Modular structure of awareness for sensorimotor disorders: Evidence from anosognosia for hemiplegia and anosognosia for hemianaesthesia

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Abstract

In the present paper, we shall review clinical evidence and theoretical models related to anosognosia for sensorimotor impairments that may help in understanding the normal processing underlying conscious self-awareness. The dissociations between anosognosia for hemiplegia and anosognosia for hemianaesthesia are considered to give important clinical evidence supporting the hypothesis that awareness of sensory and motor deficits depends on the functioning of discrete self-monitoring processes. We shall also present clinical and anatomical data on four single case reports of patients selectively affected by anosognosia for hemianaesthesia. The differences in the anatomical localization of lesions causing anosognosia for hemiplegia and anosognosia for hemianaesthesia are taken as evidence that cerebral circuits subserving these monitoring processes are located in separate brain areas, which may be involved both in the execution of primary functions and the emergence of awareness related to the monitoring of the same functions. The implications of these findings for the structure of conscious processes shall be also discussed.

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1. Introduction

Suspicion about consciousness as an object of scientific exploration may lead to the outright dismissal of any subjective report of phenomenal experience, since it is either considered to be impenetrable to external observation or even to be a non-existing problem because ‘there is no reason to grant that persons have more of an inside than particles’ (Rorty, 1982). However, counterintuitive observations in neuropsychological cases have shown (and \textit{in primis} through subjective reports) that there is an inside and a fascinating and important one (to rephrase Weiskrantz, 1997). Although acknowledging that there cannot be a science for studying those aspects of private feeling that tell us what it is like to be some other subject (Bisiach, 1992), neuropsychologists have nonetheless taken advantage of the subjective component of neuropsychological syndromes. This has resulted in consideration of consciousness as both a private experience that gives rise to personal reports (see Marcel, 2004; Weiskrantz, 1997) and as a functional property of the brain that ‘monitors any mental content through the whole spectrum of...’
available behaviour open to public observation’ (Bisiach, 1999).
The study of brain-damaged patients has not only given a more legitimate method by hoe to scientifically study consciousness, but has also contributed in a substantial way to the dismantling of some folk-psychology beliefs, suggesting alternative perspectives on the content and structure of conscious processing. In this respect, years ago, Churchland (1986) noted that when the brain functions normally, the inadequacies of common-sense theories are hidden from view. However, these inadequacies can be unmasked in the counterintuitive behaviour of brain-damaged patients. In particular, the discovery of selective disorders of conscious awareness has provided evidence for a composite nature of conscious processes, as opposed to a unitary one. Indeed, the subjective experience that people have of themselves is reported to be, in normal conditions, a feeling of unity. The ‘illusion’ of unity of the self, assumed by common-sense theories of consciousness, is evident in the normal experience of correspondence between the actual presence/absence of a stimulus and the presence/absence of a subjective experience of it. An alternative view would suggest that consciousness and self-consciousness do not have a unitary, monolithic structure, but instead have a composite nature, subserved by the activity of different brain mechanisms distributed in specialized brain areas. Such a view would predict that focal brain damage should not cause a generalized impairment of conscious experience or conscious self-monitoring, but should instead result in domain-specific disorders of awareness.

With regards to domain-specific disorders of awareness, we mainly refer to two possibilities. First, brain damage may impair the emergence of awareness for the product of the processing of a specific stimulus, without affecting the elaboration of the sensory/semantic features of the same stimulus. Crucially, the processing and awareness of stimuli presented in different domains is normal. Unilateral neglect and extinction (Berti, 2002; Berti & Rizzolatti, 1992; Marshall & Halligan, 1988; Volpe, Ledoux, & Gazzaniga, 1979), blindsight and blindtouch (Cowey, 2004; Weiskrätz, 1986, 1997) and prosopagnosia (Tranel & Damasio, 1985), are clear examples of this category of disturbances. Patients are not aware that something has been presented either in the affected field (extinction, neglect and blindsight), or on the affected side of the body (sensory extinction and blindtouch), or do not overtly recognize a familiar face (prosopagnosia × a), despite the fact that it is possible, in some cases, to demonstrate high level categorical/semantic processing of the ignored items. In these cases, stimuli outside the affected hemifield/hemisoma, or non-face stimuli, are processed normally.

Secondly, with regards to domain-specific disorders of awareness, we also refer to brain damage that can selectively affect specific self-monitoring processes (which, when normal, allow control and awareness of one’s physical and cognitive status). Anosognosia in brain-damaged patients seems to be one of the most convincing examples of the latter case. Patients with anosognosia resolutely deny the presence of some of their post-stroke deficits, and/or the consequence that those deficits may have on their behaviour. Denial may therefore affect reading, language or memory disorders (Prigatano & Schacter, 1991), or even contralesional sensorimotor impairments. In these latter cases, patients may deny being blind or plegic, and their false beliefs are strong and often cognitively intractable. However, they are able to monitor their sensorimotor status when it is not related to the affected side of the body.

