In press: *Journal of Clinical and Experimental Neuropsychology*

DOI 10.1080/13803395.2020.1843603

The downsized hand in Personal Neglect

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Abstract

Objective: Personal neglect (PN) refers to a form of hemi-inattention toward the contralesional body space and it usually occurs following a right brain lesion. Recent studies suggest that PN indicates a disorder of body representation. Specifically, patients with PN show difficulties in identifying differences between left and right hands and have an altered visuospatial body map, which is associated with disrupted mental body representations. However, the metric representation of the body, and in particular the hands, has not been systematically addressed in patients showing this form of neglect.

Method: In the present study, we have investigated this representation by testing the perceived hands' width of 11 hemiplegic patients with right hemisphere cerebral lesions (5 with PN) and 12 healthy controls on a *judgment of passability* task. Patients and controls were asked to imagine inserting their hand (left and right) through a series of vertical apertures of different sizes and to judge whether their hand could fit through. Due to the heterogeneity of the data, both parametric and non-parametric approaches were used. Furthermore, additional single-case analyses were conducted by using Crawford and Howell's (1998) method.

Results: Study findings showed that patients with PN showed a significant underestimation of the left hand compared with their right hand. In contrast, whilst the right hand was equally distorted in both patients' groups, the hemiplegic patients with no evidence of PN tended to perceive the affected hand as larger than their ipsilesional one.

Conclusions: In line with the literature, our findings confirm an underlying distorted body representation following right brain damage. However, for the first time, we report both a quantitative and qualitative difference in impact of hemiplegia and PN on body representation of the contralesional body space.

Keywords: *brain damage, hemispatial neglect, neuropsychology, personal neglect, stroke, body representation*

Introduction

Unilateral spatial neglect is an acquired neuropsychological disorder that affects spatial cognition, resulting in a defective ability to be aware and pay attention to stimuli located on the contralesional side (Halligan, Fink, Marshall, Vallar, 2003; Vallar, 1998). Research on this particular disorder highlighted that neglect can be manifested with different patterns of impairment according to specific spatial frames of reference that are selectively affected (Vallar, 1998). Neuropsychological literature supports the distinction between two major sectors of space: personal space (space occupied by the body) and extrapersonal space (space surrounding the body). The term extrapersonal neglect has often been used to classify patients who show impaired performance in tasks involving the peripersonal or reaching space (e.g., cancellation tasks, copy of drawings, line bisection etc.) whereas less attention has been devoted to form of neglect in the far-extrapersonal neglect (Lindell, Jalas, Tenovuo, Brunila, Voeten & Hämäläinen, 2007). For clarity, in this study, we refer to the peripersonal form of extrapersonal neglect.

On the contrary, patients with personal neglect (PN) show a selective "deficit relative to the side of the body contralateral to the lesion" (Guariglia & Antonucci, 1992; p.1001) whereby the contralesional half of the body is less explored (Cocchini, Beschin, Jehkonen, 2001). For example, patients showing PN may not comb the left side of their hair, or dress only the ipsilesional side of their body. Despite PN and EN being frequently co-occurring, cases of selective PN (Guariglia & Antonucci, 1992; Beschin & Robertson, 1997; Peru & Pinna, 1997; Marangolo, Piccardi, Rinaldi, 2003; Ortigue, Mégevand, Perren, Landis, Blanke, 2006; Di Vita, Palermo, Piccardi, Di Tella, Propato, Guariglia, 2016; Buxbaum et al., 2004; Guariglia, Matano, Piccardi., 2014; Rousseaux, Allart, Bernati, Saj, 2015) and double dissociations (Bisiach, Perani, Vallar, Berti, 1986; Zoccolotti & Judica, 1991; Pizzamiglio et al., 1989; Vallar, Sterzi, Bottini, Cappa, & Rusconi, 1990; Beschin & Robertson, 1997; Cocchini et al., 2001; McIntosh, Brodie, Beschin, Robertson, 2000; Bowen, Gardener, Cross, Tyrrell, Graham, 2005; Committeri et al., 2007; Spaccavento, Cellamare, Falcone, Loverre, Nardulli, 2017) have also been described, supporting a differentiation between PN and EN.

In the literature, there is a growing consensus regarding the fact that PN may be ultimately related to a disrupted body representation (see Caggiano & Jehkonen, 2018; Committeri et al., 2018 for recent reviews). Studies have reported an altered visuo-spatial body map by means of the Frontal body-evocation subtest, where patients are asked to name, localize and reconstruct specific body parts (e.g., Guariglia & Antonucci, 1992; Marangolo et al., 2003; Canzano, Piccardi, Bureca, & Guariglia, 2011; Palermo et al., 2014; Di Vita et al., 2016). Typically, PN patients are unable to place body parts' tiles in the correct position, they tend to ignore body symmetry and confuse left with right sides. This pattern of errors is not observed when PN patients are asked to reconstruct a non-body object, such as a car suggesting that in PN, rather than having a more general representational deficit, the mental representation of the body is selectively damaged with respect to that of a common object (Di Vita et al., 2016). Further disruption of body representation in PN has been shown by means of the hand laterality task (Parsons, 1987a, 1987b), in which PN patients showed an impaired performance when asked to judge laterality of left hands and of objects manipulated by left hands, such as left rear-view mirrors (Baas et al., 2011). The authors explained the results as a general dysfunction in mental representations of the contralesional limb (Baas et al., 2011; Bisiach & Berti, 1995; Bisiach & Rusconi, 1990; Bisiach & Vallar, 2000). This distortion would affect also actions performed with this limb as in the case of left rear-view mirrors.

