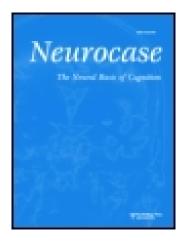
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Neurocase: The Neural Basis of Cognition

Publication details, including instructions for authors and subscription information: <u>http://www.tandfonline.com/loi/nncs20</u>

Where are your body parts? A pure case of heterotopagnosia following left parietal stroke

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To cite this article: Laurent Auclair, Marion Noulhiane, Patrick Raibaut & Gerard Amarenco (2009) Where are your body parts? A pure case of heterotopagnosia following left parietal stroke, Neurocase: The Neural Basis of Cognition, 15:6, 459-465, DOI: <u>10.1080/13554790902911642</u>

To link to this article: <u>http://dx.doi.org/10.1080/13554790902911642</u>

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Where are your body parts? A pure case of heterotopagnosia following left parietal stroke

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We studied the involvement of the parietal cortex in interpersonal body representation in a left parietal stroke patient. We used tasks assessing different types of body representations and localization of object parts. The patient performed normally on all tasks of body knowledge. However, she was unable to locate body parts on another person or on body representations. In contrast, she pointed correctly to the same body parts on herself or object representations. The data support the important role of the left parietal cortex in the transformation of intrinsic spatial coding of body parts localization in extrinsic body part coordinates.

Keywords: Parietal lobe; Body representations; Body schema; Heterotopagnosia; Autotopagnosia.

INTRODUCTION

When we act in the world, the brain must constantly monitor the position and movement of the body through space in relation to nearby objects. Different representations underlie human body processing (Schwoebel & Coslett, 2005; Sirigu, Grafman, Bressler, & Sunderland, 1991). One putative human body representation, called 'body image', provides information including the names and functions of body parts and their functional association with objects (Laiacona, Allamano, Lorenzi, & Capitani, 2006). A second type of human representation, termed 'body structural descriptions', concerns the spatial arrangement of body parts and their relationships. The third representation refers to a classic notion of neurology, the concept of 'body schema' (Gross, Webb, & Melzack, 1974; Head & Holmes, 1911), which corresponds to a dynamic, adaptable representation of the body in space, emerging from various pieces of sensory information.

The effective monitoring of the human body and its individual parts requires an integrated neural representation sustaining the 'body schema' (Gross et al., 1974; Head & Holmes, 1911; Poeck & Orgass, 1971). Disruption of this representation, which is frequently observed following parietal lesions, induces profound changes in the way that we perceive and represent our own body (i.e., egocentric perspective) or the body of another person (i.e., allocentric perspective) (Haggart & Wolpert, 2005). Previous studies in patients with left parietal lesions revealed a dissociation between body part localization related to self versus other. Selective difficulties were demonstrated in pointing to their body parts, both on verbal command and on imitation (Buxbaum & Coslett, 2001; Guariglia, Piccardi, Puglisi, & Traballesi, 2002; Ogden, 1985; Sirigu et al., 1991).

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The authors thank the patient for her patience. We are grateful to Dr J. Barra (Université Paris Descartes and CNRS UMR 8189) and Dr G. Huberfeld (INSERM U739) for previous comments on the manuscript.