The discrete, composite structure of conscious awareness proposed above would also predict:

(a) Dissociations of monitoring in relation to sensorimotor symptoms co-occurring on the affected side of the body.
(b) Different anatomical substrates for each form of denial.

In the present paper, we shall review neuropsychological data on anosognosia for sensorimotor deficits, which favors the hypothesis of selective conscious monitoring. In particular, we shall first discuss clinical data on anosognosia for hemiplegia, which suggests adopting a theoretical framework that would explain the motor denial within a model of motor awareness and motor control. We shall then turn to the problem of anosognosia for sensory (tactile and proprioceptive) disorders and to available models of sensory awareness. Finally, we shall discuss recent evidence that suggests a double dissociation between unawareness of motor impairment (anosognosia for hemiplegia, AHP) and unawareness of somatosensory deficits (anosognosia for hemianesthesia, AHA), arguing, from data collected in single case studies, for potentially different brain localizations for motor and sensory monitoring processes.

2. Anosognosia for hemiplegia

Although anosognosia is a frequent observation in brain-damaged patients (see Orfei et al., 2007; Pia, Neppi-Mihona, Ricci, & Berti, 2004 for a review), it has not been studied as extensively as other neuropsychological deficits often associated with it, such as spatial neglect. In the last century, this disregard was related to the fact that one of the most accepted interpretations of anosognosia was based on a motivational account, according to which denial is a defensive adaptation against the stress caused by the illness (motivational theories, e.g. Weinstein & Kahn, 1955). The interest in anosognosia in general, and for AHP in particular, increased at the end of the last century, thanks to a seminal paper by Bisiach and Geminiani (1991) in which the former psychodynamic interpretations of the disturbance, which prevented considering AHP as a direct consequence of a damage to a specific cognitive system, were put into question on the basis of clinical observations. For instance, Bisiach and Geminiani (1991) pointed out that the more frequent association of anosognosia with left than with right hemiplegia would not be predicted on the basis of a purely motivational account, since patients are expected to protect themselves against both left and right motor disorders. Furthermore, anosognosia can be temporarily eliminated by vestibular stimulation. Cappa, Sterzi, Vallar, and Bisiach (1987) found that the elicitation of the vestibular reflex by introducing cold water into the patient’s left ear (thus provoking left side nystagmus), can transiently ameliorate AHP, whereas a psychodynamic reaction should not be influenced by physiological manipulation. In addition, the temporal course of
anosognosia does not support a motivational explanation as it is more common in the acute phase after a stroke than in the chronic phase (Berti, Ladavas, & Della Corte, 1996). Similarly, during amytal testing, when one hemisphere is functionally suppressed by a pharmacological treatment, anosognosia appears only a few seconds after the amytal injection (Carpenter et al., 1995; Gilmore, Heilman, Schmidt, Fennell, & Quisling, 1992). A defense mechanism would, again, predict the opposite temporal pattern, insofar as time is needed for establishing a psychodynamic reaction.

Since the study by Bisiach and Geminiani (1991), many different neuropsychological interpretations have been offered to explain the disorder, most of them explaining away anosognosia as the consequence of concomitant neurological symptoms. For instance, according to some authors, the coexistence of sensory and intellectual impairments would prevent the ‘discovery’ of the deficit (e.g. Levine, Calvanio, & Rinn, 1991). However, these accounts are not supported by many studies that have demonstrated that anosognosia is not necessarily associated with sensory/intellectual disorders, and that these impairments are also not sufficient to cause anosognosia (e.g. Berti et al., 1996; Marcel, Tegnér, & Nimmo-Smith, 2004; Small & Ellis, 1996). Indeed, it has been found that although most patients with anosognosia are also affected by left side anaesthesia, it is possible to observe anosognosic patients without sensory (tactile or proprioceptive) disorders, as tested at bedside examination. On the other hand, there are patients with severe somatosenory problems without denial of hemiplegia (e.g. Berti et al., 1996; Bisiach, Vallar, Perani, Papagno, & Berti, 1986; Marcel et al., 2004; Small & Ellis, 1996). In addition, the possibility of ascribing anosognosia to a memory problem, which may prevent the acquisition and retention of new information, including that related to the disease, has been ruled out by the observation of double dissociations between these disorders (Berti et al., 1996; Marcel et al., 2004). Although these deficits may shape the severity and the way in which anosognosia manifests itself, AHP is now considered a specific cognitive problem that calls for an autonomous explanation.

