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The flubbed body: pathological body size representation in personal neglect

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ABSTRACT

Personal Neglect (PN) is a disorder in which patients fail to attend or explore the contralateral side of their body. An increasing number of studies have considered PN as a form of body representation disorder frequently observed following damage to parietal areas. The extent and the direction of the body misrepresentation is still unclear with recent studies suggesting a general reduction of contralesional hand size. However, little is known about the specificity of this representation and whether the misrepresentation also generalises to other body parts. We explored the features of the representation of the hands and face in a group of 9 right brain damaged patients with (PN+) and without PN (PN-), when compared to a healthy control group. For this, we used a body size estimation task with pictures, in which patients were required to choose the one that most closely matched the perceived size of their body part. We found that PN+ patients showed a labile body representation for both hands and face, having a larger distorted representational range. Interestingly, in comparison with PN+ patients and healthy controls, PN- patients also showed misrepresentation of the left contralesional hand which could be related to impaired motor performance of their upper limb. Our findings are discussed within a theoretical framework suggesting a reliance on multisensory integration (body representation, ownership, and motor influences) for an ordered representation of the size of the body.

Keywords: personal neglect, body representation, size distortions, hand size, face size, body size

1. INTRODUCTION

Personal Neglect (PN) is a disorder characterised by lack of awareness for and exploration of the contralesional side of the body, not explained by possible associated motor or sensory problems (Baas et al., 2011; Bisiach & Vallar, 2000; Heilman, Valenstein, & Watson, 2000). It is considered as an egocentric disorder in which the personal body space is affected (Caggiano & Jehkonen, 2018; Committeri, Piervincenzi, & Pizzamiglio, 2018; Kerkhoff, 2001) impairing patients' ability to interact with their own bodies and it is associated with longer recovery and poorer outcomes (Buxbaum et al., 2004; Chen-Sea, 2000; Iosa, Guariglia, Matano, Paolucci, & Pizzamiglio, 2016). It is usually caused by injury of the right hemisphere to inferior parietal areas (Bisiach, Perani, Vallar, & Berti, 1986; Heilman et al., 2000); temporo-parietal junction (Baas et al., 2011); the postcentral and supramarginal gyri in the parietal lobe (Committeri et al., 2007; Rousseaux, Allart, Bernati, & Saj, 2015), and white matter connections to fronto-parietal areas, causing a "within-parietal disconnection" (Committeri et al., 2018, p. 274, 2007). When recurrent associated clinical variables have been taken into account (e.g., motor impairment and extrapersonal neglect), lesions associated to PN suggest a convergence of various factors involving a medial network (Bertagnoli et al., 2022) and a multi-factorial syndrome.

Efforts have been made to understand the nature of this complex disorder. The current predominant view is that a defective contralesional body representation underlies it (Cocchini, Beschin, & Jehkonen, 2001; Coslett, 1998; Guariglia & Antonucci, 1992), intertangled with hemispatial inattention (Committeri et al., 2018). Thus, research has focused in understanding the characteristics of the representation of the body in PN patients. However, this is not a simple task as multiple 'bodies in the brain' or representations have been proposed (Berlucchi & Aglioti, 2010), which means several classification systems have been suggested. Dyadic models were one of the first classification systems on body representations and postulate the existence of two separable representations: one relating to the posture of the body, or 'body schema', and the other to the localisation of body parts, which was consequently called 'body image' (Critchley, 1979; Head & Holmes, 1911). Specifically, the body schema is a representation formed by afferent and efferent sensory and motor information that guides actions, whereas body image is a pictorial depiction of the body (Gallagher, 1986, 2005; Paillard, 1999; Rossetti, Rode, & Boisson, 1995). In reality, body image in this taxonomy accommodates all other representations that are not used for action, such as body affect, concept or percept (Gallagher, 2005), having a size estimation component (perceptual distortion, linked to body percept), and a cognitive-evaluative component (affective/emotional, linked to body concept) (Skrzypek, Wehmeier, & Remschmidt, 2001; Slade & Brodie, 1994). Due to the heterogeneity of the components of the body image in dyadic taxonomies, other authors postulated a further subdivision, becoming the triadic taxonomy (Schwoebel & Coslett, 2005). Within this framework, the body schema was still considered with the same definition, whereas the body image was divided into two: the 'visuospatial body map' or 'body structural description', which is a topological description of the map of the body (Buxbaum & Coslett, 2001; Sirigu, Grafman, Bressler, & Sunderland, 1991), and the 'body semantics', which includes conceptual and linguistic components of the body representation (Di Vita, Boccia, Palermo, & Guariglia, 2016; Schwoebel & Coslett, 2005). The visuo-spatial body map incorporates knowledge regarding the structure of the body (i.e., where body parts are located, and their boundaries), whilst the semantics of the body is involved with the conceptual and linguistic knowledge of the body. Further models have proposed supplementary disintegration of body representation in other modules (independence models); others have proposed a single unified representation (fusion models), whereas new recent proposals focus on the construction of body representation through interactions between the different components (co-construction model) (Pitron, Alsmith, & de Vignemont, 2018; Pitron & de Vignemont, 2017). However, there is currently no final agreement on how the body is represented in the brain, and more so, how many representations there are. Consequently, several studies have tried to disentangle the different components of the multidimensional body representation that are affected in PN. In our study we refer to a more general distortion of body representation mainly overlapping with the 'visuospatial body map', mentioned above, of the body image.

In one of the first studies exploring the characteristics of the body representation in PN, authors found that an impaired mental body representation caused constructional problems of the body and face, at the same time as impaired localisation of body parts on the left side of the patient's body (Guariglia & Antonucci, 1992). Coslett (1998) described PN as a selective impairment of body schema. Further, Baas et al. (2011) pinpointed body representation as the critical mechanisms of PN, as right-brain damaged patients with PN showed specific difficulties in processing stimuli representing left hands (Johnson, Sprehn, & Saykin, 2002). Likewise, other authors claimed that the impaired visuo-spatial mental representation in PN is due to an inability to construct a coherent body (Di Vita, Palermo, Boccia, & Guariglia, 2019; Palermo, Di Vita, Piccardi, Traballesi, & Guariglia, 2014). However, the understanding of how these patients represent the size of their own bodies is still limited despite the involvement of parietal areas in PN (e.g., Committeri et al., 2018, 2007), which are thought to be responsible for the metric representation of the body (e.g., Nico et al., 2010; Spitoni et al., 2013).

Thus far, only one study has attempted to understand the way patients with PN represent their own hands, by measuring their body schema with a 'judgement of passability' task in which they had to judge if their hand could fit through different sized apertures. Results showed that right-brain damaged patients with PN judged their contralesional hand as smaller, thinking their hand could fit through apertures that were too small (Caggiano et al., 2020). These results helped shedding some light into how the body is represented in PN, but further can be learnt by exploring a different aspect of body representation, to consider its multidimensionality (Committeri et al., 2018). Moreover, it seems crucial to investigate whether PN can induce a characteristic distortion linked to hands or whether it is associated to a more general blurred or imprecise body representation. A good candidate for this exploration would be the face, a body part that it is widely used to diagnose this disorder (e.g., in the Comb and Razor/Compact test).

Consequently, the main aim of this study is to explore the perceptual size estimation of both hands and face in patients with and without PN. For this, we used a depictive task (Azañón et al., 2016; Mölbert et al., 2017). We presented pictures of their hands and face distorted for them to identify the one that most closely matched the correct size of the body. The visuospatial transformation between one's own body and the presented image will require the activation of the parietal cortex (Peltz, Seifert, Lanz, Müller, & Maihöfner, 2011), an area that is part of a network damaged in PN (Bertagnoli et al., 2022; Committeri et al., 2018). Thus, it was hypothesized that patients with PN (PN+) will show a more distorted representation of the body in comparison with a control group of healthy participants and patients without PN (PN-), as PN patients may be unable to retrieve the mental representation of their own body (Mohr et al., 2010; Spitoni et al., 2013).

2. METHOD

2.1 Participants

A group of 9 right brain-damaged patients (7 males and 2 females) was recruited from 'Centro Referencia Estatal de Atención al Daño Cerebral' (CEADAC), in Madrid, Spain. All patients suffered from a first brain injury and had a right unilateral stroke (2 ischemic, 5 haemorrhagic and 2 ischemic with haemorrhagic infarction). Brain scans were not available for all patients and information about lesion site and nature was obtained from radiological and medical records. The mean time from injury onset was 256.33 days (SD = 91.5) and all had been in intensive rehabilitation for an average of 111.89 days (SD = 59.05). The exclusion criteria for this group were: history of neurological or psychiatric disease, substance abuse, previous cerebrovascular accident (CVA), neoplastic aetiology and inability to provide informed consent or perform the experimental tasks. They were all right handed except one, who was left handed (P04), as measured by the Oldfield Questionnaire (R. C. Oldfield, 1971). Demographic and clinical information is presented in Table 1.

