control in the pre-school stages of development in young children with VI (Part 2). Finally, the findings from these two investigations are synthesised in Part 3 for further insight and clarification.

PART 1: EVERYDAY EXECUTIVE FUNCTION BEHAVIOURS IN CHILDREN WITH CONGENITAL VI

INTRODUCTION

Executive functions develop continuously and rapidly through childhood and adolescence and, although many of EF aspects do not appear to mature until later childhood, it is now believed that their development begins in early infancy (Diamond, 1991; Diamond & Goldman-Rakic, 1989; Welsh & Pennington, 1988). For instance, maturational changes in the ability to hold a goal in mind in the absence of external cues, and to use that remembered goal to guide behaviour, are evident in infants between seven and twelve months of age (Diamond & Goldman-Rakic, 1989). Although inhibiting previously learned responses is difficult for infants younger than nine months of age, by twelve months, most children are able to inhibit certain behaviours and shift to a new response set (Diamond, 1985; Diamond & Doar, 1989). Importantly, such developments in EF coincide with the period of emergence of joint attention skills in young children (Butterworth & Grover, 1990). Similarly, developmental changes in attentional systems, which are an integral part of general executive functioning (P. Anderson, 2003), seem to overlap with and/or precede socially-based attention sharing in the context of joint attention; before children can co-ordinate attention between people and objects for the purpose of referential communication, they require more basic attentional capacity and the voluntary control involved in following attentional cues such as direction of eye-gaze and head-turn (Atkinson et al., 1992; Butterworth & Cochran, 1980; Hood et al., 1998). As well as with early social communication, EF has been linked to children's theory of mind development and a number of empirical studies to date have demonstrated a close relationship between flexible goal-directed behaviour in children, such as self-monitoring, working memory, planning and cognitive flexibility, and performance on theory of mind tasks (Carlson et al., 2004; Carlson & Moses, 2001; C. Hughes, 1998a, 1998b). Additionally, EF has been implicated in pragmatic language functions, which depend on adaptive responses to changes in conversational topic, planning a coherent narrative and monitoring the consequences of particular speech acts (Martin & McDonald, 2003).

Importantly, the theory of EF deficit has played a role in explaining some of the key features of autism, most notably restricted interests, behavioural stereotypes and perseveration (reviewed by Hill, 2004). Research has suggested that such behavioural features in autism mirror executive dysfunctions such as poor mental flexibility on tasks that employ the ability to shift to a mental set depending on the changes in a given situation (Ozonoff & Jensen, 1999; Ozonoff et al., 1991; Prior & Hoffmann, 1990), failure in self-monitoring on tasks that require error correction and avoidance (J. Russell & Jarrold, 1998), difficulties with strategic planning (C. Hughes, Russell, & Robbins, 1994; Ozonoff & McEvoy, 1994; Ozonoff et al., 1991), difficulties with generating novel ideas (Turner, 1999), and impairments in inhibition of prepotent responses (C. Hughes & Russell, 1993; J. Russell, Hala, & Hill, 2003). As well as accounting for some of the non-social characteristics of autism, the specific executive dysfunctions in autism have proven useful in explaining the socio-communicative problems that typify the disorder. Analogous to the demands of complex EF tasks, dealing with the demands of the social context, which is an autism-related challenge, also requires executive capacity and control (e.g., on-line planning, regulation of social behaviour, shifting of conversational topics to deal with continuously changing contextual demands, holding social information in mind while processing the dynamic features of the social world and inhibiting socially inappropriate responses). Not surprisingly, research has demonstrated connections between executive function difficulties in autism and joint attention impairments (Dawson et al., 2002), poor socio-communicative competence (Gilotty et al., 2002; McEvoy et al., 1993) and theory of mind deficits (Ozonoff et al., 1991).

Although its causality is not yet well understood, the developmental link between executive and social functioning is a relatively robust finding. This certainly has intriguing implications for children with VI, although very little is known about their executive function abilities in general. Assessing executive functions in children with VI poses a particular challenge as the existing structured measures of the relevant cognitive processes and behaviours are mostly of visual nature. While future research is required to adapt such measures for use with children with VI, the present research has employed a parental/teacher report of EF behaviours that can be observed in everyday context, in home or school environments. An advantage of sampling children's everyday EF behaviours in a naturalistic setting is that such a method has greater ecological validity and generalizability value than performance-based neuropsychological measures (V. A. Anderson, Anderson, Northam, Jacobs, & Mikiewicz, 2002; Gioia, Isquith, Kenworthy, & Barton, 2003). For the purpose of the present research, the recently developed and validated Behavioural Rating Inventory of Executive Function (BRIEF) (Gioia, Isquith, Guy, & Kenworthy, 2000) has been utilized. The BRIEF has not only been shown to be a developmentally appropriate measure for assessing day-to-day executive functioning that may have important implications for clinical diagnosis and treatment management of individual children; its utility has also been demonstrated as a comprehensive battery of wide-range developmentally-related domains, which allows for comparative description of EF profiles across different clinical populations (e.g., autism, attention deficit and hyperactivity disorder - ADHD, traumatic brain injury - TBI, reading disorder) (V. A. Anderson et al., 2002; Gioia et al., 2003; Gioia, Isquith, Retzlaff, & Espy, 2002).

Behavioural rating inventories are not without their limitations, and ideally should be utilized in conjunction with performance-based measures. However, the current investigation is an exploratory one. Its aim was to provide a preliminary insight into executive functioning in children with VI, by comparing their EF-related behavioural outcomes with those of age and ability matched children who are sighted.

Research questions and hypotheses

The following hypotheses and the related questions were addressed in the research presented in this chapter:

Hypothesis 1 (H1): The behavioural profiles of executive function, as measured by the BRIEF, in children with VI will differ from the profiles of sighted children.

Question 1 (Q1): Will this difference vary as a function of a specific BRIEF subscale?

Question 2 (Q2): Will this difference be confined to a particular BRIEF domain (e.g., behavioural vs. metacognitive)?

Question 3 (Q3): Can verbal ability, chronological age and severity of VI explain the variation in the BRIEF profiles in the VI group?

Hypothesis 2 (H2): Children with VI will be more likely to show executive function behaviours of clinical significance than their sighted developmentally matched peers.

METHOD

Participants

The participants were the same children as those who took part in the research presented in the previous chapters, except for one child in the sighted group whose parents did not complete the BRIEF. Hence, BRIEF ratings were obtained for 15 children with congenital VI and 25 children with normally developed vision. After the sighted child, whose BRIEF data were missing, was removed from the dataset, the two groups remained well matched on verbal IQ ($t_{(38)} = -.185$, $p = .855$), chronological age ($t_{(38)} = -.445$, $p = .659$) and gender ($\chi^2_{(1)} = .242$, $p = .622$). Average VIQ across the two groups was 106.4 (SD = 10.9) and the mean chronological age was 8 years and 3 months (SD = 1.7, range = 6 years and 3 months - 12 years and 11 months). In both groups there were somewhat more girls than boys (VI group gender ratio: 9/6; Sighted group gender ratio: 13/12).

Materials and procedure

The Behaviour Rating Inventory of Executive Function/BRIEF (Gioia et al., 2000) was used as a measure of executive functioning. The BRIEF is a questionnaire for parents and teachers of children ages 5-18 years, designed specifically for assessing children's everyday executive function behaviours in the home and school environments. In the present study, the BRIEF questionnaire for each child was completed by parents, except in the case of two children with VI whose BRIEF ratings were obtained by a teacher who knew the children well.

Table 6.1: *Domains measured by the BRIEF and the corresponding scales*

Measure	***Item description***	***Item example***
Behavioural Regulation Index	Sum of scales a) – c)	
a) Inhibit	Control impulses, appropriately stop behaviour at the proper time	Interrupts others
b) Shift	Move freely from one situation to another; transition; flexible problem-solving	Has trouble getting used to new situations (classes, groups, friends)
c) Emotional Control	Modulating emotional responses appropriately	Overreacts to small problems
Metacognition Index	Sum of scales d) – h)	
d) Initiate	Begin activity and generate ideas independently	Has trouble organizing activities with friends
e) Working Memory	Hold information in mind for purpose of completing a task; stick to an activity	When given three things to do, remembers only first or last
f) Plan/Organize	Anticipate future events; set goals; develop appropriate steps ahead of time; grasp main ideas	Underestimates time needed to finish tasks
g) Organization of Materials	Keep work space, play areas and materials in an orderly manner	Leaves playroom a mess
h) Monitor	Check work; assess own performance; keep track of the effect of own behaviour on others	Does not notice when his/her behaviour causes negative reactions
Global Executive Composite	Sum of BRI and MI	

The BRIEF questionnaire contains 86 items grouped into eight non-overlapping clinical scales, which are theoretically and statistically grounded, and which measure distinct aspects of executive functioning (scales a – h in Table 6.1). On each item, the rating of frequency of occurrence of a relevant behaviour is made on a 3-point scale, with 1 corresponding to *Never*, 2 corresponding to *Sometimes* and 3 corresponding to *Often*. These ratings are added to derive a raw score for each clinical scale, from which further composite scores can be calculated. More specifically, the raw scores on scales a) – c) are added to derive a Behaviour Regulation Index (BRI) and the scales d) – h) are summed to derive a Metacognition Index (MI). The sum of these two indices is in turn calculated to derive the Global Executive Composite (GEC) (Table 6.1). Raw scores on the clinical scales and the indices are then transformed into standardised (*T*) scores, using age and gender appropriate norms. The *T* scores, which are used to interpret the child's' level of executive functioning, have a mean of 50 and a standard deviation of 10. Higher raw and *T* scores indicate greater executive dysfunction. Scaled scores of 65 or above ($T \geq 65$) are considered to be of potential clinical significance.

To derive a *T* score for each clinical scale on BRIEF the parental/teacher responses on no more than two items that contribute to a scale raw score should be missing. In the case of one or two missing responses that contribute to a scale raw score, the items with missing responses are assigned the lowest value (i.e., 1 corresponding to *Never*) in order to calculate the scale raw score and proceed with deriving the *T* score for that scale. With regards to the current study, all but two items on the BRIEF were considered appropriate for use with children with VI, as a substantial number of parents of children with VI felt these two items did not apply to their child and omitted completing them. These items were Item 31 ('*has poor handwriting*') from the Monitor scale, and Item 53 ('*written work is poorly organised*') from the Plan and Organise scale. For that reason, in order to avoid systematic data skewing, each child in the study (both in the VI and Sighted groups) was automatically assigned a value of 1 for each of these two items. In cases of children where more than two items that contribute to a scale score were missing, raw and scaled scores for that scale, or the relevant indices, were not calculated. This was the case for one sighted child only, for whom the scaled scores on Organisation of Materials, and subsequently MI and GEC, could not be calculated.

Before interpreting BRIEF scores, it is possible to ensure validity of the data provided by the teachers and the parents, by considering the Inconsistency (i.e., the extent to which the respondent answers similar BRIEF items in an inconsistent manner relative to the clinical samples) and Negativity scales (i.e., the extent to which the respondent answers selected BRIEF items in an unusually negative manner relative to the clinical samples). All of the questionnaires provided by the parents and teachers in the present study passed the validity checks specified by the BRIEF manual.

Although the BRIEF is a useful screening measure of possible executive dysfunction, it is not an appropriate diagnostic tool. For the purpose of diagnosing a disorder of executive dysfunction the BRIEF should be accompanied by a full clinical assessment that includes a detailed history of the child and the family, performance-based testing and behavioural observation (Gioia et al., 2000).

In line with this, the purpose of using the BRIEF in the current study was not to establish the presence of executive dysfunction in children with VI, but to gain an insight into the children's everyday behaviours associated with specific domains in self-regulated problem solving and cognitive functioning.

RESULTS

The means and SDs of the two groups on the standardised (*T*) scores on the individual BRIEF scales, as well as the BRIEF index and composite scores, are summarized in Table 6.2. Here it can be seen that the mean *T* scores in both groups are generally within the average range ($T \leq 65$) although, relative to the sighted group, the scores of the VI group appear particularly elevated in certain domains. The SDs in the VI group are also somewhat wider than those seen in the sighted group, which is not surprising given the differing sample sizes in the two groups. However, Levene's tests of heterogeneity of variances between the two groups for each scale were not statistically significant (*p* values ranging from .09 - .86).

Table 6.2: *Mean standardised (T) scores on the BRIEF scales and indices*

Measure *Mean (SD)*	***VI*** *N=15*	***Sighted*** *N=25*	***p value***
Behavioural Regulation Index (BRI)	57.4 (11.3)	47.8 (9.3)	.007
Inhibit	52.7 (11.8)	48.3 (7.8)	.164
Shift	61.3 (11.6)	49.8 (11.1)	.003
Emotional Control	57.0 (10.8)	48.4 (8.1)	.007
Metacognition Index (MI)	53.1 (9.1)	47.8 (8.3)	.07
Initiate	55.9 (10.7)	47.4 (8.1)	.007
Working Memory	51.3 (9.5)	49.9 (9.5)	.652
Plan/Organise	52.5 (6.6)	49.3 (6.8)	.156
Organisation of Materials	52.5 (11.9)	50.9 (10.2)	.652
Monitor	50.9 (10.6)	43.1 (8.8)	.016
Global Executive Composite (GEC)	55.2 (8.4)	47.9 (8.6)	.014

First, it was of interest to examine and compare the pattern of scores (i.e., profiles) of the children in the VI and the Sighted groups across the eight distinct clinical scales and a Profile Analysis[21] was carried out with this purpose (H1 and Q1). The Profile Analysis comparing the behavioural profiles of the two groups across the BRIEF scales revealed a significant test of Flatness, indicating that when averaged across the two groups the children's performance differed across individual clinical scales (Pillai's Trace criterion: $F_{(7, 31)} = 3.011$; $p = .016$). Furthermore, the test of Levels revealed a significant difference between the two groups when their scores were averaged across BRIEF scales ($F_{(1, 37)} = 6.95$; $p = .012$). These tests are qualified by a significant test of Parallelism, indicating distinguishable executive functioning profiles between the two groups (Pillai's Trace criterion: $F_{(7, 31)} = 2.375$; $p = .046$). In summary, in line with the experimental prediction (H1), the Profile Analysis results suggest that the two groups differed significantly in terms of their executive function profiles as measured by the BRIEF. However, this between-group difference varied as a function of particular BRIEF scales on which specific executive function behaviours of the children were rated (Q1).

The differing BRIEF profiles in the two groups are graphically illustrated in Figure 6.1. This figure also highlights the results of the post hoc analyses aimed at testing which specific BRIEF subtests differentiated the two groups (Q1). More specifically, the unrelated t-tests (which were carried out following the significant test of Parallelism) revealed that the individual BRIEF scales, which differentiated children with VI as having significantly more behavioural difficulties than sighted children, were Shift ($t_{(38)} = 3.116$, $p = .003$), Emotional Regulation ($t_{(38)} = 2.852$, $p = .007$), Initiate ($t_{(38)} = 2.854$, $p = .007$), and Monitor ($t_{(38)} = 2.516$, $p = .016$; $d = .80$). Additionally, the t-tests also revealed that the children in the VI and the Sighted groups did not differ on Inhibit ($t_{(38)} = 1.419$, $p = .164$; $d = .46$), Working Memory ($t_{(38)} = .455$, $p = .652$; $d = .14$), Plan/Organise ($t_{(37)} = 3.116$, $p = .156$; $d = .47$), and Organisation of Materials scales ($t_{(38)} = .455$, $p = .652$; $d = .14$).

[21] See Chapter 3, p. 79 for details on Profile Analysis.

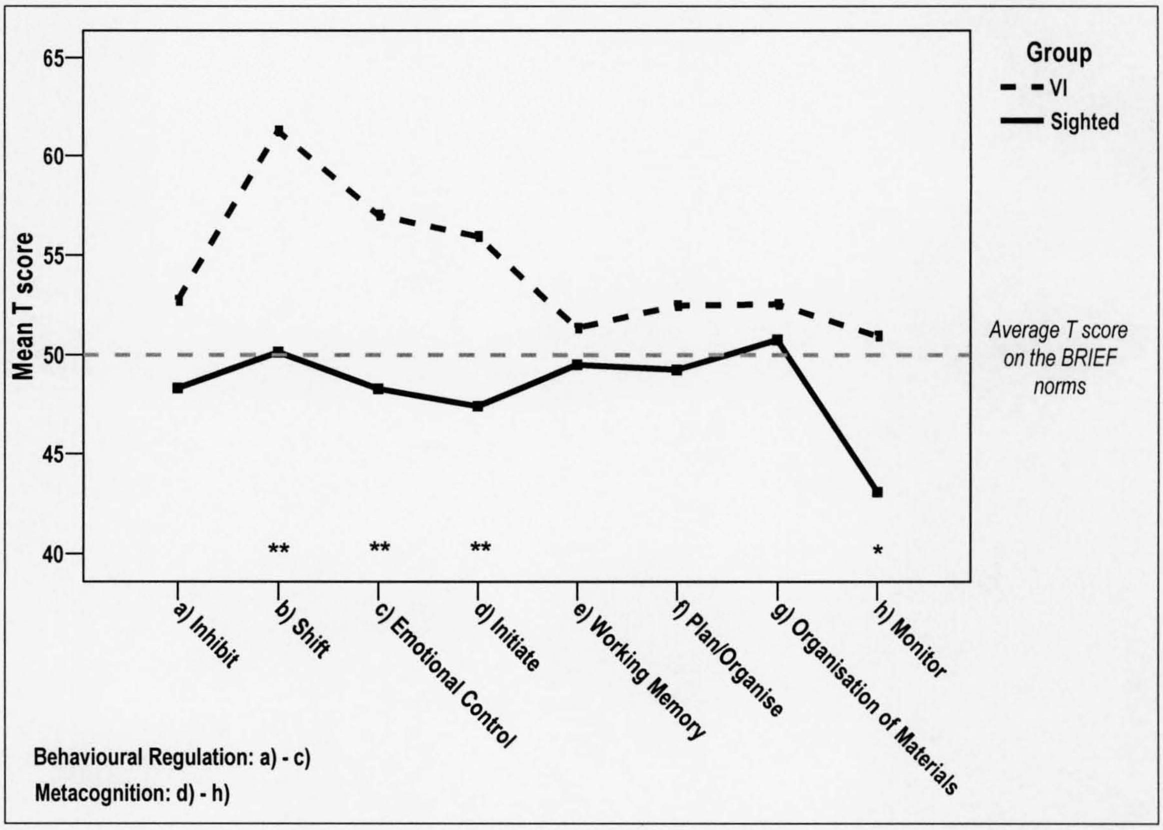

Figure 6.1: *Differing profiles between the two participant groups across the individual BRIEF scales (** p ≤ .01; and * p ≤ .05)*

In response to Q2, a mixed 2 x 2 ANOVA showed that, even thought the groups differed significantly on the BRIEF overall (i.e., BRI and MI combined) (i.e., significant main effect of Group: $F_{(1, 37)}$ = 7. 353; *p* = .010), this difference did not appear to be confined to a particular BRIEF domain (i.e., non-significant Group x Index interaction: $F_{(1, 37)}$ = 1. 716; *p* = .198). Furthermore, the BRI and MI scores were not discrepant when averaged across the two groups (non-significant effect of Index: $F_{(1, 37)}$ = 2 .080; *p* = .158). In line with these findings, it was not surprising to find a significant post hoc between-group difference on the measure of overall executive functioning, with the VI group obtaining higher GEC scores than the Sighted group ($t_{(37)}$ = 2.577, *p* = .014; *d* = .83). However, with regards to the post-hoc tests for the individual indices that form the GEC, the results were significant only with respect to the BRI ($t_{(38)}$ = 2.847, *p* = .007), while the difference on the Mi was only a strong trend ($t_{(37)}$ = 1. 842, *p* = .07; *d* = .60). This pattern of findings is graphically illustrated in Figure 6.2.

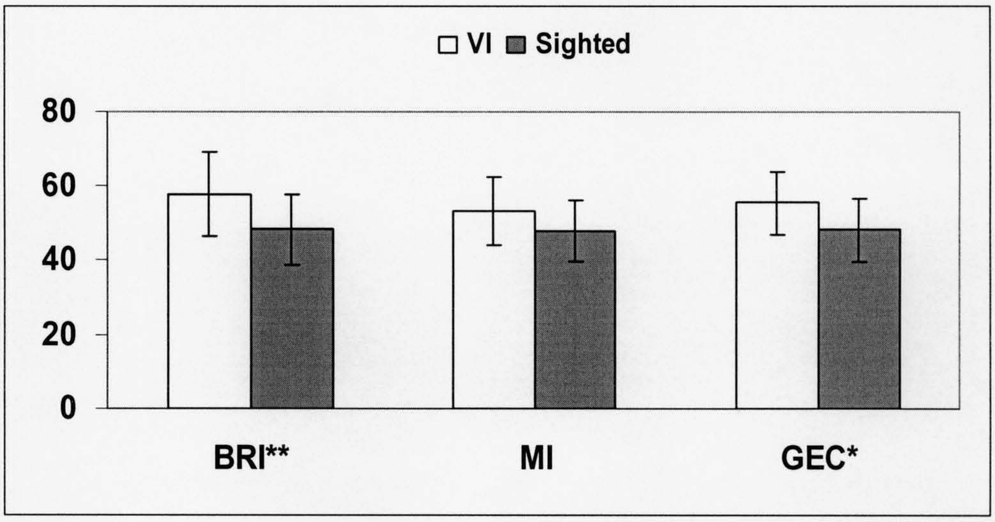

Figure 6.2: *Between-group differences on the BRIEF index and composite T scores (error bars represent the SD, ** p ≤ .01, * p ≤ .05)*

Furthermore, in response to Q3, the children's behavioural ratings on the BRIEF appeared to be independent of their verbal intelligence, as no significant correlations between the VIQ and the BRIEF *T* scores was found in either the VI group (*p* range = .254 - .987, n = 15), nor in the sighted group (*p* range = .317 - .967, n = 25) (Table 6.3). This may have been due to relative homogeneity of the two groups in terms of their intellectual levels. Interestingly, age was largely uncorrelated with BRIEF ratings in the VI group (except for the Organisation of Materials scale), unlike the sighted group where a clearer developmental pattern emerged across the BRIEF scales (i.e., negative correlations suggesting that the older the children, the better their EF skills). Thus, verbal ability and chronological age do not seem to explain the variation in the severity of the BRIEF profiles in the VI group (Q3).

Furthermore, in response to Q3, it may be worth noting that the clinically elevated scores on either one or both of the BRIEF indices were not confined to children whose VI was of greater severity, as out of the four children with clinically elevated indices, only one child had PVI based on his preschool functional vision assessment (participant ID: 19). Additionally, some of the best BRIEF outcomes overall were observed in children with PVI (i.e., participant IDs: 10 and 16). Similarly, the Shift scale (in terms of the prevalence of clinically significant scores), which most notably differentiated the VI group from the sighted, did not significantly distinguish children with PVI from those with SVI ($\chi^2_{(1)}$ = .227, *p* = .634).

Table 6.3: *Pearson coefficients for the correlations between age, VIQ and the BRIEF T scores*

		1	2	3	4	5	6	7	8	9	10	11	12	13
1	Age		.046	-.482	-.214	-.470	-.449	-.348	-.497**	-.608**	-.173	-.533**	-.519**	-.554**
2	VIQ	-.434		-.201	.052	.208	.009	-.209	.153	-.017	.071	-.143	-.046	-.031
3	Inhibit	-.169	-.005		.622**	.671**	.900**	.776**	.395	.660**	.473	.772**	.734**	.874**
4	Shift	-.115	.048	.537		.599**	.794**	.499**	.288	.216	.340	.388	.429	.674**
5	Emotional Control	.157	.025	.790**	.570		.855**	.593**	.374	.389	.357	.422	.502**	.724**
6	BRI	-.030	.018	.919**	.757**	.920**		.742**	.436	.514**	.448	.654**	.674**	.884**
7	Initiate	-.086	-.222	.425	.454	.362	.465		.500**	.493**	.520**	.548**	.737**	.796**
8	Working Memory	-.136	-.050	.349	.307	.101	.289	.740**		.543**	.709**	.533**	.858**	.740**
9	Plan /Organise	-.219	-.167	.100	.182	.018	.109	.691**	.537		.573**	.623**	.794**	.726**
10	Organising Materials	-.537	.248	.105	-.074	-.182	-.041	.343	.508	.618		.373	.825**	.741**
11	Monitor	-.381	.315	.404	.339	.211	.362	.394	.605	.465	.712**		.756**	.765**
12	MI	-.307	.033	.342	.252	.120	.277	.786**	.854**	.808**	.797**	.771**		.933**
13	GEC	-.269	-.311	.745**	.594	.586	.742**	.796**	.751**	.627	.535	.737**	.847**	

Red – VI; Blue- sighted; ** - significant at $p \leq .01$

The Pearson's correlations testing the relationship between the BRIEF scores in each group (Table 6.3) further support the differing profile pattern in the two groups (H1). In the sighted group, children's BRI and MI scores were highly correlated with one another (r = .674, $p \leq .001$, n = 24), as well as individually with the GEC (BRI: r = .884, $p \leq .001$, n = 25; MI: r = .933, $p \leq .001$, n = 24). However, in the VI group, while children's BRI and MI scores were each correlated with the GEC (BRI: r = .742, p = .002, n = 15; MI: r = .847, $p \leq .001$, n = 15), they did not correlate with one another (r = .277, p = .318, n = 15). This suggests that, relative to the presentation in the Sighted group, elevated scores on one BRIEF index did not necessarily imply elevated scores on the other in children with VI, and vice versa. This is also evident from the correlations on the individual clinical scales in Table 6.3. Although the clinical scales that form BRI and MI were inter-correlated in both groups within each domain, significant correlations across the scales in two domains were only seen in the sighted group. In the VI group, the correlations between the scales that form the BRI and those that form the MI were generally not significant, suggesting independence of the processes involved in behavioural regulation and metacognition in children with VI. However, the scatter-plot presented in Figure 6.3 implies that the discrepant pattern of EF skills across the two broader BRIEF indices in the VI group may be confined to a certain percentage of children with VI with clinically elevated scores in one index domain, but not in the other.

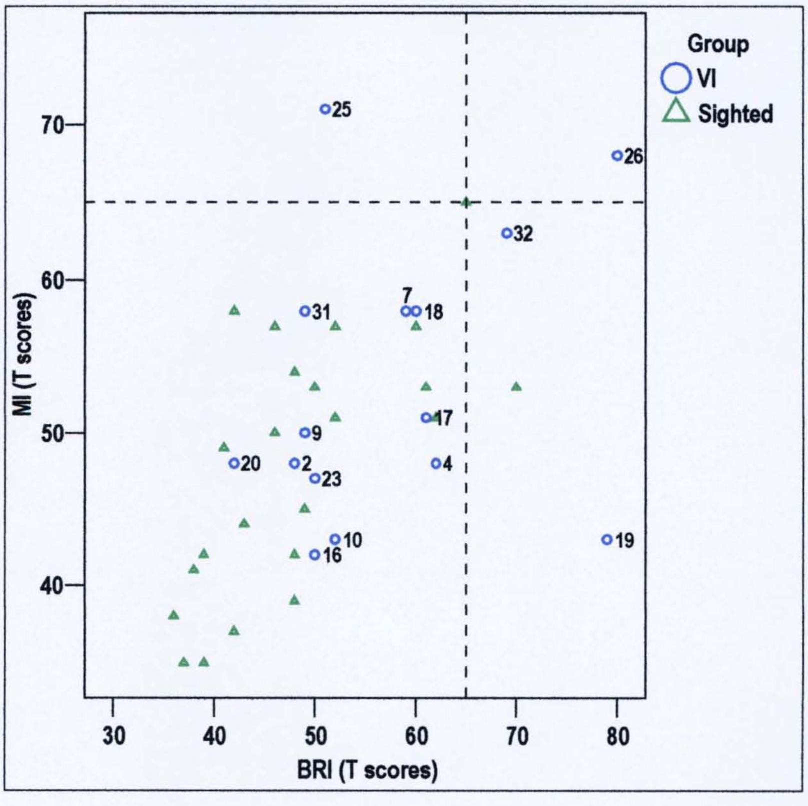

Figure 6.3: *Relationship between BRI and MI for individual children (including the clinical cut-off reference T ≥ 65)*

Following from this, the differing profiles between the two groups were compared by examining the percentage of cases within each group that reached the clinical cut-off on the BRIEF scales, in order to illustrate the relative risk in children with VI of obtaining clinically significant BRIEF scores, compared to sighted children of similar age and intelligence (H2). The children in the two groups were categorised as those whose *T* scores fell within the normal range limits ($T < 65$) and those whose *T* scores were deemed clinically elevated ($T \geq 65$). The proportions of children reaching clinically significant scores in each group are shown in Table 6.4 below. Here, it can be seen that a higher percentage of children with VI, compared to sighted children, have obtained clinically elevated scores on all BRIEF scales, except for Plan and Organise, where sighted children appeared weaker by comparison. Crucially, however, the proportions of children whose scores were the within normal range and those whose scores were above the clinical cut-off were significantly different between the two groups only on Emotional Control ($p \leq .01$) and Initiate ($p \leq .05$), where children with VI appear to be at a significantly higher risk of clinically elevated scores than sighted children.