Within the cognitive theories of anosognosia, some interpretations suggest that denial may be the consequence of damage to a central monitoring process (e.g. Goldberg & Barr, 1991). The damage to a general multi-purpose control mechanism, responsible for the inspection of subjects’ physical and cognitive capabilities, would cause a generalized impairment in detecting all possible concomitant disorders affecting either personal or extrapersonal aspects of patients’ behaviour after the stroke. However, patients with motor impairments of the left limbs may deny the presence of upper limb paresis, while being aware of the motor impairment of the lower limb and vice versa (Berti et al., 1996; Von Hagen & Ives, 1937). Moreover, patients affected by left sided hemiplegia and multiple cognitive deficits related to right brain damage, such as neglect dyslexia and drawing neglect, may deny the motor impairment while being aware of the other concomitant neurological/neuropsychological disorders (Berti et al., 1996). Awareness was also dissociated within cognitive deficits so that some patients were aware of their neglect dyslexia, but did not acknowledge neglect in drawing and vice versa.

The existence of dissociations between different kinds of unawareness (Breier et al., 1995; Jehkonen, Ahonen, Dastidar, Laippala, & Vilki, 2000) indicates that denial, in general, can be considered a selective (as opposite to generalized) disorder of monitoring and that anosognosia for hemiplegia in particular can be conceived as a monitoring disorder that selectively affects motor awareness.

### 3. Anosognosia for hemiplegia and motor awareness

According to recent models for motor control and motor awareness (e.g. Haggard, 2005; Wolpert, Ghahramani, & Jordan, 1995), when an appropriate motor command is selected and sent to the muscles for the execution of a desired movement, a prediction of the sensory consequences of the movement is formed and would be successively matched with the feedback associated with the actual execution of the intended movement by the activity of a comparator (see Fig. 1).
This prediction, based on the efference copy of the programmed motor act (i.e. on signals that, once a movement is programmed, are sent to sensory and motor structures responsible for adjustments in perception and posture required for that movement) constitutes the signal on which motor awareness is constructed, according to Blakemore, Wolpert, and Frith (2002). This may appear counterintuitive because the prediction (and therefore the motor awareness) is formed before the actual execution of the movement. However, it has been demonstrated (Libet, Gleason, Wright, & Pearl, 1983) that when neurologically intact subjects have to estimate the time at which they become aware of a voluntary motor act (the so-called ‘M’ judgment), they indicate a moment that precedes the actual initiation of the movement by 50–80 ms. Thus, Libet et al.’s findings demonstrate that motor awareness is not simply constructed on the sensory feedback from the moving muscles, but instead emerges before the afference of sensory proprioceptive inputs. It is worth noting that such a model would imply that whenever the system makes sensory predictions about a certain programmed movement, we may construct the belief that the movement is actually performed (see Berti & Pia, 2006). The comparator would then match the congruency between the belief of the intended movement and the representation of the actual status of the system. When, in normal subjects, the motor act corresponds to the representation of the intended movement, motor awareness is veridically constructed. A veridical motor awareness is also constructed when the peripheral event does not correspond to the prediction and the comparator detects the discrepancy. On the bases of this model, it has been proposed (Berti & Pia, 2006; Berti, Spinazzola, Pia, & Rabuffetti, 2007) that in hemiplegic patients without anosognosia, the comparator, still able to detect the mismatch between the prediction and the actual condition, allows the normal construction of veridical motor awareness, which in this case represents the perception of an absence. Consequently, when hemiplegic patients without anosognosia are asked to move their affected limb, they acknowledge their motor failure. On the contrary, hemiplegic anosogonic patients, with a damaged comparator, cannot detect the mismatch between the predictions and the feedbacks, so are not able to distinguish between a purely intended action and the real movement execution. This failure would lead to the construction of a non-veridical motor awareness, and consequently to the false belief of being able to move. In view of this, the patient’s verbal delusions should reflect their movement experience, which arises from the normal or quasi-normal functioning of the intentional system and of the prediction/awareness component of the model. Therefore, the intended movement is actually experienced (however see Gold, Adair, Jacobs, & Heilman, 1994, for an alternative view). According to this explanation, the activity of the areas involved in motor intentionality (such as the supplementary motor area, SMA) should be spared by the lesion, whereas the area involved in the comparator activity should be affected. This hypothesis has been confirmed in brain imaging studies. It has been shown that AHP is associated with damage to pre-motor areas and insular regions (Berti et al., 2005; Karnath, Baier, & Nagele, 2005) related to motor control and execution, but not to SMA damage. These findings further suggest that self-monitoring is not only selective for a given function, but is also implemented in areas which are responsible for the function that has to be monitored. Moreover, Berti et al. (2007), have shown that a patient affected by left side hemiplegia and anosognosia still activates the proximal muscles of the affected side as a result of the attempt to execute a purposeful movement with the plegic limb. This observation demonstrates intact intentionality and programming of the spared brain regions (note that proximal muscles are bilaterally innervated, so can be still recruited in hemiplegic patients when the distal parts are completely paralyzed).