	Gender (0= male)	**Age (years)**	**Education (years)**	**Aetiology**	**Lesion site**	**Time from injury (days)**	**Time at CEADAC (days)**
P01	0	44	8	I, H	T, P, O	326	29
P06	0	54	14	H	bg, t. ins	239	120
P07	1	39	8	H	//	423	217
P08	0	52	10	H	bg, t. ins	283	149
P09	0	40	8	H	bg, t,	150	73
P02	0	38	14	I	bg, t, ins, P, T	144	65
P03	0	53	8	H	O, c	276	168
P04	1	39	10	I, H	O, bg,	296	115
P05	0	53	8	I	ic, bg	170	71

Note: I/H: ischemic/haemorrhagic lesion.
Lesion site: F = frontal; P = parietal; T = temporal; O = occipital; ins = insula; ic = internal capsule; bg = basal ganglia; t = thalamus; c = cerebellum; ic = internal capsule; // = neuroradiological examination not available.

Table 1. Demographic and clinical information of the 9 right-brain injured patients.

Shaded rows indicate performance of patients showing PN (see later Results section 3.1).

G* Power 3.1 was used for a power analysis to determine the required sample size of the control group (Faul, Erdfelder, Buchner, & Lang, 2009), once the patient group was recruited. A power analysis based on a mixed-model ANOVA with 3 groups and 3 measurements was run, with an estimated large effect size of 0.8. Alpha was set at 0.05 and power of 0.8. The adequate sample size obtained was 9. The sample size of the patients' group was fixed due to circumstances (no more patients available at the time). Hence, we decided to have a larger sample size for the control group as research has shown this can increase power (Guo & Luh, 2013; M. Oldfield, 2016; Tichy & Chytry, 2006). We recruited a group of 16 right-handed healthy participants (mean age = 38.81 years, SD = 11.71; mean education = 10.31 years, SD = 2.12). This group was matched with the patients' group in Age [t (23) = -1.66, p = .11], Gender [t (23) = 2.01, p = .06] and Education [t (23) = .56, p = .58]. The individuals in the control group did not have any neurological or psychiatric impairments.

The study was approved by Goldsmiths Research Committee and CEADAC in line with the principles of the Helsinki Declaration (1964) and its later amendments. All participants provided informed consent and participated willingly in the study with their family support. They could withdraw at any time without any further justification.

2.2 Personal and Extrapersonal Neglect examination

The presence of PN was assessed by three different tests to account for its multidimensionality (Committeri et al., 2018; Guariglia & Antonucci, 1992). These were the Comb and Razor/Compact test (Beschin & Robertson, 1997; McIntosh, Brodie, Beschin, & Robertson, 2000), the Fluff test (Cocchini & Beschin, 2022; Cocchini, Beschin, & Jehkonen, 2001) and the One Item test (Bisiach et al., 1986).

The Comb and Razor/Compact test is a semi-structured test in which patients are required to perform actions on their own body by using common objects (Beschin & Robertson, 1997). Patients were provided a comb and a razor/compact powder case (male/females) and were asked to use each object for 30 seconds. The experimenter counted the number of strokes the patient performed on each side of the head/face, and in the middle. The bias index proposed by (McIntosh et al., 2000) was used to identify patients that will show PN (cut-off score of +11 for left PN, and -11 for right PN).

The Fluff test was also used to assess PN (Cocchini & Beschin, 2022; Cocchini et al., 2001). The test consists of 24 identical circles (2 cm in diameter), which were made of Velcro. The circles were attached on the patients' clothes, at specific locations (6 stickers on left arm, 6 on the trunk, 6 on the right leg and 6 on the left leg). Patients were blindfolded and were sat down for the whole duration of the task. Experimenter positioned all the stickers carefully to avoid tactile feedback whilst keeping patients distracted in conversation. Patients were required to remove all stickers from their body by using the right hand. The task finished when patients declared they had located all stickers. According to a new scoring system (Cocchini & Beschin, 2022), a spatial bias (i.e. the difference between the percentage of targets detached from each side) higher than 13.3 indicates ipsilesional PN and a spatial bias lower than -13.3 indicates contralesional PN. At ceiling performance or a spatial bias within the cut-offs reported above is considered as normal performance.

PN was further assessed by using the One Item test (Bisiach et al., 1986). In this test, patients are requested to touch their left hand using their right. Specifically, both hands are lying on the table, and the experimenter points to the right hand, and instructs: 'with this hand, touch your other hand'. There are four different scores for this task: 0 indicates no difficulties; 1 indicates slight difficulties (hesitation and search); 2 is awarded for interrupted search (before target is reached); and 3 indicates lack of movement towards the target hand.

Pathological performance on at least one of the three tests was considered as evidence of personal neglect. According to patient's performance on PN tests they were assigned to PN+ or PN- groups.

Extrapersonal Neglect (EN) was assessed by means of the Behavioural Inattention Test (BIT), a widely-used test to assess visual neglect (Wilson, Cockburn, & Haligan, 1987). The conventional subtests were administered, which are: line crossing, letter cancellation, star cancellation, figure and shape copying, line bisection, and representational drawing. Patients who scored below the total aggregated cut-off score of 129 out of 146 were classed as having EN.

2.3 General neuropsychological assessment

To explore possible cognitive differences between PN+ and PN- group, all patients performed a series of tests evaluating various cognitive functions.

The Digit span test was administered to measure short-term memory. In this test, a list of numbers is read aloud, and participants are required to recall it, either in direct order (forwards) or reverse (backwards). The testing stops once the participant cannot recall a full list or reaches the maximum list length (starting from 2 digits up to 9 in forward condition, and up to 8 in backwards condition). Two trials for each span were administered; hence, there were a total of 16 trials for direct presentation, and 14 in reverse. The highest number of digits recalled (span) for each presentation order was recorded. The average span for Spanish population is 6±1 digits in direct order, whereas this was 5±2 in reverse order (Tamayo et al., 2012). A span of 4 or less was considered pathological for direct order, and 3 or less in the reverse order.

Verbal learning and memory was assessed by the Hopkins Verbal Learning Test revised (HVLT-R) (Benedict, Schretlen, Groninger, & Brandt, 1998). It includes three learning trials, in which patients are read 12 words from a list and are asked to remember as many words as possible, in any order. The sequence of words remembered is recorded. Patients are told they may be asked the list at a later stage. After 20 minutes, they are asked to recall the list again. Lastly, a list of 24 words is presented, that includes the 12 target words from the previous list plus 12 nontarget words (6 are semantically related to targets). Patients must report which words were present in the previous list, and false and true answers are recorded. The total recall score is calculated as the total number of correct words remembered in the first three trials (maximum score is 36). The delayed recall is the total number of words remembered in trial 4 (maximum score of 12) (Cherner et al., 2007). The cut off for the total recall to detect memory impairment in Spanish population is < 13, whereas it is < 4 for the delayed recall (González-Palau et al., 2013).

Semantic fluency was also assessed. In this test, patients are asked to generate as many words as possible pertaining to the semantic category of animals, within 1 minute (Benton, 1968). Only correct answers are recorded, whilst perseverations (repeated words) or intrusions (words from another category) are not considered. Normative values for Spanish speakers were considered, with a cut-off score of 12.9 words (Rosselli et al., 2002).

The Controlled Oral Word Association (COWA) test was used to assess phonemic fluency (Barry, Bates, & Labouvie, 2008; Strauss, Sherman, & Spreen, 1998), in which patients were required to produce as many words as they could beginning by each letter (F, A, and S) in one minute. Proper names and repetitions are not scored. The total score is the total number of words produced for the three letters (Strauss et al., 1998). Normative scores in a Spanish speaking sample determined a cut-off of 7.6 words for F category, 7.2 in A category, and 7.6 for S category (Rosselli et al., 2002). Scores under normative performance were considered pathological.

The Wisconsin Card Sorting task (WCST) was used to measure executive functioning (Bowden et al., 1998). Patients were asked to classify 60 cards according to different criteria: colour of the symbols (red, yellow, blue, and green), their shape (stars, crosses, triangles and circles), or the number of shapes on each card (1 to 4). The rule for the classification changes every 10 cards. The task measures how people adapt to the change of rules. The number of correct matches, errors (perseverative and non-perseverative) and categories completed are recorded. The total number of errors and perseverative errors are used in the formula following Nelson (1976) to calculate the final score [(perseverative errors/ total errors) x100]. The cut off score is 50.