The hypothesis of a disruption of body representation in PN is also supported by neuroimaging studies. Committeri and colleagues (2007) observed that PN tends to be associated with lesions in the right inferior parietal areas and similar findings have been confirmed in more recent studies (Rousseaux et al., 2015; Baas et al., 2011), highlighting the relevance of inferior parietal lesions, in particular the supramarginal gyrus, in PN. These studies support the hypothesis that body centred tasks, which require conscious awareness of body representation, are affected by a functional disconnection between areas involved in processing somatosensory information. In particular, the post central gyrus and areas coding for extrapersonal spatial information, such as supramarginal gyrus (Galati, Committeri, Sanes, Pizzamiglio, 2001; Committeri et al., 2007; Committeri et al., 2018).

However, despite the growing body of behavioural and neuroimaging evidence showing a strong relationship between PN and body representation, it is still unclear in which way the metric (size) representation of the body would be affected.

In this study we aimed to investigate how PN may modulate the perceived metric of one's own body. Since the hand was specifically targeted in previous studies showing a relationship between its representation and neglect (Baas et al., 2011; Ronchi, Heydrich, Serino, Blanke, 2018), we focused our study on this body part. We then assessed the dimensions of the hands in patients showing PN, patients without PN and controls. To this aim, we adopted the aperture task in which participants are asked to judge whether their hand can fit ('passability') a series of different size apertures presented vertically. This task has been used in previous studies in the context of motor decision and affordances perception (Ishak, Adolph, & Lin, 2008; Warren & Whang, 1987). These studies have shown how body size (shoulder width specifically) is in fact used to scale the size of the apertures to judge 'passability' (Gordon & Rosenblum, 2004; Warren & Whang, 1987), thus providing an indirect measure of the metric representation of the body width. Here, we designed a set of apertures to judge the passability of the hands. Performance of PN patients was compared with that of hemiplegic patients not showing PN and healthy controls to tease apart the impact of hemiplegia and PN on representation of the contralesional hand.

Materials and methods

Participants

Eleven patients with right brain damage, admitted to Casa di Cura del Policlinico (Milan, Italy) and High View Care Services (London, UK), entered the study. Ten patients suffered a stroke (1 ischemic, 7 haemorrhagic, 2 ischemic with haemorrhagic infarction) and one patient was hospitalized after parietal meningioma resection. Lesion site was documented by clinical CT or MRI scans.

Patients had no history of previous neurological and psychiatric disorders and were all right-handed, as measured by a standard questionnaire (Oldfield, 1971). Demographic and neurological characteristics of the patients are summarized in Table 1.

Twelve right-handed healthy participants with no previous history of neurological or psychiatric disease were also considered for the study. Their mean age was 46.3 years (SD = ±16.3, range 29-76), and the mean education was 11 years (SD = ±3.11; range 5–18). None of them showed physical abnormalities on their upper limbs.

The project was approved by the local Ethical Committees and informed consent was obtained from all participants according with the Declaration of Helsinki (British Medical Journal, 302: 1194, 1991)

Personal and extrapersonal neglect assessments

PN was assessed by means of the Fluff test (Cocchini et al., 2001) and the Comb and Razor/Compact test (Beschin & Robertson, 1997). In the Fluff test, blindfolded patients are required to remove 24 circles attached on the patient's clothes on the contralesional arm, the torso and both legs. The cut-off score for this test is based on the stickers detached on the contralesional left side of the body (i.e.13 stickers detached out of 15; ≥ 86.7%). In the Comb and Razor/Compact Test patients are required to pretend to shave or apply make-up and to pretend to comb their hair. The number of strokes on each side of their face or head was calculated and transformed into an index of PN. Patient's performance was compared with the cut-off ≥ -0.11 (McIntosh et al., 2000). Patients were diagnosed with PN if they scored below the cut-off criteria on at least one of these two tests.

EN was assessed through a comprehensive neuropsychological battery, including letter (Diller & Weinberg, 1977), star and apple cancellation tasks (Wilson, Cockburn, & Halligan, 1987; Bickerton, Samson, Williamson & Humphreys, 2011), line bisection (Schenkenberg, Bradford & Ajax, 1980), clock drawing (Tuokko, Hadjistavropoulos, Miller & Beattie, 1992) and complex figure drawing (Gainotti, Messerli, Tissot, 1972). Patients were diagnosed with EN if they scored below the test cut-off on at least one of these tests.

Motor and proprioception assessment

Upper limb strength was assessed by means of the Motricity Index (Demeurisse, Demol, Robaye, 1980). Score can range from 1 (no movement) to 100 (normal motor power). Proprioception was evaluated by asking blindfolded patients: (i) to indicate whether their left finger, hand and arm were passively moved up or down by the experimenter (*movement*); and (ii) to align their healthy right finger, hand and arm to the same position as the correspondent left one, arranged by the experimenter in three different configurations (at the top, central, at the bottom; *position*). Furthermore, as part of the routine clinical assessment, in a sub-group of patients (CMG, VE, CA, BO, TW) cortical somatosensory evoked potentials (SEPs) were also recorded.