Table 6.4: *Proportions (%) of VI vs. Sighted groups reaching clinical cut-off on the BRIEF scales*

BRIEF scale	***VI***	***Sighted***	***Chi-square statistics***
a) Inhibit	13.3	8	$\chi^2_{(1)} = .296$, $p = .586$
b) Shift	26.7	12	$\chi^2_{(1)} = 1.397$, $p = .237$
c) Emotional Control	26.7	0	$\chi^2_{(1)} = 7.407$, $\mathbf{p = .006}$
d) Initiate	26.7	4	$\chi^2_{(1)} = 4.404$, $\mathbf{p = .036}$
e) Working Memory	13.3	4	$\chi^2_{(1)} = 1.171$, $p = .278$
f) Plan/Organize	0	4.2 [a]	$\chi^2_{(1)} = .641$, $p = .423$
g) Organization of Materials	20	12	$\chi^2_{(1)} = .471$, $p = .493$
h) Monitor	13.3	4	$\chi^2_{(1)} = 1.171$, $p = .278$

[a] N missing = 1

DISCUSSION

In the present investigation, the BRIEF questionnaire captured a striking profile of everyday executive function behaviours in children with VI whose verbal intelligence is in the normal range. This profile was found to be uneven in comparison to the EF profiles of developmentally matched sighted children, with specific executive domains emerging as areas of relative weakness. A spiky profile of EF strengths and weaknesses in children with VI appears to be unlike the BRIEF profiles seen in children with autism and children with ADHD, who have been found to obtain significantly poorer ratings across all the BRIEF scales compared to sighted controls (Gilotty et al., 2002; Gioia et al., 2003).

The BRIEF scales which differentiated the VI group as poorer than their sighted counterparts were Shift, Emotional Control, Initiate and Monitor. Here, it is worth noting that, judging by the BRIEF developmental norms, the means of the VI group on these specific scales still appeared to be within the limits of the normal range. For that reason, it is potentially presumptuous to conceptualise these specific EF domains of children with VI as 'dysfunctions' that characterise this clinical group. On the other hand, given that the specifically matched sighted group obtained significantly better ratings by comparison, it is crucial to understand why the children with VI in this study have not achieved the levels on these domains that would be expected from children of their age and intelligence. To address this question it may be helpful to take a closer look at the individual results and their meaning.

For instance, the Shift scale assesses mental flexibility in children, in terms of their ability to switch cognitive set from one situation or activity to another depending on the contextual demands. A possible explanation for why this domain may be vulnerable for children with VI is that the mechanisms that underlie the functioning of this domain may particularly benefit from visual processing. For instance, an important aspect of the shift domain (i.e., cognitive flexibility) is attentional control (e.g., Alexander & Stuss, 2000; Norman & Shallice, 1986). According to the executive function model proposed by Norman and Shallice (1986), attention provides higher-level 'supervision' over lower-level cognitive functions that include over-learned automatic processes or 'action schemas' (e.g., crossing the road at a pedestrian crossing), which tend to be triggered by environmental stimuli or routines (e.g., being at the same zebra crossing used every day). The 'attentional supervisory system' modulates these automatic responses in a flexible or adaptive way, for instance, in dealing with novelty or danger and in decision making (e.g., if the zebra crossing is not in use). Importantly, it is clear how visual perception can be an important vehicle for transferring attention and its executive control from one activity or stimulus to another (e.g., shifting attention away from the non-functioning zebra crossing to a traffic assistant helping the pedestrians cross the road at a different location). With regards to sighted children's everyday behaviours, visual experience may facilitate the process of shifting attention and cognitive focus onto a novel set of activities (e.g., in getting used to new situations, such as a new classroom), whereas children with VI cannot benefit from visual guidance in the same way and may be more prone to being 'stuck' on the routine-based automatic schemas.

In relation to this, perseverative and repetitive behaviours, and difficulties in modulation of motor acts in children are thought to be a result of poor shifting functions (i.e., reduced mental flexibility), the evidence for which comes from research with children with autism (e.g., Lopez, Lincoln, Ozonoff, & Lai, 2005). Importantly, researchers and clinicians have noted occurrence of these behaviours in children with VI in a social context (Brambring & Tröster, 1992; Hobson et al., 1999; Wills, 1968). This pattern is also supported by the present research in Chapter 3, where children with VI obtained significantly elevated scores on the Restricted, Repetitive and Stereotyped Patterns of Behaviour domain on the SCQ. Such behavioural outcomes of children with VI are certainly in line with their scores on the BRIEF Shift scale here.

Furthermore, it can be argued that the disruptions at the level of environmental (i.e., visual) stimuli, would also affect a child's inhibitory abilities, which may use the same attention mechanisms as shift. This is because this supervisory attentional system enables automatic responses to be suppressed when they are not satisfactory, such as impulsive actions and behaviours (e.g., crossing at the usual zebra crossing, even though it is exposed to road works). This, for instance, seems to be the case with children with ADHD (e.g., Barkley, 1997). However, the children with VI in this study did not differ from sighted children in terms of their Inhibit scores. An obvious explanation for this finding would be the lack of power, given that the direction of the group means was more favourable to sighted children. Additionally, in both groups of children in the study, the scores on Inhibit and Shift were significantly correlated, suggesting their interdependence. Similarly, 13 % of children with VI were still above the clinical cut-off on this scale, implying that inhibitory control may not be preserved in all children with VI. However, the lack of significant difference between the two groups precludes us from drawing firm conclusions.

A further BRIEF domain that differentiated the VI group as poorer than the sighted comparison group was Emotional Control. Moreover, almost a third of children with VI were reported to show levels of emotional control that are of clinical concern, compared to the sighted group where none of the children were above the clinical cut-off. This scale assesses executive functioning within the emotional domain, focussing on the child's ability to appropriately regulate emotional responses. Poor emotional regulation in a child is mirrored in angry outbursts and temper tantrums in response to seemingly minor events, the severity and frequency of which are not appropriate for that child's age. Successful regulation of emotional responses is likely to depend on the use of certain behavioural and cognitive strategies. For instance, such strategies may involve selectivity (i.e., avoiding upsetting situations/stimuli) or deployment of attention (i.e., using attention to distract oneself from situations/stimuli that provoke negative emotions) (Gross, 2002). Children with VI may not be able to benefit from such strategies in the same way as sighted children. For instance, where sighted children may be able to modulate their emotional responses by shifting attention and cognitive focus away from what causes strong emotional reactions, reduced shifting ability in children with VI may also affect their emotion regulation. Indeed, the significant correlation between the scores on the Emotion Control and Shift scales in both groups of children supports this explanation. Hence, as with shifting ability, emotional modulation strategies may also be facilitated by visual processing. In a social context in particular, feedback from other people, in terms of their facial expressions and gestures, may be particularly important in this process in that it may help children regulate their levels of distress (or laughter in inappropriate situations) so as not to cause upset to others. Lack of vision is likely to limit perception of such feedback and may provide an explanation for why children with VI may be more vulnerable to poorer emotional modulation than their sighted peers of similar developmental level.

A particularly interesting finding was the poorer initiation behaviours in the VI group. The Initiate scale on the BRIEF assesses the generativity component of EF, which relates to initiating a task or activity and independently generating ideas, responses and problem-solving strategies. Difficulties in this domain are often demonstrated in the form of difficulty with word and design fluency tasks and the need for additional cues from the adult to begin tasks. The present result of poorer Initiation in the VI group is particularly striking, considering that the VI group performed well on a performance-based test of verbal fluency presented in Chapter 3. More specifically, on the Word Association test on the expressive language domain of the CELF-3 there was a trend towards a significant between-group difference in favour of the VI group, which is in contrast to the poorer Initiate ratings on the BRIEF, according to which nearly a third of the VI group was above the clinical cut-off. Furthermore, some of the children who generated most items on the Word Association task scored in the clinical domain on the Initiate scale (e.g., participant ID: 26). Such dissociation between performance-based and questionnaire-based measures of initiative ability suggests that the two measures may tap different underlying abilities for children with VI. A closer inspection of the BRIEF items reveals that some of the Initiate items target children's initiating behaviours in free-time and in relation to activities with friends. Thus, it is possible that the poorer behavioural ratings of the VI group on the Initiate scale on the BRIEF questionnaire are a secondary outcome of their impoverished social skills.

A similar explanation may account for the poorer behavioural ratings in the VI group on the Monitor scale. This BRIEF scale assesses the habits of checking work/task performance for the purpose of effective goal achievement. Children who are poor at self-monitoring tend to rush through assigned work, making careless mistakes. Importantly, this scale also evaluates personal monitoring functions in terms of whether a child keeps track of the effect their behaviour has on other people (e.g. '*does not realize that certain behaviours bother others*'). This suggests that there is a certain pragmatic and socio-cognitive element to this scale. In fact, children with VI may find it difficult to monitor their own actions and behaviours in relation to other people because they may be perceptually unaware of other people's perceptions of such behaviours. On the other hand, sighted children may be at an advantage in this domain as they can rely on other people's non-verbal cues to know whether their behaviour is undesirable and correct it accordingly. However, while the socio-cognitive perspective explains the difference between the VI and the sighted groups on the Monitor scale, it does not explain the unusual pattern of the mean scores on this scale for sighted children. More specifically, sighted children seemed to have received particularly positive ratings from their parents on this scale, resulting in Monitor scores that were considerably better (i.e., lower) than would be expected based on the BRIEF norms. With regards to this, it is worth reiterating that one item from this scale was not included in the calculation of the *T* scores as it was considered unsuitable for children with VI (see p. 152). Hence, all the children (VI and sighted) were assigned the lowest value (i.e., best potential outcome) on this item ('*has poor handwriting*'). However, handwriting is such an important part of sighted children's daily lives. Thus, it is possible that removing this particular item also removed a certain level of variation that is an inherent part of this behaviour, resulting in particularly good Monitor outcomes in the sighted group. It is unclear how the same issue would have affected the scores of the VI group whose mean on the Monitor scale was virtually indistinguishable from the average mean in the normative data. However, it is important to highlight that the VI group was closely matched to the sighted comparison group in this study, hence their significantly poorer behaviours on this scale (relative to their sighted matches), may still have important developmental implication (e.g. see the socio-cognitive explanation above).

Thus far, it can be argued that the specific EF weaknesses of the VI group seen in this investigation are related to the possibility that visual perception (e.g., via attentional systems and socio-environmental factors) may mediate the functioning of certain executive domains. The same explanation may also account for why children with VI generally did well on other aspects of the BRIEF, like Working Memory, Plan/Organise and Organisation of Materials, which are processes that may not necessarily be vision-dependent. Working memory strength of children with VI is often evident from their Digit Span performance, where they have been found to outperform sighted children (Hull & Mason, 1995; Smits & Mommers, 1976; Tillman & Osborne, 1969). Similar superiority has been demonstrated in the present research in Chapter 3, using the Recalling Sentences task from the receptive language domain of the CELF-3. Thus, from the VI children's Working Memory scores on the BRIEF, it is clear that their short-term memory strength is likely to transpire in real life. Finally, with regards to planning, organising and organisation of materials, these processes may require similar levels of cognitive mapping that are involved in the process of spatial orientation. Importantly, research evidence suggests that children and adults with VI can achieve this efficiently by relying on alternative, non-visual strategies (e.g., tactile mapping, echolocation) (Eardley & Pring, 2007; Millar, 1994; Ungar, Blades, & Spencer, 1995a, 1995b, 1996).

However, an important issue remains to be considered. More specifically, some children with VI seemed to show more behavioural difficulties in the area of EF than others, as their scores were in the clinical domain across different BRIEF scales. Interestingly, while some of these children showed clinically significant weaknesses on some, but not other aspects of the BRIEF, only one child was above the clinical cut-off across all of the BRIEF domains. It is possible that the 'concerns' we see here in some children with VI are also picking up on the children's social difficulties (e.g., in Chapter 3), because the BRIEF targets the expression of EF behaviours in those environments which are inherently social (i.e., at school and home). It was argued in the introduction of this section that executive and social functions are not mutually exclusive, and convincing evidence for this relationship comes from considering children with autism. Thus, it is possible that the children with VI who showed more behavioural difficulties in the domain of executive functioning are those with poorer socio-communicative outcomes, because the same vulnerability that may contribute to the poorer socio-developmental outcomes of children with VI may also affect their executive function. However, as it was argued earlier in this section, it is also possible that the higher-level mechanism (e.g., the attentional system), which operates from the frontal lobes, may be particularly vulnerable when visual input is restricted, and it is disruption to this mechanism that may affect a child's executive functioning, of which triadic joint attention and metarepresentational social cognition are likely to be a part. Interestingly, the following section of this chapter (i.e., Part 2) may shed further light on this particular issue.

PART 2: ATTENTIONAL PROCESSES IN YOUNG CHILDREN WITH CONGENITAL VI

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INTRODUCTION

While the research in Part 1 looked at the broader presentation of EF in forms of everyday behaviour in school-age children with VI, the research presented in this section focuses on attention capacity and regulation as a specific aspect of general executive functioning and in young pre-school children with VI. As discussed earlier in this chapter, attentional control is thought to be central to EF; it influences the operation of other executive domains (e.g., cognitive flexibility, information processing, goal setting), which depend on it for the selection of specific stimuli, inhibition of prepotent responses, the ability to stay focussed over time, regulation and monitoring of actions, error identification and goal attainment (e.g., P. Anderson, 2003; Norman & Shallice, 1986).

Importantly, as it has previously been discussed, attention has been closely related to early socio-communicative development, which at its earlier stages involves social attention sharing (Butterworth & Grover, 1990; Corkum & Moore, 1995, 1998). Ability to co-ordinate attentional focus in the context of socially-driven joint attention experiences reflects maturation in attentional systems which undergo considerable changes in the first few months of child development (Atkinson, 1984; Butterworth & Cochran, 1980; Hood et al., 1998). Vision is likely to play a major role in these processes, given that attentional capacity and control in children is most readily measured in terms of their ability to orient to visual stimuli and follow and direct the visual focus of others (e.g., using eye-gaze as an attentional cue).

The link between the development of attention and social communication has also been demonstrated in research with children with autism. Researchers have drawn connections between social impairments that are characteristic of the disorder and attention difficulties, suggesting that disruptions to early joint attention, and subsequent deficits in higher-order cognitive processes such as theory of mind and executive functions characteristic of autism, may be preceded by early occurring attentional abnormalities (Landry & Bryson, 2004; Leekam et al., 2000; Swettenham et al., 1998). For instance, children with autism have been found to experience difficulties with orienting their attention in tasks which require them to disengage attention from one stimulus and shift it to another (Landry & Bryson, 2004; Pascualvaca, Fantie, Papageorgiou, & Mirsky, 1998), and this difficulty is particularly pronounced with stimuli of social nature (i.e., people as opposed to objects) (Leekam et al., 2000; Swettenham et al., 1998). In line with this, Leekam et al. (2000) found preschool children with autism to be less responsive than developmentally delayed children without autism in orienting to the attention bids and head-turning cues provided by another person, in line with their general joint attention impairment. It follows from this that, if adequate attention capacity and control are necessary prerequisites for joint attention skills and subsequent socio-cognitive developments, specific socio-developmental difficulties that are associated with visual impairment may also be a result of early occurring difficulties in attentional processes.

Cass et al. (1994) were amongst the first to hypothesize that early attentional processes may be a potential risk factor in the development of young children with VI. These authors (also Dale & Sonksen, 2002) identified a serious developmental outcome, which they referred to as 'developmental setback', occurring in a subgroup of children with congenital VI. They noted that the setback, which involves plateauing or loss of skills in cognitive and language development, emerged in the second and third year of life and manifested behaviourally in form of autism-related characteristics such as social withdrawal, self-directedness and resistance to social approach. According to these authors, the period when the developmental setback begins to occur coincides in childhood with the development of behavioural independence and changes in attentional control. In typical development, the second year of life is characterised by a drive for autonomy and independence, and therefore greater behavioural inflexibility, non-compliance and temper tantrums (Colson & Dworkin, 1997; Ruff & Rothbart, 1996). Furthermore, the child's attention becomes more controlled, focussed and sustained, less driven by novelty and more by what others attend to (Rothbart & Posner, 2006). For children who are visually impaired this period may be particularly vulnerable. In the absence of the integrative visual input of a sighted child, developing attentional control and responding to adult-directed attention may rely on the auditory channel and be more difficult for such children, resulting in potentially serious consequences for their subsequent developmental outcomes.

Because of the dominant role of vision in early attention development, early attentional ability may differ for those children with VI who have some, although severely degraded, functional vision (i.e., SVI), compared to children with a total absence of vision or light perception only (i.e., PVI). A distinction has often been drawn between the two groups of children, as young children with PVI, compared to those with SVI, have been found to show poorer developmental outcomes, in terms of general cognitive functioning (Dale & Sonksen, 2002; Reynell, 1978), language (McConachie & Moore, 1994), and socio-communicative competence (Preisler, 1991). Although visual impairment of significant severity (SVI, as well as PVI) is likely to have an effect on a child's developmental outcomes in general, it has been argued that presence of functional vision, however limited, can serve as a protective factor in the developmental processes of young children with VI (Sonksen & Dale, 2002). It is therefore of interest in the current investigation to examine whether the presence of functional vision in children with SVI may positively impact on their early attentional processes, compared to children with PVI.

Visual impairment in childhood is associated with a relatively high prevalence of developmental delay and global learning difficulties (Sonksen & Dale, 2002), which is likely to interfere with children's attentional processes. Even when a group of children with VI is rigorously selected for research purposes to reduce the confounding effects of additional learning difficulties (e.g., by excluding children with cortical VI), the incidence of learning difficulties is still 17% (Dale & Sonksen, 2002). In their sample of young children with autism, Leekam et al. (2000) found that the differences in specific attentional abilities between children with autism and their developmentally matched controls were largely confined to the children with autism of lower cognitive ability. Similarly, difficulties in the ability to regulate attention in young children with VI may be more prevalent in those with cognitive impairments.

Importantly, the attentional processes of children with varying levels of visual impairment have not been empirically addressed to date. The aim of the current observational study was to attempt such an investigation, by making use of existing clinical video observations of children with congenital VI undergoing a semi-structured developmental assessment. In the absence of experimental manipulation, this method allowed for the eliciting and observation of children's attentional behaviours in a naturalistic setting.

Existing models of attention divide it into unique components that involve the ability to focus on a particular stimulus, to sustain that attention over a period of time and to shift it flexibly and adaptively (Mirsky, Anthony, Duncan, Ahearn, & Kellam, 1991; Posner & Petersen, 1990). It is thought that, while integrated into a single functional entity, these processes are mediated by distinct neuroanatomical regions. Drawing on these models, attention was conceptualised in the current investigation in terms of *establishing* attentional focus on objects, *maintaining* attention on those objects over a continuous period of time and *shifting* attention flexibly onto a novel object.

Establishing attentional focus on objects occurs as the adult in interaction introduces a toy of relevance to the child and the child responds to this attentional bid by engaging with the given toy. *Maintaining* attention on objects, on the other hand, occurs as the adult continuously attempts to hold the child's attention on the toy to which attention has been established and the child responds to these adult bids by sustaining concentration on the given object. Finally, *shifting* attention to novel objects occurs as the adult in interaction tries to disengage the child's attention from the toys with which they are engaged by introducing a novel object and the child responds by orienting towards the novel objects.

Research questions and hypotheses

In line with the theoretical issues outlined so far, four research questions remain unanswered and the research presented here makes an attempt to address them:

Question 1(Q1): Do children who are visually impaired from birth experience attentional difficulties (i.e., in terms of establishing, maintaining and shifting adult-directed attention) in their early years of life?

Question 2 (Q2): Do attentional outcomes in children who are visually impaired vary as a function of severity of visual impairment?

Question 3 (Q3): Do attentional outcomes in children who are visually impaired relate to their general cognitive abilities?

Question 4 (Q4): Are different aspects of attentional behaviour (i.e., establishing, maintaining and shifting adult directed attention) in children with VI and their sighted peers independent from each other?

METHOD

Participants

The participants were 32 children with congenital VI and 17 children with normally developing vision. The children with VI were consecutively identified through a tertiary level developmental vision clinic (DVC), where they were referred for specialist assessment and management of their vision and development by paediatricians or ophthalmologists. They were included based on their age at time of assessment (10-56 months) and diagnostic classification using the diagnostic taxonomy by Sonksen and Dale (2002) (i.e., CDPVS). These children were separated into two groups depending on whether they had PVI or SVI at the time of the assessment (see Chapter 2, p.52 for details). There were 16 children with PVI: 7 girls and 9 boys, age range 15 - 53 months (Mean age = 32.2 months, SD = 10.9). Similarly, there were 16 children with SVI: 8 girls and 8 boys, age range 17 - 36 months (Mean age = 26.6 months, SD = 6.1). Individual children's visual diagnoses, as well as their vision levels, are summarised in Table 6.5.

A comparison group of 20 sighted typically developing children was recruited through the local nurseries, crèches and parent - toddler groups. Three children were subsequently excluded for the purpose of successful developmental matching, which lead to 17 sighted children being included in the study. There were 10 girls and 7 boys, age range 10 - 36 months (Mean age = 20.2 months, SD = 8.3). The sighted group was recruited and tested during the period of the present PhD research, whereas the VI sample was a clinical population whose archived clinical and video data were accessed retrospectively, for the purpose of this thesis.

Table 6.5: *Characteristics of individual children with congenital VI*

Child	***Sex***	***VI level***	***Age in months***	***Visual Diagnosis***
1	M	PVI	20	Leber's amaurosis
2	M	PVI	23	Familial exudative vitreo-retinopathy / Norrie's Syndrome[a]
3	F	PVI	45	Optic nerve hypoplasia and bilateral congenital cataracts
4	M	PVI	31	Optic nerve hypoplasia
5	F	PVI	28	Leber's amaurosis/retinal dystrophy
6	M	PVI	40	Retinal dysplasia/Norrie's Syndrome
7	M	PVI	23	Leber's amaurosis
8	F	PVI	42	Bilateral microphthalmia
9	M	PVI	30	Persistent hyperplastic primary vitreous
10	F	PVI	15	Bilateral retinal coloboma and optic nerve aplasia.
11	M	PVI	27	Bilateral microphthalmia and optic nerve aplasia
12	F	PVI	53	Bilateral optic nerve hypoplasia[a]
13	F	PVI	44	Hypoplastic optic disk (right eye) and absence of the optic disk (left eye)
14	F	PVI	37	Bilateral optic nerve hypoplasia
15	M	PVI	38	Norrie's disease
16	M	PVI	19	Bilateral optic disc and optic nerve hypoplasia
17	F	SVI	31	Leber's amaurosis
18	M	SVI	17	Corneal opacities/Microphthalmia
19	F	SVI	26	Multiple opacities/ Sclerocornea
20	M	SVI	19	Retinal dystrophy
21	M	SVI	24	Bilateral optic nerve hypoplasia
22	M	SVI	20	Congenital cataracts
23	M	SVI	32	Leber's amaurosis
24	F	SVI	22	Bilateral microphthalmia with anterior segment malformation of both eyes
25	F	SVI	36	Bilateral microphthalmia/ Sclerocornea
26	M	SVI	33	Multiple corneal opacities/ Micropthalmia
27	F	SVI	30	Retinal folds and microcephaly
28	F	SVI	24	Bilateral optic nerve hypoplasia
29	F	SVI	28	Severe retinal dystrophy and most likely Leber's amaurosis
30	M	SVI	36	Congenital bilateral Aniridia, glaucoma and Peter's anomaly in one eye
31	M	SVI	20	Leber's amaurosis
32	F	SVI	28	Leber's amaurosis

[a]Based on the functional vision assessment records these participants had a PVI status up until the age 16 months and have gained functional form vision after this age giving them SVI status. Their inclusion in the PVI group was justified based on evidence that form vision before the age of 16 months seems to exert protective effects on developmental processes of children with VI (Sonksen et al., 1991).

Materials

Cognitive characteristics of the sample

All the children, VI and sighted, were assessed on the Reynell-Zinkin Scales of Mental Development for Visually Handicapped Children (RZS) (Reynell, 1979). The RZS were specifically designed to monitor the cognitive progress of very young visually impaired children in the areas of sensory motor development, expressive and receptive language, social adaptation and exploration of environment. For the current research purposes only a subset of the test scores were used: 1) Sensory-motor understanding (SMU); 2) Verbal comprehension (VC) and 3) Expressive Language (Structure) (ELS). SMU scales show the level of learning in relation to concrete objects (e.g., exploration, manipulation, recognition and meaningful use of objects); VC scales concern the ability to understand language (i.e., recognition of familiar phrases in context, understanding of verbal labels and verbal instructions); and ELS concerns the ability to produce vocal patterns, words and sentences with appropriate use of different parts of speech.

Raw, and not age-equivalent, scores on these scales were chosen to be the most appropriate measure for a number of reasons including the fact that the RZS are only partially standardised across the subtests and according to vision level (i.e., Blind, Partially Sighted or Sighted). Furthermore, children with congenital VI typically may lag behind children who are sighted on all of the areas assessed by RZS, even though their development may fall within a 'normal' range when the severity of their VI is taken into account (Reynell, 1978). This is generally problematic when utilizing a typically developing control group by which to make comparisons. Hence, in order to achieve successful developmental matching, the raw scores were retained as they gave a direct access to the performance levels the children achieved.

The raw score profiles for the overall sample are summarised in Table 6.6. Here, it can be seen that there was a significant difference between the groups in chronological age (omnibus test: $F_{(2, 46)} = 7.909$; $p \leq .001$; individual post hoc tests: PVI vs. Sighted: $t_{(31)} = 3.56$, $p \leq .001$; PVI vs. SVI: $t_{(30)} = 1.781$, $p = .085$; and SVI vs. Sighted $t_{(31)} = 2.52$, $p = .017$) and sensory-motor understanding (omnibus test: $F_{(2, 46)} = 4.301$; $p = .019$; individual post hoc tests: PVI vs. Sighted: $t_{(31)} = -2.68$, $p = .012$; PVI vs. SVI: $t_{(30)} = -1.17$, $p = .105$; and SVI vs. Sighted $t_{(31)} = -1.401$, $p = .171$). However, there was no difference between the groups on verbal comprehension ($F_{(2, 46)} = .120$; $p = .887$) and expressive language ($F_{(2, 39)} = .305$; $p = .739$). Relatively equal verbal ability profiles in the three groups of participants suggest successful matching on verbal mental age. Gender was also found to be equally distributed across the groups ($\chi^2_{(2)} = .759$; $p = .684$).

Table 6.6: *Matching characteristics of the sample*

Matching criteria	***PVI*** *N=16*	***SVI*** *N= 16*	***Sighted*** *N=17*	***p value*** ***(F test)***
Chronological age (in months) Mean (SD)	32.2 (10.9)	26.6 (6.1)	20.2 (8.3)	.001
Gender ratio Girls/Boys	7/9	8/8	10/7	.684
Reynell - Zinkin (raw scores)				
Sensory-motor understanding (SMU) Mean (SD)	12.7 (4.3)	14.9 (3.2)	16.9 (4.6)	.019
Verbal comprehension (VC) Mean (SD)	13.2 (6.6)	14.1 (5.4)	14.2 (7.8)	.887
Expressive language (structure) (ELS) Mean (SD)	12.7 (4.8)	12.8 (3.7)	11.7 (4.9)	.739

Administration of RZS took approximately 30 minutes per child. All the assessments were video recorded with a signed parental consent.

Observational data

The administration of the RZS scale items is flexible and allows for naturalistic aspects of the joint play within which it is carried out. This semi-standardised context was chosen for the current observational coding of attention-related responses as the assessor during the assessment has to make frequent attentional demands on the child in terms of gaining child's attention on the test items (i.e., toy material), maintaining child's attention on the toys and the task, and shifting child's attention between different test items.

Equal testing conditions, as far as possible, were ensured across the three groups, in terms of i) the cognitive and/or linguistic demands they required from each child; ii) the toy materials presented to the children during the play interactions with an adult; and iii) the adult directing behaviours, which were adapted to individual children's vision levels. With regards to the adult directing behaviours, for example, where a toy may be introduced to a sighted child primarily through visual means, for a child with VI the adult uses a combination of verbal commentary (e.g., '*Look, what I've got here!*'), auditory stimulation (e.g., shaking or winding the toy to make a sound or tapping it on the table) and touch (e.g., tapping the toy on the back of the child's hand).