4. Anosognosia for hemianaesthesia

Although from the end of the last century, clinical and experimental studies have investigated the denial of sensorimotor deficits, anosognosia has mainly been evaluated for hemiplegia and hemianopia. To the best of our knowledge, anosognosia for tactile/proprionicceptive disorders was taken into account only by Marcel et al. (2004) within the context of a study on anosognosia for hemiplegia. This paper confirmed that anosognosia for hemiplegia was a specific neuropsychological disorder (not necessarily due to the concomitant presence of extrapersonal neglect, personal neglect, sensory disorders, intellectual impairment or general self-monitoring). Moreover, it was also found that unawareness for sensory deficits in general (including anosognosia for hemianopia, see Bisiach et al., 1986b) and for body sensations in particular, is much more frequent than unawareness for hemiplegia (between 66–81%). Instances of double dissociation between AHP and AHA have also been found: indeed, 18 patients were aware of their motor but not their sensory loss and one patient was aware of his sensory but not his motor impairment. However, the association/dissociation of anosognosia for sensory deficits with other concomitant disturbances (for instance, the selectivity of AHA with respect to tactile and proprioceptive deficits and their co-occurrence with neglect in patients with pure somatosensory anosognosia) was not discussed, and no explanation for AHA was offered, since anosognosia for plegia was the theoretical focus of the paper. Moreover, no data was reported about the localization of the brain damage in AHA. In the next sections of this paper, the problem of the theoretical explanation of AHA and the issue of its intrahemispheric localization will be discussed.

5. Anosognosia for hemianaesthesia and tactile awareness

Somatosensory consciousness has been defined as those aspects of neural activity elicited by the presentation of tactile stimuli (i.e. any physical stimulus that gives rise to activation of at least one class of sensory receptors located in the dermis) on the participants’ sensory receptive surface that can be reported explicitly (Gallace & Spence, in press).

As already mentioned, the dissociations between implicit processing and explicit report of external events (e.g. residual functioning in the absence of explicit knowledge), extensively described in the last century in many neuropsychological con-
ditions (e.g. Berti, 2002; Milner & Goodale, 1993; Pisella et al., 2000; Weiskrantz, 1991, 1996), have contributed to the demonstration of the selectivity of the conscious awareness process. The occurrence of similar dissociations has also been reported in the tactile domain and their study may shed light on the neurological substrates of the conscious processing of tactile events (see Gallace & Spence, in press). For instance, in the tactile domain, a phenomenon analogous to ‘blindsight’ in vision has been described, in which patients can localize the position of stimuli but are unable to consciously detect their presence or to identify their characteristics (e.g. Brochier, Habib, & Brouchon, 1994; Paillard, Michel, & Stelmach, 1983; Rossetti, Rode, & Boisson, 1995). This phenomenon was initially given the name ‘blind-touch’, but other names have been used, such as blind proprioception (e.g., Volpe et al., 1979), central de-afferentation (e.g., see Paillard et al., 1983), and numbness (e.g. Weiskrantz, 1997). Another example of processing without awareness in the tactile domain can be found in some instances of tactile extinction. The phenomenon of extinction has been studied mainly in the visual domain. In this case, patients are able to detect a stimulus presented in isolation in the ipsi- or contra-lesional side of space but often fail to detect the same stimulus on the contralateral side when another stimulus is simultaneously presented on the opposite side. Some studies have shown that extinguished stimuli can nonetheless be processed (e.g. Berti et al., 1992; Volpe et al., 1979). Extinction has also been found in the tactile modality and a dissociation between explicit and implicit processing similar to that described in the visual domain has been reported (e.g. Berti et al., 1999; Maravita, 1997).

The neurological substrate of implicit and explicit tactile processes is a matter of debate. It seems that several parietal regions outside the somatosensory cortex participate in the processing of somatosensory information (see Gallace & Spence, in press, for a review). Libet, Alberts, Wright, and Feinstein (1967) found that, in patients undergoing neurosurgery, the average evoked response to tactile stimulation could be recorded from the somatosensory cortex even when the stimulus intensity was below the threshold for producing sensory awareness. Sensory awareness was instead correlated with later components that followed the primary evoked potential. Libet et al. concluded that the primary evoked potential in SI was not a sufficient condition for perceptual awareness of tactile stimulation. Recently, Sarri, Blankenburg, and Driver (2006), used fMRI to study the neural correlates of crossmodal, left visual–tactile extinction in a patient affected by a lesion centered in the right inferior parietal cortex. Left crossmodal extinction is observed when the simultaneous stimulation of the right visual field (with a visual stimulus) and the left index finger (with a light touch stimulus) provokes extinction of left touch. The patient studied by Sarri et al. (2006) did not report the tactile stimulus on approximately half of the crossmodal double-stimulation trials, while becoming aware of left touch on the other half of the trials. Interestingly, fMRI revealed activation of contralateral primary somatosensory cortex on crossmodal trials when touch was extinguished from awareness, suggesting unconscious residual processing there. When the patient became aware of the left touch, additional activation was found in surviving right parietal cortex and in frontal regions.