The Galveston Orientation and Amnesia Test (GOAT) was administered to measure orientation to person, place and time, and memory for events preceding and following the injury. Thus, this test assesses post-traumatic amnesia (PTA) and retrograde amnesia (RA) after severe brain injury (Levin, O'Donnell, & Grossman, 1979). It includes 10 items that are verbally asked to patients (e.g., what is your name?). The number of errors in each question is recorded and subtracted from the total score (maximum score = 100 points). Scores lower than 66 indicate impaired performance; scores between 66-75 indicate borderline performance, whilst scores over 75 indicate normal performance.

The Awareness of Deficit Scale is a semi-structured scale developed to measure the level of awareness of deficit for a group of patients with acquired brain injury (Villalobos, Bilbao, Espejo, & García-Pacios, 2018). The scale considers three main areas of awareness: awareness of injury, awareness of deficit and awareness of disability. The level of awareness in each area is measured, with a range 0-6 for the awareness of injury, 0-12 for the awareness of deficit, and 0-12 for awareness of disability. The total maximum score is 30.

2.4 Motor and functional performance assessment

Patients' upper and lower extremity functioning was assessed for contralesional and ipsilesional limbs via the Motricity Index questionnaire (Demeurisse, Demol, & Robaye, 1980). This is a simple test of motor function that allows quick, valid and reliable assessment (Collin & Wade, 1990). Patients were required to perform three different tasks: pinch grip, in which they had to try to grip a 2.5cm tube using their thumb and index fingers; elbow flexion, in which patients were required to flex the elbow in 90º, and try to touch their shoulder with the hand whilst experimenter opposes some resistance at the wrist; and shoulder abduction, in which the elbow is flexed and placed against the chest, and patient is required to abduct the arm. Scores for the pinch grip are between 0 (no movement) to 33 (normal pinch grip). For elbow flexion and shoulder abduction, scores go between 0 (no movement) to 33 (normal power). The total score is calculated by adding up all scores +1 and it ranges from 1 (no movement) to 100 (normal power) (Collin & Wade, 1990).

The Barthel Index (BI) was administered to measure functional performance in activities of daily living (Mahoney & Barthel, 1965). Ten different items are scored based on the ability of the patient to perform the activity. A score of 0 is given if the patient cannot perform the activities as described in the criteria. Other scores are provided for different areas, such as continence, dressing or feeding. A score of 100 indicates independence in all the areas. Most studies consider a cut-off score of 60/61 (moderate dependency) (Shah, Vanclay, & Cooper, 1989).

Functional performance was also evaluated via the Functional Independence Measure (FIM) (Keith, Granger, Hamilton, & Sherwin, 1987). This scale consists of 18 different items that measure the level of independence in different areas, with an ordinal scale (1 = total assist and 7 = complete independence). Scores range between a minimum of 18 to a maximum of 126, any score below 6 in any given item would indicate supervision or assistance. Hence, a total score under 90 will be a sign of dependency.

2.5 Body size estimation task

The body size estimation task has been inspired by tasks in previous research (Gandevia & Phegan, 1999; Longo & Haggard, 2012; Mohr et al., 2010; Türker, Yeo, & Gandevia, 2005), where participants are presented with distorted pictures of body parts and asked to assess which one would subjectively match their perceived body size (Gardner & Boice, 2004; Kammers, Longo, Tsakiris, Chris Dijkerman, & Haggard, 2009; Miall, Afanasyeva, Cole, & Mason, 2021). Image distortion tasks are thought to measure the cognitive component of the body image (Slade & Brodie, 1994). In this study, single body parts were presented to avoid comparative judgements (Fuentes, Longo, & Haggard, 2013). Moreover, real sized pictures were used as results are susceptible to less distortion due to procedural confounds (Holder & Keates, 2006).

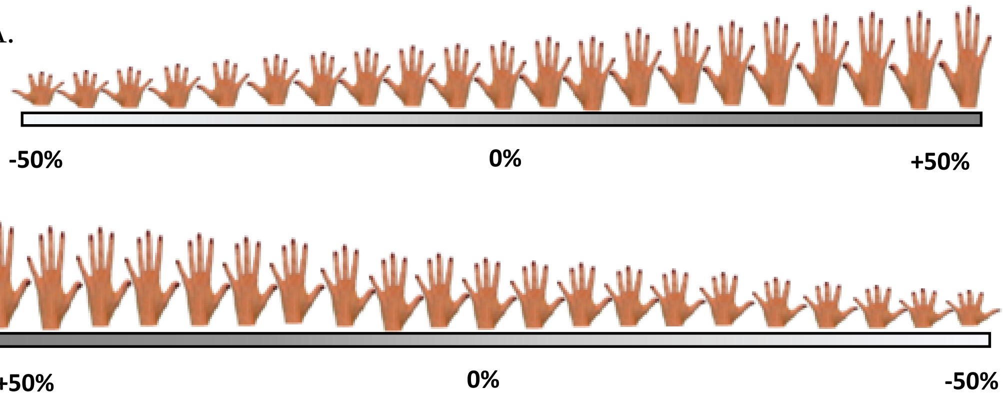
2.5.1 Stimuli

Real pictures of each participant's face and right hand (dorsal and palmar views) were taken with a Nikon 3200D camera, all at the same distance and position. By using Paint S (version 5.6.9), the background was removed from the pictures to make it standard white. Adjacent body areas (i.e., wrist for arm or neck for face) were also removed from the pictures in order to prevent providing any cues (Gardner & Boice, 2004). The mirrored image of the right hand was used as stimuli for the left hand, as patients could not open their hand to take a picture due to motor problems (e.g., hemiplegia). Thus, the same was done for healthy controls. The face image was also mirrored to present it in typical view, as seen when reflected (D'Amour & Harris, 2017). The images were then resized, for width and length (one dimension at a time), by using a bespoke-made programme (Borland C^{++} Builder, 2007). Size increases and decrements were of 5% to ensure these were not obvious and were symmetrical from the midline of each body part. The minimum size decrement was of 50% (smallest picture), and the maximum increment was of 150% (largest picture). There was a total of 21 pictures per each body part (face, right hand, and left hand) and hand view (dorsal/palmar), that is a total of 105 images where only one per body part was shown in the correct real size (100% size) (see Figure 1A).

2.5.2 Experimental procedure

Participants sat in front of a wall with a white screen, half a meter away from it, where the pictures were presented using LCD video projector (full HD, 1080 pixels, 2400 lumens) connected to a Microsoft Windows laptop. The projector was at 1.8 metres from the wall and was positioned on a table behind the participant (1 metre of height) (see Figure 1B). Images were initially adjusted in size, in such a way that the 100% picture (no distortion) matched the real size of the participant's body part when projected onto the wall. For this, a tape measure was used to size the real and projected body parts.

A.



B.

Figure 1. Depiction of hand stimuli and experimental setting.

Hand pictures for the right hand (ascending order) and left hand (descending order) with 5% size changes intervals for length (A), and pictorial representation of the experimental setting (B).

The images were projected in the right hemispace from participants' body midline, to avoid potential difficulties due to EN. Pictures were presented in ascending (from small to big) and descending (from big to small) order, one picture at a time, repeated in two rounds for each order. Presentation was counterbalanced for order (ascending and descending); dimensions (length and width); hand view (palmar dorsal), and body part (face, right hand, left hand), with a total of 8 trials for the face, and 12 trials for each hand. This method was used as other type of procedures, such as constant stimuli, require a large number of trials to ascertain the point of subjective equality (PSE) as a measure of body size estimation, which could be cumbersome for patients and increase fatigue (Gardner & Boice, 2004). For these same reasons, pictures were presented consecutively and one at a time, rather than a randomised presentation of multiple images as per previous research (Gandevia & Phegan, 1999; Kammers et al., 2009; Longo & Haggard, 2012). Single picture presentation has already been reliably used in previous studies (e.g., Gardner & Boice, 2004; Mohr et al., 2010).

Participants were required to decide if the presented image corresponded with the veridical size of each body part. If they decided a certain picture was not their actual body part size (non-veridical), the experimenter presented another picture with the 5% increment or decrement in size, depending on the presentation order. The stimuli presentation continued until the participants' response changed (i.e., changed from non-veridical to veridical), as in previous research (Caggiano et al., 2020). Participants kept their own hands on their laps, out of their view, for the whole duration of the experimental procedure.

2.6 General analyses

The perceived size of the body parts was analysed in two ways. Firstly, the overall *Representational Range* was calculated for all body parts as a measure of the certainty of the representation. For this, the absolute difference between the averaged percentage of distortion in the ascending and descending trials per participant was obtained, getting an overall measure of variability of the distortion. For example, if a participant chose an image as veridical that was 70% the size of the original one in the ascending trial, and a picture that was 125% the size of the real sized picture in the descending, the average absolute representational range was 55%. Prior preliminary paired t-tests analyses did not identify differences in the distortion of body parts depending on the dimensions (length and width) or views for the hands (dorsal and palmar). Thus, results were averaged across dimensions for the face, and dimensions and views for the hands, as a general measure of the representation of these body parts.