In order to further maximize the standardised quality of the play interactions across the sample for coding purposes, five standardised structured-play episodes were selected from the videotaped RZS assessments for their consistency of presentation across the children (Appendix D1). The average time taken for the five scenarios was approximately 20 minutes per child. During these scenarios, when children were introduced various toys to engage with, the experimenter scored the ease or difficulty that the child had in following the attentional demands of the adult by selecting the type of attentional behaviour of interest (below) and judging how successful the child was in responding to the adult's attempt to elicit that behaviour (Appendix D2).

Establishing was defined as a child's ability to respond to adult's attempt to gain the child's attention onto the toy material when the child is not attending to the adult or object of relevance. For each scenario, one of three possible success codes was given: Immediately successful (2), Successful with difficulty (i.e., requiring more prompting by the adult) (1), and Unsuccessful (0). An *Establish* score was calculated by summing the success codes over the 5 scenarios and converting this into a percentage of the maximum possible establish score (i.e., 10). For the majority of children the number of scenarios was five. However, owing to technical difficulties (e.g., child being obscured by adult in the video, or child moving outside the video framework), not all of the 5 scenarios were available for some children (n = 5). Thus, the *establish* score for these children represented the summed scenario codes as a percentage of the maximum possible establish score.

Maintaining was defined as the child's ability to respond to adult's attempt to hold the child's attention onto toy material after the child's attention on a task had been established. For every scenario in which attention was established, one of three possible maintenance success codes was given: Continuous (2); Somewhat disrupted (1), and Disrupted (0). A *Maintain* score was then calculated by summing the success codes over the scenarios in which attention had been established and converting this into a percentage of the maximum possible maintain score (i.e., 2 x the number of scenarios in which attention had been established).

Shifting was defined as the child's ability to respond to the adult's attempt to shift the child's attention from an object the child was engaged with to a novel object. For every scenario in which attention was maintained (after successful establishment of attention), one of three possible shifting success codes was given: Immediately successful (2), Successful with difficulty (1), Unsuccessful (0). A *Shift* score was then calculated by summing the success codes over the scenarios in which attention had been established and maintained and converting this into a percentage of the maximum possible maintain score (i.e., 2 x the number of scenarios in which attention had been established and maintained). Table 6.7 below summarises the frequency of participant's data upon which the Establish, Maintain and Shift scores were calculated.

Table 6.7: *Distribution of the number of valid instances upon which Establish, Maintain and Shift scores were calculated for the 49 children (VI and Sighted)*

Number of valid instances (i.e., scenarios)	***Number of children***		
	Establish (based on scenarios)	Maintain (based on established)	Shift (based on maintained)
0	-	-	2
1	-	-	-
2	1	2	4
3	2	2	5
4	2	3	9
5	44	42	29

Reliability was calculated by an independent coder who was unaware of the hypotheses of the study, for 12 randomly selected children (4 per vision group, 24.5% of the videos in total). The inter-rater Pearson correlations were r = .93, r = .75 and r = .72, for the Establish, Maintain and Shift categories respectively.

This coding system made it possible to view the children's performance on three attentional components independently. However it must be borne in mind that the scores children achieved on Maintaining were based on fewer scenarios than those they achieved on Establishing and in turn, the same was for Shifting. For instance, due to poor scores on Maintaining, two children in the PVI group could not be scored on Shifting and were excluded from subsequent analyses including this component.

General procedure

Each child was assessed by one assessor, in a quiet testing room in the presence of one or both parents. They were sat at a small table facing the assessor. The children with VI were assessed by a consultant clinical psychologist or a consultant developmental paediatrician at DVC as part of a clinical assessment which also included a paediatric assessment of functional vision. The assessors were unaware of the purpose of the study, which was conceived retrospectively. The sighted group was assessed by the PhD researcher, trained to administer the RZS by the consultant clinical psychologist who assessed the majority of the children in the VI group.

RESULTS

The attention data were not normally distributed and the variances between the three groups were heterogeneous enough to warrant the use of non parametric alternatives to test the between-subjects differences and relationships between the variables in the study.

Between-group comparisons

Table 6.8 shows the mean percentage scores the three groups of children achieved when they responded to adult-directed attention broken down into the three components. It is evident that these young children showed good sensitivity to being directed by an adult in general, considering that the majority of children in all three groups achieved relatively high percentage scores (see also Figures 6.4, 6.5 and 6.6). However, the direction of the group means (Table 6.8) suggests a difference between the three groups on all three attention categories, with a trend towards lower scores increasing with severity of visual impairment.

Table 6.8: *Performance (%) on the three attention components for the three vision groups*

Measure *Mean (SD)*	***PVI***	***SVI***	***Sighted***	***p value (F test)***
Establish	79.4 (20.5)	89.4 (12.4)	98.2 (7.27)	.001
Maintain	67.03 (32.45)	80.26 (16.59)	92.94 (7.72)	.016
Shift	80.29 (20.95) [a]	93.28 (18.72)	96.91 (6.09)	.007

[a] Missing N = 2

As the attention data were negatively skewed in the three groups and the variances between the groups were largely heterogeneous (with the heterogeneity increasing with the severity of visual impairment) the Rank Transformation approach advocated by Conover and Iman (1981) was adopted as a more appropriate alternative to a parametric analysis. This approach does not depend on normal distribution. Hence, in order to address Q1, the scores that the children obtained on the three attention categories were transformed into rank order prior to running three separate 1-way ANOVAs.

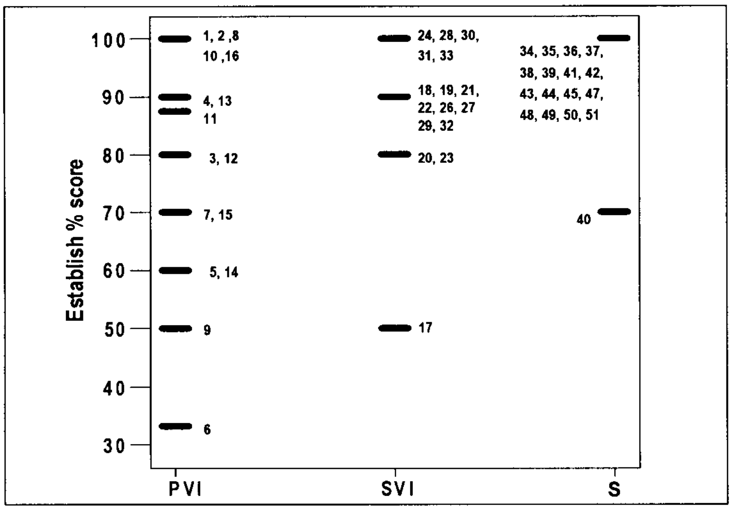

Figure 6.4: *Establish performance (%) of individual children in each group*

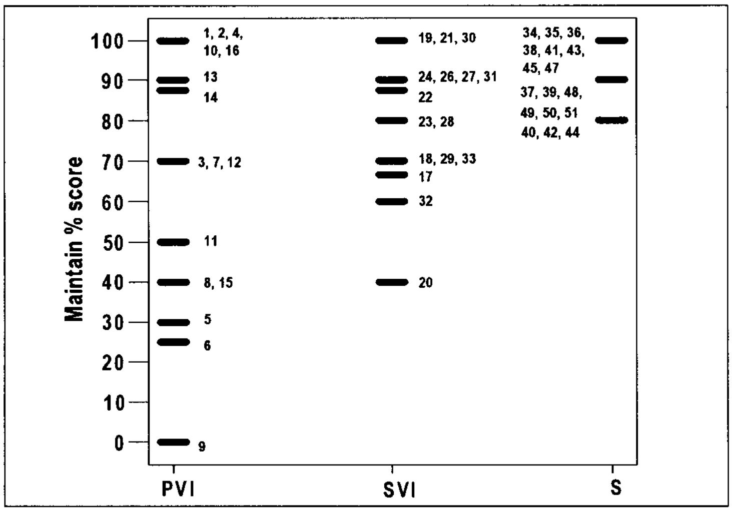

Figure 6.5: *Maintain performance (%) of individual children in each group*

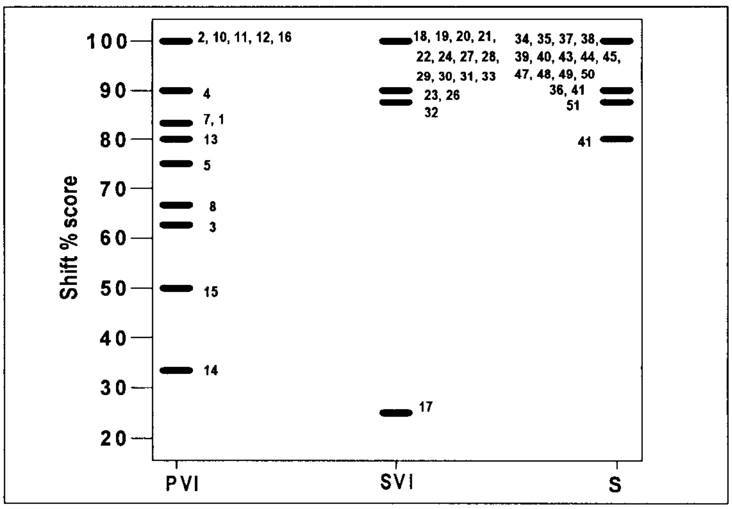

Figure 6.6: *Shift performance (%) of individual children in each group*

Firstly, an ANOVA showed a highly significant difference on *establishing* adult directed attention on toys between the three groups of children ($F_{(2, 48)}$ = 10. 909, $p \leq .001$) (Q1). It is important to note that, equal variances were not assumed for this or any of the following *F* and *t* tests. Planned contrasts, comparing the three groups individually (Q2), significantly distinguished each VI group as being significantly poorer at *establishing* adult directed attention than the sighted children in the study (PVI: $t_{(21.95)}$ = - 4.395, $p \leq .001$; SVI: $t_{(25.6)}$ = - 3.903, $p \leq .001$). However despite the directions of the scores illustrated in Figure 6.4, the children with PVI and the children with SVI in the study were not significantly different from one another ($t_{(28.3)}$ = - 1.037, p = 0.309, d = -.57).

A separate 1-way ANOVA also showed significantly different performance on *maintaining* adult directed attention on toys between the three groups ($F_{(2, 48)}$ = 4.551, p = .016) (Q1), and similar to the scores in the establish category, individual contrasts revealed that the sighted children obtained significantly higher scores at *maintaining* adult directed attention than did the children with PVI ($t_{(23.34)}$ = - 2.693, p = .013, d = - 1.09) or the children with SVI ($t_{(28.1)}$ = - 2.757, p = .01). The PVI and SVI groups did not differ between each other ($t_{(27.6)}$ = - .436, p = .67, d = - 0.51) (Q2).

Finally, a 1-way ANOVA revealed a significant between-group difference on how well children *shifted* adult-directed attention ($F_{(2, 46)}$ = 5.513, $p \leq .007$) (Q1). Interestingly based on the individual planned contrasts, the children with PVI were significantly poorer at *shifting* adult directed attention than both the SVI ($t_{(24.5)}$ = - 2.538, p = .018, d = - 0.64) and sighted children in the study (t $_{(22.25)}$ = - 2.837, p = .01), whereas children with SVI were not significantly different from the sighted ($t_{(29.92)}$ = - .210, p = .835, d = - 0.27) (Q2).

Relationship between variables

Due to the relatively small group sizes, as well as the prevalence of ceiling scores for attention, it was decided that the non-parametric correlational analyses (i.e., Spearman's rho ρ) would be more appropriate for the current data. Table 6.9 summarizes the Spearman coefficients for the correlations testing the potential relationship between the attentional outcomes and children's developmental levels (i.e., chronological ages and scores on the RZS scales) (Q3), as well as for the correlations between the scores relating to the three attentional components (Q4). For clarity, these correlations are described further in subsequent paragraphs for each vision group separately.

Table 6.9: *Spearman coefficients for correlations between age, cognitive characteristics and attention components in the three groups*

		Age	SMU	VC	EL (S)	Establish	Maintain	Shift
Age	PVI		.264	.368	.805**	.035	-.103	-.164
	SVI		.428	.520	.552	-.204	-.096	-.345
	Sighted		.978**	.972**	.976**	-.128	-.507	.006
SMU	PVI			.959**	.780**	.376	.497	.637
	SVI			.663**	.720**	.041	.136	-.146
	Sighted			.960**	.975**	-.179	-.490	.061
VC	PVI				.854**	.354	.493	.493
	SVI				.899**	.033	.011	-.262
	Sighted				.962**	-.154	-.480	.023
EL (S)	PVI					.337	.382	.330
	SVI					-.108	-.239	-.219
	Sighted					-.102	-.485	.048
Establish	PVI						.750**	.549
	SVI						.423	.542
	Sighted						.388	-.137
Maintain	PVI							.498
	SVI							.367
	Sighted							.089
Shifting	PVI							
	SVI							
	Sighted							

Red – PVI; Green – SVI; Blue- sighted; ** - significant at $p \leq .01$

Sighted children

Even though the sighted children were significantly younger than the children with VI, their attention was easy to gain and modify, so much so that their attention behaviour scores were virtually at ceiling level (Figures 6.4. 6.5 and 6.6). Unfortunately, this meant that it was methodologically problematic to analyse their attention scores through correlations and their performance needs to be interpreted with caution when compared on a statistical basis to either of the two VI groups. However, sighted children's chronological ages were significantly correlated with RZS subtest performance, in terms of SMU (ρ = .978, $p \leq .001$, n = 17), VC (ρ = .972, $p \leq .001$, n = 17), and EL(S) (ρ = .976, $p \leq .001$, n = 17), and their scores on the three RZS subtests correlated with one another (ρ coefficients = .960, .962, .975, $p \leq .001$, n = 17). The sighted children's ages and cognitive characteristics on the RZS also correlated with maintaining of attention, with the negative direction of the correlation suggesting that the younger children (and subsequently, children with lower RZS raw scores) were better at maintaining adult-directed attention; however, the prevalence of ceiling scores on the attention measures makes it difficult to further interpret attention-related correlations in the sighted group.

Children with PVI

The PVI group showed a greater heterogeneity of responses in all three categories of attention than any other group. Here, the ease with which children established adult-directed attention correlated highly with how well they maintained (ρ = .750, $p \leq$ .001, n = 16) and in turn shifted attention (ρ = .549, p = .042, n = 14). However the children's scores on maintaining and shifting of attention did not appear to be correlated, despite a very strong trend (ρ = .498, p= .07, n = 14). Interestingly, the ease with which the children maintained and shifted their attention was positively associated with their cognitive ability; the children's scores on sensory motor understanding were significantly correlated with how well they maintained attention (ρ = 0.497, p = .05, n = 16) and how well they shifted attention (ρ = .637, p = .014, n = 14). With regards to their language ability, there was a possible association between the children's verbal comprehension and how well they maintained attention (ρ = .493, p = .052, n = 16) and the correlation between verbal comprehension and how well children shifted attention showed a trend (ρ = .493, p = .073, n = 14). Similar to the Sighted group, the PVI children's sensory motor understanding, and receptive and expressive language were correlated with one another (ρ = .959, $p \leq$.001, n = 16; ρ = 0.780, p = .003, n = 12; and ρ = .854, $p \leq$ .001, n = 12). However, only the children's expressive language scores correlated with their chronological ages (ρ = .805, p =.002, n = 12).

Children with SVI

In the SVI group, the ease with which children established adult directed attention correlated with how well they shifted attention (ρ = .542, p = .03, n = 16), whereas maintaining of attention appeared to be unrelated to either establishing or shifting of attention. These children's attention behaviours did not correlate with any of their cognitive characteristics. However, there was a significant relationship between all of the aspects within the children's cognitive profiles, as their scores on the three RZS subtests (SMU, VC, ELS) were highly correlated with one another (ρ = .663, p = .005 , n = 16; ρ = .720, p = .006, n = 13; and ρ = .899, $p \leq$ .001, n = 13). Children's cognitive characteristics seemed to an extent to be associated to their chronological ages. Age was found to be correlated with verbal comprehension (ρ = .520, p = .039, n = 16) and expressive language (ρ = .552, p = .05, n = 13), whereas the correlation between age and sensory motor understanding was a trend (ρ = .428, p = .098, n = 16).

Association of attentional processes and low IQ in children with VI

The positive association of certain cognitive characteristics and some of the attentional components in children with PVI raised a question of whether their overall performance may have been related to the presence of children with low IQ in this group (Q3). Given also that the groups differed significantly in their sensory-motor understanding it was important to address potential contribution of intellectual impairment to the attention performance in the present sample.

Developmental delay is a common characteristic of the VI population. For that reason, the children with low IQ were not removed from the initial analyses, in order to retain the variance that is inherent in the presentation of congenital VI and illustrate the factors underlying the particular patterns of attentional responses shown by both the children with PVI and those with SVI in this research. However, in the second set of analyses, in order to account for potential confounding effect of intellectual impairment, it was decided to identify those children with VI who showed a cognitive delay both in the areas of sensory-motor understanding and verbal comprehension. The children's developmental levels (i.e., normal or delayed) were determined by deriving a Developmental Quotient (DQ), which is calculated based on the ratio of the children's raw-score age equivalents on the appropriate RZS norms (i.e., Blind, Partially Sighted or Sighted) and their actual chronological ages (Dale & Sonksen, 2002). Following from this, 'delayed' status does not imply a developmental delay by sighted norms, but the fact that the child's cognitive ability is at a lower lever than what would be expected even when the degree of their visual impairment is accounted for. In the study by Sonksen and Dale (2002) a cut-off of 80 was used to identify children with a cognitive delay. However, based on their findings as well as their clinical observations, these authors argued that the RZS overestimated the developmental levels of children with VI. Following from this, it was decided to adopt a more conservative cut-off of < 90 for the purpose of the present research. Based on this cut-off, 7 out of 16 children in the PVI group were found to show developmental delay compared to 3 out of 16 in the SVI group (all of the children considered as developmentally delayed had DQ < 90 both in SMU and VC).

Furthermore, the children with VI were divided into two groups, depending on whether they were performing in line with or below the level of sighted children on the three attention measures. To categorise children in this way, the 50th percentile of the sighted group was taken as a reference point because it corresponded to the ceiling performance achieved by the majority of the sighted sample. More specifically, 94% of sighted children were at or above this cut-off on Establishing, 82 % on Maintaining, and 76% on Shifting (see Figures 6.4, 6.5 and 6.6). Thus, this cut-off was used as a benchmark against which the performance of children with VI could be compared.

Importantly, while this cut-off was helpful in describing the children's performance in the present study, it must be treated with caution and is recommended that more sensitive measures should be developed in the future.

Table 6.10 shows that there were nearly equal numbers of children with low IQ and those who were not cognitively delayed in the PVI group. However, it can also be seen that low scores on all three attentional components were not found solely in the children with PVI who are cognitively delayed. Moreover, at least 40% of children with PVI (i.e., 67% in Establishing; 44% in Maintaining and 50% in Shifting) whose DQ was considered to be in the normal range, scored below the 50th percentile of the Sighted group on all three attentional components.

Table 6.10: *Number of children with PVI with and without developmental delay - a comparison with the near (or at) ceiling performance cut-off of sighted children*

		Establish cut-off		***Maintain cut-off***		***Shift cut-off*** [a]	
		at/above	below	at/above	below	at/above	below
DQ	normal	3	6	5	4	4	4
	delayed	2	5	1	6	1	5

[a]Missing data, n=2

With regards to the children with SVI (Table 6.11), even though 81% of children were within normal IQ range, a considerable number of those children scored below the 50th percentile cut-off in the Establish (69%) and Maintain category (54%), as well as a small percentage in the Shift category (23%). Additionally, out of three developmentally delayed children in this group, at least one child scored at/above the 50th percentile on all three attention components. This further confirms that lower attention scores are not confined to developmentally delayed children, nor is the higher attentional performance seen only in children with VI of normal IQ.

Table 6.11: *Number of children with SVI with and without developmental delay - a comparison with the near (or at) ceiling performance cut-off of sighted children*

		Establish cut-off		***Maintain cut-off***		***Shift cut-off***	
		at/above	below	at/above	below	at/above	below
DQ	normal	4	9	6	7	10	3
	delayed	1	2	1	2	2	1

DISCUSSION

The first question this investigation attempted to address was whether children with congenital VI experience attentional difficulties in their early years of life. When compared to typically developing sighted children with the same verbal comprehension ability, the present research has indeed shown that VI, especially if profound, does significantly reduce the capacity of a young child to regulate their attention between people and objects, in terms of establishing, maintaining and shifting attention. However, the current findings also indicate that lack of vision does not provide a sufficient explanation for why attentional processes of some children with VI are not as proficient as those of children who are sighted. Indeed, not only is the overall attention performance of the children with VI appears relatively good, a number of children both with PVI and SVI have shown ceiling performance, matching the level of children who are sighted. Crucially, this illustrates that certain attentional control is possible to achieve through other, non-visual, modalities. This is an important insight in light of known concerns about joint attention in this population. Hence, the findings support the idea of intervention as they highlight the developmental potential that should be capitalised on using alternative non-visual techniques (Dale & Salt, 2007).

The second question concerned the severity of VI and whether the attentional processes in young children with VI would vary according to vision level. The current findings were not clear cut. Contrary to other studies, where SVI has been associated with better developmental outcomes than PVI, the current findings suggest that at this developmental stage (i.e., pre-school and mainly pre-lingual) establishing and maintaining adult-directed attention is not different between children with SVI and PVI and is less well developed than in children who are sighted. It seems that the limited form vision in children with SVI is not a fully protective factor, implying that certain attentional outcomes in early childhood may be particularly sensitive to reduced vision. However, the findings also suggest that the available form vision of children with SVI may serve a protective factor in their ability to shift adult-directed attention, as this was in line with the ability of sighted children. This confirms the importance of the early promotion of functional vision to achieve form vision, wherever possible, in children with VI (Sonksen et al., 1991). Nevertheless, the current contrasting findings highlight that, despite the more promising developmental results, children with SVI should still be considered a vulnerable population.

The third research question concerned the relationship between attentional outcomes in children with VI and their general cognitive functioning. The present findings suggest that the developmental levels of the children with VI in this study must at least to some extent account for their attentional performance. With regards to children with PVI, the demands of having to maintain attention on toys and then flexibly shift this focus while being guided by an adult were related to what can be considered their performance ability level (SMU) and to an extent their verbal comprehension. However, even though higher IQ may equip a child with VI with a tool for dealing with attentional demands, the lower attentional performance seen in some of the children with VI in this study cannot be attributed solely to their lower developmental level. This is because lower attentional responses are not confined entirely to the low IQ group and a number of children with VI whose IQ is in the normal range showed lower attentional performance than some of the children with VI who are cognitively delayed. This double dissociation between IQ and attentional performance seems to suggest the independence of attention processing.

However, it must be noted that the present VI sample may have included some children with developmental setback in terms of their cognitive and language levels. The 'setback' (i.e. plateauing and/or loss of cognitive and language skills) has been identified by previous research studies as a serious developmental outcome in VI population, occurring in the second and third year of life, with a higher proportion of children with PVI (33.3%) than SVI (3%) being affected (Cass et al., 1994; Dale & Sonksen, 2002). The age range of the children with VI in the present study covers the period when the setback is first behaviourally manifested. Hence, based on previous research statistics, it is likely that the present VI sample may have included some children with setback, although it is not yet possible to identify them cross-sectionally.

It is important to emphasise some methodological limitations of the current experiment, relating not only to potentially low statistical power due to the small sample size, but also to the lack of variation and presence of the ceiling scores, which relate directly to the coding system that was used here. The limitation of the current coding schedule to produce variant responses perhaps echoes the challenge of attempting to devise a reliable coding measure, which takes the non-visual behaviours of a child with VI into account. By comparison, attention coding protocols used in studies with sighted groups of children frequently relied on vision and gaze behaviours to measure attentional capacity and control (Corkum & Moore, 1998; Leekam et al., 2000; Swettenham et al., 1998). Visual experimental stimuli as well as visually-driven attentional responses may not only be easier to measure and code, they may testify that certain attentional regulation can only be achieved by visual means, and this may partly account for the ceiling performance of sighted children in our study. In fact, it has been shown that sighted infants as young as 10-12 months are able to reliably use another person's head turn and eye-gaze to locate targets even when the targets are not visible in the visual field (Corkum & Moore, 1998), signifying the beginnings of being able to engage in joint attention. The ceiling performance of the sighted children (age range 10-36 months) in this study may therefore not be surprising.

Unfortunately, the prevalence of ceiling scores precludes us from drawing firm conclusions. With regards to the comparisons of the children with SVI and sighted children, we cannot be sure if the sighted children could have performed even higher if other measures of attention were incorporated in the study. Similarly, the apparent result suggesting no significant difference between the groups of children with PVI and SVI in establishing and maintaining attention, despite the directions of the means, may have indeed been due to the prevalence of the ceiling scores in both groups, and in particular preventing us from seeing if the children with SVI could potentially score even higher. Relating to this, we must bear in mind also the possible attrition in scores, as the children's Maintaining scores were based on fewer scenarios than their Establishing scores, and in turn their Shifting scores were based on fewer scenarios than the Maintaining scores.

Despite the methodological difficulties outlined above, the findings relating to different attentional components do seem to be in line with current models of attention (Mirsky et al., 1991; Posner & Petersen, 1990). According to these models the three attentional components are functionally related; however they are mediated by distinct brain regions, implying that dissociation between different attentional processes may not be surprising. Indeed, despite the prevalence of ceiling scores in the two VI groups (especially in establishing) the correlational analyses indeed suggest some dissociation between ability to maintain and shift attention in both the children with PVI and those with SVI. It is plausible to suggest that the shifting component of attention may particularly be perceptually driven, when compared to the process of establishing and maintaining attentional focus. When an adult in interaction brings in a novel toy, a child with SVI may be able to benefit from the visual cues of the whole object required for flexible perceptual reaction to novel stimulus. This visually-driven advantage, however, may not be sufficient to secure attention on the toy offered by an adult and hold that attention while being adult-guided. More specifically, the vision available may not be sufficient to identify what the object is or to see details of the object which might capture the child's continuing interest. This may explain why the ability of children with SVI to establish attention on toys is not in line with the ability of those children who are sighted. Furthermore, successful maintaining of attention, which is possibly the most socially driven of the attentional components in joint interaction, may be particularly difficult for any visually impaired child, since it requires shared attention to an object on which the child is focussing. Arguably, sustaining attention on a toy, while being guided by an adult, is facilitated by a level of vision that allows for eye-gaze monitoring, eye-contact and facial expression recognition. This indeed may further explain the performance of the SVI group in the current study, as in the absence of joint visual attentional cues and gaze behaviours, children with SVI may be at a similar disadvantage as children with PVI.

In summary, the present findings suggest that significant VI in childhood reduces the child's early attentional capacity and control, with those children whose VI is of greater severity (i.e., PVI) and those with lower cognitive levels being particularly vulnerable. Importantly, however, it seems that neither vision nor general cognitive level can fully account for the present pattern of findings. This is because poorer attention performance was not confined to children with VI who had lower levels of intelligence. Similarly, reduced attentional performance was not seen only in children with PVI, and some children with SVI showed poor attentional outcomes, even on attention shifting. This pattern of findings is in line with the findings in Part 1, where cognitively advanced children with varying levels of VI showed significantly poorer EF outcomes (notably in the domain of cognitive shifting) than a developmentally matched sighted group, with a certain proportion of children obtaining scores of clinical significance. It is possible that the difficulties at the level of executive functioning at school age reflect early vulnerability in attention development. This hypothesis is addressed in the following section, where connections between Part 1 and Part 2 are made.

PART 3: LONGITUDINAL CONNECTION BETWEEN EARLY ATTENTION AND LATER EXECUTIVE FUNCTION IN CHILDREN WITH CONGENITAL VI

INTRODUCTION

The children who took part in the research presented in Part 1 also participated in the research presented in Part 2.[22] This means that longitudinal connections between the two investigations could potentially be made. With this in mind, it is important to emphasise that the coding and analyses of attentional data in Part 2 had been carried out before the BRIEF ratings were obtained at school age, so the analyses of the BRIEF data had no bearing on the results relating to attentional processes.

Because of the small sample at the follow-up stage (Part 1) it was not possible to examine the longitudinal performance of the VI group cross-sectionally, separating those with PVI (who were fewer in numbers at the follow-up) and SVI. Further difficulty arose from the lack of variation and the ceiling effect associated with the attentional data, making it statistically difficult to connect this data to the children's later outcomes, which were more varied. However, establishing connections between the early attentional outcomes in the present VI sample, and their later executive function behaviours may have important clinical and developmental implications, particularly as very few longitudinal studies with children with VI exist.

Theoretically, it is expected that both the early attentional behaviours and the later executive function behaviours (i.e., BRIEF scores) are based on the same underlying processes (see the introductory section of this chapter). Hence, it would not be surprising to find a number of significant negative correlations between the children's outcomes at the two time points. However, it was of specific interest to test the hypothesis predicting the relationship between the attentional shifting in the early years of the VI group, and their performance on the Shift component on the BRIEF, which is defined by cognitive flexibility and deployment of attention.