These results seem to show that the implicit processing of tactile information requires the integrity of the somatosensory cortex, while its awareness requires the integrity of both the somatosensory cortex and the posterior parietal cortex, possibly because, as suggested by many authors, tactile information must be integrated with spatial information to gain access to consciousness (e.g. Andersen, 1997; Andersen, Snyder, Bradley, & Xing, 1997). According to this observation, we may say that ‘veridical tactile awareness’ is partially constructed by activity in the posterior parietal lobe.

What is the relation between anosognosia for somatosensory disorders and tactile awareness? Like patients with AHP, who seem to be aware of movements they do not actually perform, patients with AHA seem to be aware of tactile/propioreceptive stimulation that, because not correctly reported, we may infer are not really felt. Similar to the model we proposed for the motor domain (Berti & Pia, 2006; Berti et al., 2007), where non-veridical motor awareness is generated by the impossibility of distinguishing between a purely intended movement and its real execution, we may say that AHA patients have non-veridical sensory awareness which might be generated as an illusory experience by a failure in distinguishing between an imagined sensation and a real, physical one. A recent fMRI investigation compared activation during real tactile stimulation and tactile imagery. During real tactile stimulation, where subjects received a light touch on the right hand, bilateral activation of SI and SII, contralateral activation of superior parietal lobe (BA7), precentral gyrus (BA6), posterior insula and, subcortically, of the thalamus were observed. During tactile imagery, where subjects were asked to imagine the tactile stimulation on the same hand that was actually stimulated, contralateral primary and secondary somatosensory areas were activated, along with left precentral gyrus, and left insula. Activation in the left parietal lobe of left inferior frontal gyri (Brodmann’s area 44), the left dorsolateral prefrontal area, and the medial frontal gyrus was also observed. In the basal ganglia, activation in the left thalamus (ventral posteromedial nucleus) and putamen was found. It clearly emerged that there is a partial overlap between the areas activated during tactile imaging and those activated during actual sensation in a way that is similar to the one observed when motor imagery and motor execution are compared (e.g. Decety et al., 1994). It is worth noting that the posterior parietal lobe is activated during both experimental conditions, which suggests that its activation is correlated with the emergence of tactile awareness (see Sarri et al., 2006), whether it is ‘veridical’, like that experienced with real stimulation, or non-veridical, like that which emerges during tactile imagination. Other areas that are activated during the real stimulation and/or during the imaging condition might be related to the monitoring of the reality of the tactile sensation. Of course, the comparator that distinguishes between movement imagining and movement execution can rely on the interpretation of sensory feedback for accepting a motor awareness as veridical. It is more difficult to predict which signals must be integrated to ascribe the quality of being ‘veridical’ to sensory awareness. In any case there must be func-
6.2. Patient SC

Lesion reconstruction showed damages mainly to the insula, the external capsule, the basal ganglia, the internal capsule and the external capsule. The four patients presented in this study were tested according to the procedure indicated in the appendix.

6. Case descriptions

6.1. Patient PR

PR is a 61-year-old right-handed man with 4 years of formal education. An ischemic stroke of the right hemisphere caused a complete left hemiplegia, severe hemianesthesia and left unilateral neglect. He did not show any visual field deficit. We evaluated him 28 days after the stroke. At that time he was motivated and co-operative, although his performance in the Italian version of the mini mental state examination (MMSE) was under the cut-off score. Neuropsychological evaluation confirmed the presence of severe left extrapersonal neglect on all cancellation tests (Albert, 1973; Diller & Weinberg, 1977), as well as in drawing and reading tests, and also revealed the presence of a consistent personal neglect (Bisiach, Perani, Vallar, & Berti, 1986). He was completely aware of his hemiplegia and of the functional consequences of his motor impairment. However, he was completely unaware of his tactile and proprioceptive deficit, always convinced that he performed well in the sensory tests. Lesion reconstruction showed damages mainly to the temporal poles, the medial temporal lobe, the superior temporal lobe, the insula, the basal ganglia, the internal capsule and the external capsule.

6.2. Patient SC

SC is a 69-year-old right-handed man with 5 years of formal education. After an ischemic stroke of the right hemisphere he developed a complete left hemiplegia, severe hemianesthesia and left neglect. No visual field deficit was detected. We examined him 63 days after the acute event. He was motivated and co-operative, although presenting with a score under the cut-off in the Italian version of the MMSE. Neuropsychological evaluation revealed severe extrapersonal neglect in all the tests we used for its diagnosis. There were also signs of left personal neglect for the left upper limb (Bisiach et al., 1986a). The patient was completely aware of his left hemiplegia and of the functional consequences of his motor problem, however he did not admit his sensory deficit and was always convinced that he was able to correctly detect light touch stimuli and the position of his joints. Lesion reconstruction showed damages mainly to the superior temporal pole, the inferior, middle and superior temporal lobe, the insula, the external capsule and the basal ganglia.