Secondly, the *Body size distortion* or threshold estimates per series (ascending and descending) was computed by calculating the percentage of under/overestimation per body part (face, left hand and right hand). Previous studies have shown an influence of presentation order in size estimation of one's own body, advising against averaging size between ascending and descending conditions (Gardner & Boice, 2004; Gardner & Bokenkamp, 1996). Thus, the distortion of the size of each body part was considered in each presentation order. In this case, data was averaged across length and width dimensions to obtain overall distortion per body part.

Lastly, analyses were run to calculate the *Critical Cut-Off Scores* that would indicate impaired performance for each body part as done in previous studies (e.g., Cocchini, Beschin, & Della Sala, 2018). For this, the scores for ascending and descending conditions were averaged for each body part (left hand, right hand, and face) in the control group, to obtain a final absolute average of their performance (percentage of distortion). With this information, the highest value for each condition (body part) above which performance would be considered pathological was computed, by means of Crawford's single t-test case analyses equation (Crawford & Garthwaite, 2002; Crawford & Howell, 1998).

3. RESULTS

3.1 Personal and Extrapersonal Neglect examination

Two patients (P06 and P09) showed PN with the Comb and Razor test, whilst three showed PN with the Fluff test (P07, P08 and P09). None showed any difficulty in the One Item test, all scoring 0. Hence, a total of five patients showed PN at least on one task and were classed as having PN (PN+ group), whilst the other four did not show this disorder (PN- group). Two patients out of the nine showed EN, as assessed by the BIT battery, one in each group (P01 in PN+, and P04 in PN- group). Final patients' groups did not differ in Age [t (7) = .09, p = .93], Gender [t (7) = -.16, p = .88], Education [t (7) = -.22, p = .83], Time from injury [t (7) = 1.03, p = .34], or Time at CEADAC from admission [t (7) = .31, p = .77] (See Table 2).

3.2 General neuropsychological assessment results

Results for all neuropsychological tests are presented in Table 3. The scores between groups were compared to assess for potential differences.

In the Digit test all patients performed within the normal range in direct order, showing no impairment. In contrast all patients displayed impaired performance (< 4 span) in reverse order. Specifically, PN+ patients were able to recall 7 digits (SD = 1.58) in direct order, and 2.8 digits (SD = .84) in reserve. PN- patients recalled 6.25 (SD = 1.5) in direct presentation, and 3.25 digits (SD = .5) in reverse. Both groups performed equally in this test in direct [t (7) = .72, p = .49, d = .49] and reverse [t (7) = -.94, p = .38, d = .65] presentations.

Scores in the HVLT were considered for all patients but one (P05 in PN- group) who did not complete the test. In total recall, all patients performed over cut-off (score > 13). In delayed recall, two patients were identified as having impaired performance in PN- group (P04 and P05). Average performance was then considered to investigate differences between groups. PN+ patients were able to recall 20.6 words (SD = 6.07), whilst the PN- patients recalled 16.67 (SD = 4.62). Differences between groups did not reach significance [t (6) = .96, p = .38, d = .73]. Delayed recall did not differ between groups either (PN+: M = 6.4 words, SD = 2.3; PN-: M = 3.33 words, SD = 4.16; t (6) = 1.38, p = .22, d = .91).

Further, the scores in the phonemic fluency test (COWA) and the semantic fluency test were considered. Single patients' performance is included in Table 3, and pathological scores are denoted in bold. On average, PN+ patients produced 6.6 words for category F (SD = 3.71), 5.2 for A (SD = 3.11), and 8 for S (SD = 3.08). In PN- group, patients generated 9.25 category F words (SD = 3.86), 6.75 for category A (SD = 4.92), and 9.25 for category S (SD = 5.19).

Scores between patients' groups were compared via independent t-tests. PN+ and PN- patients did not differ in any of the scores in the different categories for FAS (F category: [t (7) = -.58, p = .58, d = .38]; A category: [t (7) = -.45, p = .67, d = .29], and S category: [t (7) = -1.05, p = .33, d = .7]. Further, on the semantic fluency test, PN+ participants produced, on average, 12 words (SD = 5.24), whereas PN- patients produced 15 (SD = 4). There were no significant differences in the semantic fluency test between groups [t (7) = -.94, p = .38, d = .64] (see Table 3).

The scores in the WSCT were then calculated. Patient P01 was unable to complete this test, whilst data for P09 was not available (both in PN+ group). Thus, the data considered was from a total of seven patients (see Table 3 for scores in this test). Considering the cut off score of 50, two patients showed impaired performance (P06 and P08), both in PN+ group. On average, patients in PN+ group were able to complete 1.67 categories in the WCST (SD = .58) whilst PN- patients completed 2.75 (SD = .96). Differences between groups were not significant [t (5) = -1.72, p = .14, d = 1.37]. Further, the patients' scores in the percentage of errors formula (Nelson, 1976) was of 51.46% (SD = 14.26) in PN+ group, whilst it was of 60.45% (SD = 5.65) in PN-. Differences were again not significant [t (5) = -1.17, p = .29, d = .83].

Two out of nine patients showed impaired performance on the GOAT (P03 in PN- group and P09 in PN+). Performance in this test did not differ between groups [t (7) = .53, p = .62, d = .34].

Similarly to previous studies using the Awareness of Deficit scale (Villalobos et al., 2018), patients in both groups showed reduced awareness (PN+ group: M = 15.6, SD = 4.34; PN- group: M = 19.5, SD = 1); however, there is no normative data for this scale. Differences between groups were not significant [t (7) = -1.74, p = .13, d = 1.24].

3.3 Motor and functional performance assessment results

Patients' scores in the Motricity Index are presented in Table 3. As in previous studies (Sunderland, Tinson, Bradley, & Hewer, 1989), patients with 'normal' scores (full marks) were identified. All patients but one (P03 in PN- group) were impaired in motor performance with their contralesional upper left limb. Scores ranged from 1 to 100 in the Motricity index test. On average, PN+ patients obtained a motricity score for the left upper limb of 29.8 ($SD = 36.95$) whilst the PN- group scored 58.75 ($SD = 42.03$). Differences between groups did not reach significance [$t (7) = -1.22$, $p = .26$, $d = .8$].

Impairments in the mobility of the contralesional lower limb were also identified for all patients but one (P03 in PN- group). The average score for the PN+ group was 45.8 ($SD = 30.98$), whilst the score for PN- group was 67 ($SD = 28.23$). Groups did not differ in the scores in the motricity index for this limb [$t (7) = -1.06$, $p = .33$, $d = .72$].

The level of independence in activities of daily living as measured by the BI showed that five patients (4 in PN+ and 1 in PN-) had a score lower than 60, indicating more dependency. Again, average scores were compared between groups, and were equivalent [$t (7) = -.8$, $p = .45$, $d = .52$]. Both groups showed partial dependency, with PN+ group obtaining, on average, a score of 46 ($SD = 18.84$), whilst the PN- group averaged a score of 60 ($SD = 33.42$). Consistently, the same patients were identified as dependent with the FIM. When group scores were compared, no significant differences were identified between groups [$t (7) = -1.17$, $p = .28$, $d = .76$], confirming their functionality level was equivalent (see Table 3 for scores).

Participant	Fluff test	Comb and razor/ Compact	One item test	BIT										
				Line cancellation		Letter cancellation		Star cancellation		Line bisection	Figure and shape copying	Drawing	Total BIT score
				L	R	L	R	L	R				
P01	**-40**	-3.03	0	**0**	**12**	**0**	7	**0**	**13**	**0**	3	3.3	**38.3**
P06	0	**31.8**	0	17	18	20	20	26	27	9	4	4	145
P07	**-20**	1.04	0	18	18	20	19	**21**	27	**6**	4	4	137
P08	**-22.2**	-1.4	0	18	18	19	19	24	25	9	4	4	140
P09	-2.2	**15.38**	0	18	18	19	**16**	25	27	8	4	3	138
P02	0	5.32	0	18	18	20	19	26	27	8	4	4	144
P03	11.1	3.1	0	18	18	20	19	27	27	9	4	4	146
P04	-13.3	-5	0	18	18	18	19	**14**	23	**4**	3	4	**121**
P05	-6.7	10.1	0	18	18	20	19	27	27	9	4	4	146

Note: Fluff test scores <-13.3 indicate contralesional PN, whereas scores >13.3 indicate ipsilesional PN; Comb and Razor/Compact cut-off score for left PN is +11, and -11 for right PN. One item test score >1 indicates contralesional PN. BIT scores for subtests represent the number of stimuli correctly reported. BIT aggregated score under 129 indicates EN.

