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# ASSESSING ANOSOGNOSIA: A CRITICAL REVIEW

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## SUMMARY

*Anosognosia is a multi-factorial syndrome whose clinical manifestations can vary considerably from patient to patient. Considering the complexity of this syndrome, its assessment represents a major challenge for the diagnostic process, and presents various methodological complications. Lack of agreement about diagnostic criteria, high exclusion rates of specific patient categories, assessment limited to certain aspects of unawareness, and the lack of normative data and methodologically robust tools may have led to contrasting, or apparently contrasting, interpretations of this syndrome. Despite the apparently greater sensitivity of the last generation of anosognosia studies to many of these issues, many aspects are still overlooked. In this manuscript, we critically review the literature on the assessment of anosognosia, mainly focusing on motor and language deficits, with a note on the issue of awareness for memory deficits.*

**Keywords:** *paresis, aphasia, amnesia*

> *Did he know this? Of course he did. He was a physician, so he must know. But he was human, so he could arrange many things for himself. Both knowing and not knowing – one of the most frequent human arrangements.*
>
> Saul Bellow, *Mr Sammler's Planet*, 1969

# INTRODUCTION

Anosognosia is a multifarious syndrome, whose clinical manifestations can vary considerably from patient to patient (see Prigatano, this issue). Patients may acknowledge their deficit but appear unconcerned (anosodiaphoria; Babinski, 1914), or deny their hemiplegia but agree to remain in the ward for treatment (e.g., Marcel et al., 2004); yet even if severely paretic, they may attempt to perform tasks that usually require both limbs, such as walking (e.g. Bisiach & Geminiani, 1991). Other patients acknowledge their deficit only if it is attributed to third parties in the same condition (e.g. Marcel et al., 2004 for hemiplegia; Maher et al., 1994 for aphasia), or partially admit their impairment though ascribing it to causes other than their brain insult, such as arthritis. Others even manifest delusions, such as denying that the paretic limb is their own (somatoparaphrenia; e.g. Ramachandran & Blakeslee, 1998). Moreover, anosognosia can be deficit-specific; that is, some patients may deny their aphasia but acknowledge their hemiplegia, or *vice versa* (e.g. Kinsbourne & Warrington, 1963 case 1; Breier et al., 1995). Dissociations have been observed even within the same type of impairment; for instance, patients may acknowledge the motor impairment of their arm but deny the paresis of their leg, or *vice versa* (Berti et al., 1996; Bisiach et al., 1986; Della Sala et al., 2009). Similarly, aphasic patients have been reported who deny their spoken errors but concede their written ones (e.g. Marshall et al., 1998 – case RMM).

# ANOSOGNOSIA FOR MOTOR IMPAIRMENTS

The reported frequency of anosognosia following brain damage varies considerably across studies, ranging from 7% to 77% (Orfei et al., 2007; see also Jehkonen et al., 2006). This variability may be due to different diagnostic criteria (Baier & Karnath, 2005) or to recruitment of patients at different post-onset intervals. Indeed, while some studies (Cutting, 1978; Starkstein et al., 1992; Stone et al., 1993; Baier & Karnath, 2005, 2008; Vocat et al., 2010) investigated the presence of anosognosia in patients in the acute phase of their disease (i.e. less than one month – Levine et al., 1991), other studies considered more heterogeneous groups of patients, including both acute and chronic patients (Nathanson et al., 1952; Berti et al., 1996; Marcel et al., 2004; Spalletta et al., 2007; Della Sala et al., 2009; Cocchini et al., 2010a).

Anosognosia for motor impairment is usually considered to be a syndrome that spontaneously resolves within a few weeks, and chronic anosognosia is thought to be associated with general cognitive impairment (McGlynn & Schacter, 1989; Levine, 1990; Levine, et al., 1991; Goldberg & Barr, 1991). However, reviews of the literature have shown that chronic anosognosia is not as rare as textbooks suggest (see Table 1 in Cocchini et al., 2002; Orfei et al., 2007; Jehkonen et al., 2006), and several studies have pointed out how lack of awareness can seriously interfere with functional recovery and rehabilitation training (Gialanella & Mattioli, 1992; Maeshima et al., 1997; Hartman-Maeir et al., 2001; Hartman-Maeir et al., 2002; Appelros et al., 2002; Gialanella, et al., 2005; Di Legge et al., 2005).

It is possible that chronic anosognosia could have been underestimated because of the diagnostic tools; indeed, relatively little attention has been paid to the assessment of anosognosia (for a critique see Jenkinson et al., 2011). Prigatano (2010) has pointed out that questionnaires or structured interviews, often used in anosognosia diagnosis, are only an indirect measure of awareness, and that the method of comparing the patients' self-evaluations on questionnaires to those of their caregivers (Prigatano & Altman, 1990; Prigatano et al., 1997) is flawed with methodological biases (see also Orfei et al., 2010a).

Marcel et al. (2004) observed that more anosognosic cases were reported when questionnaires enquiring about specific bimanual and bipedal tasks were used, rather than general interviews about motor impairments. We suggested that patients in sub-acute or chronic phases may have been overexposed to some of the more common questions used to assess anosognosia (Cocchini et al., 2009). Thus, patients may have provided the "correct" response based on what they had "learned" about themselves rather than on their actual awareness of their deficit. Interestingly, we observed that the questions that best predicted the presence of anosognosia in the less acute phases were those enquiring about activities such as "washing dishes" or "opening bottles", whereas the worst predictors were questions about "walking" or "clapping hands", both recurrent questions in the classical assessments of anosognosia, such as the structured interviews (Della Sala et al., 2009).

A further problem is posed by patients showing only a mild or moderate motor deficit. Nathanson et al. (1952) suggested that patients with mild paresis may show anosognosia; however, since their response to questions such as "Can you move your hand?" could be difficult to interpret, "the criterion of complete paralysis was necessary" (p. 381). Structured interviews rely on the fact that the patient is claiming to be able to move a paretic limb. However, some patients may actually be able to perform partial movements, rendering the interpretation of their answers ambiguous, especially if norms are not available. Moreover, some patients might not report their motor impairment spontaneously, as this may subjectively be considered less relevant than other ailments which afflict them (Baier & Karnath, 2005); this would be particularly true in the case of moderate or mild motor impairment. As a consequence, many authors have limited recruitment for their studies to acute stroke patients presenting with complete paresis (e.g. Baier & Karnath, 2008; Vocat et al., 2010).