--- Insert Table 1 about here ---

Apertures task

Stimuli and Procedure

Stimuli consisted of a series of 29 A4-sized white pieces of cardboard with apertures. Each cardboard was cut in the middle in order to obtain a rectangular aperture that varied in height across each stimulus. Following a preliminary pilot study, the apertures' height ranged from 3 cm to 18 cm with an increasing rate of 0.5 cm while the width remained constant (15 cm). To minimise the impact of possible associated EN in the clinical sample, the apertures were presented vertically (see Figure 1). Participants sat on a chair in front of the experimenter with their hands on their laps concealed from sight for the whole duration of the task. The stimuli were presented (by the experimenter) one at a time on the participants' right side, in respect to their midsagittal plane, at approximately a distance of 50 cm. The task consisted of three ascending (starting from 3 cm of height) and three descending (starting from 18 cm of height) series. Participants were asked to imagine their hands with the palm wide open and the thumb stretched up almost perpendicular to the palm and decide whether each of their hands (the palm only) could fit into the aperture shown (see Figure 1). The motor component was removed as we were interested in the representational component and we asked for a 'judgment of passability' (e.g., Warren & Whang, 1987) to indirectly evaluate the patients' subjective hands' width. Participants were instructed to judge, each time, whether their left or right hand could fit in the aperture. During the descending series (from the largest to the smallest aperture) participants had to decide when the aperture became too narrow for their hand to pass through; during the ascending series (from the smallest to the largest aperture) participants had to decide when the aperture became large enough for their palm. The stimuli presentation continued until the participants' response changed. The examiner noted the aperture size corresponding to the last 'passability' response (i.e. the size corresponding to the last 'yes' for the descending series, and the first 'yes' for the ascending series). This value was considered as the '*Imagined Width*' for that particular trial and the next presentation begun, for the same hand, in the opposite direction. Each participant was asked to consider 6 series of apertures for each hand. The presentation order of the ascending and the descending series was counterbalanced across participants as well as the order of the hands (left and right). At the end of the task, the examiner measured the real size of the participant's palm ('*Real Width*'). The percentage of under/over-estimation of the perceived hand width was calculated with the following formula:

$$\frac{Averaged\ Imagined\ Width - Real\ Width}{Real\ Width}\ x\ 100$$

--- Insert Figure 1 about here ---

Furthermore, in order to test the body-scaled nature of the aperture judgements, we calculated the ratio between the aperture height (A) and the actual hand width (H) (A/H). Previous studies have used this method to confirm that passability judgements are based on body size scaling by checking the consistency in the ratio between participants (Gordon & Rosenblum, 2004; Warren & Whang, 1987). Because the A/H ratio expresses an intrinsic relation, the critical point should be constant across individuals of different absolute size for physically similar systems (Warren & Whang, 1987). In other words, if the A/H ratio is consistent across individuals of different hand sizes, this would suggest that participants were sensitive to the aperture in body-scaled dimensions.

Statistical analysis

Because of the heterogeneity and size of the sample considered, normality was not always met. Therefore, a non-parametric approach was used when necessary; Mann-Whitney and Kruskal-Wallis tests were used when comparing two or more independents groups, respectively. Data were also examined by estimating a Bayesian factor using the statistical package JASP (JASP Team 2017; Version 0.13.1) with the purpose of overcoming the limits of frequentist statistics (Lakens, McLatchie, Isager, Scheel, & Dienes, 2018). In Bayesian statistics, BF_{10} indicates evidence in favour of the alternative hypothesis; BFs higher than 3 are interpreted as moderate evidence, between 10 and 30 as strong evidence, and above 30 as very strong evidence (Lee & Wagenmakers, 2013). Bayesian statistic requires the same assumption of frequentist statistic therefore, this approach was used when parametric tests were carried out. Furthermore, Crawford and Howell's (1998) method was used for single-case analysis. Classically, methods for single case studies tend to inflate Type I error rate due to relatively small control samples (i.e. $N < 10$). The Crawford and Howell's (1998) method controls for Type I error rate regardless of the size of the control sample by treating the control sample statistics as statistics rather than as parameters and tests whether a patient's score is significantly below that of controls. This type of analysis has been extensively used in other studies (e.g., Della Sala et al., 2009; Cocchini et al., 2010; Palermo et al., 2014; Cocchini et al., 2018; Chapman et al., in press).

Results

Personal and extrapersonal neglect assessment

Based on patients' performance on PN tests, they were divided into two groups: patients showing personal neglect (PN+) and patients not showing personal neglect (PN-). Five patients were included in the PN+ group and six patients in the PN- group.

The PN+ group consisted of 4 females and 1 male with a mean age of 68.6 years (SD = 12.14, range 53-82), and a mean education of 10.4 years (SD = 3.71, range 5-13). Mean time elapsed since brain lesion of the 5 patients was 37 months (SD = 65.14, range 1-153); however, the group mean was skewed by one patient (TW) who was tested 153 months after the brain lesion. The PN- group included 3 females and 3 males with a mean age of 54.5 years (SD = 8.80, range 39-63) and mean education of 12.0 years (SD = 3.46, range 8-17). Mean time elapsed since brain lesion was 12 months (SD = 16.85, range 1–38). Eight patients (4 PN+ and 4 PN- patients) out of 11 showed signs of EN (see Table 2). The two groups did not show differences in terms of education [PN-: mdn =

13, PN+: mdn = 13; U = 11.5, p = .54, r =.21], onset of illness [PN-: mdn = 2, PN+: mdn = 13; U = 17, p = .79, r =.12] and age [PN-: mdn = 56, PN+: mdn = 68; U = 26, p = .13, d = .52].

The lesions' site and size were compared between the two groups. The boundaries of the lesions were drawn using the MRIcro software (Rorden & Brett, 2000) onto selected horizontal slices and mapped and into a stereotactic space (Montreal Neurological Institute; Evans, Collins, Mills, Brown, Kelly, & Peters, 1993; Collins, Neelin, Peters, & Evans, 1994).

The results of the lesion analyses are presented in Figure 2, which shows the overlapped lesion maps of 10 out of 11 patients with right brain damage (PN+ and PN- groups). Scan images were unavailable for one PN+ (MW). The PN+ group showed the maximum overlap of lesions in the right insula, rolandic operculum, pre-central and post-central gyri and occipital lobe (2 patients); in the PN- group the maximum overlap was observed in the right thalamus (3 patients).

--- Insert Figure 2 about here ---

Motor and proprioception assessment

On the Motricity Index for upper limb, the PN+ patients obtained an average score of 55.4 (SD = 30.8, range 1-77), while the PN- obtained an average score of 47.0 (SD = 26.0, range 10-77). Mann-Whitney U test showed no significant difference between groups on motor impairment [PN-: mdn = 48, PN+: mdn = 65; U = 12.5, p = .66, r =.13].