Table 2. Personal and Extrapersonal Neglect assessment results.

A total of 5 patients were included in the PN+ group (shaded rows), and 4 in the PN- (unshaded rows). Scores in **bold** indicate impaired performance.

Participant	Digit span		HVLT		COWA			Semantic fluency	WSCT (% errors)	GOAT	Awareness of Deficit Scale				Motricity index		BI	FIM
	D	R	Total recall (number)	Delayed recall (number)	F	A	S				Awareness of injury	Awareness of deficit	Awareness of disability	Total	UL	LL		
P01	5	**2**	16	4	**3**	**8**	**2**	**13**	NA	90	6	8	0	14	**53**	**70**	55	64
P06	9	**3**	29	8	10	11	12	12	**41.18**	98	6	4	6	16	**29**	**53**	50	68
P07	8	**3**	24	9	**6**	11	8	20	67.74	90	6	4	6	16	**1**	**29**	45	65
P08	6	**4**	20	7	**5**	**6**	**6**	**9**	**45.45**	90	6	6	10	22	**1**	**1**	15	44
P09	7	**2**	14	4	**2**	**4**	**5**	**6**	NA	**59**	6	4	0	10	**65**	**76**	65	90
P02	8	**3**	22	8	**5**	**7**	8	17	61.53	99	6	8	6	20	**75**	**76**	80	98
P03	5	**3**	14	**0**	**3**	**7**	**7**	17	56.25	**16**	6	8	4	18	100	100	75	90
P04	5	**4**	14	**2**	**5**	**6**	**7**	**9**	56	95	6	6	8	20	**1**	**33**	10	43
P05	7	**3**	NA	NA	14	17	**15**	17	68	97	6	4	10	20	**59**	**59**	75	102

Table 3. Motor and neuropsychological assessment results.

Scores in **bold** indicate impaired performance and shaded rows refer to PN+ patients.

3.4 Body size estimation task

3.4.1 Representational range

The representational range was the absolute difference between the average percentage of distortion in the ascending and descending trials. A mixed-model ANOVA was run with two factors: Body Part as repeated measures factor (face, right hand, and left hand), and Group as between measures factor (PN+, PN- and Controls). The main effect Body Part was not significant [$F(2,44) = .97$, $p = .38$, $\eta p^2 = .04$], nor was the interaction between Body Part and Group [$F(4,44) = .87$, $p = .49$, $\eta p^2 = .07$], indicating there were no differences in the representational range across groups depending on the body part considered. In contrast, there were significant differences when considering the Group factor [$F(2,22) = 41.65$, $p < .001$, $\eta p^2 = .79$]. Bonferroni-corrected pairwise comparisons (corrected cut-off p value of .02) identified significant differences between PN+ and PN- groups [$t(7) = 3.45$, $p =.01$, $d = 2.3$], as they did between PN+ and Controls [$t(19) = 9.85$, $p < .001$, $d = 4.3$]; and between PN- and Controls [$t(18) = 3.02$, $p = .01$, $d = 1.37$]. These results confirmed there were distortions in the perceived size of all body parts in all groups, being of larger size for PN+ patients. Indeed, PN+ group showed the largest representational range (M = 64.58%, SD = 12.44) followed by PN- group (M = 35.11%, SD = 13.17) and the control group (M = 20.49%, SD = 7.45) (see Figure 2).

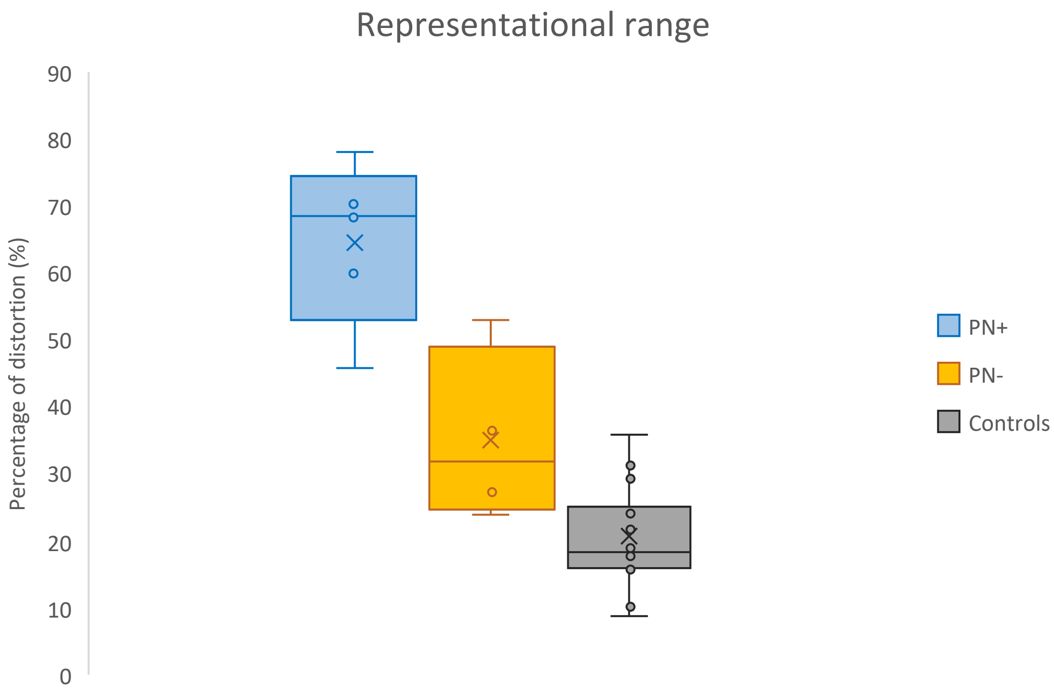


Figure 2. Box and whiskers plot with the data distributions for the representational range (%).

Representational range averaged across body parts for PN+, PN- and Control groups. The top of the rectangular box represents the 75th percentile of the sample, whilst the bottom represents the 25th percentile. The top upper whisker represents the maximum value of the sample, the bottom of the lower whisker represents the minimum value of the sample. Circles represent individual scores; x represents the sample mean and the line through the box is the median.

3.4.2 Body size distortion

Previous research has advised against averaging between ascending and descending trials as the final averaged judgement will not represent the asymmetry of these judgements within trials (Gardner & Boice, 2004). In order to identify any asymmetries in the representation (Gardner & Bokenkamp, 1996), we run further analyses considering ascending and descending order separately for each body part.

3.4.2.1 Left hand

In the ascending presentation, participants showed a general tendency to underestimate the size of their hand, but in different magnitudes. Larger underestimation was found on perceived size for the PN+ group (M = -28.88%, SD = 10.76), followed by the PN- patients (M = -19.22%, SD = 7.44). Controls also underestimated the size of the left hand but were more accurate (M = -5%, SD = 5.42) (see Figure 3). A one-way ANOVA was run to investigate differences in the perceived size of the left hand between groups. Significant differences were found between Groups [F (2,22) = 24.85, $p < .001$, $\eta p^2 = 1.25$]. Post-hoc Bonferroni corrected multiple comparisons identified significant differences between the size of the left hand in the PN+ group and Controls [t (19) = -6.68, $p < .001$, d = 2.8], and between PN- and Controls [t (18) = -3.65, $p = .004$, d = 2.19]. However, differences between PN+ and PN– did not reach significance [t (7) = -2.06, $p = .15$, d = 1.04]. In the descending condition there was, instead, overall overestimation of size, supporting the decision not to average across order conditions. Significant differences in size perception were also discovered when running a one-way ANOVA [F (2,22) = 11.38, $p < .001$, $\eta p^2 = 1.97$]. Multiple post hoc comparisons revealed these differences appeared between PN+ and PN- groups [t (7) = 3.44, $p = .007$, d = 2.1]. Differences were also significant when comparing PN+ patients to Controls [t (19) = 4.69, $p < .001$, d = 2.99], as Controls were far more accurate. Lastly, differences between PN- and Controls did not reach significance [t (18) = .15, $p = 1$, d = .07]. Specifically, PN+ patients showed larger overestimation (M = 36.88%, SD = 5.36) than PN- patients (M = 15.31%, SD = 13.47), whilst Controls showed 14.45% overestimation of size (SD = 9.15) (see Figure 3).