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[22] It is worth mentioning that one child with SVI from the present VI sample of 15 children (i.e., participant ID: 25) was not included in the research in Part 2, in order to achieve successful developmental matching between the participant groups. However, her data on attention behaviours was available and was included in the analyses presented here.

RESULTS

Spearman's coefficients resulting from correlational analyses are presented in Table 6.12. First, it is worth noting that the early ability to establish and maintain adult-directed attention was found to be uncorrelated with any of the BRIEF behaviours at school age. On the contrary, as predicted, ability to shift adult-directed attention in early development was found to be negatively correlated with the Shift, although only at a $p \leq .05$ level (ρ = -.570; p = 033, n = 14). The Initiate scale on the BRIEF at school age was also found to be highly correlated with the early attention shifting (ρ = -.674; p = 008, n = 14). Interestingly, it may also be worth noting that there were trends towards a significant negative correlation with Emotional Control (ρ = -.488; p = 077, n = 14) and the Behavioural Regulation Index (ρ = -.491; p = 074, n = 14).

Table 6.12: *Spearman coefficients between the children's early attentional performance and their BRIEF ratings at school age*

	Establish	***Maintain***	***Shift***
Inhibit	-.036	.075	-.250
Shift	.006	.144	-.570
Emotional Control	-.240	-.071	-.488
BRI	-.082	.130	-.491
Initiate	-.342	-.313	**-.674****
Working Memory	.114	.039	-.180
Plan / Organise	-.146	-.396	-.336
Organisation of Materials	.008	-.262	.145
Monitor	-.197	-.057	-.056
MI	-.003	-.025	-.025
GEC	-.178	-.309	-.361

** - significant at $p \leq 0.01$

Although interpretation of these results requires caution, it is intriguing to establish a potential association between attention shifting in early childhood and those BRIEF domains which, at school age, differentiated the VI group from sighted children as having significantly more behavioural difficulties. The scatter graphs below (Figures 6.7 and 6.8), showing individual children's data points, graphically illustrate the negative correlations between attention shifting and the Shift and Initiate BRIEF scales, and show that those children who in fact scored below ceiling on the shift component (and did not match the performance of the majority of the sighted comparison group in pre-school period) were the ones who were more likely to show elevated scores on the two BRIEF domains later in childhood. Remarkably, the same non-parametric correlations remain significant at $p \leq .05$ level even after the outlier child (ID: 17) is removed from the analyses (Attention Shift and BRIEF Shift: ρ = -.606; p = 028, n = 13; Attention Shift and BRIEF Initiate: ρ = -.626; p = 022, n = 13). Based on these findings, it is plausible to suggest that the measure of attention shifting tapped the same processes as those involved in specific BRIEF domains, lending some support to the attention coding methodology in Part 2.

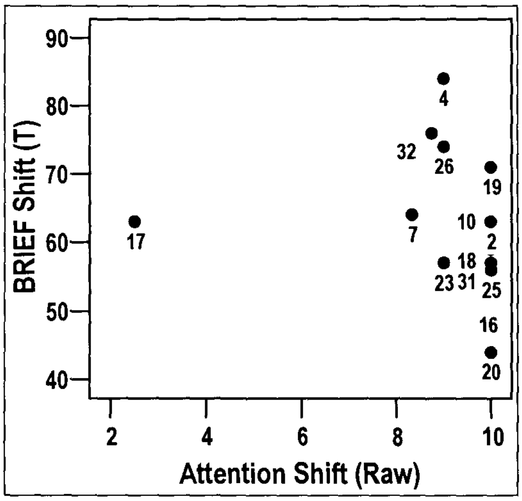

Figure 6.7: Relationship between the Attention Shift and the BRIEF Shift for individual children

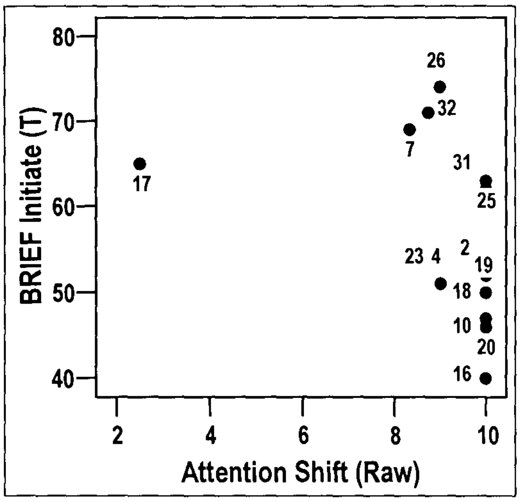

Figure 6.8: Relationship between the Attention Shift and the BRIEF Initiate for individual children

GENERAL DISCUSSION

The present findings are the first to date to provide evidence relating to neuro-cognitive processes of executive functioning and attention of children with congenital VI. In Part 1, it was found that, in the school-age period of development (i.e., 6-12 years), children with congenital VI with normal verbal intelligence showed irregular profiles of everyday executive function behaviours, with the domains of cognitive shifting, emotional regulation, initiating and monitoring emerging as areas of weakness. A potential explanation for the lower levels of ability in these particular EF domains in children with VI is that the functioning of these domains may be mediated by, although not dependent on, visual perception. This is because visual perception may be facilitative to the functioning of the systems (i.e., attention) that provide higher-level executive control of cognitive actions and behaviours.

In relation to this, in Part 2, an observational investigation of early attentional processes of young children with VI (including also the children with VI who participated in research in Part 1) was carried out. The results revealed that early attention may be a risk factor for some children with VI, particularly those with lower cognitive levels and whose VI is of greater severity. However, although related to general cognitive functioning, reduced attentional capacity and control was not found to be exclusive to children with low IQ and children whose VI is of greater severity. This may explain why in Part 1, some children with SVI in their early years and at normal levels of intelligence showed behavioural difficulties in broader executive functioning at school age.

These findings received some support from the findings in Part 3, where a longitudinal examination between early attentional and later executive functioning outcomes unearthed some significant correlations. More specifically, in the present sample of children with varying levels of VI and with verbal intelligence that is within the normal range, those children who showed poorer ability to shift adult-directed attention in their early childhood also seemed to show higher levels of behavioural problems in the area of cognitive shifting and initiation at school age, as reported by their parents. These findings seem to lend support to the measures of attention behaviours developed for this study. More importantly, the findings imply that, in some children with VI, early vulnerability in attention development, specifically in terms of attention shifting, may be a potential marker for long-term difficulties in certain executive functions.

What are the general implications of such findings? With regards to executive function behaviours at school age, it is important to emphasise that the scores of clinical significance were present only in some children with VI. Additionally, the areas of specific weakness were not consistent across the group, and varied across different executive functions measured by the BRIEF. Hence, it is clear that specific executive 'dysfunctions' are not a universal feature of the VI population. However, given the differences with the sighted group, visual impairment does seem to impose a degree of vulnerability in these processes, the impact of which may be evident very early on in development (i.e., in early attention). Considering this vulnerability may be crucial in furthering our understanding of autistic-like presentation in this population.

Firstly, reduced functions in specific executive domains (e.g., shifting) may provide an explanation for certain behavioural patterns seen in children with VI, like repetitive and stereotyped behaviours and restricted interests[23]. Second, such behaviours, as well as underlying EF difficulties, may be a concurrent outcome of the primary socio-developmental vulnerability that is associated with significant vision loss in childhood. Understanding of the children's early attention

[23] It is important to emphasise that certain behavioural characteristics in this domain (e.g., stereotypies or 'blindisms' such as eye-poking) are considered to be normal and unique to VI population (Cass, 1998), although the elevated levels in a wider presentation of perseverative, repetitive and sterotyped behaviours may have clinical significance.

development may prove useful in unpacking the nature of this vulnerability in children with VI, as well as its potentially complex relationship with overall executive functioning. Involvement of attention in both executive and social processes has been a recurrent theme throughout this chapter, and it is easy to appreciate how the early disruptions to its visual manifestations (e.g., eye-gaze cueing, directing and following) may be detrimental to the development of higher-order social and cognitive functions later on (e.g., executive control and theory of mind), both in children with VI and in children with normally developing vision (i.e., as evident from sighted children with autism). However, with regards to children with VI in particular, it is possible that these early vulnerabilities in individual children may need to interact uniquely with the influences of other variables (e.g., socio-environmental and neurological factors, as well as general intelligence levels and the severity of vision loss) to determine the severity of the autism-like picture that is seen later in childhood in this population. To capture the aspects of such potential interactions, the performance of individual children is considered in the following chapter through correlational analyses across the various measures reported so far and additional qualitative observations.

Chapter 7

Cross-study considerations

SUMMARY

The goal of this chapter was to consider the relationships between the outcomes that were examined in the empirical chapters thus far. This was to help sum up the developmental picture provided by the current sample of children with congenital VI throughout this thesis. First, specific research questions were raised regarding the connections between the children's pre-school language and attentional outcomes (i.e., the data presented in the Part 2 of Chapter 6) and their developmental patterns obtained from their assessments at school age (Chapters 3 - 6). Then, specific relationships were considered across the outcomes obtained at school age. Subsequently, the findings presented in this chapter and their brief discussion helped to bring together the findings across the thesis before they are discussed in greater detail in the final discussion chapter.

INTRODUCTION

In the present chapter an attempt was made to provide some further correlational patterns across the experimental chapters thus far. Hence, specific questions arising from these chapters have been addressed as follows:

Question 1 (Q1): Is structural language strength stable over time in children with VI?

The findings in Chapter 3 demonstrated structural language strength of children with VI at school-age. These findings are of particular significance in light of some early language irregularities and delays in structural language domain reported in other research studies (e.g., Andersen et al., 1984; McConachie & Moore, 1994). Hence, they support the idea that children with VI are able to overcome the early language concerns by middle childhood (e.g., Landau & Gleitman, 1995). For this reason, in this chapter it was of interest to determine if there is a longitudinal relationship between the early language and cognitive outcomes and later linguistic competence in the present VI sample of children.

Question 2 (Q2): Are early attentional behaviours markers of later socio-communicative competence in children with VI?

Socio-communicative difficulties in children with VI have been attributed to the breakdown in joint attention in early childhood (Hobson, 1993; 2002), although the longitudinal relationship between early joint attention capacity and poor outcomes in social communication in later childhood in such children has not been established empirically. In fact, given its largely visual nature, very little is known about the developments of early joint attention in visual impairment. Research presented in Chapter 6 has offered some insight into these processes by highlighting some specific challenges in the ability to respond to adult-directed attention in pre-school children with VI. Similarly, the research in Chapter 3 has emphasised ongoing socio-communicative concern in a group of children with VI at school age, some of whom are the same children whose early attentional behaviours were assessed in Chapter 6. Therefore, it is helpful to determine if there is a potential longitudinal relationship between the early attentional behaviours in the present sample children with VI and their later socio-communicative outcomes.

Question 3 (Q3): Are characteristics of mother-child discourse influenced by the child's own linguistic and socio-communicative competence?

The findings in Chapter 5 emphasized some specific strengths of maternal language input to children with VI, and highlighted the potential contribution of sensitive and mind-minded language input provided by mothers to developing socio-cognitive knowledge of their children with VI. However, given that the children with VI in this research all have good and potentially advanced linguistic abilities, it remains uncertain if their mothers' language input is independent or is a reflection of the children's s own language ability and socio-communicative competence. For this reason, it was of interest to investigate a potential relationship between the mother's (as well as the child's) discourse characteristics during the joint-book reading task with the child's language and socio-communicative outcomes, although the causality of these factors may not be possible to establish at the present time.

Question 4 (Q4): What evidence is there for the potential role of short-term memory in structural language skills of children with VI?

In Chapter 3, it was argued that the structural language strength in children with VI may, at least partly, be explained by their strong (potentially superior) short-term memory. Although the children with VI group did not excel at Digit Span, they showed superior performance on a test of verbal recall (i.e., Recalling Sentences), in comparison to their sighted peers. This performance was also found to correlate with the children's verbal IQ. Whilst unpacking the relationship between verbal ability and short-term memory in children with VI is likely to be difficult, here it was of interest to examine what other evidence is there to support it. Thus, the measures of structural language were correlated with the measure of working memory in a behavioural context (i.e., BRIEF).

Question 5 (Q5): Can the pattern of stereotypical and repetitive behaviour and restricted interests in children with VI be explained in terms of specific executive function difficulties?

As discussed in Chapter 6, the pattern of repetitive and stereotypical behaviours and restricted interests that characterise children with autism have been explained in terms of the children's difficulties at a level of executive functioning (López, et al., 2005). As similar behaviours have been observed in children with VI (in the current as well as in the previous research) it is plausible to suggest that their manifestation may be related to challenges with specific executive functions. Hence, it was of interest to investigate a potential relationship between the children's behavioural ratings on the BRIEF and the two socio-communicative checklists used in this research. Given the theoretical framework in the discussion of Part 1, Chapter 6, a particular focus was placed on the possible correlations between the cognitive flexibility component (i.e., Shift) on the BRIEF and the CCC-2 and the SCQ scales assessing stereotypical and repetitive behaviours, and restricted interests.

Question 6 (Q6): Did the same children with VI who showed executive function difficulties also show poor socio-communicative outcomes?

As discussed in Chapter 6, evidence from children with autism demonstrates that executive and social functions are not mutually exclusive. Thus, it is possible that those children with VI who show more behavioural difficulties in the domain of executive functioning are those with poorer socio-communicative outcomes. Here, it was of interest to test this hypothesis by examining if the same children with VI who showed clinically elevated scores on the behavioural rating of executive function on the BRIEF also showed clinically significant scores on the behavioural ratings of social communication on the SCQ and the CCC-2.

RESULTS

Small sample size and high data variability imposed certain constraints on correlational data analyses. With this in mind, while Pearson coefficients were used to correlate the performance on all the structured assessment, Spearman coefficients were used on those occasions where structured assessments were correlated with proportional data. Additionally, given the issues of power, in some circumstances the correlations were carried out only on composite/index scores rather than on individual scales that make up those composites.

In order to investigate stability of language competence over time in children with VI (Q1) correlational analyses were carried out between the language and cognitive outcomes at pre-school age, as assessed by the Reynell-Zinkin Scales (RZS), and at the follow up stage in primary school years, as assessed by the WISC-III and the CELF-3. There was a potential issue with correlating the children's' performance on the RZS in their early development with their later language outcomes, as the RZS are not a standardised measure and provide only raw scores or age equivalents. In relation to this, as discussed previously (Chapter 6), Dale and Sonksen (2002) proposed a derivation of RZS developmental quotients (DQs), which can be calculated as a ratio of the children's raw-score based age equivalents on the appropriate RZS norms (i.e., Blind, Partially Sighted, or Sighted) and their chronological ages. The DQs are not without their problems, as they are, like raw scores and age equivalents, affected by differing developmental progress of children with differing vision levels. However, statistically they resemble scaled scores (by taking into account where the child's performance lies relative to their chronological age), so they were seen as a more appropriate measure for the current correlation analyses.

Pearson's correlations were calculated between the DQs on the three RZS domains (i.e., Sensory Motor Understanding, Verbal Comprehension and Expressive Language Structure) that children were tested on between the ages 17-36 months and the scaled scores on the WISC-III and CELF-3 at school age (i.e., 6-12 years) (Q1). First, it may not be surprising to find a lack of correlations between the sensory-motor domain of the RZS, and the WISC-III and the CELF-3. This is because the latter tests are language-based, compared to the Sensory-Motor

Understanding scale of the RZS, which is performance-based. Second, no highly significant correlations (i.e., p values $\leq .01$) have been detected between the language outcomes at the two time points. However, it is interesting to point out that two correlations were significant at $p \leq .05$ between the children's early and later language outcomes, namely those concerning the two verbal domains on the RZS and the Word Associations on the CELF-3 ($r = .588$, $p = .021$, n = 15; $r = .601$, $p = .030$, n = 13). While it is not clear why only the Word Associations (i.e., test of verbal fluency) produced significant correlations between the RZS and the language-based measures in later development, these correlations are not surprising, given that Word Associations is a vocabulary-based test. However, in answer to Q1, those correlations suggest possible stability of verbal ability levels over time in this sample of children with VI, and imply that certain scales across different assessment measures are likely to tap the same linguistic/cognitive skills. Interestingly, the trend towards a significant correlation between the Verbal Comprehension DQ on RZS and Comprehension Scaled score on the WISC-III ($r = .477$, $p = .072$, n = 15), and the trend between the Expressive Language Structure DQ on RZS and Expressive Language Composite score on the CELF-3 ($r = .524$, $p = .066$, n = 13) may also be supportive of this pattern.

With regards to addressing Q2, correlations between the children's early attentional performance and their scores on the measures of social communication (as assessed by the SCQ and the CCC-2) were calculated. Rather than examining the correlations with the individual CCC-2 scales, the sums of scaled scores across the three broad CCC-2 domains (Structure, Pragmatics, Social) were used instead. Interestingly, no highly significant correlations (i.e., p values $\leq .01$) have been established between the early attentional performance and later socio-communicative outcomes. However, it is of importance to highlight a number of meaningful correlations found to be significant at $p \leq .05$. First, the ability to establish adult-directed attention in preschool stages of development was positively related with the children's summed scaled scores on the CCC-2 Social domain ($\rho = .557$, $p = .039$, n = 14). This suggests that the children with better attention establishing outcomes early on achieved more positive parental ratings in terms of their social competence later in childhood. Second, in line with this result was the negative correlation of attention establishing and the children's total SCQ scores ($\rho = -.516$, $p = .049$, n = 15), indicating that the better outcome on early attention establishing was related to lower incidence of socio-communicative difficulties later in childhood. Importantly, it is worth noting that these non-parametric correlations remained significant at the $p \leq .05$ level after the outlier child (ID: 17, see

Chapter 6, Part 3) was removed from the analyses (CCC-2: ρ = .564, p = .044, n = 13; and SCQ: ρ = - .556, p = .039, n = 14).

In order to address Q3, correlations were calculated between the mother-child discourse outcomes in Chapter 5 and the children's language and their socio-communicative outcomes in Chapter 3. From Table 7.1 it can be seen that the mothers' mentalistic language use (potentially independent of their overall verbosity) was found to be related to the children's language competence, particularly in terms of their comprehension (i.e., mothers' Mental State proportion scores and the children's Receptive and Total Language on the CELF-3: ρ = .800, p = .002, n = 12; and ρ = .733, p = .007, n = 12). Further correlations significant at $p \leq .05$ further support this pattern. Mothers' overall use of mentalistic language during the joint book-reading session was related to the children's VIQ levels on the WISC-III (ρ = .585, p = .046, n = 12), although this did not take into account the total number of elaborations produced by the mother.

Additionally, the children's own mentalistic language during the joint book-reading session was found to be related to the children's VIQ levels on the WISC-III, although only at $p < .05$ (Child: ρ = .612, p = .035, n = 12). Even though this did not take into account the total number of elaborations produced by the child, it is possible that the total number of elaborations was not a factor in the relationship between children's verbal ability and their use of mentalistic elaborations, as their proportion Mental State scores correlated highly with their VIQ's (ρ = .720, p = .002, n = 12). Interestingly, children's discourse during the book reading session appeared unrelated to their language performance on the CELF-3. Overall, this pattern of correlations suggests a general relationship between the quality of the mother-child conversational dialogue and the children's own language levels in the present VI sample (Q3).

Importantly, both children's and mothers' mentalistic language during the joint book-reading task was correlated to the children's pragmatic and social competence on the CCC-2. As expected, the children's proclivity to use mental state language (i.e., Mental States proportion scores) appeared was correlated to their scores on the pragmatic CCC-2 domain (ρ = .639, p = .034, n = 12). However, this may not be independent of their overall elaboration level, considering a trend with Mental States total score (ρ = .575, p = .065, n = 12). The same may be said for the mothers' language characteristics, given the correlations of their total number of Mental State elaborations, as well as their total number of all elaborations, with their children's pragmatic levels (ρ = .633, p = .036, n = 12; ρ = .629, p = .038, n = 12). Similarly, general proclivity to elaborate in both children and mothers correlated with the children's social competence on the CCC-2 (ρ = .755, p = .007, n = 12; ρ = .687, p = .02, n = 12). Overall, in response to Q3, these findings imply that those children with VI who showed better pragmatic and social outcome on the CCC-2 were the ones whose mothers, as well as the children themselves, showed a greater tendency to elaborate on the book content, using also mentalistic language.

Table 7.1: *Spearman coefficients for the correlations between the mother-child discourse elements, and the children's language and socio-communicative outcomes*

	Mother-child discourse Child			***Mother-child discourse Mother***		
	Mental States (total)	Mental States (proportion)	All elaborations (total)	Mental States (total)	Mental States (proportion)	All elaborations (total)
WISC-III						
VIQ	.612	**.720****	.262	.585	.067	.459
CELF-3						
Receptive Language	-.076	-.180	-.056	.095	**.800****	-.225
Expressive Language	.196	.023	.216	.245	.459	.070
Total Language	.177	.045	.125	.288	**.733****	.000
SCQ						
Total	-.145	-.075	-.171	-.158	.218	-.137
CCC-2						
Structure	.254	.428	.194	.205	-.310	.337
Pragmatics	.575	.639	.447	.633	-.196	.629
Social	.507	.263	**.755****	.572	-.206	.687

** - significant at $p \leq .01$

With regards to Q4, correlations between the children's scores on Working Memory scale of the BRIEF and their scores on the structural language scales on the CCC-2, as well as their scores on the standardised tests of verbal ability (CELF-3 and WISC-III) were calculated. Firstly, no significant correlations were found between the Working Memory BRIEF scores and the scores on either the WISC-III or the CELF-3 (*p* values > .05). However, there seemed to be a consistent pattern of significant negative correlations between the children's Working Memory levels on the BRIEF and their scores on certain CCC-2 structural language scales (Speech: $r = -.657$, $p = .01$, n = 14; Coherence: $r = -.558$, $p = .038$, n = 14; summed Structure score: $r = -.562$, $p = .037$, n = 14). This pattern of results does seem to support the idea that short-term (i.e. working) memory may play a role in structural language skills of children with VI, although unpacking the nature of this relationship may need to be investigated further.

In response to Q5, it is quite surprising that, potentially owing to the lack of power, the BRIEF Shift scale did not correlate with the scales that tap the behavioural stereotypes and repetitions on either SCQ or CCC-2 (*p* values > .05, Table 7.2), particularly as cognitive inflexibility (e.g., high Shift scores) has been implied in these behaviours (e.g., Chapter 6). However, a striking pattern of correlations in Table 7.2, which suggests associations of these behaviours with the other behavioural scales that are interdependent with BRIEF Shift, may be meaningful in this respect. More specifically, there was a notable pattern of correlations between the behavioural stereotypes, repetitions and restricted interests on the SCQ and the BRIEF Behavioural Regulation/BRI ($r = .649$, $p = .009$, n = 15), BRIEF Inhibit ($r = .650$, $p = .009$, n = 15) and BRIEF Emotional Control ($r = .571$, $p = .026$, n = 15). Similarly, there were significant negative correlations between BRIEF Emotional control and CCC-2 Stereotyped language ($r = -.680$, $p = .007$, n = 14), and BRIEF Emotional control and CCC-2 Interests ($r = -.566$, $p = .035$, n = 14). Additionally, BRIEF BRI correlated with Stereotyped language on the CCC-2 ($r = -.560$, $p = .037$, n = 14).

Table 7.2: *Pearson coefficients for the correlations between the socio-communicative behaviours on the CCC-2 and SCQ, and the BRIEF*

	Inhibit	***Shift***	***Emotional Control***	***BRI***	***Initiate***	***Working Memory***	***Plan***	***Organising Materials***	***Monitor***	***MI***	***GEC***
CCC											
Speech	-.321	-.183	-.269	-.310	-.355	**-.657****	-.072	-.161	-.339	-.422	-.489
Syntax	-.176	.279	-.140	-.045	-.047	-.089	.341	.196	.145	.095	.048
Semantics	-.489	-.170	-.307	-.392	-.273	-.523	-.057	-.093	-.297	-.316	-.454
Coherence	-.369	-.381	-.298	-.384	-.396	-.558	.082	-.024	-.349	-.320	-.455
Initiation	-.438	-.357	-.555*	-.512	-.150	-.158	.380	.121	-.233	-.032	-.314
Stereotype	-.406	-.382	**-.680****	-.560	-.050	.078	.433	.472	.204	.272	-.135
Context	-.486	-.158	-.300	-.385	-.245	-.431	.005	.093	-.040	-.173	-.346
Non-verbal	-.537	-.263	-.286	-.429	-.345	-.622	.005	-.324	**-.643****	-.487	-.590
Social	-.380	.094	-.379	-.298	-.299	-.433	-.038	-.256	-.485	-.401	-.451
Interests	-.301	-.195	-.566	-.415	.049	.015	.508	.210	-.119	.146	-.135
GCC	-.523	-.264	-.479	-.497	-.301	-.472	.204	.067	-.236	-.209	-.438
Structure	-.415	-.131	-.313	-.345	-.329	-.562	.099	-.021	-.253	-.293	-.411
Pragmatic	-.609	-.401	-.639	-.639	-.240	-.322	.313	.168	-.195	-.088	-.431
Social	-.395	-.049	-.539	-.407	-.154	-.254	.254	-.041	-.359	-.163	-.348
SCQ											
Total	.351	.201	.352	.365	.472	.433	.236	.527	.479	.540	.572
Social Interaction	-.074	-.119	-.135	-.105	.273	.356	.258	.611	.412	.483	.266
Communication	.165	.165	.395	.281	.403	.162	.481	.319	.282	.398	.418
Stereotyped Behaviours	**.650****	.441	.571	**.649****	.262	.258	-.145	.121	.226	.187	.506

** - significant at $p \leq .01$

In relation to these correlations, it was of interest to see if the BRIEF was sensitive to the same children who scored in the clinically significant domain on the SCQ and the CCC-2 and (Q6). Table 7.3 illustrates the individual children's profiles on the three parental questionnaires. The areas shaded in dark grey indicate that the child's profile on that measure is in the clinical domain. The areas shaded in light grey indicate that the child's profile, although not reaching the clinical cut-off, is in line with the bottom 10% of the children in the sighted comparison group (only possible to calculate for the SCQ and the BRIEF). Here, it can be seen that only one child with VI obtained a profile that was in the clinical domain on all three measures. This was an 8 year old girl with SVI in her early years and VIQ = 95 (i.e., participant ID: 26). Interestingly, it may be worth noting that, although her attentional performance in early childhood was near the ceiling, it was still below the level of the sighted comparison group. Additionally, although she did not have an additional diagnosis at the time of her participation in this research, it may be worth emphasising that she is the only participant in the present VI sample who did not attend a mainstream school at the time of the present research, but had been placed in a specialist school for children with VI. Thus, it is possible that her behavioural characteristics that were captured here have already been identified professionally outside of the context of this research.

Table 7.3: *Individual children's profiles*

Child ID	SCQ	CCC – 2	BRIEF - BRI	BRIEF - MI	BRIEF - GEC
02					
04					
07					
09					
10					
16					
17					
18					
19					
20					
23					
25					
26					
31					
32					

Area shaded in black represents missing data

The profiles of other children were not as clear cut. However, they may be worth discussing very briefly, because it was possible to discern three broad profile patterns. The first pattern encompasses children whose profiles are not within the area of clinical concern on any of the measures (participant IDs: 02, 10, 16, and 31). As discussed previously in Chapter 3, these children do not provide any clues, in terms of their cognitive level, family background or visual level and diagnosis, into their seemingly better outcomes. It may be worth noting that the early attention performance of three of these children was at ceiling. Interestingly, three of these children (participant IDs: 02, 10, and 16) were in the PVI group at the time of the attention study, although the participant 02 obtained some useful functional vision (i.e., SVI) after the age of 16 months.

The second pattern encompasses the profiles of children whose socio-communicative profiles may be of clinical concern, but their overall BRIEF ratings appear to be within the normal range (e.g., participant IDs: 04, 09, 17, 20 and 23). However, despite the BRIEF index and composite *T* scores that were in the normal range, a closer examination of the individual BRIEF scales reveals the Shift scores to be an area of relative weakness in three of these children. One child was chosen here to illustrate this developmental pattern (i.e., participant ID: 17). This boy with SVI in his early years showed particularly low attentional performance at an earlier age, especially in attention shifting (Chapter 6, Figures 6.7 and 6.8). During the observational analyses of this child's attentional behaviours in his early years, it was observed that he showed strong negative emotional reactions to being guided by an adult during the assessment and was distressed at the assessor's attempts to guide him away from the toys he was engaging with. Closer examination of his BRIEF profile at school age revealed that, although his overall BRIEF ratings were generally within a normal range, his Shift scores were just below the clinical cut-off ($T = 63$) and substantially elevated in comparison to the sighted group's mean. More importantly, his scores on the Emotional Control scale on the BRIEF exceeded the clinical cut-off of 65 and above ($T = 69$). In the previous chapter, it was shown that emotional regulation and cognitive shifting are related in both children with VI and those who are sighted. It was also discussed that executive control may facilitate emotional modulation, in that the ability to shift attention away from upsetting events may help to reduce negative emotions and vice versa. This child's data at an early, as well as at older age, may be illustrating this pattern.