6.3. Patient GC

GC is a 76-year-old right-handed woman, with 8 years of formal education. The patient had a right hemisphere ischemic stroke that left her with left hemiplegia, left hemianesthesia, and left neglect, without evidence of visual field deficits. We tested her 23 days after the onset, and she was oriented and co-operative. She had no global reasoning or language problems and performed above cut-off values on standardised test of verbal intelligence (the verbal judgements test, Spinler & Tognoni, 1987) but her performance in the MMSE was under the cut-off. GC had normal performance in verbal memory tests and her object recognition was preserved. Neuropsychological evaluation revealed severe extrapersonal and mild personal neglect (Bisiach et al., 1986a). She was fully aware of her hemiplegia and of its functional consequences. Nonetheless, she presented with complete anosognosia for her hemianesthesia. Lesion reconstruction showed damages mainly to the frontal inferior/orbital gyrus, the superior temporal lobe, the insula, the external capsule, the internal capsule, and the basal ganglia.

6.4. Patient RS

RS is a 46-year-old right-handed man with 18 years of formal education. He suffered a right hemisphere haemorrhage which left him with a complete left hemiplegia and severe proprioceptive hemianesthesia, while being perfectly able to detect light touch stimuli on the affected side of the body. Visual fields were also intact. We evaluated him 50 days after the stroke. He was fully co-operative, oriented in time and space and aware of his hemiplegia, which he reported immediately at the beginning of the evaluation. He was also concerned about the consequences of his motor impairment. As reported, light touch sensation was perfectly normal, however, the patient’s proprioception on the affected side was severely impaired: his answers were completely at chance for all the examined joints. Although fully aware of his motor problem, he was convinced that his capacity for detecting the position of the different body parts, passively moved by the examiner, was perfect. Lesion reconstruction showed damages mainly to the basal ganglia, the external capsule, the putamen, and the caudate nucleus.
In summary, all these patients were affected by contralateral hemiplegia and somatosensory deficits. They were perfectly aware of their motor problems, but denied the presence of any sensory problems affecting the contralateral part of their body. In this respect they represent ‘pure’ cases of anosognosia for hemianesthesia. None of them showed visual field deficits. Three out of four patients showed left visuospatial neglect, personal neglect, and mild cognitive impairment, while patient RS was a real pure case of anosognosia for AHA, because he was not affected by any form of spatial disorder, intellectual impairment, memory or language problems.

7. Anatomo-clinical data

Patients’ brain lesions were mapped in the stereotaxic space of Talairach and Tournoux (1988) using a standard MRI volume that conformed to that space as redefined by the Montreal Neurological Institute. Image manipulations were performed with MRlcro software (Rorden & Brett, 2000). Fig. 2 upper part shows the superimposed image of the three patients affected by somatosensory anosognosia and neglect, while in Fig. 2 lower part, patient RS’s damage is shown. All patients had a lesion involving the putamen, the insular cortex, the internal and the external capsule. In SC, GC and PR the superior temporal gyrus was also affected—an area whose damage has been associated with the presence of left spatial neglect, especially when, as in these cases, there are no visual field defects (for a review see Karnath, Berger, Kuker, & Rorden, 2004). On the contrary, in RS this brain region was spared.

8. Discussion

The aim of the present paper was to review clinical evidence and theoretical models related to anosognosia for sensorimotor impairments that may help in understanding the normal processing underlying conscious awareness. The initial assumption was that available neuropsychological data on syndromes like neglect, blindsight, numbness, blind touch and extinction, which shows selective disorders of monitoring of external events, would suggest a composite nature of perceptual awareness that might also apply to self-monitoring processes, like the ones involved in the awareness of post-stroke consequences.

The data we reviewed show how anosognosia for hemiplegia can be considered not only a specific disorder of awareness, but also a highly selective one, often associated with damages of cortical areas related to the programming of motor behaviour. Moreover, the evidence emerging from the study of patients suffering from anosognosia for hemianesthesia suggests that self-monitoring of sensory deficits is also both specific and highly selective, possibly associated with lesions of brain areas related to sensory processing. Indeed, Marcel et al. (2004) found that anosognosia for hemianesthesia can be dissociated from anosognosia for hemiplegia. In the present paper we also reported four patients affected by both left side hemiplegia and left side hemianesthesia who, while being absolutely aware of their motor condition, denied their sensory deficits, thus confirming the finding of Marcel et al. Interestingly, one of these patients did not show any sign of personal or extrapersonal neglect, demonstrating that denial of somatosensory disorders, like denial of motor disorders, is not the consequence of a spatial impairment where the patients do not pay attention to, or do not consider as belonging to them, the left parts of their body, but instead, that also anosognosia for hemianesthesia can be a specific disorder of self-monitoring.