It is generally accepted that anosognosia for hemiplegia is caused by lesions in the right hemisphere. However, most diagnostic measures rely heavily on the patients' verbal report. This is a clear limitation in studies attempting to assess anosognosia in aphasic patients, and has led to high exclusion rates of left brain damaged patients, and possible underestimation of anosognosia for right hemiplegia (e.g., Cutting, 1978; Morin et al., 2007; Cocchini et al., 2009). Studies capitalising on the intra-carotid sodium amobarbital procedure (Wada test) did show a higher frequency of anosognosia for left hemiplegia after anesthesia of the right hemisphere; however, they also indicated that when the left hemisphere was inactivated, the frequency of anosognosia for right hemiplegia varied greatly, ranging from zero (Gilmore et al., 1992) to 86% (Durkin et al., 1994). It follows that the apparent strong hemispheric asymmetry that has guided research and theoretical interpretations may suffer from a methodological bias, and the left hemisphere may play some role in awareness, even if probably less crucial than or different from that of the right hemisphere.

Awareness is a general term which encompasses different levels. Typical questions in structural interviews are: “Are your hands equally strong?” or “Do you have any problem in walking?”. Hence, this method may not be suitable to assess less explicit manifestations of anosognosia. For example, Ramachandran and Blakeslee (1998) described a patient severely anosognosic for his hemiplegia. However, during a conversation with the authors, the patient commented “I can't wait to get back to two-fisted drinking” (p. 143). Another patient (CC) described by Berti et al. (1998) showed a “backwards awareness” for a deficit which was never directly acknowledged. When the examiner asked her how her left hand was, CC stated: “it was very disobedient […] I wanted to lift it up…but it… nothing!” (p. 31) despite claiming soon after that now her limb was fine and could be lifted up without any problem. Investigation of this less explicit anosognosia may reveal important information about awareness mechanisms and guide rehabilitation training.

## METHODS TO ASSESS ANOSOGNOSIA FOR MOTOR IMPAIRMENT

### Explicit anosognosia

The assessment of anosognosia is usually based on meta-cognitive tasks, where patients are asked to reflect upon their own condition and provide some type of self-evaluation. This is what we will refer to as “explicit anosognosia”. The most common diagnostic tools are structured interviews and self-evaluation questionnaires (e.g., Jehkonen et al., 2006; Orfei et al., 2007, 2010a; also summarised in Table 1).

*The structured interview*

Interviews engage the patients in a more or less structured conversation in order to understand whether they are aware of their deficit or if they can become aware after demonstration of the impairment. The examiner often assigns a score indicating the patient's degree of awareness. Typically, these interviews begin with general questions about the patient's situation and then narrow down to a specific deficit. If the patient denies his/her deficit, then they are "forced" to face a demonstration (e.g. "Can you touch the examiner's nose with your left

Table 1. Most common tests and methods to assess various types of anosognosia for motor deficits

| Awareness type | Method/ Test | Original paper/s | Summary of procedures |
|---|---|---|---|
| EXPLICIT AWARENESS | ***Structured Interview Method*** | | |
| | Nathason et al.'s Interview | Nathason et al. (1952) | Clinician's evaluation and comparison with patient's actual performance; dichotomous diagnosis of denial |
| | Cutting's Scale | Cutting (1978) | Clinician's evaluation and comparison with patient's actual performance; classification of anosognosia and related phenomena (See also Table 2) |
| | Bisiach et al.'s Scale | Bisiach et al. (1986); Bisiach & Geminiani (1991) | Clinician's evaluation and comparison with patient's actual performance; scores range 0 (aware) - 3 (severe anosognosia) |
| | Starkstein et al.'s Questionnaire | Starkstein et al. (1992) | Clinician's evaluation based on 6 questions; if the patient shows denial then comparison with patient's actual performance; score for motor and visual deficits from 0 (aware) to 3 (unaware) |
| | Berti et al.'s Scale | Berti et al. (1996) | Clinician's evaluation and comparison with patient's actual performance; scores range from 0 (aware) to 2 (severe anosognosia) |
| | Feinberg et al.'s Scale | Feinberg et al. (2000) | Clinician's evaluation and comparison with actual performance; questions for anosognosia embedded in 10 more general question about motor impairment; score range for each question: 0 (aware); 0.5 partially aware 1 (complete unawareness) |
| | Structured Awareness Interview | Marcel et al. (2004) | Clinician's evaluation but also comparison with patient's actual performance; responses are scored on a 3-point scale and then classified as "aware", "unaware" or "inapplicable"; a post-performance evaluation is also required after patient's actual performance of tasks; the clinician evaluates patients' rating of their post-performance as correct (score =1); minor overestimation (score =2) and gross overestimation (score = 3) |
| | ***Self-rating questionnaires*** | | |
| | Patient Competency Rating Scale (PCRS) and PCRS-revised | Prigatano et al. (1986); Prigatano et al. (2005) | Self-rating questionnaire; 5-point Likert scale; comparison with caregivers; discrepancy =evidence of anosognosia; no normative data |
| | Self-rating Questionnaire | Berti et al. (1996) | Self-rating questionnaire; 15 bi-manual and bi-pedal questions plus 4 monomanual questions; 10-point Likert scale; rating equal or higher than 6 = evidence of anosognosia; section 2: comparison with patient's actual performance |

[paretic] hand?"; Bisiach et al., 1986, Berti et al., 1996). Responses to this question can provide interesting information. For example, when Welman (1969) asked his patient to lift the paretic hand, the patient replied "I'm not superman!" while one of Ramachandran and Blakeslee's (1998) patients (RM) uttered "Done" despite the fact that her arm remained motionless. Both these patients showed lack of awareness of their hemiplegia, but it is interesting to consider the patients' differing reactions to the task. Welman's patient acknowledged, to some extent, that his hand did not move, whereas RM denied clear evidence that she could not move her arm, suggesting that very different mechanisms might underlie these patients' anosognosia. For example, one of our own patients, NL, when asked to lift his left (paretic) hand, kept raising his right unimpaired hand. NL's