The third pattern encompasses the profiles of three children, who showed socio-communicative profiles of potential clinical significance and specific weaknesses on the metacognitive aspect of the BRIEF (i.e., participant IDs: 07, 18, and 25). It is not easy to summarise the developmental pattern in this group, as the children did not show weaknesses in the same metacognitive areas. However, in at least two children, the scores on the BRIEF Initiate and Monitor scales were near or above the clinical cut-off. These children perhaps illustrate the point made in the preceding chapters that the poorer outcomes on these BRIEF scales may be a secondary outcome of the child's socio-communicative difficulties. It may also be worth noting that two of these children (participants IDs: 07 and 18) showed relatively low levels of attention maintaining in early childhood (Chapter 6, Figure 6.6), and attention maintaining was argued to be most socially-driven of the three attentional behaviours assessed here.

Finally, the developmental picture of the remaining child (participant ID: 32), who obtained highly elevated scores on the BRIEF despite the socio-communicative profiles in the normal range are somewhat challenging to explain in terms of the underlying socio-communicative vulnerabilities. However, it is worth emphasising that this child's early attention behaviours were not as proficient as those in the majority of children in her vision group (i.e., SVI) (Chapter 6, Figures 6.4 - 6.6), and the parental ratings of her behaviours on the BRIEF appeared sensitive to the same underlying vulnerabilities.

DISCUSSION

The first question this chapter attempted to address concerned the stability of structural language strength in children with VI over time. In relation to this, significant correlations between the children's early RZS outcomes and their later performance on the CELF-3 have been detected. These results suggest that the verbal competence in children with VI is likely to be stable over time as different assessment measures presented at different time points may tap the same linguistic/cognitive skills. Even though the RZS are not a standardised tool, they have a widely recognised value in assessing the developmental progress of young children with varying levels of VI. Hence, despite their low psychometric properties, it is encouraging to see their potential to predict children's verbal levels in subsequent development.

Second aim of the research presented in this chapter was to establish whether there was a longitudinal relationship between the early attentional ability and later socio-communicative outcomes in children with VI. In relation to this, significant correlations were found between the children's ability to establish adult-directed attention in their pre-school years and their scores on the parental measures of social competence at school-age. These correlations may reflect the fact that the children's attentional performance was assessed in a socio-interactive context, which was likely to mediate child's attentional responses. More specifically, establishing attention onto a toy offered by the assessor in the context of this particular assessment may involve a certain level of intersubjectivity from the child's side and acknowledgement of the assessor as the social partner. Despite the methodological difficulties with the measure of early attentional behaviours, it was of value to find that aspects of this measure were sensitive enough to unearth a longitudinal relationship between the ability to respond to adult-directed attention in early childhood and the children's later social competencies. This implies an attentional basis to socio-communicative skills in children with VI and is in line with theories and empirical findings that emphasise a developmental link between attention development, joint attention skills and subsequent socio-communicative achievements, both in typically developing sighted children and sighted children with autism (e.g., Butterworth & Cochran, 1980; Leekam & Ramsden, 2006). Thus, early difficulties with establishing adult-directed attention may potentially be an early marker of long-term socio-communicative problems in children with congenital VI, although this needs to be substantiated with further research.

In this chapter it was also of interest to examine if the characteristics of mother-child discourse were influenced by the child's own linguistic and socio-communicative competence in children with VI. In relation to this, significant correlations between mothers' discourse (including their mentalistic language) and the children's linguistic, pragmatic and social outcomes were obtained in this chapter. Interestingly, the children's own discourse and mentalistic language were found to be correlated with their linguistic abilities. These correlations provide evidence for an enduring, yet complex, relationship between language and socio-communicative development in children with VI, and the language input that such children receive in their daily lives. However, it was not possible to unearth the causality within this relationship. While it is likely that the language that mothers direct to their children with VI (including also certain levels of mind-mindedness) plays an important role in the ways that their children communicate and interact with others, it is also possible that the mothers' own levels of language input (including their mentalistic discourse) to their children are affected by their children's own language levels and social responsiveness. Considering that the verbal quality of mother-child interaction is intertwined with the linguistic ability of the child with VI, as well as their levels of social and pragmatic competence, mother-child dialogue seems to be a clear target for future interventions.

Following from the research presented in Chapter 3, it was of interest to examine further evidence for a potential role of short-term memory in structural language in children with VI. In relation to this, there was as significant correlation between the working memory component on the BRIEF and the structural language composite score on the CCC-2, as well as its individual aspects such as speech and coherence. On the other hand, the BRIEF working memory was uncorrelated with the standardised language measures (CELF-3 and WISC-III). This pattern of findings makes it difficult to fully unpack the nature of the relationship between short-term memory and language. However, they may reflect the complexity of the short-term measures and a possibility that its outcomes are context dependent. This potentially explains the significant correlation pattern between the two variables of interest on two questionnaire measures, but not when standardised language measure is concerned. More specifically, the two questionnaires (where working memory and structural language skills were found to be related) tap those skills in terms of the children's everyday behaviours as perceived by their parents. Similarly, despite the lack of relationship between the short-term memory in the context of everyday behaviours (the BRIEF Working Memory) and structural language as assessed by the standardised language measures (WISC-III and CELF-3), this relationship was indeed established when short-term memory was assessed in a context of a structured assessment (Recalling Sentences). The possibility that short-term memory is contextually manifested may potentially explain the general lack of correlations between the scores on Digit Span, Recalling Sentences and Working Memory, all of which are measures of short-term memory.

An additional question this chapter attempted to address was whether the pattern of stereotypical and repetitive behaviours and restricted interests in children with VI could be explained in terms of specific difficulties at a level of executive functioning. With regards to this, it was surprising to find that the BRIEF Shift scale scores (i.e., a behavioural measure of cognitive flexibility) did not correlate with the relevant behavioural scales on either socio-communicative questionnaire. This finding may potentially be down to reduced experimental power, particularly as there were correlations with other related BRIEF scales which have been found to correlate with the Shift component in Chapter 6 (i.e., emotional regulation and inhibition scales, which together with the Shift, form the behavioural regulation composite on the BRIEF). Unfortunately, no conclusion can be drawn regarding the relationship between cognitive shifting and behavioural repetitions, sterotypies and limited repertoire of interests. However, it is clear how high scores on shift-related scale such as Emotional Control may also account for these behaviours in children with VI. For instance, those children with VI who are more perseverative and repetitive in their behaviours and interests may also be more likely to show negative emotional responses (e.g., tantrums) when those behaviours and interests are interrupted, because of their inability to flexibly shift cognitive set. Interestingly, the correlation between the BRIEF Inhibit and repetitive and stereotypical behaviour on the SCQ (suggesting that children with VI who have poorer motor inhibition may present with higher levels of stereotyped behaviour) may have implications for our understanding of specific 'blindisms' in children with VI, the behavioural characteristics that are often considered to be a 'normal' aspect of such children's behavioural phenotype (i.e., unusual and repetitive motor movements, such as hand-flapping) (Cass, 1998).

Finally, the last question this chapter attempted to address was whether the same children with VI who showed more behavioural difficulties in the domain of executive functioning on the BRIEF also showed poorer socio-communicative outcomes on the SCQ and the CCC-2. Here, an individual-differences approach was adopted, highlighting the behavioural profile of each child with VI on the three parental-report measures of everyday behaviours. It is clear from the presentation of these profiles that there is a certain level of consistency in terms of behavioural outcomes in both social communication and executive function, confirming previous argument that the two may not be mutually exclusive. However, it was also evident that less desirable scores on socio-communicative measures did not necessarily imply poorer scores on the BRIEF and vice versa. In relation to this, the comparison of these outcomes with the children's early attentional behaviours shed some light on this pattern of findings, suggesting that the poorer behavioural outcomes at school age can potentially be traced to lower attentional levels in some children with VI. This is also in line with the significant relationship between attention establishing in early childhood and social competence at school age demonstrated earlier in this chapter. Importantly, it was not possible to define the present sample of children with VI with a clear distinction between the children with and those without significant socio-communicative and executive function challenges. While this may potentially be owing to the small sample, this pattern of heterogeneity further highlights the remarkable nature of children with VI, the challenges of conducting research studies involving such children, and the subsequent difficulties in conceptualising their unique strengths and weaknesses. The discussion of these important issues is developed further in the concluding chapter of this thesis.

Chapter 8

General Discussion

At the outset of this thesis it was noted that children who are visually impaired from birth are vulnerable in their achievement of important social milestones. It has been argued that this vulnerability may arise from early childhood disruptions to vision-dominated behaviours (e.g., eye-gaze directing and following) in the context of joint attention. Moreover, subsequent socio-communicative and socio-cognitive difficulties experienced by such children have been likened to the social impairments seen in sighted children with autism. While such serious effects of vision loss are likely to carry important implications for the children's social lives long term, the goal of the present thesis was to consider the potential of children with congenital VI to overcome these vulnerabilities, with a specific emphasis on language as a possible compensatory mechanism. The verbal domain has generally been highlighted as an area of strength in children with VI, so it is intriguing to consider what language, as an information channel, may provide for such children. However, to date, empirical research has not been able to establish whether language can compensate for the role of vision in dealing with the demands of the everyday, vision-dominated, and inherently social world of which children with VI are a part. It is now important to reflect on how the present findings link in with the existing knowledge of this area and to consider the unique contribution of the current research.

The findings presented in Chapter 3 provide important original evidence on the role of language in the development of children with congenital VI. First, it was shown that children with VI, whose intelligence levels are within the normal range, may have a language-specific strength that distinguishes them from sighted children, as their performance on a standardised test of language function was shown to exceed that of sighted peers. Interestingly, what is striking about this potential language advantage of children with VI is that it a) may be confined to the domain of structural language, b) might be driven by their strong verbal short-term memory, and c) may be evident only when assessed in a structured context. This has been argued in some depth in Chapter 3, and the correlation between everyday working memory ability on the BRIEF and structural language skills on the CCC-2 in Chapter 7 further supports this idea. Additionally, the solid nature of structural language trends across development (i.e., Chapter 7) provides heartening evidence that strength in this language domain is likely to be stable over time in those children with VI who show early developmental promise.

However, in contrast to the strong structural language function in children with VI, their pragmatic language development seems to be an area of an ongoing concern at primary school age, even in those children with advanced intellectual and verbal capabilities. This is the first piece of research that has systematically demonstrated such a serious discrepancy in the language presentation of children with VI. Importantly, this suggests that solid cognitive grounding in verbal intelligence and structural language in children with VI may not be sufficient to fully compensate for the processes and behaviours that are required for dealing with the complexity of everyday demands of pragmatic communication and social interaction. This is potentially because these processes and behaviours may be particularly dependent on the successful development of visually-driven precursors, such as joint attention and triadic, interpersonal engagement. These arguments support the theory of Hobson (1990; 1993; 2002), who emphasised the role of visual behaviours that are at the heart of a child's developing intersubjectivity. According to Hobson, the visual means of observation and imitation allow the expression of a child's innate, affectively-driven predisposition to identify with and relate to others (who the child perceives to be like him/her), and to comprehend their unique relatedness with other agents in the environment. Similar to Hobson, the present argument postulates that it is not visual perception per se, but the 'easy' route to intersubjectivity that vision may provide to pre-lingual children, which allows their spontaneous integration into the social world that surrounds them. Thus, it may be the breakdown of the 'main vehicle' along this route that may predispose a child with VI to socio-communicative vulnerabilities, the nature of which is reminiscent of the difficulties that characterise autism in sighted children.

The breakdown in visual means does not imply that intersubjectivity cannot be achieved by an individual with VI, because many adults with congenital VI are known to lead independent and fulfilling social lives. In the present school-age sample, some children with VI appeared to be more or less like sighted children on the current measures, while in others, there was a wide range of severity of socio-communicative difficulties. There clearly seems to be a potential in children with VI, who do not have any additional impairments, to overcome such developmental obstacles, even though these may still pose an ongoing challenge, in varying degrees, in their primary school years. It is possible that the early acquisition of language (the developmental process that seems relatively spared in VI), in addition to the sensitive language input by the primary caregiver(s), kick-starts alternative developmental pathways for children with VI. The workings of these pathways may still be establishing throughout the primary school age, which is a prolific period of knowledge acquisition for any child. It seems an intriguing prospect to follow such children from the earliest stages of development, when primary intersubjectivity first emerges, into adulthood, by which time they would have reached full socio-cognitive potential, in order to capture their unique transitions between the crucial social milestones. However, at this point in time, the present findings may provide an invaluable snapshot into the developing social knowledge of such children at school age.

The research presented in Chapter 4 may provide a useful context within which socio-cognitive understanding in children with VI can be considered. The previous studies (e.g., Green et al., 2004; Peterson et al., 2000), which have relied on false-belief tasks, have been invaluable in bringing socio-cognitive concerns in children with VI into the spotlight. However, the dichotomous nature of the false-belief paradigm used in these studies may have somewhat obscured and underestimated the emerging mentalising capabilities of such children. On the contrary, by targeting the social understanding of children with VI in their dominant (i.e., verbal) domain, the research presented in Chapter 4 has captured their mentalising potential more subtly. Here, children with VI demonstrated good sensitivity to the subjective desires and beliefs of story protagonists, by spontaneously referring to these states in their explanations of the protagonists' emotions, and doing so as frequently as their sighted peers. Without a doubt, such language use is likely to be indicative of an active theory of mind and an insight into the subjective mental states of others (in line with Bartsch & Wellman, 1995; Symons, 2004). This, in turn, is likely to be suggestive of socio-pragmatic language potential in children with VI.

However, while the mentalistic language use of children with VI may reveal their existing social understanding, it still remains uncertain how well such children can apply this knowledge implicitly for the purpose of social interaction, where it is not 'scripted'. Judging by the parental reports of their everyday socio-communicative capabilities presented in Chapter 3, it is plausible to predict that representing mental states in real-life social circumstances would still be a challenge for many of these children. Nevertheless, language ability is thought to play an important role in scaffolding children's social understanding in general, providing them with both the semantic basis for various mental state concepts, and the grammatical basis for understanding how such concepts are embedded in everyday language (Ruffman et al., 2003; Slade & Ruffman, 2005). Therefore, the structural language strengths of children with VI (as seen in Chapter 3) are likely to have provided a foundation for their mentalistic understanding, as is evident from their ability to use mental state language in their discourse, and may prove to be a useful bootstrapping mechanism in these processes in the long term. However, the quality and nature of such understanding is likely to differ from that of sighted children, at least while it is actively developing, as it is acquired through inherently different experiences.

The social and communicative environment within which children's social understanding is acquired is where the experiential differences between children with VI and sighted children notably come to light. The research in Chapter 5 highlighted some key aspects of this environment. For instance, this research demonstrated children with VI to have dependence on language input provided by the interacting parent, and prominently so in those circumstances where children who are sighted would typically benefit from their own visual experiences (i.e., the descriptive characteristics of the story-book content). More importantly, while this research pinpointed the strong inter-dependence between the children's own and their mothers' overall mentalistic language in general, the mothers of children with VI seemed more prone to verbalising the subjective mental states of the story characters than were the mothers of sighted children. This subtle finding may offer an important snapshot into parental sensitivity towards their child's socio-cognitive needs. Learning about other people's mental states may come very spontaneously to sighted children, through observing and imitating others' facial expressions, gestures and eye-gaze direction. However, similar knowledge in children with VI may need to be achieved more explicitly, most notably through language. As discussed above, the child's own structural language may provide a solid basis for acquisition of social understanding. Interestingly, the findings in Chapter 5 suggest that the primary caregiver's involvement in this process is likely to play an even more significant role. The mothers of children who are sighted may not always feel a need to point out to their children what seems 'obvious'. On the other hand, the mothers of children who cannot see other people's facial or gestural clues to their inner mental states may adopt the role of a compensatory information channel for their child. As well as interacting with the child's unique sensory needs, such maternal sensitivity was shown to be related to the language levels of children with VI (Chapter 7). Similarly, the mothers' overall language provision was shown to be related to the socio-pragmatic competence of their child with VI (Chapter 7). Disentangling the complex relationship between the characteristics that tie together the mother-child social interaction and communication is challenging, particularly in such a unique population. However, it is possible that the quality of a mother's input to her child with VI may have causal value in this relationship, given that the child is the learner. This would certainly be in keeping with Ruffman at al. (2002) and Meins et al. (2003) who showed that maternal mind-mindedness is a unique predictor of the developing social knowledge in sighted children, even when the children's language and their own mentalising levels are taken into account.

Thus far, it can be argued that language can potentially make a very significant contribution to the development of social functioning in children with VI both through sensitive verbal input by the caregiver and the children's productive output. This contribution may, to an extent, interact with general cognition, as low verbal IQ has been previously identified as a confounding factor in socio communicative and socio-cognitive outcomes in children with VI (Green et al., 2004; Hobson et al., 1999). In this research, examining the outcomes of children with VI whose verbal intelligence levels are within the normal range enabled us, to a certain degree, to appreciate the role of language over and above general cognitive capacity. However, such language contribution does not sufficiently explain why some children with VI show better socio-developmental outcomes than others, and more importantly, why some children with VI, despite their cognitive and language proficiency, appear to have more profound socio-pragmatic problems. While these problems may be reminiscent of autism-related difficulties in sighted children, the nature of the autistic-like clinical picture in children with congenital VI is bound to be multi-factorial and intrinsically different from autism in the sighted child.

Naturally, the inherently unique factor within this structure is the children's sensory impairment. The role and potency of visual provision has been specified earlier in this discussion, with an emphasis on the vision-driven route towards the development of interpersonal engagement in the early, pre-lingual child. The present research suggests that the role of such provision is potentially even greater than was previously thought, with far-reaching effects on a child's wider behavioural and cognitive presentation. For instance, in Chapter 6, children with VI showed vulnerable outcomes in certain domains of executive functioning, particularly in shifting, emotional modulation and initiative ability. Rather than implying a neuro-cognitive impairment per se in children with VI, it was suggested in Chapter 6 that these specific executive functions may be more vulnerable for such children because visual impairment may hinder the mechanism that drives these functions. More specifically, vision may play an important facilitatory role in attentional control. The retrospective study of young children with congenital VI (including those who were subsequently seen at school age) supports this argument, by providing unique evidence that early attentional regulation (i.e., establishing, maintaining and shifting adult-directed attention) is indeed an area at risk for children born with a significant sight loss, particularly in those children whose VI is of greater severity. The deployment of attention has generally been given a central role in the execution of cognitive actions and behaviours (e.g., P. Anderson, 2003; Norman & Shallice, 1986), as well as socio-communicative and socio-cognitive achievements (e.g., Leekam et al., 2000; Leslie, Friedman, & German, 2004). Therefore, it was fascinating to find a longitudinal relationship between the early ability to shift adult-guided attention, and the BRIEF correlates of shifting capacity (as well as initiating and, potentially, emotional regulation) at school age (Chapter 6). Of similar value was the longitudinal relationship found between the early attention establishing capacity and parent-reported levels of social interaction later in childhood (Chapter 7).

Executive and social behaviours and processes can be very difficult to separate, because of their developmental and functional overlap (Carlson & Moses, 2001; Carlson et al., 2002). Such overlap in children with VI has been implied by some of the correlational patterns in Chapter 7. Based on the present research, it could be hypothesised that the complex interaction between executive and social functions may be owing to the involvement of attention in both domains. Thus, the early attentional behaviours may be an early indicator of long term strength and vulnerability in these processes in all children, including those with VI. Similar developmental patterns can also be implied from the research with children with autism, who have been associated with vulnerable outcomes in social and executive functioning, as well as in attention development. Crucially, while such developmental pattern in autism has been suggested to link in with a core frontal lobe deficit (e.g., Ozonoff et al., 2004), children with VI may differ in that their underlying risk factor might be their impoverished visual input. Naturally, further investigation is required to help us better understand these issues.

Another crucial factor within the multi-faceted, autistic-like presentation in children with VI is the children's unique social environment, of which language is an important part. The nature and the role of this environment have already been discussed in much detail. The possibility that the developmental vulnerability imposed by VI in childhood could be somewhat ameliorated through sensitive parental language contribution, as well as through the children's own linguistic knowledge, has a great value from an intervention perspective. These aspects are likely to further interact with other important socio-cultural characteristics of the child's life, such as the influence of peers, siblings and other family members, and even their parents' and other people's continuous reactions to the demands of their disability (Warren, 1994). Although such variables were not examined in the present research, they certainly present an exciting prospect for future studies. Additionally, further research might benefit from considering older children and young adults with congenital VI who have gone through the trajectory described in this thesis, and the insight these individuals can provide us with when reflecting upon their childhood.

While the child's own verbal ability (i.e., verbal IQ) may be an important mediator of the severity the autistic-like phenotype in VI (R. Brown et al., 1997; Hobson et al., 1999), the most severe presentation of autistic features in VI does not seem to be confined solely to children with learning difficulties. This brings into focus another factor that may be influential in shaping the nature of the autism-like clinical picture in children with VI: the child's innate, biological predisposition to specific personal qualities (e.g., temperament). In the general population, some children may be inherently more empathic, socially insightful, and confident in communicating and interacting with other people, compared to others who naturally may be socially withdrawn. There is no reason to believe that the same diversity of personal characteristics would not be seen also in the wider population of children with congenital VI. Thus, it seems that the unique interaction across time, between i) the vulnerability intrinsically created by the children's visual impairment, ii) the powerful socio-environmental and linguistic influences, and iii) the child's innate predisposition towards certain qualities is what may determine the severity of the autistic-like picture in children with VI. Through development, and the social experiences that come naturally with different developmental stages, the severity of such a picture may change in individual children across time, and may potentially even be overcome by adulthood. Ultimately, however, in some children with VI, the interaction between these variables may produce a disorder that is very much like autism in the sighted. Certain conditions, such as the presence of learning difficulties, may exacerbate this developmental picture, given that the child may lack the cognitive capacity to explicitly adopt appropriate learning and compensatory strategies. Finally, some children with VI indeed may have the core disorder of autism itself.

The discussion above accentuates just how unusual and unique children with congenital VI are. In the present research, this uniqueness was potentially most notably captured in Chapter 7, where it was not possible to identify suitable groupings within the current VI sample. The unique nature of the VI-related developmental outcome is a stark reminder of the theoretical, as well as methodological, difficulty of comparing children with congenital VI to those who are sighted. As discussed earlier, the quality of the experiences in the two groups of children, and subsequently, in their overall learning process, must be genuinely different. This, in turn, may not only impact on the expression of their developmental trajectories, but also on the research methods which attempt to capture those trajectories. For instance, at the outset of this research it was deemed important to adapt the available tests to the sensory needs of children with VI, for example, by removing individual items that did not seem suitable for such children (e.g., in the CCC-2 and the BRIEF), and by relying only on a partial assessment of specific developmental domains (e.g., the CELF-3 and the WISC-III). However, while trying to impose a certain level of rigour to suit the experiences of children with VI, such methods may be disadvantageous to the performance profiles of sighted children, for whom such tests are generally designed. Finally, as they are not developed with children with VI in mind, the same tests are likely to be less sensitive to the developmental strengths of this group, while heavily loading on the domains that may emphasise their inherent weaknesses. This clearly illustrates the difficulty of contrasting children with VI with those who are sighted.

Comparing the outcomes between children with differing levels of sight loss (e.g., PVI vs. SVI) may prove to be a more realistic and informative method in the long term, than a comparison with the sighted. The children's ability to adapt to the various developmental tasks is likely to vary with the degree of their VI, and there is substantial evidence for this variation across different developmental domains (e.g., Dale & Sonksen, 2002; McConachie & Moore, 1994; Preisler, 1991). In line with the earlier discussion of the fulfilling role of vision, it is not surprising to find that even a small amount of vision can be facilitative of certain developmental outcomes, most notably motor behaviour, spatial orientation and object permanence (Warren, 1994). Although it was not possible to carry out comparisons between children with differing degrees of VI systematically at all time points in the present research, the study of attentional behaviours in a larger group of pre-school children with VI was able to capture the advantage of residual visual capability in children with SVI in their early years (e.g., in attention shifting). Such advantage in children with some, however limited, functional vision is likely to have significant developmental implications in the long term. Promoting residual vision in children with SVI should be an important element of subsequent clinical and educational intervention (Sonksen et al., 1991).

In empirical terms, however, even the cross-sectional consideration of children with differing degrees of VI can be problematic for several reasons. Visual impairment is not a categorical condition, but one that lies on the continuum of decreasing visual function. Very few children who are born with a serious sight loss have no functional vision at all, while the levels of residual vision in those who do (e.g., SVI) tend to vary immensely. Additionally, VI does not present as a stable variable across an individual's developmental trajectory, owing to maturational changes of the visual system in early childhood, so it is not uncommon to detect changes in the levels of residual vision in individual children across time. This creates a serious challenge for categorising children according to VI level at any given developmental assessment. This is particularly true when evaluating the long-term impact of VI on a child's developmental process, as this sub-grouping cannot control for the potential change in the VI levels in individual children prior to or following any given assessment. Moreover, even when differences are found between the developmental outcomes of children with residual functional vision and those who suffer from complete sight loss, this picture can be difficult to interpret, because of the intermediate influence of other variables that are found to play a significant part (e.g., learning difficulties and the child's unique socio-cultural environment) (Warren, 1994). Certainly, even in the present research (e.g., an advantage of children with SVI in early attentional shifting) learning difficulties imposed a degree of confounding that could not be explained by the presence (or lack) of form vision.

Nevertheless, the present research provides evidence that even those children with some limited levels of functional vision, despite promising cognitive outcomes, are vulnerable to socio-communicative difficulties that bear resemblance to autism in the sighted. Although children with SVI in their early years may be able to locate objects and people in their immediate visual environment, and even perceive broad bodily gestures, their available vision may not be sufficiently sensitive to the subtleties of eye-gaze behaviours and facial expressions that are at the heart of human intersubjectivity. In this respect, all children with a serious congenital sight loss may be at a similar disadvantage. Eye-gaze behaviours are thought to have an evolutionary origin and their role has been identified in virtually all living beings (Emery, 2000). However, the rich information provided by the subtle expressions of the eyes and the face may have particular significance for humans alone. This is because the elaborate system of visual signalling through eyes and faces forms part of the sophisticated representational socio-cognitive system, which uniquely distinguishes humans from other species (Povinelli & Preuss, 1995; Tomasello & Rakoczy, 2003). Thus, it is easy to appreciate how disruptions to this process, as seen from autism and visual impairment, may have a profound impact on an individual's social life.

Naturally, such concerns call for the implementation of appropriate intervention strategies, wherever possible, at an early as well as at an older age. Based on the current findings, there was clearly a spectrum of vulnerability that is open to intervention, even in children with congenital VI but proficient cognition and linguistic skills. Given that the nature of such vulnerabilities in these children appears to be social rather than cognitive, intervention programmes for such children may need to follow the same pattern. However, implementing an intervention strategy with school-age children, whose socio-communicative vulnerabilities have already left a profound mark on their adjustment and integration into the social world, is likely to be challenging. In the UK, it is normal practice to integrate children with VI into mainstream educational settings, unless they have complex educational needs (e.g., as a result of severe learning difficulties). Early socio-emotional difficulties have been found to affect integration into mainstream nursery and preschool settings in young children with severe and profound levels of VI (Brambring, 2001; Preisler, 1993). Such integration may be even more difficult in middle and later childhood, which is the period marked by the emergence of a greater need for independence and the development of the self (e.g., Erikson, 1968). Interestingly, the school context may provide a useful framework within which the effectiveness of an intervention programme with school-age children with VI, with a possible emphasis on peer-assisted socio-interactive learning, might be evaluated.

Without a doubt, it is in the individual child's best interest to have access to appropriate intervention as early as possible. The present findings imply that, in order to establish an effective early intervention framework for children with VI, it is crucial to identify early risk and protective factors and processes that have long-term implications for a child's socio-emotional and behavioural adjustment, with a view to reducing their negative impact (also Dale & Salt, 2007). Developed through such an approach (e.g., Salt, Dale, Osborne, & Tadić, 2005), new materials for helping and guiding the parents of infants and young children with congenital VI, and the professionals who closely work with them, are now available in the UK (Developmental Journal for Babies and Children with Visual Impairment, DfES, 2006), although their effectiveness has yet to be evaluated systematically.

In addition to informing an appropriate intervention framework, the current findings also emphasise the need for establishing a suitable diagnostic process for children with VI. Diagnostically, distinguishing core autism in children with VI from the VI-related syndrome that is similar to autism in the sighted may not be possible. However, from a clinical perspective, the present findings highlight the need to identify children with VI on a spectrum of socio-communicative abnormalities that require professional attention. To achieve this, the development of appropriate diagnostic procedures for assessing a broader clinical phenotype in children with VI (whether this phenotype be described as 'autistic-like' or as 'VI-related socio-communicative impairment') is of utmost importance, in order to meet the children's specific clinical and educational needs. The current diagnostic criteria for autism in sighted children may provide a useful clinical guide, even though the existing assessment process is not fully satisfactory, given its heavy reliance on visual testing materials. Therefore, in order to enhance the diagnostic process for children with VI, it is essential to establish an assessment framework that is sensitive to their unique sensory experiences.