One PN+ patient and 4 PN- patients showed impaired proprioception for movement and position conditions on the clinical assessment.

All the five patients tested with evocated potentials presented altered somatosensory evoked potentials. PN- patients (CMG, VE, BO) and the PN+ patient TW showed normal median and ulnar N20 conduction time and amplitude for the right side of the body, while responses were absent on the left side. In one PN- patient (CA), responses were recorded bilaterally, normal on the right side, and altered in both latency and amplitude on the left one.

--- Insert Table 2 about here ---

Apertures task

Before running any statistical analyses, we tested our grouping criteria (based on presence/absence of PN) by correlating the patients' performance for the left hand with all quantitative demographic variables (reported in Table 1) and individual scores on the personal and extrapersonal neglect tasks (reported in Table 2). This was done to consider whether other possible factors were contributing to the aperture performance for the left hand. For this reason, we ran a series of Spearman correlations, which showed that the judgment of passability for the left hand significantly correlated with the Comb/Razor test only. Fluff test and Letter Cancellation task showed a trend (Table 3).

--- Insert Table 3 about here ---

We then used the median split approach to divide participants (both Controls and patient groups) into two major groups, *large*- (mean = 9.1 cm, SD = 1.16) and *small*-hand width (mean = 7.2 cm, SD = .33), and considered the A/H ratios for the right “healthy” hand. Results showed that the A/H ratio for the *small*-hand participants (mdn = .91) did not statistically differ from that of *large*-hand participants (mdn = .95) [U = 62.5, p = .83, r = .001]. We also considered A/H ratios for each group. The three groups judged an aperture, for right hands, shorter than their hands width to be the boundary for hand passage (Control: mdn = .93; PN-: mdn = .93; PN+: mdn = .87); Kruskal-Wallis test did not indicate a significant difference [$\chi 2(2) = 2.69$, $p = .26$, $\eta 2 = .12$]. Overall, the consistency of this ratio suggests the use of body-scaled information for judging the apertures.

In light of the preliminary analysis discussed above, the under/overestimation for both hands and each of the three groups were then considered (see Figure 3). Descriptive statistics showed that Controls and PN+ tended to underestimate both right (Controls: mean = -6.65%, SD = 8.96; PN+: mean = -17.3%, SD = 20.56) and left (Controls: mean = -5.21%, SD = 9.54; PN+: mean = -24.58%, SD = 15.29) hands, while PN- underestimated the right (mean = -13.31%, SD = 12.6) but not the left hand (mean = 1.22%, SD = 15.23).

Kruskal-Wallis test for the three groups showed a significant difference for the left hand [$\chi^2(2) = 8.24$, $p = .016$, $\eta^2 = .37$], but not for the right [$\chi^2(2) = 1.93$, $p = .38$, $\eta^2 = 09$]. Dunn-Bonferroni pairwise comparisons for the left hand highlighted a significant difference between PN+ (mdn = -17.4%) and Controls (mdn = -6.05%) (p = .033), PN+ and PN- (mdn = 2.31%) (p = .026) but not between PN- and Controls (p = 1).

--- Insert Figure 3 about here ---

We transformed the data from the passability judgment into *delta* values by calculating the difference of under/overestimation between left and right hands (i.e. left hand *minus* the right hand) to highlight the degree of discrepancy among the two hands in the three groups (Figure 4). Specifically, negative *delta* values indicate that the left hand was perceived smaller than the right, positive values that left hand was perceived larger than the right, zero indicates no difference between left and right. Data were normally distributed by performing a Shapiro-Wilk test for each group [Controls: $W(12) = .94$, $p = .51$; PN+: $W(5) = .96$, $p = .81$; PN-: $W(6) = .97$, $p = .91$], so a parametric approach was adopted. Firstly, three one-sample t-test against zero were conducted to assess the significance of the discrepancy. Results were statistically significant for PN+ (mean = -7.89%, SD = 5.23) [$t(4) = -3.37$, $p = .028$, $d = 1.51$, $BF_{10} = 3.35$] and PN- (mean = 14.52%, SD = 10.51) [$t(5) = 3.39$, $p = .020$, $d = 1.38$, $BF_{10} = 4.18$], but not for Controls (mean = 1.44%, SD = 3.29) [$t(11) = 1.51$, $p = .16$, $d = .44$, $BF_{10} = .71$]. The Bayes factor indicated that while there is moderate evidence in favour of significant difference from zero (i.e. left and right hands equally distorted) for both patient groups, the opposite was true for Controls with evidence in favour of H_0.

A one-way ANOVA was carried out to test differences between groups which resulted significant [$F(2, 7.14) = 11.18$, $p = .006$, $\eta 2 = .65$, $BF_{10} = 447.09$]. The estimated Bayes factor, to compare the fit of the data under the null hypothesis and the alternative hypothesis, confirmed the frequentist statistics indicating strong evidence of that data were more likely to occur under the alternative hypothesis. Post-hoc analyses adjusted for Games-Howell correction were conducted comparing *delta* values between groups. Results revealed that *delta* values for the PN+ group were significantly different from PN- [$t(7.58) = 4.59$, $p = .005$, $d = 2.61$, $BF_{10} = 16.81$] and Controls [$t(5.38) = 3.68$, $p = .028$, $d = 2.38$, $BF_{10} = 53.53$]. The difference between Controls and PN- was not significant [$t(5.49) = 2.98$, $p = .061$, $d = 2.02$, $BF_{10} = 30.02$] however, the effect size was large ($d = 2.02$) and BF_{10} suggested evidence of significant difference.