Table 1(cont.). Most common tests and methods to assess various types of anosognosia for motor deficits

| | | | |
|---|---|---|---|
| | Estimation of current abilities | Marcel et al. (2004) | Self-rating questionnaire; 13 bimanual and bipedal questions; comparison with caregiver's rating; overestimation in 5 or more questions = anosognosia; section 2: comparison with actual patient's performance |
| | Visual-Analogue Test for Anosognosia for motor deficit (VATAm) * | Della Sala et al. (2009) | Self-rating questionnaire: 12 bimanual and bipedal questions; 4-point Likert scale; comparison with caregivers; normative data and cut-offs for mild, moderate and severe anosognosia; check questions available to ensure reliability of patient's data; suitable for aphasic patients (see also Figure 1) |
| **IMPLICIT AWARENESS** | 1st- and 3rd-person estimates | Marcel et al. (2004) | Rating of self (1st-person condition) and others (3rd-person condition) performance on 13 bimanual and bipedal actions ; comparison of patients' rating in the two conditions for each question: if 1st-person rating is greater than 5 and 3rd-person rating is half or less, the response is classified as 1st>3rd; the opposite is classified as 3rd>1st |
| | Dot-probe test | Nardone et al. (2007) | Response latencies in a decision task are compared when a "hemiplegia-related word" versus "non-hemiplegia related word" is presented with the target; higher latencies indicate denial mechanisms in progress |
| | Behavioural Motor Task (BMT) | Cocchini et al. (2009) | Patient's actual performance on 8 bimanual tasks; error score indicates lack of use of aware strategies; cut-off available and normative data available; suitable for aphasic patients; possibility to ask for second performance to test empirical learning; diagnoses only anosognosia for upper limb severe or moderate motor impairment |
| | Implicit test for anosognosia | Fotopoulou et al. (2010) | Latencies during completion of sentences taken from section 2 of the Hayling Test; 10 emotionally neutral; 10 emotionally negative; 10 emotionally negative deficit-specific sentences; an explicit task can be also associated where patients are asked to rate from 1 (not related) to 10 (extremely related) how much each sentence was related to their current situation |
| | | | |
| See text for specific terms | | | |
| * A similarly structured assessment (VATAmem) is in preparation to assess anosognosia for one's memory disorders. | | | |

behavior supported the idea that his anosognosia might have been caused by an associated disorder of body schema and personal neglect (Cocchini et al., 2002). An attempt to consider different patients' "reactions", and not just lack of awareness, can be found in Cutting et al.'s (1978) study, where the authors provided a classification of the patients' responses to specific questions/situations, identifying a set of "anosognosic phenomena", which could be associated with a general lack of awareness (see Table 2).

### *Self-evaluation of motor abilities*

Another method often used to evaluate explicit anosognosia for motor impairment consists in asking the patients to rate their ability to perform various everyday tasks which require the use of both hands or legs, such as clapping hands

Table 2. Anosognosia questionnaire (Cutting, 1978)

Anosognosia

General questions:

- Why are you here?
- What is the matter with you?
- Is there anything wrong with your arm or leg?
- Is it weak, paralysed or numb?
- How does it feel?
-

Procedure if denial elicited on general questions
(Arm picked up)

- What is this?
- Can you lift it?
- You clearly have some problem with this?
  (Asked to life arms)
- Can't you see that the two arms are not at the same level?

Anosognosia phenomena

| Phenomenon | Questions |
|---|---|
| Anosodiaphoria | Is it a nuisance? How much trouble does it cause you? What caused it? |
| Nonbelonging | Do you ever feel that it doesn't belong? Do you feel that it belongs to someone else? |
| Strange feelings | Do you feel the arm is strange or odd? |
| Misoplegia | Do you dislike the arm? Do you hate it? |
| Personification | Do you ever call it names? |
| Kinaesthetic hallucinations | Do you ever feel it moves without your moving it yourself? |
| Overestimation | How's the other arm? |
| Phantom supernumerary limb | Do you ever feel a strange arm lying beside you separate from the real arm? |

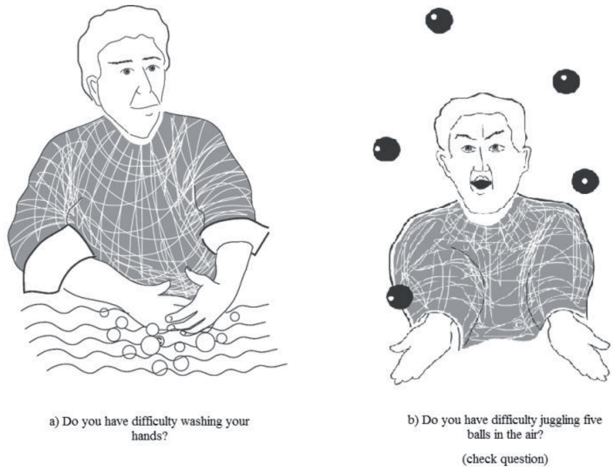


Figure 1. Examples of questions and illustrations from the VATAm. Redrawn from Della Sala et al., 2009

or walking (e.g. Berti et al., 1996; Marcel et al., 2004; Della Sala et al., 2009). The patient's possible overestimation of their motor abilities, as contrasted with a caregiver's rating of their actual performance, is interpreted as evidence of anosognosia. Compared to the structured interviews, this type of assessment allows clinicians and researchers to better identify the various degrees of unawareness, and this method can also be used with patients with moderate motor impairments. However, the patients are asked to "predict" what *would* be their performance in specific situations. Moreover, this method relies heavily on the patients' verbal competency, which leads to high rates of exclusion of left-brain damaged patients, resulting in underestimation of anosognosia following left-brain damage.