Despite the evident challenges in research with children with VI, the present work demonstrates that such research can be successfully achieved, and needs to continue to make an even bigger impact on our understanding of their development. Needless to say, there is an outstanding necessity both in research and clinical practice for appropriate standardised measures for assessing developmental outcomes of pre-school and school-aged children with VI. However, it is hoped that the present findings may provide a helpful guide for some future empirical work. For instance, it would be useful to develop performance-based measures of such children's pragmatic capabilities that capitalise on their strengths in the language domain and consider their own social lives. This could be achieved by eliciting narratives about people who are an important part of the child's social environment (e.g., siblings, best friends, parents), and by cuing such narratives in relation to a particular social context that is meaningful to the child (e.g., a birthday party or a school trip). As argued previously in this discussion, considering the experiences and knowledge of adolescents and adults with congenital VI may be particularly revealing in this respect. Given that these individuals have come from the same trajectory, their insight into the social experiences of children with VI may be especially enlightening. Research may especially benefit from targeting such individuals' own awareness of their sensory impairment and their perception of how it might (or might not) affect their social adjustment and ability to relate to others. The same approach might shed light on how VI affects the functioning of other important domains in these individuals, and the strategies they may explicitly rely on in dealing with the cognitive and physical demands of everyday, as well as experimental, tasks.

In conclusion, the research presented in this thesis is believed to offer a unique contribution to understanding the social development of children with congenital VI. This research brings to the forefront vulnerable processes in the developmental trajectory of such children, as well as the factors that may play a protective role along this pathway, with a potential to enhance the developmental climate in children with VI in the long term. As well as carrying important implications for intervention and diagnostic consideration with children with congenital VI, the current findings are believed to have significant bearing on our general understanding of the mechanisms that drive the development of social functioning in childhood. They suggest that the role of vision-guided experience in typical child development is far-reaching and show that the impact on a child's life, when access to this experience is restricted, can be substantial. Crucially, however, they also remind us that child development is an exceptionally resilient and adaptive process, and one which strives to make learning possible even in the most unusual conditions.

References

Adamson, L. B., Bakeman, R., & Deckner, D. F. (2004). The development of symbol-infused joint engagement. *Child Development, 75*(4), 1171-1187.

Alexander, M. P., & Stuss, D. T. (2000). Disorders of frontal lobe functioning. *Seminars in Neurology, 20*(4), 427-437.

Andersen, E. S., Dunlea, A., & Kekelis, L. S. (1984). Blind children's language: Resolving some differences. *Journal of Child Language, 11*(3), 645-664.

Andersen, E. S., Dunlea, A., & Kekelis, L. S. (1993). The impact of input: language acquisition in the visually impaired. *First Language, 13*(37), 23-49.

Anderson, P. (2003). Assessment and Development of Executive Function (EF) During Childhood. *Child Neuropsychology (Neuropsychology, Development and Cognition: Section C), 8*(2), 71-82.

Anderson, V. A., Anderson, P., Northam, E., Jacobs, R., & Mikiewicz, O. (2002). Relationships Between Cognitive and Behavioral Measures of Executive Function in Children With Brain Disease. *Child Neuropsychology (Neuropsychology, Development and Cognition: Section C), 8*, 231-240.

Astington, J. W. (2001). The future of theory-of-mind research: Understanding motivational states, the role of language, and real-world consequences. *Child Development, 72*(3), 685-687.

Astington, J. W., & Baird, J. A. (2005). *Why language matters for a theory of mind.* New York: Oxford University Press.

Astington, J. W., & Jenkins, J. M. (1999). A longitudinal study of the relation between language and theory-of-mind development. *Developmental Psychology, 35*(5), 1311–1320.

Atkinson, J. (1984). Human visual development over the first 6 months of life. A review and a hypothesis. *Human Neurobiology, 3*(2), 61-74.

Atkinson, J., Hood, B., Wattam-Bell, J., & Braddick, O. (1992). Changes in infants' ability to switch visual attention the first three months of life. *Perception, 21*(5), 643 – 653.

Baird, G., & Moore, A. T. (1993). Epidemiology. In A. R. Fielder, A. B. Best & M. C. O. Bax (Eds.), *The management of visual impairment in childhood* (pp. 1-7). London: Mac Keath Press.

Bakeman, R., & Adamson, L. (1984). Coordinating Attention to People and Objects in Mother-Infant and Peer-Infant Interaction. *Child Development, 55*(4), 1278-1289.

Barkley, R. A. (1997). Behavioral Inhibition, Sustained Attention, and Executive Functions. Constructing a Unifying Theory of ADHD. *Psychological Bulletin, 121*(1), 65-94.

Baron-Cohen, S. (1987). Autism and symbolic play. *British Journal of Developmental Psychology, 5*(2), 139-148.

Baron-Cohen, S. (1989). The Autistic Child's Theory of Mind: a Case of Specific Developmental Delay. *Journal of Child Psychology and Psychiatry, 30*(2), 285-297.

Baron-Cohen, S. (1995a). The Eye Direction Detector (EDD) and the Shared Attention Mechanism (SAM): Two Cases for Evolutionary Psychology. In C. Moore & P. J. Dunham (Eds.), *Joint attention: Its origins and role in development* (pp. 41-61). Hove, UK: Earlbaum.

Baron-Cohen, S. (1995b). *Mindblindness: An essay on autism and theory of mind.* London: The MIT Press.

Baron-Cohen, S. (2000). Theory of mind and autism: A fifteen year review. In S. Baron-Cohen, Tager-Flusberg, H., Cohen, D.J. (Ed.), *Understanding other minds: perspectives from developmental cognitive neuroscience.* (pp. 1-20). Oxford, UK: Oxford University Press.

Baron-Cohen, S. (2002). I am loved, therefore I think. Book rewiew of the *Cradle of Thought* by R.P. Hobson. *Nature, 416*, 791-792.

Baron-Cohen, S., Jolliffe, T., Mortimore, C., & Robertson, M. (1997). Another Advanced Test of Theory of Mind: Evidence from Very High Functioning Adults with Autism or Asperger Syndrome. *Journal of Child Psychology and Psychiatry, 38*(7), 813-822.
Baron-Cohen, S., Leslie, A. M., & Frith, U. (1985). Does the autistic child have a "theory of mind" ? *Cognition, 21*(1), 37-46.
Baron-Cohen, S., & Swettenham, J. (1996). The relationship between SAM and ToMM: Two hypotheses. In P. Carruthers & P. S. Smith (Eds.), *Theories of Theory of Mind* (pp. 158-168). Cambridge: Cambridge University Press.
Bartsch, K., & Wellman, H. M. (1995). *Children talk about the mind.* New York: Oxford University Press.
Bauman, M. (1999). Autism: Clinical features and neurobiological observations. In H. Tager-Flusberg (Ed.), *Neurodevelopmental Disorders* (pp. 383-399). Cambridge, MA: MIT Press/Bradford Books.
Behl, D. D., Akers, J. F., Boyce, G. C., & Taylor, M. J. (1996). Do mothers interact differently with children who are visually impaired? *Journal of Visual Impairment & Blindness, 90*(6), 501-511.
Benjamini, Y., & Hochberg, Y. (1995). Controlling the false discovery rate: a practical and powerful approach to multiple testing. *Journal of the Royal Statistical Society, Series B, 57*(1), 289-300.
Bigelow, A. E. (1992). Locomotion and Search Behavior in Blind Infants. *Infant Behavior & Development, 15*(2), 179-189.
Bigelow, A. E. (1995). The effect of blindness on the early development of the self. In P. Rochat (Ed.), *The self in infancy: Theory and research. Advances in psychology,* (pp. 327-347). Amsterdam, Netherlands: North-Holland/Elsevier Science Publishers.
Bigelow, A. E. (2003). The development of joint attention in blind infants. *Development and Psychopathology, 15*(2), 259-275.
Bishop, D. V. M. (2003). *The Children's Communication Checklist (CCC) - 2.* London: The Psychological Corporation.
Bishop, D. V. M. (2005). *Uncommon Understanding: Development and Disorders of Language Comprehension in Children* (5 ed.). Hove: Psychology Press.
Bishop, M., Hobson, R. P., & Lee, A. (2005). Symbolic play in congenitally blind children. *Development and Psychopathology, 17*(2), 447-465.
Bland, J. M., & Altman, D. G. (1995). Multiple significance tests: the Bonferroni method. *British Medical Journal, 310*(6986), 170.
Botting, N., & Conti-Ramsden, G. (2003). Autism, primary pragmatic difficulties, and specific language impairment: can we distinguish them using psycholinguistic markers? *Developmental Medicine and Child Neurology, 45*(8), 515-524.
Bowler, D. M. (1992). Theory of Mind in Aspergers Syndrome. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 33*(5), 877-893.
Brambring, M. (2001). Integration of children with visual impairment in regular preschools. *Child: Care, Health and Development, 27*(5), 425-438.
Brambring, M., & Tröster, H. (1992). On the Stability of Stereotyped Behaviors in Blind Infants and Preschoolers. *Journal of Visual Impairment and Blindness, 86*(2), 105-110.
Brown, J. R., Donelan-McCall, N., & Dunn, J. (1996). Why talk about mental states? The significance of children's conversations with friends, siblings and mothers. *Child Development, 63*(3), 836-849.
Brown, R., Hobson, R. P., Lee, A., & Stevenson, J. (1997). Are there 'autistic-like' features in congenitally blind children? *Journal of Child Psychology and Psychiatry, 38*(6), 693-703.
Bruner, J. (1983). *Child's Talk: Learning to Use Language.* New York: Oxford University Press.
Butterworth, G., & Cochran, E. (1980). Towards a Mechanism of Joint Visual Attention in Human Infancy. *International Journal of Behavioral Development, 3*(3), 253-272.

Butterworth, G., & Grover, L. (1990). Joint visual attention, manual pointing, and preverbal communication in human infancy. In M. Jeannerod (Ed.), *Attention and performance XIII* (pp. 605–624). Hillsdale, NJ: Erlbaum.

Butterworth, G., & Jarrett, N. (1991). What minds have in common is space: Spatial mechanisms serving joint visual attention in infancy. *British Journal of Developmental Psychology, 9*(1), 55-72.

Campbell, M. J., Julious, S. A., & Altman, D. G. (1995). Estimating sample sizes for binary, ordered categorical, and continuous outcomes in two group comparisons. *British Medical Journal, 311*(7013), 1145-1148.

Capps, L., Kehres, J., & Sigman, M. (1998). Conversational Abilities Among Children with Autism and Children with Developmental Delays. *Autism, 2*(4), 325-344.

Capps, L., Losh, M., & Thurber, C. (2000). 'The Frog Ate the Bug and Made his Mouth Sad': Narrative Competence in Children with Autism. *Journal of Abnormal Child Psychology, 28*(2), 193-204.

Carlson, S. M., Mandell, D. J., & Williams, L. (2004). Executive Function and Theory of Mind: Stability and Prediction From Ages 2 to 3. *Developmental Psychology, 40*(6), 1105-1122.

Carlson, S. M., & Moses, L. J. (2001). Individual Differences in Inhibitory Control and Children's Theory of Mind. *Child Development, 72*(4), 1032.

Carlson, S. M., Moses, L. J., & Breton, C. (2002). How specific is the relation between executive function and theory of mind? Contributions of inhibitory control and working memory. *Infant and Child Development, 11*(2), 73-92.

Carpendale, J. I. M., & Lewis, C. (2004). Constructing an understanding of mind: The development of children's social understanding within social interaction. *Behavioral and Brain Sciences, 27*(1), 79-96.

Carpenter, M., Nagell, K., & Tomasello, M. (1998). Social cognition, joint attention, and communicative competence from 9 to 15 months of age. *Monographs of the Society for Research in Child Development, 63*(4), 1-143.

Cass, H. D. (1998). Visual Impairment and Autism: Current Questions and Future Research. *Autism, 2*(2), 117-138.

Cass, H. D., Sonksen, P., & McConachie, H. R. (1994). Developmental Setback in Severe Visual Impairment. *Archives of Disease in Childhood, 70*(3), 192-196.

Charman, T. (2003). Why is joint attention a pivotal skill in autism? *Philosophical Transactions of the Royal Society of London Series B-Biological Sciences, 358*(1430), 315-324.

Charman, T., Baron-Cohen, S., Swettenham, J., Baird, G., Cox, A., & Drew, A. (2000). Testing joint attention, imitation, and play as infancy precursors to language and theory of mind. *Cognitive Development, 15*(4), 481-498.

Charman, T., Baron-Cohen, S., Swettenham, J., Baird, G., Drew, A., & Cox, A. (2003). Predicting language outcome in infants with autism and pervasive developmental disorder. *International Journal of Language & Communication Disorders, 38*(3), 265-285.

Charman, T., & Shmueli-Goetz, Y. (1998). The relationship between theory of mind, language, and narrative discourse: an experimental study. *Cahiers De Psychologie Cognitive-Current Psychology of Cognition, 17*(2), 245-271.

Charman, T., Swettenham, J., Baron-Cohen, S., Cox, A., Baird, G., & Drew, A. (1997). Infants With Autism: An Investigation of Empathy, Pretend Play, Joint Attention, and Imitation. *Developmental Psychology, 33*(5), 781-789.

Charman, T., Taylor, E., Drew, A., Cockerill, H., Brown, J.-A., & Baird, G. (2005). Outcome at 7 years of children diagnosed with autism at age 2: predictive validity of assessments conducted at 2 and 3 years of age and pattern of symptom change over time. *Journal of Child Psychology and Psychiatry, 46*(5), 500-513.

Chase, J. B. (1972). *Retrolental Fibroplasia and Autistic Symptomatology*. New York: American Foundation For Blind.

Chess, S. (1971). Autism in children with congenital rubella. *Journal of Autism and Developmental Disorders, 1*(1), 33-47.
Cohen, J. (1992). Power primer. *Psychological Bulletin, 112*, 155-159.
Cohen, J. (1994). The Earth is round (p< .05). *American Psychologist, 49*, 997-1003.
Colson, E. R., & Dworkin, P. H. (1997). Toddler Development. *Paediatrics in Review, 18*(8), 255-259.
Conover, W. J., & Iman, R. L. (1981). Rank Transformations as a Bridge Between Parametric and Nonparametric Statistics. *The American Statistician, 35*(3), 124-129.
Conti-Ramsden, G., Botting, N., & Faragher, B. (2001). Psycholinguistic Markers for Specific Language Impairment (SLI). *Journal of Child Psychology and Psychiatry, 42*(6), 741-748.
Conti-Ramsden, G., & Pérez-Pereira, M. (1998). Conversational interactions between blind infants and their mothers. *Infant Behavior and Development, 21*, 354.
Conti-Ramsden, G., & Pérez-Pereira, M. (1999). Conversational interactions between mothers and their infants who are congenitally blind, have low vision, or are sighted. *Journal of Visual Impairment & Blindness, 93*(11), 691-703.
Corkum, V., & Moore, C. (1995). Development of Joint Visual Attention in Infants. In C. Moore & P. J. Dunham (Eds.), *Joint visual attention: Its origins and role in development.* (pp. 61–84). Hove, UK: Erlbaum.
Corkum, V., & Moore, C. (1998). The origins of joint visual attention in infants. *Developmental Psychology, 34*(1), 28-38.
Cullinan, T. R. (1987). The epidemiology of blindness. In S. Miller (Ed.), *Clinical ophthalmology* (Vol. 571-578). Bristol: Wright.
Cutting, A. L., & Dunn, J. (1999). Theory of mind, emotion understanding, language, and family background: Individual differences and interrelations. *Child Development, 70*(4), 853-865.
Dale, N., & Salt, A. (2007). Early support developmental journal for children with visual impairment: the case for a new developmental framework for early intervention. *Child: Care, Health and Development, 33*(6), 684-690.
Dale, N., & Sonksen, P. (2002). Developmental outcome, including setback, in young children with severe visual impairment. *Developmental Medicine and Child Neurology, 44*(9), 613-622.
Dannenberg, J. (2000). *First day jitters.* Watertown, Mass: Charlesbridge.
Dawson, G., Munson, J., Estes, A., Osterling, J., McPartland, J., Toth, K., et al. (2002). Neurocognitive Function and Joint Attention Ability in Young Children with Autism Spectrum Disorder Versus Developmental Delay. *Child Development, 73*(2), 345-358.
Dawson, G., Toth, K., Abbott, R., Osterling, J., Munson, J., Estes, A., et al. (2004). Early social attention impairments in autism: Social orienting, joint attention, and attention to distress. *Developmental Psychology, 40*(2), 271-283.
Day, S. (1997). Normal and abnormal visual development. In D. Taylor (Ed.), *Paediatric Opthalmology* (2 ed., pp. 13-28). London: Blackwell.
de Rosnay, M., & Hughes, C. (2006). Conversation and theory of mind: Do children talk their way to socio-cognitive understanding? *British Journal of Developmental Psychology, 24*(1), 7-37.
de Rosnay, M., Pons, F., Harris, P. L., & Morrell, J. (2004). A lag between understanding false belief and emotion attribution in young children: Relationships with linguistic ability and mothers' mental state language. *British Journal of Developmental Psychology, 22*(2), 197–218.
de Villiers, J. G. (2000). Language and theory of mind: what are the developmental relationships? In S. Baron-Cohen, H. Tager- Flusberg & D. J. Cohen (Eds.), *Understanding other minds: perspectives from developmental cognitive neuroscience* (2 ed., pp. 83-121). Oxford, UK: Oxford University Press.

de Villiers, J. G., & Pyers, J. E. (2002). Complements to cognition: a longitudinal study of the relationship between complex syntax and false-belief-understanding. *Cognitive Development, 17*(1), 1037-1060.
Dekker, R. (1993). Visually impaired children and haptic intelligence test scores: intelligence test for visually impaired children (ITVIC). *Developmental Medicine & Child Neurology, 35*(6), 478-489.
Dennis, M., Lazenby, A. L., & Lockyer, L. (2001). Inferential language in high-function children with autism. *Journal of Autism and Developmental Disorders, 31*(1), 47-54.
DfES. (2006). *Department for Education and Skills. Developmental Journal for Babies and Children with Visual Impairment*. London: Crown.
Diamond, A. (1985). Development of the ability to use recall to guide action, as indicated by infants' performance on AB. *Child Development, 56*(4), 868-883.
Diamond, A. (1991). Developmental time course in human infants and infant monkeys, and the neural bases of inhibitory control in reaching. *Annals of the New York Academy of Sciences: Pt. VII. Inhibition and executive control, 608*, 637-704.
Diamond, A., & Doar, B. (1989). The performance of human infants on a measure of frontal cortex function, the delayed response task. *Developmental Psychobiology, 22*(3), 271-294.
Diamond, A., & Goldman-Rakic, P. S. (1989). Comparison of human infants and rhesus monkeys on Piaget's AB task: evidence for dependence on dorsolateral prefrontal cortex. *Experimental Brain Research, 74*(1), 24-40.
Dote-Kwan, J. (1995). Impact of Mothers' Interactions on the Development of Their Young Visually Impaired Children. *Journal of Visual Impairment & Blindness, 89*(1), 46-58.
Drew, A., Baird, G., Baron-Cohen, S., Cox, A., Slonims, V., Wheelwright, S., et al. (2002). A pilot randomised control trial of a parent training intervention for pre-school children with autism - Preliminary findings and methodological challenges. *European Child & Adolescent Psychiatry, 11*(6), 266-272.
DSM-IV-TR. (2000). *Diagnostic and Statistical Manual of Mental Disorders - Text Revision* (4 ed.). Washington DC: American Psychiatric Association.
Duncan, J., Emslie, H., Williams, P., Johnson, R., & Freer, C. (1996). Intelligence and the Frontal Lobe: The Organization of Goal-Directed Behavior. *Cognitive Psychology, 30*(3), 257-303.
Dunlea, A. (1989). *Language and the emergence of meaning: Blind and sighted children's early language.* Cambridge: Cambridge University Press.
Dunn, J., Brown, J., Slomkowski, C., Tesla, C., & Youngblade, L. (1991). Young Children's Understanding of Other People's Feelings and Beliefs: Individual Differences and Their Antecedents. *Child Development, 62*(6), 1352-1366.
Dyck, M. J., Farrugia, C., Shochet, I. M., & Holmes-Brown, M. (2004). Emotion recognition/understanding ability in hearing or vision-impaired children: do sounds, sights, or words make the difference? *Journal of Child Psychology and Psychiatry, 45*(4), 789-800.
Dyer, J. R., Shatz, M., & Wellman, H. M. (2000). Young children's storybooks as a source of mental state information. *Cognitive Development, 15*(1), 17-37.
Eardley, A. F., & Pring, L. (2007). Spatial processing, mental imagery, and creativity in individuals with and without sight. *European Journal of Cognitive Psychology, 19*(1), 37-58.
Ek, U., Fernell, E., Jacobson, L., & Gillberg, C. (1998). Relation between blindness due to retinopathy of prematurity and autistic spectrum disorders: a population-based study. *40, 5*(297-301).
Emery, N. J. (2000). The eyes have it: the neuroethology, function and evolution of social gaze. *Neuroscience & Biobehavioral Reviews, 24*(6), 581-604.
Erikson, E. H. (1968). *Identity, youth and crisis.* New York: W. W. Norton Company.

Erin, J. N. (1986). Frequencies and Types of Questions in the Language of Visually Impaired Children. *Journal of Visual Impairment & Blindness, 80*(4), 670-674.
Faul, F., Erdfelder, E., Lang, A.-G., & Buchner, A. (2007). G*Power 3: A flexible statistical power analysis program for the social, behavioral, and biomedical sciences. *Behavior Research Methods Instruments & Computers, 39*, 175-191.
Fisher, N., Happé, F., & Dunn, J. (2005). The relationship between vocabulary, grammar, and false belief task performance in children with autistic spectrum disorders and children with moderate learning difficulties. *Journal of Child Psychology and Psychiatry, 46*(4), 409-419.
Flavell, J. H. (1999). Cognitive Development: Children's Knowledge About the Mind. *Annual Review of Psychology, 50*(1), 21-45.
Florey, C. D. (1993). Sample size for beginners. *British Medical Journal, 306*(6886), 1181–1184.
Fodor, J. A. (1992). A theory of the child's theory of mind. *Cognition, 44*(3), 283-296.
Folger, R. (1989). Significance tests and the duplicity of binary decisions. *Psychological Bulletin, 106*(1), 155-160.
Fraiberg, S. (1977). *Insights from the blind*. London: Souvenir.
Fraiberg, S., & Adelson, E. (1977). Self-representation in language and play. In S. Fraiberg (Ed.), *Insights from the blind* (pp. 248-270). London: Souvenir Press.
Frith, U. (1996). Cognitive explanations of autism. *Acta Paediatrica, 85*, 63-68.
Frith, U. (2003). *Autism: Explaining the enigma* (2 ed.). Malden, MA: Blackwell.
Galati, D., Miceli, R., & Sini, B. (2001). Judging and coding facial expression of emotions in congenitally blind children. *International Journal of Behavioral Development, 25*(3), 268-278.
Gilotty, L., Kenworthy, L., Sirian, L., Black, D. O., & Wagner, A. E. (2002). Adaptive Skills and Executive Function in Autism Spectrum Disorders. *Child Neuropsychology (Neuropsychology, Development and Cognition: Section C), 8*(4), 241-248.
Gioia, G. A., Isquith, P. K., Guy, S. C., & Kenworthy, L. (2000). *The Behavior Rating Inventory of Executive Function (BRIEF)*. Odessa, FL: Psychological Assessment Resources.
Gioia, G. A., Isquith, P. K., Kenworthy, L., & Barton, R. M. (2003). Profiles of Everyday Executive Function in Acquired and Developmental Disorders. *Child Neuropsychology (Neuropsychology, Development and Cognition: Section C), 8*(2), 121-137.
Gioia, G. A., Isquith, P. K., Retzlaff, P. D., & Espy, K. A. (2002). Confirmatory Factor Analysis of the Behavior Rating Inventory of Executive Function (BRIEF) in a Clinical Sample. *Child Neuropsychology (Neuropsychology, Development and Cognition: Section C), 8*(4), 249-257.
Gnepp, J. (1983). Children's social sensitivity: inferring emotions from conflicting cues. *Developmental Psychology, 19*, 805-814.
Good, W. V., Jan, J. E., Burden, S. K., Skoczenski, A., & Candy, R. (2001). Recent advances in cortical visual impairment. *Developmental Medicine & Child Neurology, 43*(1), 56-60.
Gopnik, A., & Meltzoff, A. (1997). *Words, Thoughts, and Theories*. Cambridge, MA: MIT Press.
Gopnik, A., & Wellman, H. M. (1994). The Theory-Theory. In L. Hirschfeld & S. Gelman (Eds.), *Mapping the Mind: Domain Specificity in Cognition and Culture* (pp. 257-293). New York: Cambridge University Press.
Green, S., Pring, L., & Swettenham, J. (2004). An investigation of first-order false belief understanding of children with congenital profound visual impairment. *British Journal of Developmental Psychology, 22*(1), 1-17.
Green/née Cupples, S. (2001). *Social cognition in children with visual impairment*. Unpublished PhD Thesis, Goldsmiths, Univeristy of London.
Gross, J. J. (2002). Emotion regulation: Affective, cognitive, and social consequences. *Psychophysiology, 39*(3), 281-291.

Happé, F. G. E. (1994). An advanced test of theory of mind: Understanding of story characters' thoughts and feelings by able autistic, mentally handicapped, and normal children and adults. *Journal of Autism and Developmental Disorders, 24*(2), 129-154.
Happé, F. G. E. (1995). The Role of Age and Verbal-Ability in the Theory of Mind Task-Performance of Subjects with Autism. *Child Development, 66*(3), 843-855.
Harris, P. L. (1989). *Children and Emotion*. Oxford: Blackwell.
Harris, P. L. (1992). From Simulation to Folk Psychology: The Case for Development. *Mind and Language, 7*(1-2), 120– 144.
Harris, P. L. (1996). Desires, beliefs and language. In P. Carruthers & P. K. Smith (Eds.), *Theories of theories of mind*. Cambridge: Cambridge University Press.
Harris, P. L. (2000). *The work of the imagination*. Oxford, UK: Blackwell.
Harris, P. L., de Rosnay, M., & Pons, F. (2005). Language and children's understanding of mental states. *Current Directions in Psychological Science, 14*(2), 69-73.
Hatton, D. D., Bailey, D. B., Burchinal, M. R., & Ferrell, K. A. (1997). Developmental growth curves of preschool children with vision impairments. *Child Development, 68*(5), 788-806.
Hermelin, B. (1972). Locating events in space and time: Experiments with autistic, blind, and deaf children. *Journal of Autism and Developmental Disorders, 2*(3), 288-298.
Hill, E. L. (2004). Evaluating the theory of executive dysfunction in autism. *Developmental Review, 24*(2), 189-233.
Hirst, C., Poole, J. J., & Snelling, G. S. (1993). Liverpool visual assessment team 1985-1989: 5 years on. *Child: Care, Health and Development, 19*(3), 185-195.
Hobson, R. P. (1990). On the origins of self and the case of autism. *Development and Psychopathology, 2*, 163-181.
Hobson, R. P. (1991). Against the theory of 'Theory of Mind'. *British Journal of Developmental Psychology, 9*, 33-51.
Hobson, R. P. (1993). *Autism and the development of mind*. Hove: Earlbaum.
Hobson, R. P. (2002). *The Cradle of Thought*. London: Macmillian.
Hobson, R. P. (2005). Why connect? On the relation between autism and blindness. In L. Pring (Ed.), *Autism and Blindness: Research and Reflections* (pp. 10-25). London: Whurr.
Hobson, R. P., & Bishop, M. (2003). The pathogenesis of autism: insights from congenital blindness. *Philosophical Transactions of the Royal Society of London Series B-Biological Sciences, 358*(1430), 335-344.
Hobson, R. P., Lee, A., & Brown, R. (1999). Autism and congenital blindness. *Journal of Autism and Developmental Disorders, 29*(1), 45-56.
Hood, B. M., Willen, J. D., & Driver, J. (1998). Adult's Eyes Trigger Shifts of Visual Attention in Human Infants. *Psychological Science, 9*(2), 131-134.
Howell, D. C. (2002). *Statistical Methods for Psychology* (5 ed.). Pacific Grove, CA: Duxbury.
Hughes, C. (1998a). Executive function in preschoolers: Links with theory of mind and verbal ability. *British Journal of Developmental Psychology, 16*, 233-253.
Hughes, C. (1998b). Finding your marbles: Does preschoolers' strategic behavior predict later understanding of mind? *Developmental Psychology, 34*(6), 1326-1339.
Hughes, C., & Dunn, J. (1997). "Pretend you didn't know": Preschoolers' talk about mental states in pretend play. *Cognitive Development, 12*(4), 381-403.
Hughes, C., & Dunn, J. (1998). Understanding mind and emotion: Longitudinal associations with mental-state talk between young friends. *Developmental Psychology, 34*(5), 1026-1037.
Hughes, C., & Russell, J. (1993). Autistic Children's Difficulty With Mental Disengagement From an Object: Its Implications for Theories of Autism. *Developmental Psychology, 29*(3), 498-510.
Hughes, C., Russell, J., & Robbins, T. W. (1994). Evidence for executive dysfunction in autism. *Neuropsychologia, 32*(4), 477-492.