--- Insert Figure 4 about here ---

Finally, a Spearman's correlation between the *delta* values and the Motricity Index scores was conducted to further assess whether the discrepancy between the left and right hand might have been influenced by motor impairments. Results showed a weak non-significant correlation ($\rho = -.153$, $p = .65$) as the PN- and PN+ findings showed opposite trends.

Single case analyses

We also considered patients' individual performance on the Aperture task to explore whether specific patients showed different patterns of responses. Individual results are illustrated in Figure 5 and showed that PN+ patients tend to underestimate both hands but more the left hand with the exception of one patient (ME) who underestimated equally both hands. PN- patients showed a more heterogeneous performance in terms of under-over estimation but they tended to overestimate their left hand except for one patient (GL) who underestimated equally both hands.

To statistically investigate the difference between hands in PN+ and PN- patients, we then ran Crawford's analysis on the left hand *minus* the right hand differences using SINGLIMS_ES.exe (Crawford & Garthwaite, 2002; Crawford & Howell, 1998).

In the PN+ group, four patients out of five [TW: $t(1,12) = -2.49$, $p = .030$; TA: $t(1,12) = -4.58$, $p = .001$; PM: $t(1,12) = -2.66$, $p = .022$; MW: $t(1,12) = -3.42$, $p = .006$] showed a significant negative difference between left and right hand compared to controls. On the other hand, four patients out of six in the PN- group [CMG: $t(1,12) = 7.62$, $p < .0001$; VE: $t(1,12) = 6.43$, $p < .0001$; CA: $t(1,12) = 4.72$, $p = .001$; MG: $t(1,12) = 3.22$, $p = .008$] showed a significant positive difference between left and right hand compared to controls.

--- Insert Figure 5 about here ---

Discussion

Patients and controls were asked to imagine their hands with the palm wide open and the thumb stretched up almost perpendicular to the palm and decide whether the palm of their hands could fit into a series of vertical apertures. Results showed that healthy controls exhibited an altered representation of their hands. They equally underestimated by about 7% the width of both their left and right hands, showing no evidence of hand dominance. Previous studies have reported that even healthy adults can misjudge the metric of their own body parts (Longo & Haggard, 2010; Longo, Azañón, Haggard, 2010, Longo & Haggard, 2012; Fuentes, Longo, Haggard, 2013; Linkenaguer et al. 2015; Cocchini, Galligan, Mora, Kuhn, 2018; Caggiano & Cocchini, 2020) and the extent of the underestimation is in line with previous studies adopting a similar method of 'passability' (Warren & Whang, 1987; Ishak et al., 2008). Indeed, previous studies using apertures have found a tendency to undershoot the size of the hand in passability judgements, with participants trying to squeeze through apertures 7% smaller than their hand size (Ishak et al., 2008), and in whole body judgements, were narrower apertures were deemed as passable (Warren & Whang, 1987).

In general, the majority of the patients showed a larger error than controls when estimating both hands. This could be due to various attentional and visuo-spatial factors that may have affected the patients' ability to judge the apertures or to perform estimate tasks more generally. These factors may have affected their overall performance providing larger errors in estimating both hands.

Interestingly, PN+ and PN- groups showed a profoundly different distortion of their left hand. The analysis of *delta* values showed that while PN- patients perceived their left hand larger than their right hand, PN+ patients showed the opposite pattern of data. It is, however, of particular interest the significant difference between the PN+ and both groups (i.e. Controls and PN-). On the other hand, PN- showed a different representation of left and right hands (i.e. *delta* different from zero) but it was not significantly different from that of Controls. However, the associated large effect size, suggested a difference between the two groups. This assumption was also confirmed by the Bayesian analysis which seemed to indicate an appreciable difference. Crucially, the single-case analysis supported a significant difference of the clinical sample from the Control group (at least for the majority of the patients) highlighting the opposite pattern of misestimation of the left compared to the right for the two patient groups (see Figure 5).

The discrepancy between the representation of the left and right hands in PN+ and PN- may be due to the combination of motor impairment and PN. Indeed, a recent study conducted by Muroi and colleagues (2017) on stroke sufferers patients, indicates that motor deficits and limited availability of proprioceptive inputs from the paretic side of the body, significantly impact on the ability to effectively walk through a narrow aperture (Muroi, Hiroi, Koshiba, Suzuki, Kawaki, & Higuchi, 2017). Critically, patients with hemiplegia not only make imprecise movements, but may have difficulties in correctly estimating the affected body size. Previous studies have shown that lack of somatosensory information, as in cases of hemiplegia or anesthetisation, may lead to a perceived enlargement of the affected body part (Wallgren, 1954; Miles, 1956, Prevoznick & Eckenhoff, 1964; Melzack & Bromage, 1973; Nelles et al. 1999; Isaacson, Funderburk, Yang, 2000; Gentili Verton, Kinirons, Bonnet, 2002; Paqueron et al., 2003). Furthermore, some studies have reported that patients do have an illusion of swelling (i.e. increased width) of their entire upper or lower limb(s) following deafferentation. In these circumstances, perceived changes in body size may be explained by the removal of inhibitory background activity at different stages of the somatosensory pathway (Dykes & Lamour, 1988; Dykes & Craig, 1998), leading to a reorganization of overlapping sensory maps in the somatosensory cortex (Calford & Tweedale, 1988, 1991). Similarly, in hemiplegic patients the motor deficit associated with 'underused' limb, impact on the cortical representation of somatosensory and motor body maps, resulting in a form of maladaptive plasticity (Dohle et al., 2009).

In line with this literature, it is possible that hemiplegia may have led to a maladaptive plasticity effect, inducing a subjective 'enlargement' effect of the affected limb. As a result, PN- tended to represent their left hand larger than the right one. This aspect is important as PN+ patients also showed motor impairment and, based on this possible effect, we would expect to see a similar trend in the PN+ group, too. On the contrary, we observed the opposite result with PN+ patients, who showed a significant underestimation of the affected hands. Individual results (see Figure 5 and single case analyses) show a consistent result across the PN+ patients who judged the left hand as smaller than the right one. It seems therefore that, despite the possible 'enlargement' effect of the affected limb due to the associated motor impairment, PN has induced an opposite and larger impact on the representation of the neglected side of the body.