Disclosure: The authors report no conflict of interest

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For example, patients pointed to a body part that was spatially close to the target (hand \rightarrow forearm) or one that was functionally similar to the target (knee \rightarrow elbow). This impairment, called autotopagnosia, may reflect damage to the structural descriptions of the body and its parts that define the position of body parts relative to other parts (Buxbaum & Coslett, 2001; Corradi-Dell'acqua, Hesse, Rumiati, & Fink, 2008). Defective imitation of the examiner's body movements, often called ideomotor apraxia, is also frequently reported after left parietal lesion. Some studies (Goldenberg, 1995, in press) suggested that ideomotor apraxia is due to a lack of general topographic knowledge about the human body and could be directly related to autotopagnosia. More rarely, patients with left parietal damage may have a selective deficit in pointing to the examiner's body parts and sometimes to parts of a representation of human body, whereas they can point correctly to their own body parts; this is called heterotopagnosia (Degos, Bachoud-Levi, Ergis, Petrissans, & Cesaro, 1997; Felician, Ceccaldi, Didic, Thinus-Blanc, & Poncet, 2003). When asked to point to the examiner's shoulder, some of these patients point to their own shoulder, suggesting an impairment in the spatial integration of another person's body parts (Degos et al., 1997; Haggard, 2003). Body part localization involves both an intrinsic spatial coding of body parts relative to one another and an extrinsic coding of the body parts in order to update the visual information on oneself (Buxbaum & Coslett, 2001; Buxbaum, Giovannetti, & Libon, 2000; Creem-Regehr, Neil, & Yeh, 2007). Previous studies have suggested that the left parietal lobe plays a predominant role in intrinsic but not extrinsic spatial coding. Overall, these data indicate that different processes are involved in the perception of one's own body and the body of another person. Functional MRI (fMRI) studies have also indicated activation of a focal region of the lateral occipitotemporal cortex called the EBA (Extrastriate Body Area) when healthy participants were instructed to observe a human body (static or not) or body parts (Downing, Jiang, Shuman, & Kanwisher, 2001; Peelen & Downing, 2007). Interestingly, the right hemisphere EBA is more activated when body parts are presented from an allocentric (other's body) than from an egocentric perspective (one's own body), whereas there is no modulation of body perspective on the left hemisphere EBA (Chan, Peelen, & Downing, 2004; Saxe, Jamal, & Powell, 2006). Based on these results, the right EBA appears to be crucial to the body allocentric perspective. Yet, it is not known which specific cerebral networks are involved in spatially binding body parts to the self or to another person.

We report here on the case of the patient PA following a left parietal stroke. We used tasks especially designed to assess the distinct body representations of self and others in this patient.

CASE REPORT

Patient PA is a 72-year-old right-handed woman who had been employed in banking and had no history of neurological or psychiatric disorders. She was admitted to the emergency ward owing to the sudden onset of headache and acute confusional syndrome associated with hypertension and spatio-temporal disorientation. A CT scan performed in the following hours showed a 50-mm left parieto-occipital hemorrhage associated with a small intraventricular hemorrhage. Upon admission, the neurological examination showed an equilibrium deficit but no motor deficit. She had a right hemianopia, right spatial hemineglect without head and gaze deviation toward the side of the lesion, and a right-side tactile extinction. A T1-weighted MRI performed 3 months after the stroke showed a large parieto-occipital hematoma with a slight left ventricular mass effect and a slight subcortical atrophy (see Figure 1 for details).

A detailed neuropsychological assessment was carried out at this time. PA's language assessment, including standard comprehension, picture naming and reading tasks, did not show any sign of aphasia. She performed transitive and intransitive limb gestures normally upon verbal command or imitation. The patient initially presented the four elements of Gerstmann's syndrom. There was no visual agnosia or prosopagnosia. Moreover, the identification of objects presented from an unusual angle was perfect. Spatial hemineglect was assessed with a French test battery (Batterie d'Evaluation de la Négligence, Azouvi et al., 2002) including assessment of personal neglect and gaze orientation, several paper-and-pencil tests for extrapersonal neglect (i.e., bell cancellation test, two line-bisection tasks, overlapping figure identification) and related disorders such as anosognosia and extinction. The neuropsychological examination indicated no signs of spontaneous head and gaze deviation toward the right side of the space. Furthermore, PA did not show personal neglect: she could touch her left hand with her right hand, with her eyes open or closed. without hesitation. Concerning the extrapersonal

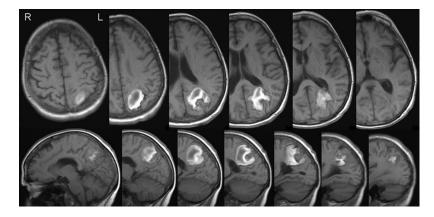


Figure 1. Axial and sagittal slides (in a rostro-caudal direction) from T1-weighted MRI of PA showing that the location of the hematoma concerned the left parietal lobe, precuneus, part of the superior parietal gyrus and the posterior forceps of the corpus callosum. *Abbrevations:* L, left; R, Right.

neglect, the right spatial hemineglect was reduced: there were no signs of spatial neglect on overlapping figure or line bisection tests and, PA omitted only two right-side items on the cancellation test. However, she had left spatial dysgraphia (a tendency to write only on the right-hand side of the page, to produce wavy lines and to duplicate some letters), and left spatial dyslexia in text reading (when skipping from line to line). Apart from the visuospatial domain, retrieval in verbal episodic memory was impaired. A neuropsychological assessment focusing on the types of body representations was carried out.