Hughes, M., Dote-Kwan, J., & Dolendo, J. (1998). A Close Look at the Cognitive Play of Preschoolers with Visual Impairments in the Home. *Exceptional Children, 64*(4), 451-462.
Hull, T., & Mason, H. (1995). Performance of Blind Children on Digit-Span Tests. *Journal of Visual Impairment and Blindness, 89*(2), 1666-1169.
James, D. M., & Stojanovik, V. (2007). Communication skills in blind children: a preliminary investigation. *Child: Care, Health and Development, 33*(1), 4-10.
Jolliffe, T., & Baron-Cohen, S. (1999). The Strange Stories Test: A Replication with High-Functioning Adults with Autism or Asperger Syndrome. *Journal of Autism and Developmental Disorders, 29*(5), 395-406.
Kasari, C., Paparella, T., Freeman, S., & Jahromi, L. B. (2008). Language Outcome in Autism: Randomized Comparison of Joint Attention and Play Interventions. *Journal of Consulting and Clinical Psychology, 76*(1), 125–137.
Keeler, W. R. (1956). Autistic patterns and defective communication in blind children with retrolental fibroplasia. In P. H. Hoch & J. Zubin (Eds.), *Psychopathology and Communication* (pp. 64-83). New York: Grune & Stratton.
Kekelis, L. S., & Andersen, E. S. (1984). Family communication styles and language development. *Journal of Visual Impairment and Blindness, 78*(2), 54-65.
Kekelis, L. S., & Prinz, P. M. (1996). Blind and sighted children with their mothers: The development of discourse skills. *Journal of Visual Impairment & Blindness, 90*(5), 423-436.
Klin, A. (2000). Attributing Social Meaning to Ambiguous Visual Stimuli in Higher-functioning Autism and Asperger Syndrome: The Social Attribution Task. *Journal of Child Psychology and Psychiatry, 41*(7), 831-846.
Landa, R. (2000). Social language use in Asperger syndrome and high-functioning autism. In A. Klin, F. Volkmar & S. S. Sparrow (Eds.), *Asperger syndrome* (pp. 125-155). New York: Guildford Press.
Landau, B., & Gleitman, L. R. (1985). *Language and Experience: Evidence from the Blind Child.* Cambridge: Harvard University Press.
Landau, B., Spelke, E., & Gleitman, H. (1984). Spatial knowledge in a young blind child. *Cognition, 16*(3), 225-260.
Landry, R., & Bryson, S. E. (2004). Impaired disengagement of attention in young children with autism. *Journal of Child Psychology and Psychiatry, 45*(6), 1115-1122.
Leekam, S. R., López, B., & Moore, C. (2000). Attention and Joint Attention in Preschool Children with Autism. *Developmental Psychology, 36*(2), 261-273.
Leekam, S. R., & Perner, J. (1991). Does the autistic child have a metarepresentational deficit? *Cognition, 40*(3), 203-218.
Leekam, S. R., & Ramsden, C. A. (2006). Dyadic orienting and joint attention in preschool children with autism. *Journal of Autism and Developmental Disorders, 36*(2), 185-197.
Leslie, A. M. (1987). Pretense and Representation. The Origins of "Theory of Mind". *Psychological Review, 94*(4), 412-426.
Leslie, A. M. (1994). ToMM, ToBY and agency: core architecture and domain specificity. In Mapping the Mind: Domain Specificity in Cognition and Culture, ed. , pp. 119– 48. Cambridge, UK: Cambridge Univ. Press. In L. A. Hirschfeld & S. A. Gelman (Eds.), *Mapping the Mind: Domain Specificity in Cognition and Culture* (pp. 119– 148). Cambridge, UK: Cambridge University Press.
Leslie, A. M., Friedman, O., & German, T. P. (2004). Core mechanisms in `theory of mind'. *Trends in Cognitive Sciences, 8*(12), 528-533.
Leslie, A. M., & Thaiss, L. (1992). Domain specificity in conceptual development: Neuropsychological evidence from autism. *Cognition, 43*(3), 225-251.

Lewis, C., Freeman, N. H., Kyriakidou, C., Maridaki-Kassotaki, K., & Berridge, D. M. (1998). Social Influences on False Belief Access: Specific Sibling Influences or General Apprenticeship? *Child Development, 67*(6), 2930-2947.

Lewis, V., Norgate, S., Collis, G. M., & Reynolds, R. (2000). The consequences of visual impairment for children's symbolic and functional play. *British Journal of Developmental Psychology, 18*, 449-464.

Lloyd, H., Paintin, K., & Botting, N. (2006). Performance of children with different types of communication impairment on the Clinical Evaluation of Language Fundamentals (CELF). *Child Language Teaching and Therapy, 22*(1), 47-67.

Lopez, B. R., Lincoln, A. J., Ozonoff, S., & Lai, Z. (2005). Examining the Relationship between Executive Functions and Restricted, Repetitive Symptoms of Autistic Disorder. *Journal of Autism and Developmental Disorders, 35*(4), 445-460.

Lord, C., & Paul, R. (1997). Language and communication in autism. In D. J. Cohen & F. R. Volkmar (Eds.), *Handbook of Autism and Pervasive Developmental Disorders* (pp. 195-225). New York: John Wiley & Sons Inc.

Lord, C., Rutter, M., DiLavore, P., & Risi, S. (1999). *Autism Diagnostic Observation Schedule (ADOS)*. Los Angeles, CA: Western Psychological Society.

Loukusa, S., Leinonen, E., Kuusikko, S., Jussila, K., Mattila, M. L., Ryder, N., et al. (2007). Use of context in pragmatic language comprehension by children with Asperger syndrome or high-functioning autism. *Journal of Autism and Developmental Disorders, 37*(6), 1049-1059.

Markus, J., Mundy, P., Morales, M., Delgado, C. E. F., & Yale, M. (2000). Individual differences in infant skills as predictors of child-caregiver joint attention and language. *Social Development, 9*(3), 302-315.

Marschark, M., Green, V., Hindmarsh, G., & Walker, S. (2000). Understanding theory of mind in children who are deaf. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 41*(8), 1067-1073.

Martin, I., & McDonald, S. (2003). Weak coherence, no theory of mind, or executive dysfunction? Solving the puzzle of pragmatic language disorders. *Brain and Language, 85*(3), 451-466.

Maxfield, K. E., & Buchholtz, S. A. (1957). *A Social Maturity Scale for Blind Pre-school Children*. New York: The American Foundation for the Blind Inc.

McAlpine, L. M., & Moore, C. L. (1995). The Development of Social Understanding in Children with Visual Impairments. *Journal of Visual Impairment & Blindness, 89*(4), 349-358.

McCartney, K., & Rosenthal, R. (2000). Effect Size, Practical Importance, and Social Policy for Children. *Child Development, 71*(1), 173-180.

McConachie, H. R., & Moore, V. (1994). Early Expressive Language of Severely Visually-Impaired Children. *Developmental Medicine and Child Neurology, 36*(3), 230-240.

McEvoy, R. E., Rogers, S. J., & Pennington, B. F. (1993). Executive Function and Social Communication Deficits in Young Autistic-Children. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 34*(4), 563-578.

McGinnis, A. R. (1981). Functional Linguistic Strategies of Blind Children. *Journal of Visual Impairment and Blindness, 75*(5), 210-214.

Meins, E., Fernyhough, C., Johnson, F., & Lidstone, J. (2006). Mind-mindedness in children: Individual differences in internal-state talk in middle childhood. *British Journal of Developmental Psychology, 24*, 181-196.

Meins, E., Fernyhough, C., Russell, J., & Clark-Carter, D. (1998). Security of Attachment as a Predictor of Symbolic and Mentalising Abilities: A Longitudinal Study. *Social Development, 7*(1), 1-24.

Meins, E., Fernyhough, C., Wainwright, R., Clark-Carter, D., Das Gupta, M., Fradley, E., et al. (2003). Pathways to Understanding Mind: Construct Validity and Predictive Validity of Maternal Mind-Mindedness. *Child Development, 74*(4), 1194-1211.
Meins, E., Fernyhough, C., Wainwright, R., Das Gupta, M., Fradley, E., & Tuckey, M. (2002). Maternal Mind-Mindedness and Attachment Security as Predictors of Theory of Mind Understanding. *Child Development, 73*(6), 1715-1726.
Meltzoff, A. (1999). Origins of theory of mind, cognition and communication. *Journal of Communication Disorders, 32*(4), 251-269.
Meltzoff, A., & Gopnik, A. (1993). The role of imitation in understanding persons and developing a theory of mind. In S. Baron-Cohen, Tager-Flusberg, H., Cohen, D.J. (Ed.), *Understanding other minds: Perspective from autism* (pp. 335-366). Oxford: Oxford University Press.
Meltzoff, A., & Moore, M. K. (1977). Imitation of facial and manual gestures by human neonates. *Science, 198*, 75–78.
Meltzoff, A., & Moore, M. K. (1983). Newborn infants imitate adult facial gestures. *Child Development, 54*(3), 702-709.
Mervis, C. A., Boyle, C. A., & Yeargin-Allsopp, M. (2002). Prevalence and selected characteristics of childhood vision impairment. *Developmental Medicine & Child Neurology, 44*(8), 538-541.
Mervis, C. B., & Klein-Tasman, B. P. (2004). Methodological Issues in Group-Matching Designs: alpha Levels for Control Variable Comparisons and Measurement Characteristics of Control and Target Variables. *Journal of Autism and Developmental Disorders, 34*(1), 7-17.
Millar, S. (1988). Models of sensory deprivation: The nature/nurture dichotomy and spatial representation in the blind. *International Journal of Behavioural Development, 11*, 69-87.
Millar, S. (1994). *Understanding and Representing Space*. Oxford: Oxford University Press.
Mills, A. E. (1993). Visual handicap. In D. Bishop & K. Mogford (Eds.), *Language development in exceptional cicumstances* (pp. 150–164). Hove: Earlbaum.
Minter, M., Hobson, R. P., & Bishop, M. (1998). Congenital visual impairment and 'theory of mind'. *British Journal of Developmental Psychology, 16*, 183-196.
Minter, M., Hobson, R. P., & Pring, L. (1991). Recognition of Vocally Expressed Emotion by Congenitally Blind-Children. *Journal of Visual Impairment & Blindness, 85*(10), 411-415.
Mirsky, A. F., Anthony, B. J., Duncan, C. C., Ahearn, M. B., & Kellam, S. G. (1991). Analysis of the elements of attention: A neuropsychological approach. *Neuropsychology Review, 2*(2), 109-145.
Moeller, M. P., & Schick, B. (2006). Relations between maternal input and theory of mind understanding in deaf children. *Child Development, 77*(3), 751-766.
Moore, C., & Dunham, P. J. (1995). *Joint attention: Its origins and role in development*. Hove, UK: Earlbaum.
Moore, C., Pure, K., & Furrow, D. (1990). Children's understanding of the modal expression of speaker certainty and uncertainty and its relation to the development of a representational theory of mind. *Child Development, 61*(3), 722-730.
Moore, V., & McConachie, H. R. (1994). Communication between Blind and Severley Visually-Impaired Children and Their Parents. *British Journal of Developmental Psychology, 12*, 491-502.
Mukaddes, N. M., Kilincaslan, A., Kucukyazici, G., Sevketoglu, T., & Tuncer, S. (2007). Autism in visually impaired individuals. *Psychiatry and Clinical Neurosciences, 61*(1), 39-44.
Mulford, R. (1983). Referential development in blind children. In A. E. Mills (Ed.), *Language Acquisition in the Blind Child: Normal and Deficient*. London: Croom Helm.
Mulford, R. (1988). First words of the blind child. In M. Smith & J. Locke (Eds.), *The emergent lexicon: The child's development of a linguistic vocabulary* (pp. 293–338). New York: Academic Press.

Mundy, P. (1995). Joint Attention and Social-Emotional Approach Behavior in Children with Autism. *Development and Psychopathology, 7*(1), 63-82.
Mundy, P., Fox, N., & Card, J. (2003). EEG coherence, joint attention and language development in the second year. *Developmental Science, 6*(1), 48-54.
Mundy, P., & Gomes, A. (1998). Individual differences in joint attention skill development in the second year. *Infant Behavior & Development, 21*(3), 469-482.
Mundy, P., Kasari, C., & Sigman, M. (1992). Nonverbal-Communication, Affective Sharing, and Intersubjectivity. *Infant Behavior & Development, 15*(3), 377-381.
Mundy, P., Kasari, C., Sigman, M., & Ruskin, E. (1995). Nonverbal-Communication and Early Language-Acquisition in Children with Down-Syndrome and in Normally Developing-Children. *Journal of Speech and Hearing Research, 38*(1), 157-167.
Mundy, P., Sigman, M., & Kasari, C. (1990). A longitudinal study of joint attention and language development in autistic children. *Journal of Autism and Developmental Disorders, 20*(1), 115-128.
Mundy, P., Sigman, M., & Kasari, C. (1994). Joint Attention, Developmental Level, and Symptom Presentation in Autism. *Development and Psychopathology, 6*(3), 389-401.
Nash, G. (2008). *Music and Language Processing in Children with Autism and those with Specific Language Impairment.* Unpublished PhD Thesis, Goldsmiths, University of London.
Neisser, U. (1991). Two perceptually given aspects of the self and their development. *Developmental review, 11*(3), 197-209.
Newland, T. E. (1979). The Blind Learning Aptitude Test. *Journal of Visual Impairment & Blindness, 73*, 134-139.
Ninio, A., & Snow, C. E. (1996). *Pragmatic development.* Boulder: Westview Press.
Norbury, C. F., & Bishop, D. V. M. (2002). Inferential processing and story recall in children with communication problems: A comparison of specific language impairment, pragmatic language impairment and high-functioning autism. *International Journal of Language and Communication Disorders, 37*(3), 227-251.
Norgate, S., Collis, G. M., & Lewis, V. (1998). The developmental role of rhymes and routines for congenitally blind children. *Cahiers De Psychologie Cognitive-Current Psychology of Cognition, 17*(2), 451-477.
Norman, D. A., & Shallice, T. (1986). Attention to action: Willed and automatic control of behavior. In R. J. Davidson, G. E. Schwartz & D. Shapiro (Eds.), *Consciousness and self-regulation: Vol. 4. Advances in research and theory.* (pp. 1-18). New York: Plenum Press.
Olson, D. R. (1988). On the origins of belief and other intentional states in children. In J. W. Astington, P. L. Harris & D. R. Olson (Eds.), *Developing theories of mind* (pp. 414-426). New York: Cambridge University Press.
Ozonoff, S., Cook, I., Coon, H., Dawson, G., Joseph, R., Klin, A., et al. (2004). Performance on Cambridge Neuropsychological Test Automated Battery Subtests Sensitive to Frontal Lobe Function in People with Autistic Disorder: Evidence from the Collaborative Programs of Excellence in Autism Network. *Journal of Autism and Developmental Disorders, 34*(2), 139-150.
Ozonoff, S., & Jensen, J. (1999). Brief Report: Specific Executive Function Profiles in Three Neurodevelopmental Disorders. *Journal of Autism and Developmental Disorders, 29*(2), 171-177.
Ozonoff, S., & McEvoy, R. E. (1994). A longitudinal study of executive function and theory of mind development in autism. *Development and Psychopathology, 6*(3), 415-431.
Ozonoff, S., & Miller, J. N. (1996). An Exploration of Right-Hemisphere Contributions to the Pragmatic Impairments of Autism. *Brain and Language, 52*(3), 411-434.
Ozonoff, S., Pennington, B. F., & Rogers, S. J. (1991). Executive Function Deficits in High-Functioning Autistic Individuals: Relationship to Theory of Mind. *Journal of Child Psychology and Psychiatry, 32*(7), 1081-1105.

Pascualvaca, D. M., Fantie, B. D., Papageorgiou, M., & Mirsky, A. F. (1998). Attentional capacities in children with autism: Is there a general deficit in shifting focus? *Journal of Autism and Developmental Disorders, 28*(6), 467-478.

Pennington, B. F., & Ozonoff, S. (1996). Executive Functions and Developmental Psychopathology. *Journal of Child Psychology and Psychiatry, 37*(1), 51-87.

Pérez-Pereira, M. (1994). Imitations, Repetitions, Routines, and the Child's Analysis of Language: Insights from the Blind. *Journal of Child Language, 21*(2), 317-337.

Pérez-Pereira, M., & Castro, J. (1992). Pragmatic functions of blind and sighted children's language: a twin case study. *First Language, 12*(34), 17-37.

Pérez-Pereira, M., & Castro, J. (1997). Language acquisition and the compensation of visual deficit: New comparative data on a controversial topic. *British Journal of Developmental Psychology, 15*, 439-459.

Pérez-Pereira, M., & Conti-Ramsden, G. (1999). *Language development and social interaction in blind children*. Hove: Psychology Press.

Pérez-Pereira, M., & Conti-Ramsden, G. (2001). The use of directives in verbal interactions between blind children and their mothers. *Journal of Visual Impairment & Blindness, 95*(3), 133-149.

Perneger, T. V. (1998). What's wrong with Bonferroni adjustments. *British Medical Journal, 316*(7139), 1236–1238.

Perner, J. (1997). *Understanding the Representational Mind*. Cambridge, MA: MIT Press.

Perner, J., Frith, U., Leslie, A. M., & Leekam, S. (1989). Exploration of the Autistic Child's Theory of Mind: Knowledge, Belief, and Communication. *Child Development, 60*(3), 689-700.

Perner, J., Leekam, S. R., & Wimmer, H. (1987). Three-year-olds' difficulty with false belief: The case for a conceptual deficit. *British Journal of Developmental Psychology, 5*(2), 125-137.

Perner, J., & Wimmer, H. (1985). John Thinks that Mary Thinks that... Attribution of second-order beliefs by 5-to 10-year-old children. *Journal of Experimental Child Psychology, 39*(3), 437-471.

Peters, A. M. (1994). The interdependence of social, cognitive and linguistic development: Evidence from a visually impaired child. In H. Tager- Flusberg (Ed.), *Constraints on Language Acquisition: Studies of Atypical Children* (pp. 195-220). Hillsdale, NJ: Erlbaum.

Peterson, C. C., Peterson, J. L., & Webb, J. (2000). Factors influencing the development of a theory of mind in blind children. *British Journal of Developmental Psychology, 18*(3), 431-447.

Peterson, C. C., & Siegal, M. (1995). Deafness, Conversation and Theory of Mind. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 36*(3), 459-474.

Peterson, C. C., & Siegal, M. (1998). Changing focus on the representational mind: Deaf, autistic and normal children's concepts of false photos, false drawings and false beliefs. *British Journal of Developmental Psychology, 16*, 301-320.

Peterson, C. C., & Siegal, M. (1999). Representing inner worlds: Theory of mind in autistic, deaf, and normal hearing children. *Psychological Science, 10*(2), 126-129.

Peterson, C. C., & Siegal, M. (2000). Insights into theory of mind from deafness and autism. *Mind & Language, 15*(1), 123-145.

Posner, M. I., & Petersen, S. E. (1990). The Attention System of the Human Brain. *Annual Review of Neuroscience, 13*, 25-42.

Povinelli, D. J., & Preuss, T. M. (1995). Theory of mind: evolutionary history of a cognitive specialization. *Trends in Neurosciences, 18*(9), 418-424.

Preisler, G. M. (1991). Early patterns of interaction between blind infants and their sighted mothers. *Child: Care, Health and Development, 17*(2), 65-90.

Preisler, G. M. (1993). A descriptive study of blind children in nurseries with sighted children. *Child: Care, Health and Development, 19*(5), 295-315.

Pring, L. (2005). *Autism and Blindness: Research and Reflections*. London: Whurr.
Pring, L., Dewart, H., & Brockbank, M. (1998). Social cognition in children with a visual impairment. *Journal of Visual Impairment & Blindness, 92*(11), 754-768.
Pring, L., & Tadic, V. (2005). More than meets the eye: blindness, talent and autism. In L. Pring (Ed.), *Autism and Blindness: Research and Reflections* (pp. 50-73). London: Whurr.
Prior, M., & Hoffmann, W. (1990). Brief report: Neuropsychological testing of autistic children through an exploration with frontal lobe tests. *Journal of Autism and Developmental Disorders, 20*(4), 581-590.
Rabbit, P. (1997). *Methodology of frontal and executive function*. Hove: Psychology Press.
Rahi, J. S., & Cable, N. (2003). Severe visual impairment and blindness in children in the UK. *The Lancet, 362*(9393), 1359-1365.
Rahi, J. S., & Dezateux, C. (1998). Epidemiology of visual impairment in Britain. *Archives of Disease in Childhood, 78*(4), 381-386.
Rapin, I. (1996). Practitioner Review: Developmental Language Disorders: A Clinical Update. *Journal of Child Psychology and Psychiatry, 37*(6), 643-655.
Rapin, I., & Dunn, M. (2003). Update on the language disorders of individuals on the autistic spectrum. *Brain and Development, 25*(3), 166-172.
Recchia, S. L. (1997). Establishing intersubjective experience: Developmental challenges for young children with congenital blindness and autism and their caregivers. In V. Lewis & G. M. Collis (Eds.), *Blindness and Psychological Development in Young Children* (pp. 116-129). Leicester, UK: The British Psychological Society Books.
Reynell, J. (1978). Developmental patterns of visually handicapped children. *Child: Care, Health and Development, 4*(5), 291–303.
Reynell, J. (1979). *Manual for the Reynell-Zinkin scales for young visually handicapped children*. Windsor: NFER.
Reynell, J., & Zinkin, P. (1979). *Reynell-Zinkin developmental scales for visually handicapped children*. Chicago, IL: Stoelting Co.
Rieffe, C., & Meerum Terwogt, M. (2000). Deaf children's understanding of emotions: Desires take precedence. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 41*(5), 601-608.
Rieffe, C., Meerum Terwogt, M., & Cowan, R. (2005). Children's understanding of mental states as causes of emotions. *Infant and Child Development, 14*(3), 259-272.
Rieffe, C., Meerum Terwogt, M., & Stockmann, L. (2000). Understanding atypical emotions among children with autism. *Journal of Autism and Developmental Disorders, 30*(3), 195-203.
Rieser, J. J., & Rider, E. A. (1991). Young Children's Spatial Orientation with Respect to Multiple Targets When Walking without Vision. *Developmental Psychology, 27*(1), 97-107.
Robinson, G. C. (1977). Causes, ocular disorders, associated handicaps, and incidence and prevalence of blindness in childhood. In J. E. Jan, R. D. Freeman & E. P. Scott (Eds.), *Visual Impairment in Children and Adolescents* (pp. 22-47). New York: Grune and Stratton.
Roch-Levecq, A. (2006). Production of basic emotions in children with congenital blindness: Evidence for the embodiment of Theory of Mind. *British Journal of Developmental Psychology, 24*, 507-528.
Rock, I. (1985). *The Logic of Perception*. Cambridge, MA: The MIT Press.
Röder, B., Rösler, F., & Spence, C. (2004). Early Vision Impairs Tactile Perception in the Blind. *Current Biology, 14*(2), 121-124.
Rogers, S. J., & Newhart-Larson, S. (1989). Characteristics of infantile autism in five children with Leber's congenital amaurosis. *Developmental Medicine & Child Neurology, 31*(5), 598-608.

Rogers, S. J., & Puchalski, C. B. (1984). Social Characteristics of Visually Impaired Infants' Play. *Topics in Early Childhood Special Education, 3*(4), 52-56.
Rogers, S. J., & Puchalski, C. B. (1988). Development of Object Permanence in Visually Impaired Infants. *Journal of Visual Impairment & Blindness, 82*(4), 137-142.
Rothbart, M. K., & Posner, M. I. (2006). Temperament, attention, and developmental psychopathology. In D. Cicchetti & D. Cohen (Eds.), *Developmental psychopathology: Vol. 2. Developmental Neuroscience* (2 ed., pp. 465-501). New York: Wiley.
Rowland, C. M. (1983). Patterns of interaction between three blind infants and their mothers. In A. E. Mills (Ed.), *Language Acquisition in the Blind Child: Normal and Deficient*. London: Croom Helm.
Rowland, C. M. (1984). Preverbal Communication of Blind Infants and Their Mothers. *Journal of Visual Impairment and Blindness, 78*(7), 297-302.
Ruff, H. A., & Rothbart, M. K. (1996). *Attention in early development.* Oxford: Oxford University Press.
Ruffman, T. (2004). Children's understanding of mind: Constructivist but theory-like. *Behavioral and Brain Sciences, 27*, 79-151.
Ruffman, T., Garnham, W., Import, A., & Connolly, D. (2001). Does Eye Gaze Indicate Implicit Knowledge of False Belief? Charting Transitions in Knowledge. *Journal of Experimental Child Psychology, 80*(3), 201-224.
Ruffman, T., Slade, L., & Crowe, E. (2002). The relation between children's and mothers' mental state language and theory-of-mind understanding. *Child Development, 73*(3), 734-751.
Ruffman, T., Slade, L., Rowlandson, K., Rumsey, C., & Garnham, A. (2003). How language relates to belief, desire, and emotion understanding. *Cognitive Development, 18*(2), 139-158.
Russell, J., Hala, S., & Hill, E. (2003). The automated windows task: the performance of preschool children, children with autism, and children with moderate learning difficulties. *Cognitive Development, 18*(1), 111-137.
Russell, J., & Jarrold, C. (1998). Error-Correction Problems in Autism: Evidence for a Monitoring Impairment? *Journal of Autism and Developmental Disorders, 28*(3), 177-188.
Russell, P. A., Hosie, J. A., Gray, C. D., Scott, C., Hunter, N., Banks, J. S., et al. (1998). The Development of Theory of Mind in Deaf Children. *Journal of Child Psychology and Psychiatry, 39*(6), 903-910.
Rutter, M., Bailey, A., & Lord, C. (2003). *The Social Communication Questionnaire (SCQ)*. Los Angeles: Western Psychological Services.
Salt, A., Dale, N., Osborne, J., & Tadic, V. (2005). Development of a national monitoring protocol (developmental guide) for children with visual impairment 0-36 months. *International Congress Series. Vision 2005 - Proceedings of the International Congress held between 4 and 7 April 2005 in London, UK, 1282*, 293-297.
Sandler, A.-M., & Hobson, R. P. (2001). On Engaging with People in Early Childhood: The Case of Congenital Blindness. *Clinical Child Psychology and Psychiatry, 6*(2), 205-222.
Scaife, M., & Bruner, J. S. (1975). The capacity for joint visual attention in the infant. *Nature, 253*(5489), 265-266.
Scholl, B. J., & Leslie, A. M. (1999). Modularity, Development and 'Theory of Mind'. *Mind & Language, 14*(1), 131-153.
Scholl, G. T. (1986). *Foundation of education for blind and visually handicapped children and youth: Theory and practice*. New York: American Foundation for the Blind.
Schopler, E., Reichler, R. J., & Renner, B. R. (1988). *The Childhood Autism Rating Scale (CARS)*. Los Angeles: Western Psychological Services.
Semel, E., Wiig, E. H., & Secord, W. (2000a). *Clinical Evaluation of Language Fundamentals (CELF)* (UK, 3 ed.). London, UK: The Psychological Corporation.