The recently developed VATAm (Visual-Analogue Test for Anosognosia for motor deficits – Della Sala et al., 2009) contains some innovations that allow for a more reliable investigation of anosognosia in brain damaged patients in general, and specifically in patients presenting with language impairments. In this task, patients are asked to rate their own performance on a series of bimanual and bipedal tasks. To account for possible verbal communication difficulties, each question is illustrated by a drawing (see Figure 1a) and a 4-point visual-analogue Likert scale facilitates the patients' rating. Finally, the patient's reliability is monitored by means of 4 "check questions" requiring obvious answers regardless of the impairment (e.g. ease in juggling five balls in the air -– see Figure 1b). Patients' ratings are then compared with those of their caregivers, and normative data allow the interpretation of possible discrepancies with cut-offs which identify various degrees of anosognosia (i.e. mild, moderate and severe). This test has recently been compared with the structured interview method in assessing anosognosia in left-brain damaged patients showing various degree of motor impairment (Cocchini et al., 2009). By means of the VATAm up to 70% of the left-brain-damaged patients were reliably assessed, and 40% of them showed some degree of anosognosia. By means of the structured interview, on the other hand, only half of the patients could be assessed and of these only 10% were identified as anosognosics. Interestingly, a high number of anosognosic patients diagnosed by means of the VATAm consisted of patients showing severe and moderate, not mild, degrees of anosognosia.

However, this test also relies partly on verbal competence, and the reliability of the patients' responses is indirectly inferred from check questions. Moreover, the patients have different degrees of direct experience of the situations depicted. For example, some may have already experienced difficulty in washing their hands but not in washing dishes. Some authors have also pointed out some gender biases in the patients' responses that should be considered (Marcel et al., 2004).

The role of the caregiver's evaluation is also debatable. Despite some studies showing that caregivers' ratings reflect data from more objective measures of the patient's deficit (e.g. Fleming et al., 1996; Della Sala et al., 2009), caregivers are offering a subjective evaluation, which may be affected by personal, as well as medical, factors (Prigatano et al., 2005; Godfrey et al., 2003). Therefore, the comparison with actual performance may be preferable (Berti et al., 1996; Marcel et al., 2004).

### *Implicit awareness*

Answers to questionnaires or structured interviews may not be the best instruments to reveal phenomenological experience (Prigatano, 2010). There may be some discrepancy between what is acknowledged verbally and what is less consciously believed by the patient, yet responsible for their actual behavior. House and Hodges (1988) described an anosognosic patient who, amongst a series of pictures of people, identified those on a wheelchair as most similar to her. These observations suggest that some knowledge about the deficit can be processed without reaching consciousness, and may become apparent only in the patient's behavior, conversation or responses to indirect questions.

Several recent studies have attempted to address implicit processing in anosognosia with novel methods that allow for a systematic investigation (see Table 1). Marcel et al. (2004) developed the "1$^{st}$- and 3$^{rd}$ -person estimates task" whereby patients are asked to rate their own ability in performing a specific bi-manual or bipedal task and then to rate the same ability by somebody in the same condition (see also Berti et al., 1996). Under these conditions, some anosognosic patients rated the ability of the other person significantly lower than their own. Nardone et al. (2007; see also Fotoupoulou et al., 2010) reported further evidence for implicit awareness in anosognosic patients. By means of the Dot probe paradigm, whereby patients are asked to perform a speedy decision task about the color of a target, these authors observed that anosognosic patients showed longer latencies than aware patients if a word related to a motor tasks (e.g. walking) was displayed simultaneously with the target. No significant differences were observed between the two groups if the word was not related to a motor task (e.g. reading) (Figure 2). The authors suggested that longer latencies by anosognosic patients in association with motor words reflected a denial process triggered by the presence of threatening information.

Inspired by Ramachandran and Blakeslee's (1998) work, we have recently developed the Behavioural Motor Task (BMT – Cocchini et al., 2010a), which assesses anosognosia considering the patient's actual behavior while performing simple bimanual motor tasks. Although these tasks are usually performed better using both hands (e.g. holding a two-handle tray placing one hand at each extremity), they can also be performed using only one hand if the motor action is re-organized ("aware strategy" – e.g. placing the unimpaired hand underneath the tray, at the center). We observed that about a quarter of our sample (i.e., 7 out of 30; 23%) did not adopt "aware strategies." Interestingly, some of them started to adopt aware strategies when asked to perform the same tasks a second time (empirical learning). Despite the fact that this benefit was transient for several of them, these findings suggest that a process of empirical learning may not only modify the patient's verbal evaluations, as reported by Marcel et al. (2004) and by Berti et al. (1996; cases CF and CG), but also the patient's behavior. This change of awareness after actual performance is intriguing, as it suggests that in some cases unawareness may be due to lack of information, or to difficulty in understanding the feedback, about their motor impairment.

Double dissociations between implicit and explicit anosognosia, reported in some studies (e.g. Marcel et al., 2004), imply that different mechanisms might

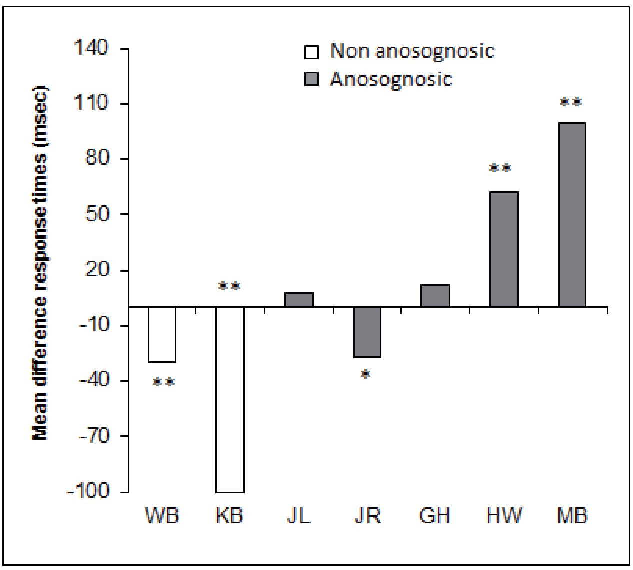


Figure 2. Mean difference response times (DRT) obtained by two patients aware and five patients unaware of their motor deficits. DRTs were based on the mean response latencies to a color target when motor related word conditions vs. non-motor related word conditions were considered. Significance values are represented as: *p < .05; **p < .01. Redrawn from Nardone et al., 1997

be responsible for different types of anosognosia. Limiting the assessment to one aspect, the more explicit one, may have led clinicians and researchers to overlook other forms of anosognosia. Less explicit information can be crucial to better account for anosognosia, as it suggests a possible contribution of motivational and psychological mechanisms in anosognosia (Weinstein & Kahn, 1955; Weinstein, 1991; Turnbull & Solms, 2007; Nardone et al., 2007). One direct implication of these accounts is that patients should possess information about their motor deficit for a denial mechanism to be triggered, but this information may have not reached full consciousness. Marcel et al.'s (2004) "1$^{st}$- and 3$^{rd}$ - person estimates task" could be easily adopted in clinical settings, but it still relies to some extent on explicit and verbal report. On the other hand, the BMT is not suitable for patients showing mild motor impairments, as they may still be able to perform the task, although clumsily.