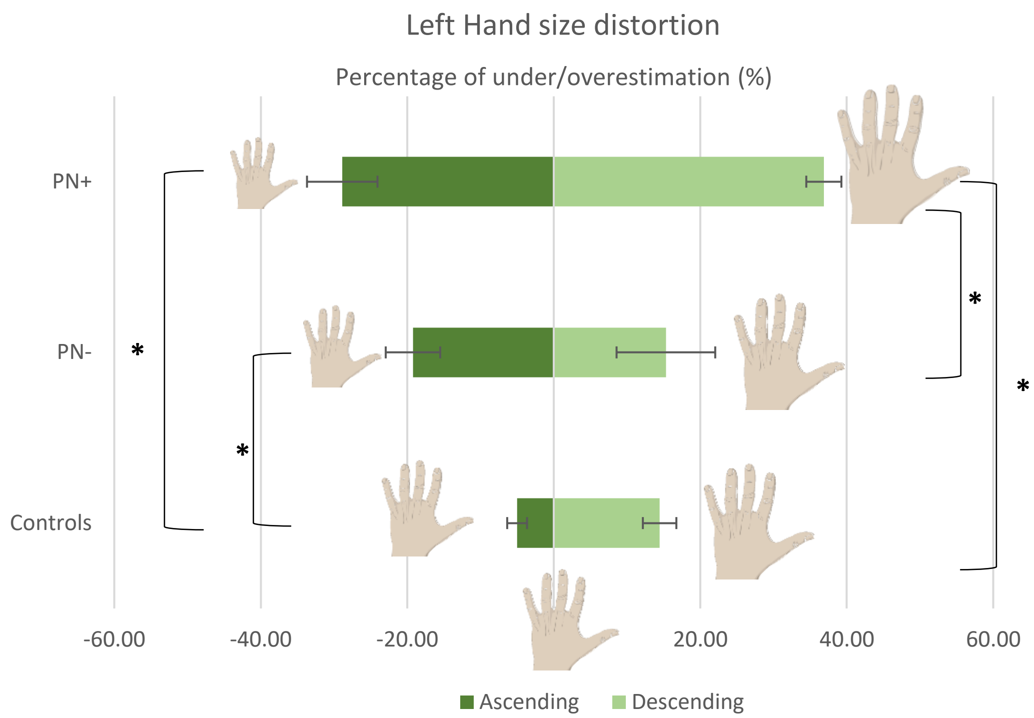


Figure 3. Left hand distortion.

Percentage of under/overestimation (%) of the perceived size of the left hand for all groups (PN+, PN- and Controls), for ascending and descending conditions. Hands depict the pictorial size distortion. * Denote significant differences.

3.4.2.2 Right hand

In the ascending condition there was, again, overall tendency to underestimate the size of the hand in all groups (see Figure 4). In this case, PN+ underestimated the size of their right hand by -25.75% (SD = 10.86), followed by PN- (M = -16.41%, SD = 10.82), and controls (M = -5.2%, SD = 6.45). A one-way ANOVA yielded significant results [$F(2,22) = 13.27$, $p < .001$, $\eta p^2 = 1.83$], indicating these differences in size perception were significantly different between groups. In ascending condition, there were not significant differences between PN+ and PN- patients [$t(7) = -1.72$, $p = .3$, $d = .86$], as both groups did underestimate the size of their right hand. In contrast, differences between PN+ and Controls were significant [t (19) = -

4.95, $p < .001$, $d = -2.3$], as Controls showed smaller underestimation of size. Lastly, differences between PN- and Controls were not significant [t (18) = -2.47, p =.07, d = 1.26].

For the descending condition, there was again overall overestimation of size. PN+ showed larger overestimation (M = 34%, SD = 5.53) than PN- (M = 18.44%, SD = 11.21), whilst Controls were slightly more accurate (M = 15.47%, SD = 9.35). These differences in size perception between groups were significant [F (2,22) = 7.99, p = .002, ηp^2 = 2.38]. Bonferroni corrected post-hoc tests showed significant differences between patients' groups [t (7) = 2.56, p = .05, d = 1.76], and between PN+ and Controls [t (19) = 3.99, p = .002, d = 2.41], confirming the distortion for PN+ was much larger than the other two groups. Lastly, no differences were found between PN- and Controls [t (18) = .59, p = .1, d = .29].

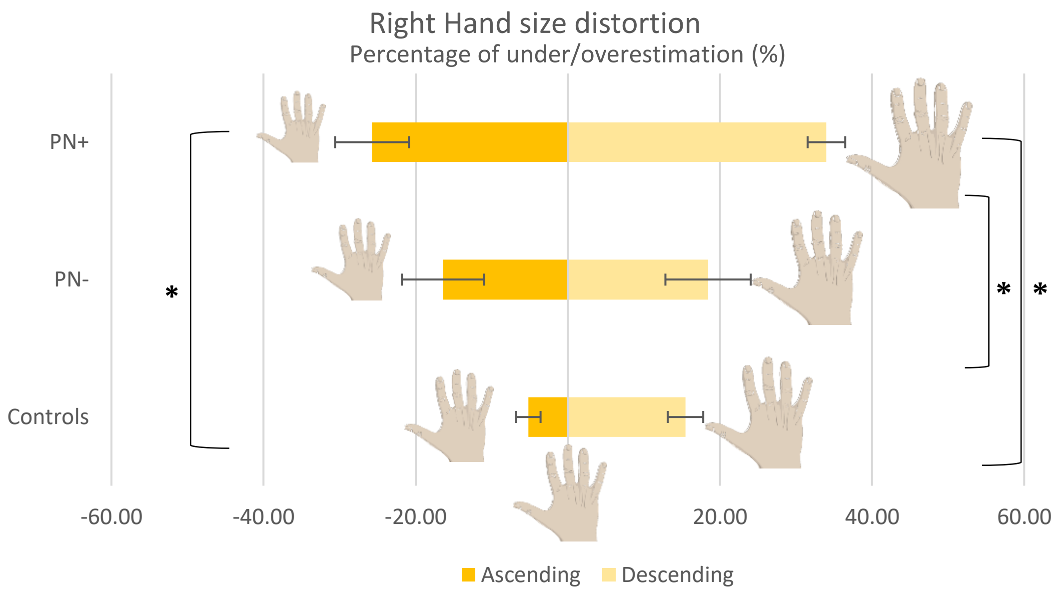


Figure 4. Right hand distortion.

Percentage of under/overestimation (%) of the perceived size of the right hand for all groups (PN+, PN- and Controls), for ascending and descending conditions. Hands depict the pictorial size distortion. * Denote significant differences.

3.4.2.3 Face

Size distortion for the face followed the same pattern as the hands. That is, there was perceived underestimation of size in the ascending condition, and underestimation in descending (see Figure 5). PN+ patients underestimated the size of their face more (M = -33.75%, SD = 7.02) than PN- (M = -10.94%, SD = 7.09) or Controls (M = -3.13%, SD = 6.06). A one-way ANOVA confirmed significant differences in size estimation for the ascending condition [$F(2,22) = 43.78$, $p < .001$, $\eta p^2 = 1.25$]. Post-hoc comparisons revealed differences between PN+ and PN- were significant [$t(7) = -5.32$, $p < .001$, $d = 3.23$]. Similarly, PN+ showed significantly larger underestimation than Controls [$t(19) = -9.36$, $p < .001$, $d = 4.67$]. PN- and Controls similarly distorted their face [$t(18) = 2.19$, $p = .12$, $d = 1.18$].

In the descending condition, differences were identified between groups in the overall ANOVA [F (2,22) = 13.73, $p < .011$, $\eta p^2 = 1.8$]. Post-hoc analyses revealed significant differences between PN+ and PN- patients [t (7) = 2.69, p = .04, d = 1.62], as PN+ patients showed larger overestimation of size (PN+: M = 34.25%, SD = 7.43; PN-: M = 23.13%, SD = 6.25). When compared with Controls, PN+ performed significantly worse [t (19) = 4.58, $p < .001$, d = 2.48], since Controls overestimated in less magnitude (M = 17.73%, SD = 5.78). Differences between PN- and Controls did not reach significance [t (18) = 2.06, p = .4, d = .9].

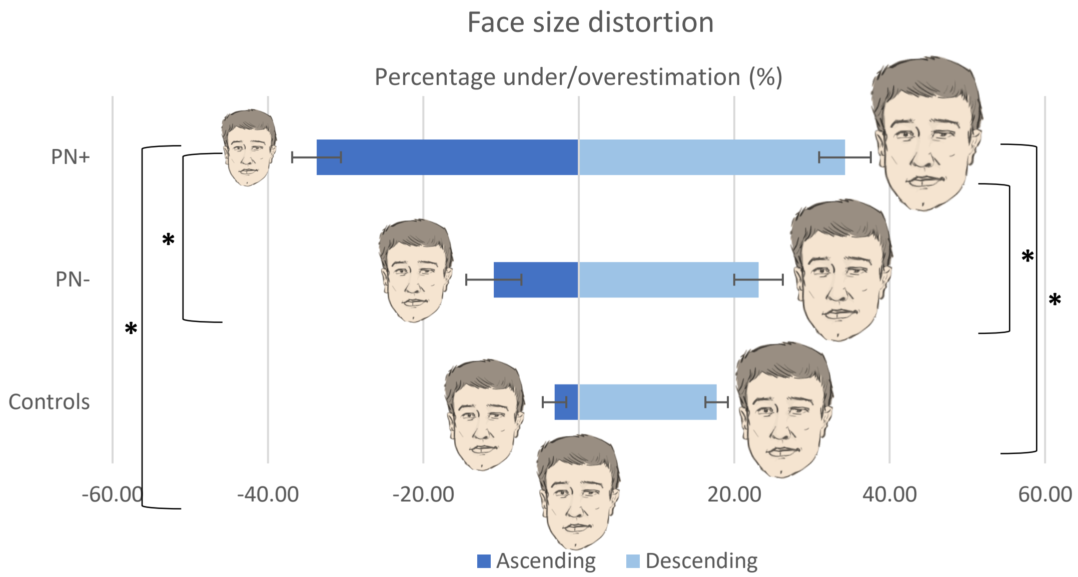


Figure 5. Face distortion.