The specificity and selectivity of anosognosia for somatosensory impairments is also suggested by the intrahemispheric localization of patients’ brain damage, which seems partially different (being more inferior and posterior) from that causing anosognosia for hemiplegia. While AHP is associated with damage to the frontal pre-motor cortex (Berti et al., 2005) and insular
region (Karnath et al., 2005), AHA seems to be related to insular, temporal and subcortical lesions mainly affecting basal ganglia (especially the putamen). Although the insular regions seems to be damaged both in AHP and AHA, it is worth noting that the presence of anosognosia for hemianesthesia was not evaluated in studies by Berti et al. (2005), and Karnath et al. (2005), therefore the fact that the insula was found to be affected might be due to the presence of AHA in those patients’ sample. Indeed, many patients involved in those studies were affected by sensory problems and might have developed sensory denial as well as motor denial.

Further research is needed in order to draw firm conclusions about the neuroanatomy of the monitoring of sensory awareness. Nonetheless, we may advance the hypothesis that the insular regions and the subcortical nuclei, selectively damaged in the patients described in this paper and related to sensory processing in previous works (e.g. Yoo, Freeman, McCarthy, & Jolesz, 2003) may be the neural bases of sensory self-monitoring. Their integrity might be necessary for distinguishing between ‘veridical’ and ‘non-veridical’ sensory awareness. Because the emergence of sensory awareness has been related to activity in posterior parietal areas (Gallace & Spence, in press; Sarri et al., 2006), which can arise even in the absence of real stimulation (Yoo et al., 2003), normal subjects and brain-damaged patients without AHA can correctly detect ‘veridical’ sensory awareness through the normal activity of the areas related to sensory self-monitoring. On the contrary, patients affected by anosognosia for hemianesthesia may develop a false belief of being able to feel somatosensory stimuli, because the activity in parietal regions (which are spared in our patients) is no more controlled by the damaged areas related to sensory self-monitoring.

Three of the four patients described in the present study had lesions in the superior temporal gyrus, damage to which has been associated with the presence of unilateral neglect (Karnath et al., 2005), especially when neglect is not accompanied by visual field deficits. Because these patients had neglect and no visual field deficit (while the other patient, SR, who did not have temporal damage did not show neglect), the temporal damage might be more related to the presence of a spatial impairment than to AHA.

In conclusion, the data reviewed in the present paper and the observations in patients with anosognosia for hemianesthesia seem to confirm that (1) awareness of sensory and motor deficits depends on the functioning of discrete self-monitoring processes that, when selectively affected by brain damage, cause domain-specific disorders of awareness, and (2) the cerebral circuits of self-monitoring processes for primary sensory and motor functions are located in different brain areas involved both in the execution of the primary functions and in the emergence of awareness related to the self-monitoring of the same functions. These facts reinforce the idea that self-consciousness is not a function of a central executive system hierarchically superimposed on other information processing systems, but rather is a function that is implemented in the same neural networks involved in the processes that have to be monitored (e.g., Berti et al., 2005).

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Appendix A

A.1. Intellectual impairment

As an index of patients’ general intellectual capacity, the Italian version of mini mental state examination (MMSE) is employed (Folstein, Folstein, & McHugh, 1975). Patients’ scores are corrected for age and educational level. In this test, the maximum score is 30 and a score of less than 23.80 is taken as an index of cognitive impairment.

A.2. Unilateral neglect

Extrapersonal neglect is diagnosed on the basis of the performance on cancellation tests (Albert, 1973; Diller & Weinberg, 1977; Gauthier, Deahault, & Joanette, 1989) and reading tasks (40 words and 9 sentences, Kinsbourne & Warrington, 1962). Patients are considered to be affected by extrapersonal neglect if one of these tests reveals a pathological performance. The results of cancellation tests are considered an index of left spatial neglect when the difference between left and right omissions exceeds three elements. Personal neglect is tested following Bisiach et al.’s methodology (Bisiach et al., 1986a), where patients are asked to reach their left hand with the right hand (eyes open). A four level scale is used, ranging from 0 (normal performance) to 3 (no attempt to reach the target).

A.2.1. Assessment of anosognosia for hemiplegia (AHP)

Anosognosia for hemiplegia is assessed through the traditional ‘brief interview’ described in other papers (Berti et al., 1996; Bisiach et al., 1986b; Marcel et al., 2004) and by more specific questions about the potential capability of performing specific movements and actions.