METHOD AND RESULTS

Three months post-onset, we used various tasks that assess distinct types of body representations (Schwoebel & Coslett, 2005) including body image (lexical-semantic knowledge), body structural description and body schema. The data reported here were collected in compliance with the ethical principles outlined in the Helsinki Declaration. Several successive replications of each task were done in order to assess their reliability. The results are summarized in Table 1.

Assessment of body image – semantic body representation

Naming body parts

The patient was asked to name 11 body parts (e.g., hand, ear, shoulder, foot) singled out by the examiner on her own body, the examiner's body

 TABLE 1

 PA's performance on the different body-related tasks

 (S1= session 1 and S2 = session 2)

Body-related tasks	PA's performance
Assessment of body image representation	
Naming body parts	100 (11/11)
Body part semantic knowledge	100 (11/11)
Assessment of body structural description	
Structural knowledge of body parts	100 (10/10)
Localization of named body parts	
On PA	S1: 100, S2: 100
On examiner	S1: 30, S2: 40
Assessment of body schema	
Localization of pictures of body parts	
On PA's body	100 (10/10)
On examiner's body	25 (2/10)
On drawing of body	37.5 (3/10)
Pointing to parts of human face	14 (1/7)
Localization of objects on the body	
surface (Sirigu's task)	
Examiner's body	100 (10/10)
Recall of body location	90 (9/10)
Pointing to parts of non-human representa	tions
Animals	96 (22/23)
Non-living objects	91 (21/23)

Results (% of correct responses) in body-related tasks.

and body pictures. She was also instructed to name 19 individually presented pictures of body parts. There was no time limit to answer. The patient performed perfectly in each condition.

Semantic knowledge of body parts

An item of clothing or an accessory (e.g., watch, scarf, necklace, ring, shoes, trousers, hat, etc.) was named verbally. The patient was asked to indicate

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the body part with which the item was most closely associated. The task consisted of 11 trials repeated twice. She responded successfully on both administrations of the task.

Assessment of body structural description

Structural knowledge of body parts

A target body part was pointed out and then two other body parts were verbally indicated (e.g., 'is the wrist next to the shoulder or the elbow?'). The patient was instructed to say which of the two body parts mentioned was either just below or beside the target body part, with no time limit to respond. In 10 trials, repeated twice, PA was accurate 100% of the time.

Localization of named body parts

The name of a body part was auditorily presented and the patient was asked to point to the named part on her own body, the examiner's body and a picture of a human body viewed from the front. There were 10 trials in each condition and each condition was repeated twice with no time limit to respond. A response was considered incorrect when the body part was not pointed to accurately. In another 10 trials, PA was instructed to point to her own or the examiner's body parts while keeping her eyes closed. She had no difficulty pointing to her own body parts on verbal command (100% correct responses for each session). The gesture was correct and unhesitating. In contrast, she failed to localize body parts on the examiner's body (session 1: 30% correct responses [3/10]; session 2: 40% correct responses [4/10]) or on a representation of the human body (session 1: 30% correct responses [3/10]; session 2: 30% correct responses [3/10]). When the examiner informed the patient of her error, she nevertheless persisted and searched the examiner's whole body, often saying 'the part is somewhere else but I don't know where'. A qualitative analysis of the localization errors obtained across the different sessions was carried out (Sirigu et al., 1991). The patient made 22% contiguous errors, corresponding to a response to a part close to the target (e.g., forearm for elbow); 19% nearby errors, that is, pointing to the same region of the body (e.g., bicep for hand); there was no substitution of functionally similar parts (e.g., elbow for knee). There were 41% unrelated errors (e.g., eyebrow for chest) and 19% no response, meaning that she searched around the body but did not point to any specific part.

Assessment of body schema

Localization of pictures of body parts

A picture of a body part was presented on the center of a sheet of A4 paper. The patient was requested first to name the body part and then to point to the corresponding part on her own body, the examiner's body and a picture of a human body. There were 10 trials in each condition. As in the previous task, the patient was able to point to her own body part without fail but was unsuccessful at localizing body parts on the examiner's body (2 correct answers out of 10) or on a drawing of a human body (3 correct answers out of 10).