Semel, E., Wiig, E. H., & Secord, W. (2000b). *Clinical Evaluation of Language Fundamentals (CELF). Technical Manual.* (UK, 3 ed.). London, UK: The Psychological Corporation.
Sigman, M., & Ruskin, E. (1999). Continuity and change in the social competence of children with autism, Down syndrome, and developmental delays. *Monographs of the Society for Research in Child Development, 64*(1), 1-10.
Slade, L., & Ruffman, T. (2005). How language does (and does not) relate to theory of mind: A longitudinal study of syntax, semantics, working memory and false belief. *British Journal of Developmental Psychology, 23*, 117-141.
Slaughter, V., Peterson, C. C., & Mackintosh, E. (2007). Mind What Mother Says: Narrative Input and Theory of Mind in Typical Children and Those on the Autism Spectrum. *Child Development, 78*(3), 839-858.
Smits, B. W. G. M., & Mommers, M. J. C. (1976). Differences between blind and sighted children on WISC verbal subtests. *New Outlook for the blind, 70*, 240-246.
Sonksen, P. (1983). The assessment of 'vision for development' in severely visually handicapped babies. *Acta Ophthalmologica, 61*(157), 82-90.
Sonksen, P. (1993). The assessment of vision in the preschool child. *Archives of Disease in Childhood, 68*(4), 513-516.
Sonksen, P., & Dale, N. (2002). Visual impairment in infancy: impact on neurodevelopmental and neurobiological processes. *Developmental Medicine and Child Neurology, 44*(11), 782-791.
Sonksen, P., Petrie, A., & Drew, K. J. (1991). Promotion of Visual Development of Severely Visually-Impaired Babies - Evaluation of a Developmentally Based Program. *Developmental Medicine and Child Neurology, 33*(4), 320-335.
Spiedel, G. E. (1989). Imitation: A bootstrap for learning to speak? In G. E. Spiedel & K. E. Nelson (Eds.), *The many faces of imitation in language learning* (pp. 151-179). Berlin, Germany: Springer-Verlag.
Stern, D. N. (1985). *The interpersonal world of the infant*. New York: Basic Books.
Stone, W. L., & Yoder, P. J. (2001). Predicting Spoken Language Level in Children with Autism Spectrum Disorders. *Autism, 5*(4), 341-361.
Stuss, D. T., & Knight, R. T. (2002). *Principles of frontal lobe function*. Oxford: Oxford University Press.
Swettenham, J., Baron-Cohen, S., Charman, T., Cox, A., Baird, G., Drew, A., et al. (1998). The frequency and distribution of spontaneous attention shifts between social and nonsocial stimuli in autistic, typically developing, and nonautistic developmentally delayed infants. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 39*(5), 747-753.
Symons, D. K. (2004). Mental state discourse, theory of mind, and the internalization of self-other understanding. *Developmental Review, 24*(2), 159-188.
Symons, D. K., Peterson, C. C., Slaughter, V., Roche, J., & Doyle, E. (2005). Theory of mind and mental state discourse during book reading and story-telling tasks. *British Journal of Developmental Psychology, 23*, 81-102.
Tabachnik, B. G., & Fidell, L. S. (2001). *Using Multivariate Statistics* (4 ed.). Needham Heights: Allyn & Bacon.
Tager- Flusberg, H. (1992). Autistic Children's Talk about Psychological States: Deficits in the Early Acquisition of a Theory of Mind. *Child Development, 63*(1), 161-172.
Tager-Flusberg, H. (1992). Autistic Children's Talk about Psychological States: Deficits in the Early Acquisition of a Theory of Mind. *Child Development, 63*(1), 161-172.
Tager-Flusberg, H. (1993). What Language Reveals about the Understanding of Minds in Children with Autism. In S. Baron-Cohen, Tager-Flusberg, H., and Cohen, D. J. (Ed.), *Understanding other minds. perspectives from autism* (pp. 138-157). Oxford, UK: Oxford University Press.

Tager-Flusberg, H. (1995). Once Upon a Ribbit - Stories Narrated by Autistic Children. *British Journal of Developmental Psychology, 13*, 45-59.
Tager-Flusberg, H. (1999). An introduction to research on neurodevelopmental disorders from a cognitive neuroscience perspective. In H. Tager- Flusberg (Ed.), *Neurodevelopmental disorders* (pp. 3-24). Cambridge, MA: MIT Press.
Tager-Flusberg, H. (2000). Language and understanding minds: Connections in autism. In S. Baron-Cohen, Tager-Flusberg, H., Cohen, D.J. (Ed.), *Understanding other minds: Perspectives from developmental cognitive neuroscience* (pp. 124-149). Oxford, UK: Oxford University Press.
Tager-Flusberg, H. (2001). A reexamination of the theory of mind hypothesis of autism. In J. Burack, T. Charman, N. Yirmiya & P.R. Zelazo (Eds.), *The development of autism: Perspectives from theory and research* (pp. 173– 193). New Jersey: Lawrence Erlbaum Associates.
Tager-Flusberg, H., & Sullivan, K. (1994a). Predicting and explaining behavior: A comparison of autistic, mentally retarded and normal children. *Journal of Child Psychology and Psychiatry, 35*(6), 1059–1075.
Tager-Flusberg, H., & Sullivan, K. (1994b). A second look at second-order belief attribution in autism. *Journal of Autism and Developmental Disorders, 24*(5), 577-586.
Tager-Flusberg, H., & Sullivan, K. (1995). Attributing mental states to story characters: A comparison of narratives produced by autistic and mentally retarded individuals. *Applied Psycholinguistics, 16*(3), 241-256.
Taumoepeau, M., & Ruffman, T. (2006). Mother and infant talk about mental states relates to desire language and emotion understanding. *Child Development, 77*(2), 465-481.
Teplin, S. W. (1995). Visual Impairment in Infants and Young Children. *Infants and Young Children, 8*(1), 18-51.
Tillman, M. H., & Bashaw, W. L. (1968). Multivariate analysis of the WISC scales for blind and sighted children. *Psychological Reports, 23*, 523-526.
Tillman, M. H., & Osborne, R. T. (1969). The performance of blind and sighted children on the Wechsler Intelligence Scale for Children: Interaction effects. *Education of the Visually Handicapped, 1*(1-4).
Timer-Van de Vosse, H., & Hamers, J. H. M. (1994). Reynell-Zinkin Scales for Young Visually Handicaped Children: A Retrospective Study. In K. A. C., L. P.L., W. J.A. & V. D. W. G.J. (Eds.), *Low Vision: Research and New Developments in Rehabilitation*. Oxford: IOS Press.
Tomasello, M. (1995). Joint attention as social cognition. In C. Moore & P. J. Dunham (Eds.), *Joint Attention: Its Origins and Role in Development.* (Vol. 103-130). Hillsdale, NJ: Erlbaum.
Tomasello, M., & Farrar, M. J. (1986). Joint Attention and Early Language. *Child Development, 57*(6), 1454-1463.
Tomasello, M., & Rakoczy, H. (2003). What Makes Human Cognition Unique? From Individual to Shared to Collective Intentionality. *Mind & Language, 18*(2), 121-147.
Tomasello, M., & Todd, J. (1983). Joint attention and lexical acquisition style. *First Language, 4*(12), 197-211.
Trevarthen, C. (1979). Communication and cooperation in early infancy: a description of primary intersubjectivity. In M. Bullowa (Ed.), *Before Speech: The Beginning of Interpersonal Communication* (pp. 321–347). Cambridge, MA: Cambridge University Press.
Trevarthen, C., & Hubley, P. (1978). Secondary intersubjectivity. In A. Lock (Ed.), *Confidence, confiding and acts of meaning in the first year: Action, gesture, and symbol* (pp. 183-229). London: Academic Press.
Tröster, H., & Brambring, M. (1992). Early social-emotional development in blind infants. *Child: Care, Health and Development, 18*(4), 207-227.

Tröster, H., & Brambring, M. (1994). The Play Behavior and Play Materials of Blind and Sighted Infants and Preschoolers. *Journal of Visual Impairment & Blindness, 88*(5), 421-432.

Tröster, H., Brambring, M., & Beelmann, A. (1991). Prevalence and situational causes of stereotyped behaviors in blind infants and preschoolers. *Journal of Abnormal Child Psychology, 19*(5), 569-590.

Turner, M. A. (1999). Generating novel ideas: Fluency performance in high-functioning and learning disabled individuals with autism. *Journal of Child Psychology and Psychiatry and Allied Disciplines, 40*(2), 189-201.

Ungar, S., Blades, M., & Spencer, C. (1995a). Mental rotation of a tactile layout by young visually impaired children. *Perception, 24*(8), 891-900.

Ungar, S., Blades, M., & Spencer, C. (1995b). Visually impaired children's strategies for memorising a map. *British Journal of Visual Impairment, 13*(1), 27-32.

Ungar, S., Blades, M., & Spencer, C. (1996). The Construction of Cognitive Maps by Children with Visual Impairments. In J. Portugalli (Ed.), *The Construction of Cognitive Maps* (pp. 247-273). Dordrecht: Kluwer Academic Publishers.

Urwin, C. (1978). The development of communication between blind infants and their parents. In A. Lock (Ed.), *Action, gesture and symbol: The emergence of language* (pp. 79-108). London: Academic Press.

Urwin, C. (1983). Dialogue and cognitive functioning in the early language development of three blind children: Normal and deficient. In A. E. Mills (Ed.), *Language Acquisition in the Blind Child* (pp. 142-161). London: Croom Helm.

Warren, D. H. (1994). *Blindness and Children: an individual differences approach.* Cambridge: Cambridge University Press.

Warren, D. H., & Hatton, D. D. (2003). Cognitive development in children with visual impairment. In I. Rapin & S. Segalowitz (Eds.), *Handbook of neuropsychology, Vol. 8, Part II. Child neuropsychology* (2 ed., pp. 439-458). New York: Elsevier.

Watson, A. C., Painter, K. M., & Bornstein, M. H. (1998). Longitudinal Relations Between 2-Year-Olds' Language and 4-Year-Olds' Theory of Mind. *Journal of Cognition and Development, 2*(4), 449 - 457.

Wechsler, D. (1989). *Wechsler Preschool and Primary Scales of Intelligence - Revised (WPPSI-R).* San Antonio, TX: The Psychological Corporation.

Wechsler, D. (1992). *Wechsler Intelligence Scale for Children (WISC)* (UK, 3 ed.). London, UK: The Psychological Corporation.

Wellman, H. M. (1990). *The child's theory of mind.* Cambridge, MA: MIT Press.

Wellman, H. M., Cross, D., & Watson, J. (2001). Meta-analysis of theory-of-mind development: The truth about false belief. *Child Development, 72*(3), 655-684.

Wellman, H. M., & Liu, D. (2004). Scaling of theory-of-mind tasks. *Child Development, 75*(2), 523-541.

Welsh, M. C., & Pennington, B. F. (1988). Assessing frontal lobe functioning in children: Views from developmental psychology. *Developmental Neuropsychology, 4*(3), 199-230.

WHO. (1980). *World Health Association. The International Classification of Diseases, 9th revision, Clinical Modification, ICD-9-CM.* Ann Arbor, MI: Commission of Professional and Hospital Activities.

Wills, D. M. (1968). Problems of play and mastery in the blind child. *British Journal of Medical Psychology, 41*(213-222).

Wills, D. M. (1979). Early Speech Development in Blind Children. *The Psychoanalytic Study of the Child, 34*, 85-117.

Wimmer, H., & Perner, J. (1983). Beliefs about beliefs: Representation and constraining function of wrong beliefs in young children's understanding of deception. *Cognition, 13*(1), 103-128.

Wing, L. (1981). Asperger's syndrome: a clinical account. *Psychological Medicine, 11*(1), 115-129.

Wing, L., & Gould, J. (1979). Severe Impairments of Social-Interaction and Associated Abnormalities in Children - Epidemiology and Classification. *Journal of Autism and Developmental Disorders, 9*(1), 11-29.

Woolfe, T., Want, S. C., & Siegal, M. (2002). Signposts to development: Theory of mind in deaf children. *Child Development, 73*(3), 768-778.

Appendices

Appendix A1 - Pearson coefficients for correlations between the WISC-III and the CELF-3

	VI group – CELF-3 (scaled score)							*Sighted group - Celf-3 (scaled score)*						
	Word Classes	Recalling Sentences	Listening to Paragraphs	Word Associations	Receptive Language	Expressive Language	Total Language	Word Classes	Recalling Sentences	Listening to Paragraphs	Word Associations	Receptive Language	Expressive Language	Total Language
WISC - III														
VIQ	.524	.509	.074	.609	.426	.589	.559	.478	.640**	-.036	.604**	.387	.734**	.715**
Information	.238	.274	-.191	.434	.145	.370	.322	.374	.522**	.181	.574**	.491**	.642**	.687**
Similarities	.572	.360	.322	.399	.526	.388	.458	.371	.456	-.042	.295	.250	.467	.461
Vocabulary	.537	.402	.051	.541	.432	.481	.491	.376	.522**	.047	.354	.392	.534**	.559**
Comprehension	.553	.186	-.053	.693**	.389	.557	.571	.166	.320	.021	.560**	.149	.476	.435
Digit Span	-.161	.409	.105	.069	-.112	.252	.081	.325	.280	-.409	.255	-.057	.296	.193

** - significant at $p \leq .01$

Appendix A2 - Pearson coefficients for correlations between parental-report measures (SCQ and CCC-2) and structured assessment measures (WISC-II and CELF-3)

		WISC-III						*CELF-3*						
		VIQ	Information	Similarities	Vocabulary	Compreh.	Digit Span	Word Classes	Recalling Sentences	Listening Paragraphs	Word Associat.	Receptive Lang.	Expressive Language	Total Language
SCQ														
Total Score	VI	.094	-.179	.159	.140	-.139	.316	.056	.233	-.080	.110	.024	.119	.043
	Sighted	-.175	-.019	-.070	-.211	-.073	-.147	.123	.059	-.099	-.106	.011	-.070	-.028
CCC														
Speech	VI	-.244	.136	-.309	-.285	.059	-.454	-.357	-.085	-.183	-.031	-.333	-.046	-.139
	Sighted	.270	.371	.200	.218	.355	-.136	.032	.076	.046	.201	.062	.115	.092
Syntax	VI	.363	.399	.294	.413	.287	-.022	.158	.253	-.113	.466	.063	.404	.324
	Sighted	.263	.089	.007	.414	-.101	.393	.314	.025	.035	.318	.308	.149	.200
Semantics	VI	.184	.622	-.278	.118	.159	-.055	-.307	.126	-.336	.156	-.324	.155	.014
	Sighted	.376	.250	.272	.256	.240	.274	.043	-.071	-.216	.277	-.165	.062	-.015
Coherence	VI	-.191	.011	-.222	-.046	.012	-.278	-.192	-.008	-.122	.074	-.177	.043	.012
	Sighted	.008	.042	-.042	.074	-.080	.054	-.146	-.107	-.185	.112	-.301	-.023	-.185
Initiation	VI	-.367	-.261	-.117	-.064	-.184	-.559	-.060	-.291	-.241	-.093	-.151	-.210	-.154
	Sighted	.009	-.155	-.092	-.018	-.088	.357	-.130	-.221	.052	.039	-.056	-.109	-.126
Stereotype	VI	-.132	.052	-.257	.053	.316	-.516	.181	.034	-.338	.322	.032	.197	.196
	Sighted	-.095	-.107	-.176	-.085	.013	.087	-.153	-.412	-.004	-.071	-.152	-.337	-.352
Context	VI	.464	.729**	.137	.409	.234	-.166	-.102	.222	-.327	.295	-.178	.271	.137
	Sighted	.108	.076	-.004	-.038	.176	.185	-.190	-.171	-.139	.401	-.320	.111	-.082
Non-Verbal	VI	-.090	.299	-.195	-.089	-.080	-.266	-.472	-.337	-.222	-.246	-.431	-.303	-.344
	Sighted	-.204	.005	-.296	-.092	-.130	-.166	-.184	-.219	.192	.131	.011	-.089	-.119
Social	VI	-.179	.231	-.319	-.159	-.043	-.227	-.326	-.227	-.318	-.041	-.394	-.127	-.225
	Sighted	.277	.362	.062	.324	-.065	.140	.134	.056	.312	.349	.412	.256	.309
Interests	VI	-.439	-.129	-.396	-.203	-.032	-.594	-.012	-.170	-.219	.013	-.067	-.090	-.034
	Sighted	-.315	-.432	-.239	-.227	-.130	-.024	-.081	-.381	.019	-.056	-.090	-.293	-.271
GCC	VI	-.017	.306	-.163	.078	.143	-.395	-.172	-.007	-.309	.173	-.234	.098	.024
	Sighted	.123	.102	-.030	.109	.087	.177	-.106	-.232	-.046	.254	-.145	-.044	-.139

** - significant at $p \leq 0.01$

Appendix B1 - The Six stories from the Emotion task

1) This is a story about a boy called Max. One day Max comes home from school and his mother says: "Max, I have a surprise for you", and she gives him a little package. He doesn't know what is inside the package.

2) This is a story about a girl called Anna. Anna hears that the children outside are playing hide-and-seek so she goes outside to join them.

3) This story is about a girl called Linda. Linda's parents had told her that they would be going to the zoo today. But now, Linda's mother says that they can't go, and that they will have to stay at home.

4) This story is about a boy called Walter. Walter has a dog that he usually plays with. But today, Walter's dog is not very well and he lies in his basket.

5) This story is about a girl called Maggie. One day Maggie comes home from school. There is no one home and it's very quiet. Suddenly, Maggie can hear that someone is moving in the living room.

6) This story is about a girl called Nadia. Nadia is lying in her bed because she is going to sleep. It is night time and all the lights in the house are already switched off. Suddenly, Nadia hears a strange noise.

Appendix B2 - Example scoring sheets for the Emotion task

Table 1: *A boy with VI, age 6 years and 6 months*

					Typical emotion condition						*Atypical emotion condition*				
					Child's explanation	**Explanation category**					**Child's explanation**	**Explanation category**			
	Story	**Typical Emotion**	**Child's emotion prediction**	**N of valid stories**		**Belief**	**Desire**	**Situation**	**Don't know**	**Atypical Emotion (fixed)**		**Belief**	**Desire**	**Situation**	**Don't know**
1	Present	**Happy**	Surprised		**He didn't know** that he would have a surprise	1				Angry	Because **he didn't want** one		1		
2	Hide-and-seek	**Happy**	Happy		Because **she likes** playing		1			Afraid	Because outside is **dangerous** sometimes			1	
3	Zoo	**Angry / Sad**	Sad		Because she felt excited to go and **now she can't go**[1]			1		Happy	**She might think** there might be crocodiles and **might think** she'll be eaten	1			
4	Dog	**Sad**	Sad		Because he can't play with him anymore			1		Afraid	Because he **might catch the illness**			1	
5	Person	**Scared**	Scared		**She doesn't know** what it is	1				Happy	Because when she came in she didn't hear something, **now she can hear it**			1	
6	Strange noise	**Scared**	Scared		**She doesn't know** what it is	1				Angry	Because **it's waking her up**			1	
	Total			6		3	1	2	0			1	1	4	0

[1] Note that reference to emotions are not coded as mental states as they are self evident in this context.

Appendix B2 (continued)

Table 2: *A sighted boy, age 6 years and 4 months*

					Typical emotion condition						***Atypical emotion condition***				
					Child's explanation	**Explanation category**					**Child's explanation**	**Explanation category**			
	Story	**Typical Emotion**	**Child's emotion prediction**	**N of valid stories**		**Belief**	**Desire**	**Situation**	**Don't know**	**Atypical Emotion (fixed)**		**Belief**	**Desire**	**Situation**	**Don't know**
1	Present	**Happy**	Happy		**Because he**'s really excited and **thinks** it's gonna be a really cool toy	1				Angry	**Because he didn't want** a surprise, he **wanted** something else, like a chocolate		1		
2	Hide-and-seek	**Happy**	Happy		**Because** people are **letting her play game** with them			1		Afraid	I don't know.				1
3	Zoo	**Angry / Sad**	Angry		**Because she** didn't get what she **wanted**		1			Happy	**Because she didn't want** to go to the zoo and see the animals because she **doesn't like** animals		1		
4	Dog	**Sad**	Sad		**Because he likes** his dog a lot and now his dog gets to be sick and he cant play with him		1			Afraid	**That his dog is gonna die** and he won't be able to play with him any day.			1	
5	Person	**Scared**	Frightened		**Because she thinks** it's a burglar	1				Happy	**Because she thinks** it's her mum	1			
6	Strange noise	**Scared**	Scared		**Because she thought** it was a monster	1				Angry	**Because she doesn't want** to go to sleep.		1		
	Total			6		3	2	1	0			1	3	1	1

Appendix C1 - Mother-child discourse coding instructions

Category	*Example of utterance*	*Note*
1. Mental State Elaborations Refers to the mental state attributions in the story.	**Desire:** Want, like, love, hope, wish, dream, prefer, keen on **Emotion:** Happy, sad, unhappy, feel, cross, angry, grumpy **Modulation of assertion:** Sure, guess, figure, reckon, certain, suppose, wonder, expect, curious, bet **Think and Know:** Do you know what it is? She knows what's going to happen. They're thinking hard. Let me think. I think it's lovely. (I think so! Used to mean yes or no are coded as conversational rather than genuine uses) **Other mental state:** We did that, remember? I understand that. She didn't mean it like that.	**1. To character:** Why is she nervous? She is thinking? **2. To self:** I think, I want, I wonder, I recon, I like **3. To partner:** What do you think? Do you remember? You know that. **4. Other:** We were thinking she was a child. It's like pretending.
2. Behavioural and physical Behavioural elaborations: Refers to actions and physical aspect of the story and the book which have add to the descriptive value to the conversation	**Behavioural:** That boy is running; The cat is sitting on the bed; They are looking for her; What is she doing? **Physical:** The girl has brown hair; There is a big blue duvet on the bed; I have shoes like those; She's a child. She is smiling; She is crying; How many children are there? On this picture there is a dog. The illustrations are beautiful.	Also relating to the aspects of child's life: It's like me when I get up in the morning; Our cat does that; I would sit on the steps
3. General (other) utterances: Refers to any statement that could not be classified in any of the categories above.	What did you say?; What is that? ; We will find out? What?; No; Yes; Alright; Let's see; Lets' turn the page; Also utterances introducing the book text, e.g.: So this is called: First day jitters. So the author is: Judy Dannenberg	The statements such as: 'I don't know, I've no idea, I know' are placed into this category, they do not count as mental states

Some utterances will have both mental state and descriptive elaborations. If an utterance contains both a mental state and a descriptive elaboration, they are counted as follows:

- I think she is going to school
  - 1 utterance, 2 elaborations (1 mental state, 1 behavioural/physical).
- A picture of, I think it's either gonna be, a little girl or a little boy
  - 1 utterance, 2 elaborations (1 mental state, 1 behavioural/physical)
- I guess she is a bit scared
  - 1 utterance 2 mental state elaborations
- I think she worried about what they'll think about her
  - 1 utterance, 3 mental state elaborations

Appendix C2 - An example section of a coded transcript of mother-child discourse involving a child with VI

	Child						Parent					
Time: hrs;min	**Mentalistic**				**Non-mentalistic**		**Mentalistic**				**Non-mentalistic**	
	To character	To self	To partner	Other	Description	General	To character	To self	To partner	Other	Description	General
"First day jitters"...												
Child: What is it, mum?						1						
Mother: Uh, we will find out.												1
Ok, so this is called: **First day jitters!**												1
Child: Yeah?						1						
Page 1: The dog holding the lunch box												
Mother: You've got a doggy with ahm...sitting there											1	
and he has a lunch box.											1	
Page2: Introductory page with a big illustration												
Page 3: Acknowledgement page and the first sentence in the book:												
Mother: Yeah, and it is written by Juddy Dannenberg.												1
And the pictures are by Juddy Love.												1
And on the front of the page there's a picture of a dog											1	
and a picture of a cat											1	
and a picture of, *I think it's either gonna be*, a little girl or a little boy.								1			1	
And they are looking at the calendar.											1	
Child: Yeah?						1						
Mother: Yeah, and it's got on the calendar: **September.**											1	

Appendix C2 (continued)

And by one of the numbers it's got: **First day of school.**											1	
Child: Oh, my god, this is gonna be…						1						
Mother: (Laugh) And you know what?									1			
The little girl…/ it's either gonna be a little girl or a little boy…/ she looks really sort of scared.							1				1	
Do you know what jitters means?									1			
Child: No.						1						
Mother: You don't?												1
Jitters means you are a bit worried,										1		
like when I make a dip with the car.												1
Ok, and it's: **First day jitters.**												1
Let's see how it gets on												1
Page 3:												
"Sarah, dear, time to get out of bed." Mr. Hartwell said, poking his head through the bedroom doorway. "You don't want to miss the first day at your new school, do you?"												
Child: Oh, my, I do…						1						
Mother: Oh, dear, she is still in her bed.											1	
/OK, so…												1
Child: Is it bigger than my bed?					1							
Mother: Ahm, it's higher,											1	
it looks higher than your bed.											1	

Appendix D1 - Standardised scenarios from the RZS assessment from which attentional behaviours were coded

	Scenario description
Scenario 1	Child is presented with an item from the Sensory-Motor understanding subscale of the RZ, usually the item assessing the child's cause-effect understanding (e.g., Music box: the child has to open and shut the lid on the box to start and stop the music). Minority of (older) children may not have been presented this particular item and an available substitute scenario (one of the subsequent items from the scale) with equivalent cognitive demands is used with these children (e.g., opening/closing a screw top jar in order to get a sweet out).
Scenario 2	The child is presented with a subsequent item from the Sensory-Motor subscale, usually an item following the item given in Scenario 1. In majority of the children this scenario involves object combination play, for instance up to 3 pots of different size and colour, with neutral colour lids are introduced on the table and the child is required to put the lid(s) on. A substitute scenario of equivalent cognitive demands may be used for the minority of (older) children (e.g., object sorting task with beads and cups of different shape and size).
Scenario 3	This scenario involves presenting the child with a single or a combination of objects in a task requiring recognition of every day objects through a) meaningful or adaptive use of the objects or b) meaningful labelling or selection of objects (usually observed in older children). Even though this scenario may be derived from the Sensory Motor subtest for younger children and from the Verbal comprehension subtest for older children, it is chosen as it involves the same toys and places equivalent cognitive demands on the children. For instance, younger children are presented with up to 3 objects (e.g., cup, spoon and brush, usually one after another) and are assessed on their ability to recognise these objects by their adaptive use or labelling them correctly. Older children are presented with up to 2 arrays of 3 objects (e.g., cup, spoon and brush; shoe, sponge and comb) which they are asked to label, select based on the labels provided or show their adaptive use.
Scenario 4	In this scenario the examiner engages the child in a combination play with every day objects, which follow up from the items in Scenario 3. The examiner's aim within this scenario is to assess further meaningful recognition of the objects through their adaptive use and pretend play where possible; imposing the same cognitive demands like Scenario 3.
Scenario 5	An additional scenario where the examiner engages the child in free play with a single or multiple toys (e.g., telephone on its own or in combination with a doll/ teddy bear). This scenario places the cognitive demands required for meaningful recognition and adaptive use of objects from the Sensory Motor subscale and usually incites pretend play in children.

Appendix D2: Attention behaviours coding schedule

Attention category	*Code*	*Behaviours*
Establishing: The baseline criterion: Child's attention is directed by the examiner using visual, tactile and/or auditory means onto a task involving single or multiple objects.	Immediately successful (scores 2)	Begins manipulating introduced objects (e.g., prolonged looking, listening or reaching out to objects leading to exploring the objects using hands or mouth; manifesting adaptive use of the objects; manifesting pretend play)
	Delayed/ Successful with difficulty (scores 1)	Child's attention is gained onto the task after some effort, usually if the child is passive initially and/or is reluctant to engage with the introduced toys straight away (even though child's attention may be shifted onto these toys – see Attention Shift below)
	Unsuccessful (Scores 0)	Child does not establish attention onto the task after numerous attempts by the assessor.
Maintaining: The baseline criterion: Child's attention has been established by the examiner	Continuous (Scores 2)	Child's attention is successfully maintained on the task by the assessor until a new task (involving single or multiple objects) is introduced (i.e. child plays with or explores the toys). Some distraction may be observed allowing for other things or people in room, and the child may try and direct the assessor's attention within the task but child's attention can generally be redirected onto the task without difficulty.
	Somewhat disrupted (Scores 1)	Child shows some difficulty in holding attention on the task (.e., child may get easily distracted and may engage in stereotypical behaviours but the assessor can generally with some effort redirect the child's attention on the task). Also, child may maintain attention well for quite a while but then breaks the interaction and cannot be redirected onto the task, leading to the assessor having to introduce new toys.
	Disrupted (Scores 0)	Child does not hold attention on the task or maintaining is very brief; there may be extensive use of stereotypical behaviours (e.g. whistling, casting) or distress at the assessor's attempt to engage the child.
Shifting: * Baseline criterion: Adult directs child's attention away from the object they are engaged in onto a novel object using visual, tactile and/or auditory means.	Immediately successful (Scores 2)	Child shifts attention onto the novel object straight away (i.e., child 'stills' while listening or looking at novel objects; releasing the object they are engaged in and or/ reaching for the novel object). Children who have completed the task (maintained continuously) and are waiting for the examiner to introduce a new toy should not be penalised on Shift.
	Successful with difficulty /Delayed (Scores 1)	Assessor has to re-introduce the novel object before the child switches attention (including multiple modalities in children with VI)
	Unsuccessful (Scores 0)	Child does not shift attention onto novel object after numerous attempts by the assessor

*In contrast to Attention Establish, which requires the child to engage with a novel object, this measure takes into account the flexibility of switching attention between objects and tasks without having to engage with them. For instance, child may momentarily 'still' and listen (PVI children) or look (SVI and S) at the new toy, but may not engage with it.