It should be noted that the frequency of patients showing sensory/proprioceptive deficits was larger in the PN- group compared to the PN+. This observation could suggest that PN- hand's distortion is the result of a primary motor and sensory deficit. Similarly, PN+ performance may have been affected in a similar way (i.e. overestimation of the affected hand); however, PN must have had the opposite impact (i.e. underestimation). Therefore, we believe that motor and somatosensory impairments may lead toward an overestimation of the affected hand, whereas PN may have the opposite effect (i.e. underestimation). In this case, PN- showed mainly the impact of motor and sensory deficit; whereas the PN+ group showed an underestimation of the affected hand, which was at the net of the opposite trend due to motor impairment. Furthermore, it is reasonable to argue that the hand distortion observed in both patient groups cannot be the result of a general attentional deficit (de Vignemont, 2010) or allocentric neglect of the aperture, as in this case we would have observed a similar impact of passability responses for both hands and both patient groups. Also, the results cannot be interpreted as an effect of EN components as we would have observed the same pattern of distortion in both groups. On the contrary, the findings are better explained as a primary disturbance of body representation.

Lesion analysis on the PN+ sample highlighted an involvement of the right insula, rolandic operculum, pre- and post-central gyri and occipital lobe. Recent studies have suggested that PN can be the consequence of a functional disconnection between regions that code motor and somatosensory input, such as the postcentral gyrus, and those which encode more abstract representation of the body in space (Coslett, 1998; Galati, Committeri, Sanes, & Pizzamiglio, 2001; Committeri et al., 2018). As a consequence, PN patients find difficult to create (or update) a mental model of *body image* and be aware of the configuration and motion of body in space (Galati et al., 2001). These observations, combined with evidence from recent studies (Commiteri et al. 2007; Rousseaux et al. 2015; Baas et al., 2011) seem to further strengthen the hypothesis that a deficient body representation is the major mechanism underlying PN.

To summarize, we asked a group of controls and two groups of patients to imagine whether their hands could fit into a series of vertical apertures and used their "*judgment of passability*" to estimate the perceived subjective hand size. While the controls showed some degree of underestimation similar for both left and right hands, the patients' groups showed a larger degree of distortion of the contralesional hand and in opposite directions, suggesting the presence of different underlying mechanisms. It remains unclear whether the underestimation of their hand width was mainly due to a misjudgement of the apertures or of the hand (Warren & Whang, 1987). Despite controls' data do not offer any insight on this issue, findings on the clinical sample seem to suggest that the misjudgement mainly reflects a misrepresentation of the body part as patients showed different performance for each hand. In fact, even though both groups underestimated their right hand, they showed an opposite pattern of misrepresentation for the left hand.

Limitations

The present study was an original attempt to investigate the effect of PN on subjective body size, in particular the hand. We acknowledge that the relatively small group size and demographic factors, such as differences on age, education and lesion size, might have contributed to the different performances among groups; hence, results should be taken with caution. However, were these factors pivotal in the passability task, we would have expected to observe a different performance also for the ipsilesional hand. The correlation analyses confirmed that the presence/absence of PN was the variable that better explained the difference between hands on the task. In detail, considering the right (healthy) hand, both patients' groups performed similarly; however considering the left hand, the PN- group showed a trend in the opposite direction than controls, whereas the PN+ group showed underestimation in the same direction than controls, though more pronounced.

The single-case analyses allowed to minimize the risks of Type I error which could not be fully controlled with a non-parametric approach. Nonetheless, the results seem to be in line with the most recent literature on PN. They open a new window for possible future studies on body metrics and size in this particular clinical samples; it would indeed be desirable to investigate further this aspect considering larger patients' samples. Also, further studies may want to analyse the responses considering separately responses for ascending and descending order to explore whether clinical samples may show a qualitatively different impact (e.g. Gardner & Boice, 2004; Gardner & Bokenkamp, 1996).

Acknowledgment

We are grateful to William Amarteifio for proofreading the manuscript.

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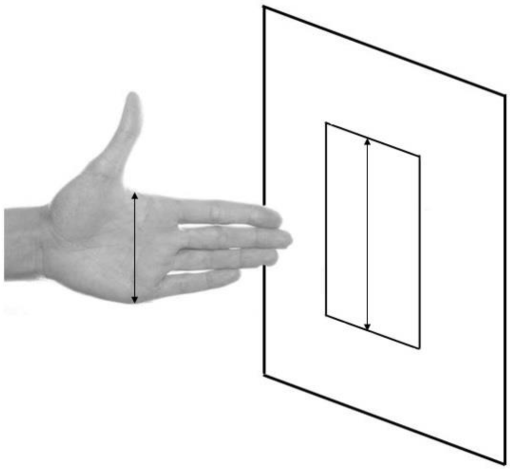

Figure 1. Schematic representation of the apertures task. Participants were asked to imagine whether the palm of their hand, indicated by the arrows, could fit into a series of vertical