Localization of objects on the body surface (Sirigu's task)

Ten pictures of familiar objects were pinned to the examiner's clothing at various locations on his body. The following pairs were made: car-right shoulder, bed-right hand, phone-left elbow, garbage-left forearm, bicycle-abdomen, butterflyright knee, rooster-left thigh, saucepan-right foot, shoes-left shoulder, glass-right elbow. A set of 10 similar pictures were placed in the same way on the patient's body. The patient was requested to point to the same object on the examiner's body that the examiner had touched on her patient's body. The patient's performance on this task was perfect. After the pictures were removed, the patient was instructed to verbally recall the different locations where the objects had been placed and then to replace the pictures in their initial positions on the examiner's body. The verbal recall of body locations was nearly perfect (9/10) but she was unable to correctly arrange the different pictures on the examiner's body from memory (0/10).

Pointing to parts of the human face and construction

Seven parts of the face were verbally named and the patient was asked to point to their location on a representation of a human face and name the selected parts (ear, nose, eyebrow, eye, mouth, neck, cheek). She was unsuccessful at localizing the various parts on the drawing of a face (1 correct answer out of 7); only the eye was correctly localized. On another occasion, she was instructed to put small pictures of facial parts in the right places on a drawing of a head. She correctly placed the eyebrow, mouth and nose; although the eyes were placed in the right location, they were reversed. Furthermore, the ears were placed just below the eyes.

Pointing to parts of non-human representations (complementary task)

The patient was asked to point to parts of different drawings: animals (cow, cat, elephant, dog, rooster) or non-living objects (house, car, cup, coat, bicycle). Each drawing was placed on the desk in front of the patient and she was instructed to point to the named parts. For each drawing, 3 to 6 parts were selected, for a total of 23 parts for drawings of animals and 23 parts for non-living objects. She obtained quite similar performances for animals (21 correct responses out of 23) and non-living objects (22 correct responses out of 23). On the animal drawings, she confused the tail with the leg for both the cat and the elephant. For the non-living objects, the patient made only one error on the drawing of a bicycle (headlight for handlebars).

DISCUSSION

PA's performance supports the proposal that distinct representations underlie human body processing: body image, body structural representation and body schema (Schwoebel & Coslett, 2005; Sirigu et al., 1991). First, our patient's performance on the tasks assessing the verbal and semantic knowledge of the human body provides strong evidence that body image was preserved. The data also indicate that PA's knowledge of the position on her own body of the body parts relative to one another was functional, suggesting that there was no impairment of the body structural system. Thirdly, the data are also consistent with the integrity of the own body schema since PA was able to point to body parts on herself, which requires her to determine the relative localization of the body part according to the whole body position; her localization of objects placed on the body surface was also efficient. Thus, PA had no impairment of her body schema.

Additionally, Patient PA had no problems in localizing body parts on herself attesting that egocentric body representation was preserved. In contrast, allocentric body presentation was impaired since PA was unable to localize body parts on another person. These data, consistent across the

successive sessions, revealed heterotopagnosia. This impairment, which was observed on both non-verbal and verbal tasks, could not be related to syntactic comprehension difficulties. The inability to localize another person's body parts could not be attributed to visuo-motor impairment since the patient was able to effortlessly point to parts of non-human representations (objects, animals) or to objects situated in the peripersonal space. Furthermore, she was able to point to objects attached to different parts of the examiner's body although she could not localize these body parts on the examiner or on a body picture. She could also verbally recall the objects placed on the various body locations, whereas her memory of their arrangement on the examiner's body was impaired. These findings emphasize that the patient's localization difficulties were specific to human body parts. Few cases of heterotopagnosia have previously been described (Degos et al., 1997; Felician et al., 2003). Unlike our patient, these previous cases reported greater impairment in localizing other persons' body parts than in pointing to parts on a human representation (picture of a body, puppet). Our data suggest that the structural knowledge of the body and its parts is not limited to real or 3D human bodies. However, this discrepancy between results is not clear-cut. For instance, Felician et al. (2003) reported that their patient often failed on the first trial when the patient was instructed to point to a part of a body representation. Degos et al. (1997) investigated the localization of body on dolls but not systematically on body pictures since only four patients out of nine have been tested. Furthermore, few details have been reported on pointing performances to pictures of body and doll. Hence, future studies must be done to specify whether the pointing deficit associated to heterotopagnosia is limited to real 3D human bodies.