## ANOSOGNOSIA FOR LANGUAGE IMPAIRMENT

Aphasics show anosognosia for various aspects of their language difficulties, including comprehension, production and even pragmatics (i.e., ability to respect the rules of effective communication; Kertesz et al., 2010; see also Rubens & Garrett, 1991; Vuilleumier, 2000; Adair et al., 2003; Kertesz, 2010). Lack of awareness of language impairment has been considered a key feature of jargon aphasia, to such an extent that this type of aphasia has been defined as "a mixture of aphasia and anosognosia" (Weinstein et al., 1966; p. 187). Anosognosia for aphasia has been associated with bilateral lesions (e.g., Weinstein et al., 1966; Ruben & Garett, 1991) suggesting a crucial role of the right hemisphere also in the denial of language disorders. However, lack of awareness of aphasia has been observed also in patients whose lesion was confined to the left hemisphere (Kertesz & Benson, 1970; Gainotti, 1972; Cocchini et al., 2010b; Cocchini et al., submitted). Within this framework, it has been suggested that anosognosia for aphasia results from a lack of comprehension or disturbed feedback of one's own speech (e.g. Leburn, 1987). Gainotti (1972) investigated anosognosia in a group of 19 non-fluent, 16 fluent and 24 amnesic aphasic patients. He found that only 4 fluent aphasics appeared unaware of their language deficits. Therefore, the association of unawareness with jargon and sensory aphasia, and the fact that monitoring relies on intact comprehension, has led researchers to maintain that poor comprehension would prevent accurate monitoring and discovery of the deficit (e.g., Boller et al., 1978; Wernicke, 1874; Heilman, 1991). Other studies, however, reported that some anosognosic patients could recognise their own speech errors when listening to their recorded performance (Shuren et al., 1995; Marshall et al., 1998) or if the errors were attributed to other persons (Alajouanine, 1956; Kinsbourne & Warrington, 1963; Maher et al., 1994), suggesting other possible causes for anosognosia than a comprehension deficit.

# METHODS TO ASSESS ANOSOGNOSIA FOR LANGUAGE DISORDERS

The assessment of anosognosia for language deficits is particularly complex due to the difficulty in interpreting aphasics' responses to questions related to their deficits. In this context, information about the patient's explicit self-evalua-

Table 3. Most common tests and methods to assess various types of anosognosia for language deficits

| Awareness type | Method/ Test | Original paper/s | Summary of procedures |
|---|---|---|---|
| | ***Clinical scales*** | Kertesz et al. (2000) | Clinician evaluates patient's verbal ability based on observation; often awareness for more than one deficit is assessed. |
| **EXPLICIT ANOSOGNOSIA** | ***Error detection method*** | | |
| | | Marshal et al. (1998), Gainotti (1972); Maher et al. (1994); Weinstein et al. (1966); Alajouanine (1956); Shuren et al. (1995) | Number of errors detected during conversation, verbal descriptions or naming tasks |
| | | Maher et al., 1994; Marshall et al. 1998 | The number of errors detected by the patients while they are speaking (on-line condition) is compared with the number of errors detected when patients are listening to their own tape-recorded performance (off-line condition); this provides indication of possible depletion of attention resources. It is also possible to compare number of errors detected by the patients while they are listening to their own performance (own-condition) with the number of errors detected by the patients when they are listening to other persons' recorded performance (other-condition); this provides information about possible psychological denial |
| | ***Self-rating assessments*** | | |
| | Visual-Analogue Test for Anosognosia for Language Disorders (VATA-L) * | Cocchini et al. (2010b) | Self-rating questionnaire: 14 questions concerning various language tasks; 4-point Likert scale; comparison with caregivers' ratings; normative data and cut-offs are available for diagnosis of anosognosia for comprehension and expressive language impairment; check questions are available to ensure reliability of patient's data; suitable for aphasic patients (see also Figure 2) |
| **IMPLICIT ANOSOGNOSIA** | ***Self-correction method*** | | |
| | | Marshall & Tompkins (1982); Hofmann & Cohen (1979); Schlenck et al. (1987); Oomen et al., 2001 | Patients are asked to describe a complex scene or perform a naming task; self-corrections are analysed in terms of pre-pair and repairs; analyses of pre-pairs include various types of pre-articulatory attempts (including latencies); analysis required expertise on aphasia. |
| See text for specific terms | | | |
| * A similarly structured assessment (VATAmem) is in preparation to assess anosognosia for one's memory disorders. | | | |

tion, so relevant for anosognosia for motor impairment, is rarely considered (e.g. Breier et al., 1995; see also Kertesz's 2010) or is reported anecdotally (e.g. Kinsbourne & Warrington, 1963; Kertesz 2010). The difficulty in engaging patients with language difficulties in relatively complex conversations about their deficits and the risk of collecting unreliable data, have led clinicians and researchers to explore alternative methods of assessing anosognosia for aphasia either by means of clinical scales or by indirect methods, such as the evaluation of the patients' self-corrections (See Table 3).

Clinical scales devised to assess lack of awareness in patients presenting with language difficulties are mainly based on clinicians' evaluations (e.g. Kertesz et al., 2000). Some of these scales contain several questions related to various deficits (e.g., apathy, inhibition) together with questions enquiring about the patients' insight into their language deficit (e.g. "Is s/he aware of any problems or changes in behavior, or does s/he seem unaware of them or deny them when discussed?"; Kertesz et al., 1997).