Percentage of under/overestimation (%) of the perceived size of face for all groups (PN+, PN- and Controls), in ascending and descending conditions. Faces depict the pictorial size distortion. * Denote significant differences between groups.

3.4.3 Cut-off scores and individual performances

Cut-off scores were calculated per body part, as explained in the general analyses section. The critical value that indicated impaired performance was 19.38% for the left hand; 23.82% for the right hand and 22.35% for the face. The individual averaged absolute distortion per participant and body part was then calculated. Results indicated that all patients in PN+ group (100%) were above the cut-off for face and left-hand size perception, whilst 80% of them were above for right hand distortion. In contrast, in the PN- group only 25% of participants went over cut-off for the left and right hands, whilst none showed pathological performance in the face task (see Figure 6A, B and C for bar graphs of individuals' performance).

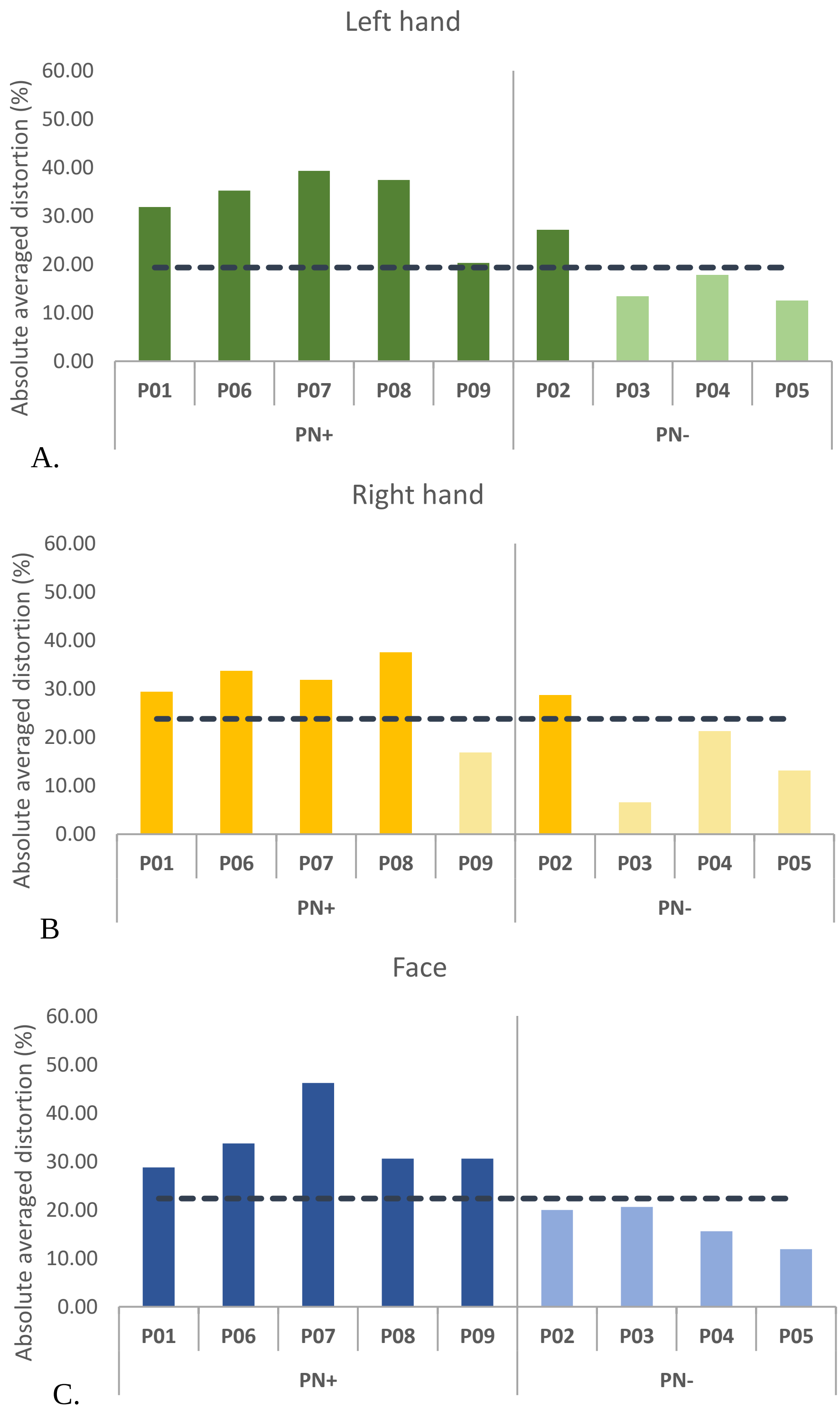


Figure 6. Absolute averaged distortion in the body size estimation task per participant.

Representation of the absolute averaged distortion of the left hand (A); right hand (B), and face (C) for all patients. The dashed lines indicate the cut-off for each body part. Abscissa axis indicates the patient's groups (PN+ and PN-) and the numbers for each patient. Pathological performance is indicated by darker-coloured bars (over cut-off).

4. DISCUSSION

In this study, the distortions in the size representation of hands and face were investigated in a group of patients with PN (PN+) and compared with patients without PN (PN-) and age-matched healthy controls. Pictures of their own hands and face were presented gradually distorted in either ascending or descending trials so they could choose the one that best fitted their own representation. Except for the PN tests, the patient groups showed a similar performance pattern on the cognitive and motor tests. On the contrary, group differences were evident when we considered the Representational Range (absolute difference between the averaged percentage of distortion in the ascending and descending trials), showing the magnitude of the representational error. In this analysis, we observed a larger error range in PN+ patients ($\simeq 65\%$), compared to PN- ($\simeq 35\%$), and Controls ($\simeq 21\%$). This was associated with less accuracy for those patients with PN, suggesting more ambiguity in their responses and a hazier representation of their body, which led to larger range of possible sizes. Comparatively, this is also seen in patients with eating disorders (ED) (Slade & Brodie, 1994), characterised by this lack of accuracy, ample range, and a more labile or tenuous view of their bodies (Holder & Keates, 2006; Touyz, Beumont, Collins, McCabe, & Jupp, 1984). Analogously with PN patients, patients with ED show more variability in their representation (Espeset, Gulliksen, Nordbø, Skårderud, & Holte, 2012; Mussap, McCabe, & Ricciardelli, 2008), with greater overestimation and underestimation (Gardner & Bokenkamp, 1996). This may be due to an erroneous stored body image, which cannot be updated by experience (Riva, 2012), giving ED patients the impression to live with a 'wrong body' (Osman, Cooper, Hackmann, & Veale, 2004; Riva & Dakanalis, 2018; Riva, Gaudio, & Dakanalis, 2015). Given that PN has been considered as a disconnection syndrome that compromises a network of cortical and subcortical structures involved in body representation (Bertagnoli et al., 2022), it is possible that the visuospatial transformation and comparison between one's own body and the presented image required in this task (Peltz et al., 2011) may be impaired in PN, due to inability to access one's own body representation (Mohr et al., 2010; Spitoni et al., 2013). Hence, PN patients become locked to a distorted body representation (Di Vita, Palermo, Piccardi, & Guariglia, 2015).