Moreover, PA's ability to point to body parts on herself demonstrated that she could code body part locations in intrinsic coordinates but not extrinsic ones. Her visuospatial transformation procedures were preserved since she was able to localize animal and object parts from pictures and identify unusual views of picture representations. The patient's inability to point to the body of other person also contrasts with her preserved ability to point at objects on the examiner's body. The fact that the same position is localized correctly when it corresponds to an external object and inaccurately when it coincides with a body part is not in agreement with the hypothesis of a deficit in the representation of the allocentric space. These findings suggest that PA presented specific difficulties with the transformation of intrinsic spatial body part coordinates into extrinsic ones. Difficulties pointing to another person's body parts also suggest an inefficient projection of body ownership inside the body of another individual (spatial embodiment) (Arzy, Thut, & Mohr, 2006).

Our results converge with those of previous neuroimaging studies (Bonda, Petrides, Frey, & Evans, 1995; Felician et al., 2004) and neuropsychological case reports (Felician et al., 2003; Haggard & Wolpert, 2005; Schwoebel, Coslett, & Buxbaum, 2001) in reinforcing the strong involvement of the parietal cortex in coding the position of body parts. It has previously been shown that left-braindamaged patients made more errors when they had to identify other people's body parts than with their own body parts, while no difference was found for right-brain-damaged patients (Frassinetti, Maini, Romualdi, Galante, & Avanzi, 2008). PA's pointing performance is therefore consistent with the claim that the left parietal cortex participates in a cortical network involved in the processing of other people's body parts (Felician & Romerguère, 2008). Some evidence also suggests the crucial role of the right parietal cortex in body representation mechanisms. Patients with somatoparaphrenia, frequently associated to extensive right parietal lobe damage, claim either that their contralesional arm belongs to another person ('alien' limb) or that an another person's arm belongs to them (Gerstmann, 1942; Vallar & Ronchi, 2009; see also Ramachandran & McGeoch, 2007). Likewise, focal electrical stimulation within the right inferior parietal lobule induces illusory perception of one's own body such as autoscopic phenomena characterized by seeing a second body in the extracorporeal space (Blanke, Ortigue, Landis, & Seek, 2002; Blanke & Mohr, 2005).

Finally, data from our patient also address the question of the role of the EBA in the perception of the self and others. Functional MRI studies found that the temporal end of the temporal sulcus responded strongly to images of human bodies presented in a variety of formats (e.g., photographs, silhouettes). Some studies have reported that the right EBA was more activated in allocentric views than egocentric ones (Chan et al., 2004; Saxe et al., 2006). Other studies did not find differential activation of the EBA according to the body point of view (Hodzic, Muckli, Singer, & Stirn, 2008). In addition, neither left nor right EBA was sensitive to the identity of the human body (subject's own

body or other). Our patient's lesion included a large area in the left parietal lobe. Since she presented an allocentric body impairment, our findings suggest that the right EBA may participate in the self-other body distinction but may not be sufficient to account for this distinction. Recently, an fMRI study of Hodzic et al. (2008) shows that the perception of body related information involves a large cortical network, in addition to EBA and the ventral part of the fusiform gyrus (FBA), including different cortical region in the left hemisphere (extrastriate visual cortex) and in the right hemisphere (extrastriate visual cortex, parietal cortex and the precentral gyrus). The size and the extension of the lesion of our patient mainly concerned the left parietal lobe, precuneus, part of the superior parietal gyrus and the posterior forceps of the corpus callosum (see Figure 1). Thus, cortical regions outside of EBA could contribute to the self-other distinction reported here, as well as the corpus callosum. Indeed, the integration deficits observed in PA patient could be related to interconnecting deficits following the corpus callosum damage. The apparent discrepancy between results obtained with fMRI and our data might be explained by methodological aspects: the pointing tasks presented here were very different from those used in the fMRI studies. Future studies using fMRI could be relevant to specify this question. For example, it could be useful to compare blood oxygen level dependent (BOLD) signal when patients are instructed to point either to body parts on their own body or on a whole body picture. These two conditions must be compared to a control situation in which patients have to point to different space localization.

> Original manuscript received 20 November 2008 Revised manuscript accepted 4 March 2009 First published online 17 June 2009

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