# EXPLICIT ANOSOGNOSIA

The patients' explicit knowledge of their own language deficits can currently only be assessed by means of crude and over-simplified questions, hence the "development of methods for investigating unawareness in different aphasic groups is clearly necessary" (McGlynn & Schacter, 1989; p. 180).

*Error detection method*

The error detection method (Marshall et al., 1998) has been used in several studies to assess explicit awareness of aphasia (see Table 3). Patients are typically asked to detect their errors during the description of a complex scene or during a naming task (e.g. Alajouanine, 1956; Gainotti, 1972; Maher et al., 1994; Marshall et al., 1998).

The ability to detect errors implies a flawless monitoring process, which requires a correct analysis of feedback information (Boller et al., 1978; Peuser & Temp, 1981). However, comprehension or feedback disorders *per se* may not be the only cause of monitoring failures. Failures in error detection may result from various causes, including the patients' inability to accurately compare the actual with the intended output (Marshall et al., 1998). In line with this hypothesis, some studies (Shuren et al., 1995; Marshall et al., 1998; Maher et al., 1994) have reported on patients whose detection of speech errors was better when they were asked to listen to their own previously tape-recorded performance (off-line condition), than they did while speaking (on-line condition). In particular, Maher et al. (1994) systematically investigated error detection ability in one aphasic patient (case AS) during on-line and off-line conditions. AS recognized only about 25% of his own errors in the on-line condition, but he identified as many as 65% of his errors in the off-line condition. Maher et al. (1994) interpreted the different performance in the two conditions as a reduced attentional capacity that would compromise the patient's ability to speak and monitor simultaneously (see also Lebrun, 1987; Shuren et al., 1995; Oomen & Postma, 2002). This type of interpretation implies that the monitoring process is not directly linked to the language deficit *per se,* as it would be in the case of a lack of comprehension of one's own speaking errors. Also Nozari et al. (2011) reported poor correlation between error detection and comprehension, and they suggested that speech error detection may depend mainly on production, rather than comprehension, processes.

Further analyses of types of errors have provided interesting data. Some jargon aphasics may acknowledge their oral and written mistakes only when these are believed to be another person's errors (Alajouanine, 1956; Kinsbourne & Warrington, 1963; Maher et al., 1994). Maher et al.'s (1994) patient AS was asked to listen to a tape-recorded examiner's performance and to detect possible errors. The errors were the same as those previously committed by the patient. AS identified a higher percentage (88%) of the "examiner's errors" than of his own recorded errors (i.e. 65%), suggesting a higher sensitivity in detecting others' errors than their own. Maher et al. (1994) did not exclude the possibility that in addition to a reduced attentional capacity, AS's unawareness of speech errors could also be caused by adaptive denial mechanisms. Successful monitoring of speech depends on a number of intact processes, while "a deficit in any of these mechanisms may be sufficient to cause anosognosia for aphasia" (Maher et al., 1994; p. 415). Therefore, as for anosognosia for motor impairment, anosognosia for aphasia should also be considered a multi-factorial phenomenon, which may require different diagnostic tools tapping different aspects of awareness.

### *Self-rating method*

To our knowledge the VATA-L (Visual-Analogue Test for Anosognosia for Language Disorders – Cocchini et al., 2010b) is the only method that attempts to assess anosognosia for aphasia requiring self-rating of language impairments (see Table 3). The VATA-L format is very similar to the VATAm previously described. It consists of a series of questions about the patients' ability to perform common verbal tasks requiring language production and comprehension deficits. Drawings illustrate each question to facilitate comprehension (see Figure 3a), the visual-analogue Likert scale facilitates the patient's rating task, response reliability is monitored by means of check questions (see Figure 3b), and normative data allow interpretation of possible rating discrepancy (index of anosognosia) between patients and caregivers. Our recent study showed that most of the aphasics manage to reliably complete the test and that 20% of them showed pathological underestimation of their language deficits (i.e. anosognosia for aphasia). Interestingly, when language performance on comprehension and production tasks is compared with anosognosia sub-scales for related language disorders, we observed that unaware patients performed significantly worse on production than comprehension sections of the language test (see Figure 4), questioning, as seen in the discussion of the Error detection method, the role of verbal production in awareness.

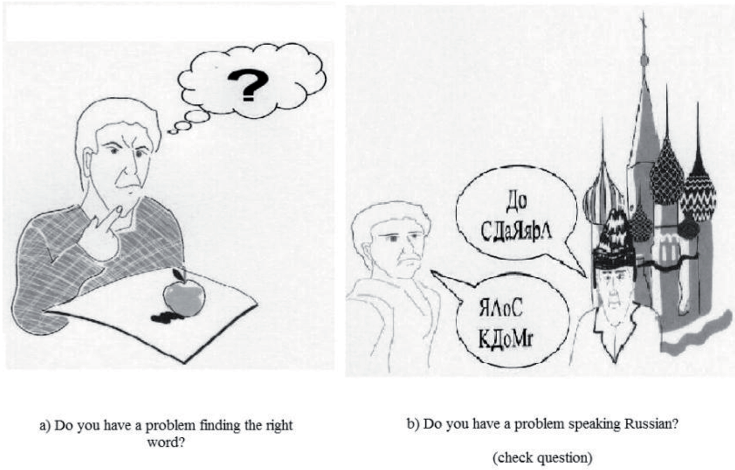


Figure 3. Examples of questions and illustrations from the VATA-L. Redrawn from Cocchini et al., 2010b

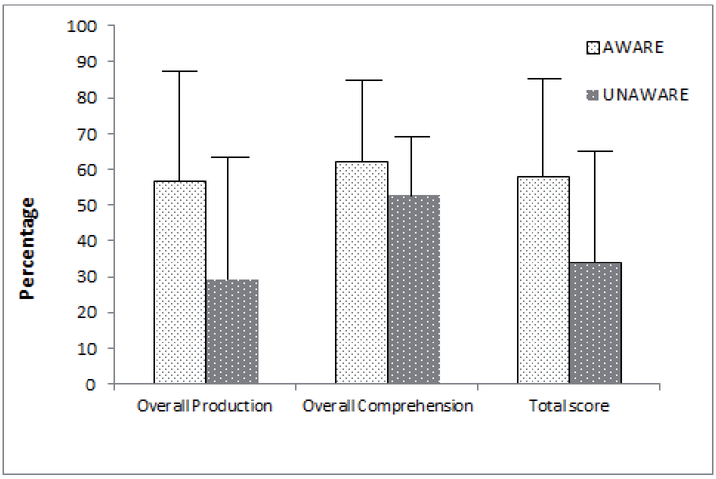


Figure 4. Performance of patients (percentage of correct responses) showing anosognosia for production and comprehension language deficits. Redrawn from Cocchini et al., 2010b

VATA-L shares the same main innovations: i) non-verbal communication is encouraged and facilitated; ii) check questions allow one to exclude potentially unreliable responses; iii) normative data allow us to extend assessment of anosognosia to patients with various degrees and types of verbal impairment.