The hypothesis of a blurred body representation in PN+ was further supported when exploring the results on the ascending and descending trials. Firstly, we considered the representation in health to understand the effect of PN. Controls showed an asymmetry in their representation, characterised for a tendency to overestimate the size of all body parts in descending trials, whereas they were more accurate in the ascending ones. This has been previously reported in literature as an error of anticipation (Gardner & Boice, 2004), found preferentially after enlarged pictures when using size adjustment methods (Gardner & Bokenkamp, 1996). Preference to larger body parts is seen in embodiment (Haggard & Jundi, 2009; Pavani & Zampini, 2007), as an adaptive mechanism to accommodate body growth (De Vignemont, Ehrsson, & Haggard, 2005), suggesting that body size is closely associated to body ownership (Di Vita et al., 2015). The opposite effect is seen after visual reduction of the size of a body part which causes loss of the sense of ownership of that limb (Ramachandran & Rogers-Ramachandran, 2007). Hence, a shrunken image of the hand would not be associated to one's own body (not 'owned'), wiping out any influences in grasping (Marino, Stucchi, Nava, Haggard, & Maravita, 2010). Similarly, owing to reduced ownership over a shrunken limb, chronic pain is reduced, confirming the rehabilitative potential of visual size manipulation (Moseley, Parsons, & Spence, 2008). To sum up, that a preference for bigger pictures was found in Controls may be part of general preferential processing of the body, where larger body parts are embodied, whereas smaller ones are not.

In contrast, PN+ patients did not show this asymmetry, and appeared equally influenced by the initial image presented in both directions (small and large). That is, they showed inaccurate responses in both ends of the size ‘spectrum’, underestimating the size of their body parts more in ascending order ($\simeq$-29%) and overestimating more in descending ($\simeq$ 37 %) when compared with Controls. Following a previous hypothesis, it is possible that usual mechanisms leading to preferential ownership of enlarged body parts are particularly affected in PN patients, due to uncertainty or disintegration of body representation (Razmus, 2017). Indeed, body ownership depends of the interaction between multisensory input and internal body models (Tsakiris, 2010). In PN there is a disconnection in multisensory integration of somatosensory/proprioceptive information with representations of the body space (Coslett, 1998; Galati, Committeri, Sanes, & Pizzamiglio, 2001). Supporting this, studies investigating embodiment and ownership in neglect patients have shown that these patients are more susceptible to the rubber hand illusion, owing to a pathological reliance on visual information (Llorens et al., 2017), ‘incomplete’ body representation (Ronchi, Heydrich, Serino, & Blanke, 2018) and a more malleable body representation (van Stralen, van Zandvoort, Kappelle, & Dijkerman, 2013).

Bayesian casual inference models could also provide an interesting framework to explain these findings. These models postulate that the brain binds multisensory information that comes from different sources under certain optimal conditions, accepting a degree of incongruence (Blanke, Slater, & Serino, 2015; Rohe & Noppeney, 2015). The size estimation task in this study requires the brain to combine the visual template provided in each trial to the stored mental representation of the body part (Mohr et al., 2010; Spitoni et al., 2013). It is then possible that results in this task indicate the level at which multisensory inputs are integrated at optimal levels to construct the body representation, as seen with the rubber hand illusion (Pamplona, Gruaz, Mauron, & Ionta, 2022). Hence, results from Controls could indicate the degree of incongruence accepted to match visual images of body parts to the inner template of the body. For PN+, this integration may be disrupted as they seem more tolerable to incongruence, increasing the 'inferential error' and suggesting a disrupted multisensory causal inference.

Following, PN- group performance did appear in line to that of Controls, except for the left (contralesional) hand in ascending order presentation, in which they appeared more impaired. This finding suggests a potential influence of associated deficits, such as motor impairment (all patients but one showed motor impairment for the contralesional limb as measured by the motricity index). For example, increased malleability in the incorporation of the rubber hand is also seen in hemiplegics (Burin et al., 2015) who may also show distorted agency even for the ipsilesional limb (Cocchini et al., 2022). Sensory and motor information are relevant in memory retrieval, as memory of a particular stimuli or event will be stored in the same underlying 'machinery' that processed it (Leemhuis, De Gennaro, & Pazzaglia, 2019). Hence, it is possible that there is an influence of motor performance in the incidence of body representation disorders (Llorens et al., 2017). Indeed, limb immobilisation is associated with shrinkage of cortical representation (Hallett, 2001; Punt, Cooper, Hey, & Johnson, 2013). Changes in cortical representation, in turn, distort the representation of the size of the affected body area (Johnson et al., 2002; Lotze & Moseley, 2007; Matamala-Gomez, Nierula, Donegan, Slater, & Sanchez-Vives, 2020). Hence, use-dependent plasticity (Johnson et al., 2002), affects connectivity and structure of the deprived cortex (Leemhuis et al., 2019; Makin et al., 2013). In healthy adults, short-term immobilization causes a reduction of the size of the peripersonal space, whereas the overused limb is perceived as larger (Bassolino, Finisguerra, Canzoneri, Serino, & Pozzo, 2015). This may explain why PN- patients showed some disruption in the representation of the left contralesional hand in comparison with Controls, as all but one (P03) showed some degree of motor impairment. Furthermore, deficits in body representation are common after unilateral (left and right) brain damage even in absence of PN (Raimo et al., 2022) and different aspects of body representation may be affected (Razmus, 2017; Schwoebel

& Coslett, 2005). Thus, it is possible that motor, somatosensory and other body related deficits affect perceived body size, whilst maximum distortion is instead found in PN+, due to a combination of factors, such as distorted stored body representation, attentional influences and hemiplegia (Committeri et al., 2018). Still, when considering single patients' performance, only one patient in PN- group (P02) showed impairment enough to exceed the cut-offs for both hands, whereas all PN+ patients did (including the face). Hence, our findings confirm that the motor impairment per se can increase the uncertainty in estimating one's own body size, whereas PN can have a further detrimental impact rendering even more blurred the representation of the body part to be represented. Indeed, a combination of factors, namely PN and motor impairment, may lead to a distorted representation of body parts (Caggiano et al., 2020). Moreover, differences between patients' groups appeared for all the descending order trials, but only for the face in ascending ones. These results confirm that pathological overestimation of body parts is associated with PN whereas pathological underestimation can be present due to motor impairments and other body representation deficits.

Lastly, we presented cut-off data to consider the severity of the misrepresentations. It is here where the PN+ appeared clearly impaired when compared with PN-, confirming that pathological distorted representation characterises PN. Indeed, asymmetries in the size representation of hands in PN+ were not found, which were also equivalent to the face, confirming the premise of an overall pathological body representation (Di Vita et al., 2017; Guariglia & Antonucci, 1992; Palermo et al., 2014).

We wanted to note that our results may seem in contrast to Caggiano et al.'s (2020) study where PN patients showed a specific downsized representation of the contralesional hand. A possible reconciliation is that in Caggiano et al.'s study, patients were required to imaging a stable image of their hand moving toward a visible but changeable aperture, measuring the body schema. In our current study, PN+ patients were asked to imagine either their hand or face on the screen and compare it with a series of distorted images of their hand or face. This task aimed to assess a more explicit component of their representation (i.e., the visuospatial body map of the body image). Similarly, previous studies have shown different representations of body schema depending on the task used (Matsumiya, 2022).

It is important to highlight that this study presented with some limitations. Despite a small sample size is not uncommon in clinical studies (Caggiano & Jehkonen, 2018), it represents a potential limitation to generalization of findings. Hence, results need to be taken with caution, even though differences in performances were found. A larger sample of participants could help solidifying these findings and strengthening the conclusions. Moreover, we only employed one representation task (size estimation) disregarding other aspects of body representation that may be impaired (Raimo et al., 2022). However, we decided to consider size estimation here as previous studies have looked into other aspects of the body more frequently (see Caggiano & Jehkonen, 2018; Committeri et al., 2018 for reviews). In the future, a more holistic approach to body representation research in clinical populations could be achieved by employing several body representation tests concurrently, in order to understand the disintegration of body representations that occurs after stroke (Razmus, 2017).

As shown in Table 1, both PN+ and PN- groups show lesions encompassing the basal ganglia. Recent studies have postulated that PN should be considered as a disconnection syndrome in which white matter tracks that underly body representation are damaged, rather than discrete modular lesions (Bertagnoli et al., 2022; Committeri et al., 2007; see also Committeri et al., 2018 for a revision). It is also possible that basal ganglia, known to change their role depending on the pathway involved in a specific function (Zeugin & Ionta, 2021), work differently in patients with PN who present with an impaired body representation network. Further anatomical studies can help shed light on this aspect.

Taking together these findings, PN appears to be due to an underlying deficit in all body representations, which includes distortions of size as reported here, impaired body schema (Baas et al., 2011; Caggiano et al., 2020) and deficient topological body map (Palermo et al., 2014).

5. DECLARATIONS

The authors report there are no competing interests to declare. This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

6. CRediT AUTHOR STATEMENT

Laura Mora: Conceptualization, Methodology, Software, Validation, Formal analysis, Investigation, Resources, Data curation, Writing – Original draft, Visualization, Project administration. **Carlos Gonzalez Alted:** Resources, Writing – Review & Editing. **Gianna Cocchini:** Conceptualization, Writing – Review & Editing, Supervision, Visualization.

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