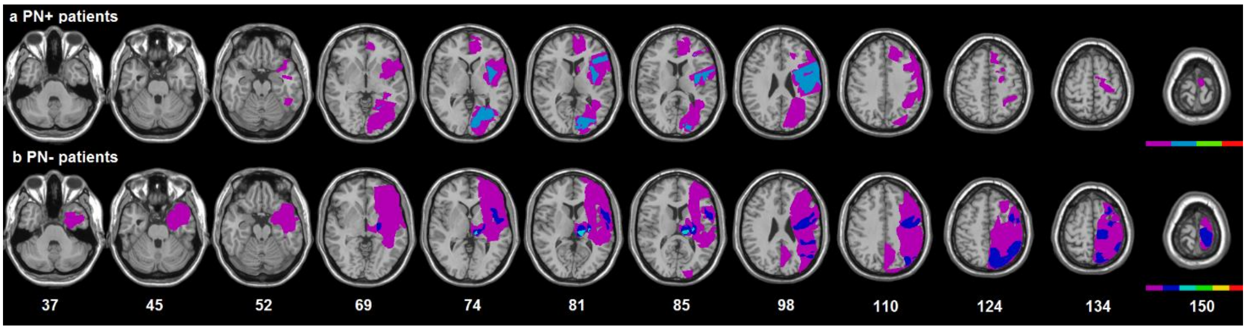

Figure 2. Superimposition of the right-hemispheric lesions in 4 right-brain-damaged PN+ patients (a) and in 6 right-brain-damaged PN- patients (b). Scan images were unavailable for PN+ patient MW. Montreal Neurological Institute (MNI) Z-coordinates for the shown axial slices are given. The number of overlapping lesions is indicated by different colours, coding increasing frequencies from violet to red.

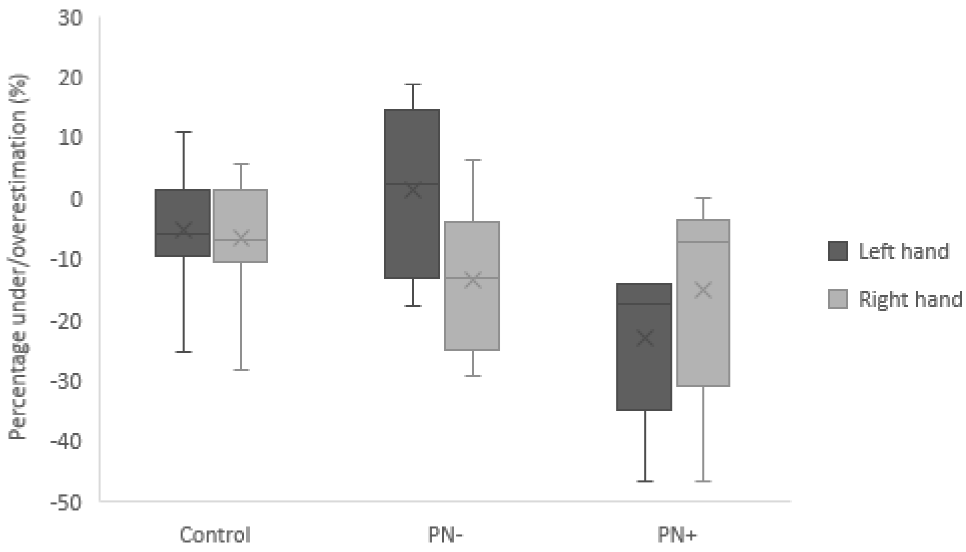

Figure 3. Graphic representation of both left and right hands distortion for the three groups. The middle line of the boxes represents the median, the x in the box represents the mean. The bottom line of the boxes represents the lower quartile, the top line the upper quartile. The vertical lines indicate the upper and lower extremes of each data set. * Significant difference between groups.

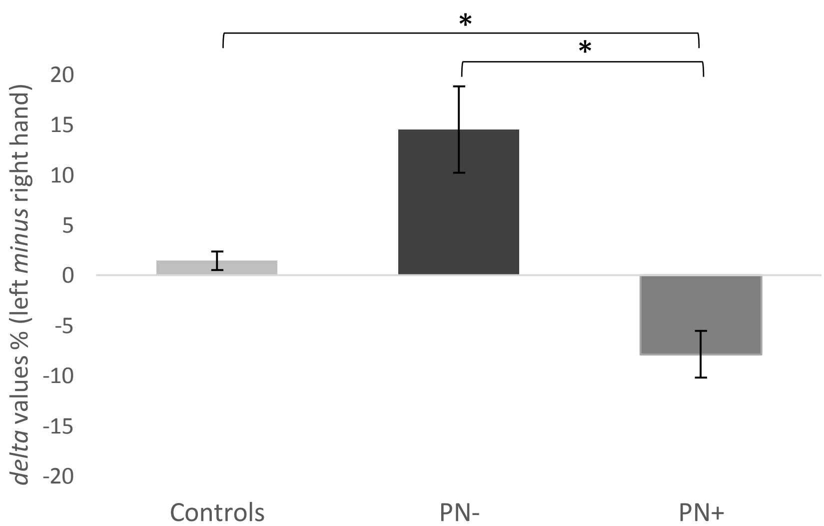

Figure 4. Percentage difference (±SE) between left and right hands in the two patients' groups.