# IMPLICIT/INDIRECT METHODS

*The self-correction method*

Some aphasics do not self-correct their speech errors. These patients may hold conversations with little, if any, attempt to repair their mistakes, so that their communication is largely incomprehensible (e.g. Alajouanine, 1956; Kinsbourne & Warrington, 1963; Weinstein et al., 1966; Levelt, 1983; Marshall et al., 1985; Maher et al., 1994; Wernicke, 1874). Self-correction mechanisms may be intentional, but they may also occur under less conscious control than, for example, error detection; for this reason we labelled this method as an "implicit/indirect method" (see Table 3).

The basic assumption of the self-correction method is that the rate of self-corrections should reflect awareness of speaking errors: if aphasics are not aware of their language errors, they will not attempt to correct them. Hence, lack of self-corrections has been considered as evidence of anosognosia for language impairments (e.g. Rubens & Garrett, 1991; Adair et al., 2003; Kertsez, 2010 for a review). However, while self-corrections clearly indicate some type of awareness of an error, lack of corrections does not necessarily imply its unawareness. Marshall and Tompkins (1982) pointed out the importance of considering as index of awareness only successful self-corrections (i.e. the speaking error is correctly amended). Therefore, the method of self-corrections has recently been focused on the types of self-correction carried out rather than assessing whether or not they occurred. In particular, recent studies have carried out detailed analyses of healthy volunteers' speech, underlining that self-corrections may occur even before production and potential errors can be "detected" and "corrected" before they reach the post-articulatory phase (Laver, 1980; Oomen et al., 2001). Accordingly, Hofmann and Cohen (1979) noticed that aphasics showed latencies preceding an error and there were self-corrections of different natures, implying different mechanisms. Schlenck et al. (1987; see also Buckingham & Kertesz, 1974; Butterworth, 1979; Keller, 1979) proposed a classification of self-corrections encompassing "repairs", defined as attempts to correct errors which just occurred, and "prepairs", which consist of various searching behaviors (such as pauses) that are not preceded by an error. This distinction is relevant, as these two types of self-correction seem to act as indexes of good functioning of post-articulatory and pre-articulatory monitoring systems, respectively. Interestingly, repairs are far less frequent than prepairs, and clearly their scoring is more complex.

# ANOSOGNOSIA FOR OTHER DEFICITS

Some of the causes of anosognosia may be deficit-specific; for example, lack of verbal comprehension may account for anosognosia for aphasia (Lebrun, 1987), personal neglect may prevent patients from becoming aware of hemiplegia (Cocchini et al., 2002) and, similarly, memory impairment could *per se* prevent patients from remembering their memory failures, resulting in a lack of awareness of their amnesia (Hannesdottir & Morris, 2007). Accordingly, we would expect to observe the phenomenon of anosognosia limited to a specific deficit and not extending to other possible associated deficits. However, findings in the literature and various theories (e.g. McGlynn & Schacher, 1989; Levine, 1990;

Weinstein, 1991; Turnbull & Solms, 2007; Marshall et al., 1998; Maher et al., 1994) suggest that common mechanisms (e.g. monitoring deficits or motivational denial) may be responsible for unawareness of various deficits. For example, outcomes of studies on anosognosia for language disorders (e.g., Marshall et al., 1998), suggested interesting overlaps with possible interpretations of anosognosia for motor impairments, and some theoretical models do not exclude shared mechanisms for awareness of different deficits (e.g. Levine, 1990; McGlynn & Schacter, 1989; Weinstein, 1991). Therefore, investigating anosognosia for all the patient's deficits would provide important insights into the deficit-specific or shared mechanisms underlying lack of awareness (Davies et al., 2005). However, anosognosia for different deficits is usually assessed with very different methods, and this has led to a partial segregation of the literature according to the deficit denied or the type of syndrome shown by the patient, making it difficult to explore anosognosia across different domains and deficits.

An important anosognosia is that for one's own **memory impairment.** This has been often investigated in patients in early stages of Alzheimer's Disease – AD (for a recent review see Kaszniak & Edmonds, 2010). The results indicate that over 40% of AD patients show anosognosia for their amnesia (Orfei et al., 2010b) and its presence is associated with decreased functional activation of medial prefrontal and anterior temporal areas (Zamboni et al., in press). Lack of awareness of memory deficits in dementia has also been sketched in cognitive models. For instance, Hannesdottir and Morris (2007; See also Agnew & Morris, 1998) distinguished between a form of anosognosia secondary to memory or executive dysfunction, and primary anosognosia, which affects the system of self-awareness directly.

Less is known about anosognosia for memory deficits in patients with focal brain lesions, and our knowledge is mainly anecdotal or derived from observations of single cases (Schacter et al., 1990). However, several authors have pointed out that lack of awareness of memory impairment is a common occurrence also in patients suffering from traumatic brain injury (TBI - Prigatano et al., 1990; Sherer et al., 1998; Bach & David, 2006; Trahan et al., 2006; Malec et al., 2007; Sherer et al., 2003; Hart et al., 2009; see also Prigatano, this issue). Tinson and Lincoln (1987) maintained that unawareness of memory problems is frequent also in stroke patients; similarly, Wilson et al. (2008) pointed out that these patients often show lack of insight. Usually, assessment of anosognosia for amnesia in focal patients is based on questionnaires consisting in a series of questions about various possible deficits, including amnesia (e.g., the Patient Competency Rating Scale - Prigatano et al., 1986; the Awareness Questionnaire – Sherer et al., 1998). These represent a valuable approach to evaluate lack of awareness across different deficits, but they are clearly limited to the most evident aspects of memory deficits.

Some authors have observed that prospective, rather than retrospective, memory difficulties may be more relevant in everyday life (Smith et al., 2000; Maylor et al., 2002; Salthouse et al., 2004) and that patients may have different degrees of awareness for different types of amnesia (Baddeley, 1990). Smith et al. (2000) devised the Prospective and Retrospective Memory Questionnaire (PRMQ), which consists of sixteen questions, eight enquiring about prospective, and eight about retrospective memory failures. Participants are asked to rate how often, from 1 (never) to 5 (very often), they would experience memory failures. Patients' ratings are then compared with proxy-ratings (i.e. ratings of caregivers evaluating the patients' memory ability) and discrepancies higher than the cut-off derived from norms are considered abnormal (Crawford et al., 2003; Crawford et al., 2006); therefore, such discrepancies can be considered as an index of anosognosia.

Using the PRMQ, we have investigated (unpublished study) awareness for memory deficits in a group of 26 (18 males and 8 females) brain damaged patients (17 stroke, 7 TBI and 2 anoxic patients), diagnosed as global amnesiacs by means of a comprehensive neuropsychological test battery. Their mean age and education were 56.23 years (sd= 15.22) and 11.53 years (sd= 4.26), respectively. We found that 20 out of 26 (77%) of these amnesic patients had a discrepancy score well above the cut-off indicated by Crawford et al. (2006), thus showing some lack of awareness of their amnesia (see Figure 5), but we did not find a significant difference in awareness between prospective and retrospective amnesia. In this sample the severity of amnesia, as assessed by means of the Rivermead Behavioural Memory Test (RBMT-3; Wilson et al., 2008) correlated positively with the degree of anosognosia ($r= .40$ ; $p<.05$). This finding is in line with one of the three types of anosognosia for amnesia identified by Agnew and Morris (1998) as *mnemonic anosognosia.* According to these authors, mnemonic anosognosia would be directly caused by amnesia *per se*, which would prevent a correct updating of the patient's semantic personal knowledge with information about memory failures. It would be therefore interesting to challenge this diagnosis by exploring whether similar patients would show anosognosia for other possible deficits and, if so, by considering other explanations that may contemplate a common mechanism, such as an impairment of the Conscious Awareness System (CAS), also identified by Hannesdottir and Morris as *primary anosognosia* (see also Agnew & Morris, 1998).

Assessment of anosognosia across different deficits with comparable measures should be encouraged. Some studies (e.g. Marcel et al., 2004) have attempted to investigate various levels of awareness for more than one deficit in the same clinical sample. In response to this necessity, we are also validating a new measure to assess anosognosia for amnesia (the VATAmem: Visual-Analogue Test for Anosognosia for memory impairment; still unpublished). The format, procedures and basic methodology of this new tool are very similar to those of the VATAm and VATA-L described above. The VATAmem consists of a series of questions enquiring about different aspects of memory (prospective and retrospective, in particular). Vignettes (see Figure 6) illustrate each question, not only to facilitate comprehension in general, but also to support memory, as wordy questions themselves can tax memory. Check questions ensure reliability and

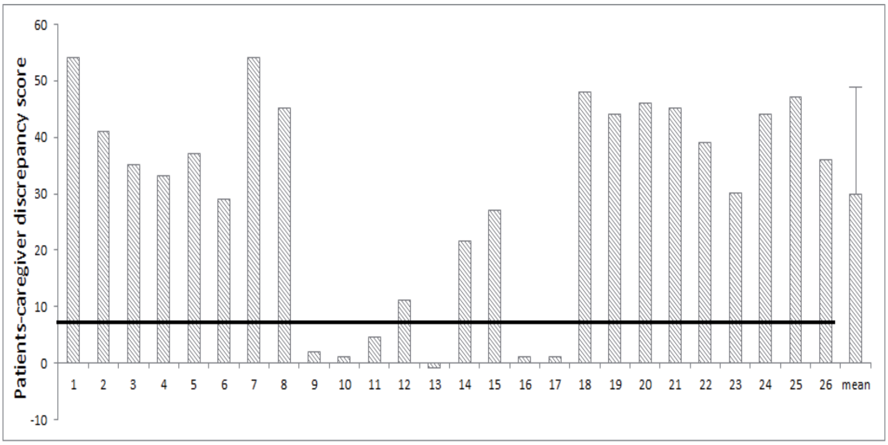


Figure 5. Patient-caregiver discrepancy values and overall mean of 26 amnesic patients. Discrepancy is calculated using true scores. Horizontal line indicates the cut-off score of 7 (Crawford et al., 2006)


Figure 6. Examples of questions and vignettes from the VATAmem test

normative data allow interpretation of possible patient-caregiver discrepancies.

These features should facilitate the assessment of anosognosia for memory impairment in large clinical populations with various degrees of amnesia. It should then be possible to reliably assess explicit awareness for three different deficits by means of very similar methods, facilitating comparison of findings and providing insight about the underlying mechanisms.

# CONCLUSIONS

The complexity of anosognosia represents an evident challenge for its assessment, which presents with various methodological hurdles (e.g. Prigatano, 2010). Lack of consensus amongst researchers and clinicians about the methods of assessment results in difficulties in comparing findings across different studies (Vuilleumier, 2000; 2004; Adair et al., 2003; Baier & Karnath, 2005; Jehkonen et al., 2006; Vallar & Ronchi, 2006; Orfei et al., 2007; Jenkinson et al., 2011), and it may have produced contrasting findings (Cocchini & Della Sala, 2010). It seems indeed likely that while similar mechanisms may underlie anosognosia for different deficits, different causes may determine anosognosia for a specific deficit. Hence, on one hand, anosognosia studies should explore different aspects and levels of awareness for a specific deficit to better characterize the possible reasons for lack of awareness. On the other hand, it is important to extend the investigation to awareness to the gamut of deficits shown by the patient.

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