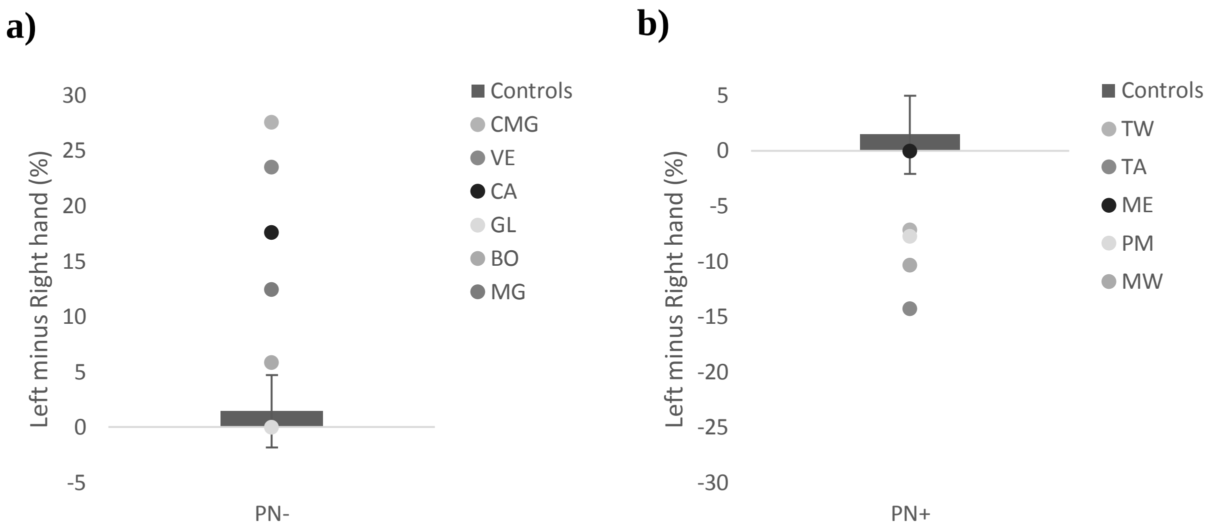

Figure 5. Individual left/right hands differences for a) PN- group and b) PN+ group.

Table 1. Demographic and neurological information of 11 right brain-damaged patients.

	Gender/Age/ Education	Oldfield	Duration of disease (months)	Etiology/ Lesion site	Lesion volume (cc)	Motricity Index (upper extremity)	Neurological examination			Proprioception	
							M	*SS*	*V*	*Movement*	*Position*
TW	F/61/13	.89	153	I/F P T ins	53.0	65/100	+	-	-	-	-
TA	F/79/5	.89	1	IH/F T O	73.5	1/100	++	-	++	-	-
ME	F/82/8	.68	1	H/P O	45.4	77/100	+	-	++	-	-
PM	F/68/13	1	19	H/F T P	59.4	64/100	+	ext	ext	+	++
MW	M/53/13	1	13	H/P T	na	70/100	-	-	+	-	-
CMG	F/51/13	1	3	IH/F P	167.3	26/100	++	ext	ext	-	+
VE	M/39/13	.95	30	H/F T P Ins Bg	266.2	10/100	++	+	++	++	++
CA	M/57/17	1	1	H/t	8.1	48/100	+	ext	ext	+	+
GL	F/63/13	1	1	H/t ic	3.4	73/100	+	ext	-	-	-
BO	M/55/8	.95	38	H/t ic	2.4	48/100	+	-	-	+	+
MG	F/62/8	.79	1	N/P	11.4	77/100	+	-	-	-	-

M/F: male/female; Age and formal education are expressed in years.
I/H/N: ischemic/haemorrhagic/neoplastic lesion. F: frontal; P: parietal; T: temporal; O: occipital; Ins: insula; ic: internal capsule; Bg: basal ganglia; t: thalamus.
Neurological examination: M/SS/V, motor/somatosensory/visual half-field deficit contralateral to the damaged hemisphere; ext: extinction to double simultaneous stimulation (for visual and somatosensory deficit). +/++, deficit; -, no deficit.
na: not available for mapping

Table 2. Assessment for personal and extrapersonal neglect.

	Comb and Razor/ Compact Test	Fluff test	Letter cancellation		Star cancellation		Line bisection		Apple task	Complex Figure Drawing	Clock Drawing
PN+ patients	*%bias*		*L*	*R*	*L*	*R*	*80mm*	*160mm*	*Asymmetry score for allocentric neglect*		
TW	-0.21*	100	53/53	51/51	30/30	26/26	-3	5,2	0	0/5	0/12
TA	-0.43*	73.3*	29/53*	49/51	22/30*	26/26	-9.5*	7.3*	-1	1.5/5*	6/12*
ME	-0.20*	93.3	44/53*	51/51	20/30*	24/26	4.8	32.6*	50*	1.5/5*	1/12*
PM	-0.38*	86.7	46/53*	49/51	18/30*	21/26	-7.0	1.3	3*	2.5/5*	3/12*
MW	-0.21*	73.3*	0/53*	36/51	0/30*	21/26	na	na	na	na	0/12
PN- patients											
CMG	0.04	100	53/53	46/51	25/30*	26/26	4.5	5.3	0	0.5/5*	0/12
VE	0.06	93.3	53/53	51/51	27/30*	25/26	-9*	-1.2	0	0.5/5*	0/12
CA	-0.02	100	52/53	49/51	26/30	23/26	4	7.1*	0	0/5	0/12
GL	-0.08	93.3	53/53	51/51	28/30*	26/26	2.5	4.6	0	0.5/5*	0/12
BO	-0.06	86.7	53/53	51/51	30/30	26/26	3.3	0.5	0	0/5	0/12
MG	0.01	100	53/53	51/51	30/30	26/26	0	0.2	1	0/5	0/12

* Defective performance, as compared with normative data.
Cancellation tasks: number of correct detections in the left/right (L/R) hand-side of the display.
Line bisection: percentage deviation (-/+leftward/rightward deviation) for lines of different length: 80 and 160 mm; complex figure and clock drawing: 0/5 and 0/12 indicate errorless performances, respectively.

Table 3. Correlation between LH performance and variables

Variables	Spearman correlation (n =11)	
	ρ coefficient	p value
Age	- .456	.159
Education	.203	.549
Duration of disease	-.220	.516
Lesion volume	.169	.620
Motricity Index	-.156	.648
Hand Laterality	.101	.768
Comb and Razor/Compact test	**.683**	**.021***
Fluff Test	.587	.058
Letter Cancellation	.599	.052
Star Cancellation	.469	.146
Line bisection 80mm[a]	-.292	.413
Line bisection 160mm[a]	-.377	.283
Apple task[a]	-.291	.226
Complex figure drawing[a]	-.421	.226
Clock drawing	-.491	.125

[a] data from 1 patient were not available (n =10)

* indicates significant correlation