A.2.1.1. Brief interview. Patients were requested to answer a few preliminary questions about their present condition. These questions assessed what is usually referred to as verbal awareness of the disease and is a modified version of Bisiach’s test (Berti et al., 1996; Bisiach et al., 1986a, 1986b; Marcel et al., 2004).

A.2.1.2. Anosognosia score for the brief interview. Based on the interview, patients are classified as having no anosognosia, moderate anosognosia or severe anosognosia, as follows (Bisiach & Geminiani, 1991; Bisiach et al., 1986b):

0: the disorder is spontaneously reported or mentioned by the patient in reply to a general question about his complaints (no anosognosia);
A.2.1.3. Analytic and functional questions. We also evaluate motor awareness through more analytic questions about the potential ability of moving the left limbs. The patients are asked these questions before the actual execution of the movements. There are 15 questions (nine for the upper limb and six for the lower limb) (see Appendix B). The questions examine specific limb movements (analytic movements, three for the upper limb and three for the lower limb) and functional actions (three unimanual, three bimanual and three for the lower limb). Patients have to self-evaluate their motor abilities as follows: normal movement, movement with difficulty, no movement. A preliminary question was asked about the motor abilities of healthy side.

A.2.1.4. Anosognosia score for the analytic and functional questions. After requesting the actual execution of the movements (see below), the examiner judges the patient’s movements as normal movement, movement with difficulty, no movement. Awareness of the potential ability to perform movements is evaluated comparing the examiner’s judgment with the patient’s self-evaluation, as follows:

0: full accord in all questions (no anosognosia);
1: disagreement in one or two questions (moderate anosognosia);
2: disagreement in all questions (severe anosognosia).

A.2.2. Assessment of anosognosia for hemianaesthesia (AHA)

This section was concerned with patients’ explicit knowledge of the integrity of sensory perception. We used four questions: two questions were about tactile hemianaesthesia (one for the upper limb and one for the lower limb) and two questions about proprioceptive defects (one for the upper limb and one for the lower limb) (see Appendix B). For each question patients have to self-evaluate their perceptual abilities using a verbal judgment as follows: normal perception, perception with difficulty, no perception. A preliminary question was asked about the sensory abilities of the intact side.

A.2.2.1. Anosognosia score for hemianaesthesia. After the examination of patients’ sensory functions (see below), the examiner judged the patients’ performance as normal perception, perception with difficulty, no perception. Awareness of the potential ability to feel sensations was scored comparing the examiner’s judgment with the patient’s self-evaluation, as follows:

0: full accord in all questions (no anosognosia);
1: disagreement in one or two questions (moderate anosognosia);
2: disagreement in all questions (severe anosognosia).

In summary, the sequence of the tests was a brief interview, questions about awareness of hemiplegia, evaluation of sensory–motor conditions, scoring.

A.2.3. Motor and somato-sensory evaluations

A.2.3.1. Somato-sensory evaluation. Somato-sensory functions are tested with vision precluded.

A.2.3.2. Tactile anaesthesia. To examine tactile hemianaesthesia, subjects are asked to report light touch stimuli applied either on the hands or on the feet (in random order) answering “right” or “left”. Ten stimuli were delivered on each limb for a total of 40 stimuli (10 on the left arm, 10 on the right arm, 10 on the left foot, and 10 on the right foot).

The score is defined on the basis of performance of healthy controls:

0: normal perception. Ten or nine contralesional stimuli are detected;
1: moderate impairment. From 5 to 8 contralesional stimuli are detected;
2: severe defect. From 0 to 4 contralesional stimuli are detected.

It is worth noting that when a sensory ability is scored one, we do not evaluate the patient as having anosognosia to avoid any ambiguity related to the fact that the patient can actually report some stimulation.

A.2.3.3. Proprioceptive anaesthesia. To assess the patients’ sense of their joint position, the examiner placed their wrist and ankle in different positions and subjects had to report the direction their wrist and ankle were facing.

The score was defined on the basis of performance of healthy controls as follows:

0: normal perception. Ten or nine contralesional limb positions are detected;
1: moderate impairment. From 5 to 8 contralesional limb positions are detected;
2: severe defect. From 0 to 4 contralesional limb position is detected

It is worth noting that when sensory abilities are scored one, we do not evaluate that the patient has anosognosia to avoid any ambiguity related to the fact that the patient can actually report some stimulation.

A.2.3.4. Motor evaluation. Analytic/functional evaluation of motor functions. Motor abilities are examined by employing the 15 movements indicated in the questionnaire (nine arms and six legs). The examiner evaluates the patients motor abilities on the basis of their capacity to carry out the requested movements on a three level scale, as follows:

0: normal movements;
1: movements executed with difficulty; 2: movements absent.

As for the sensory domain, when the score for sensory abilities is one, we do not classify anosognosia to avoid any ambiguity related to the fact that the patient can actually make some movements.

Appendix B. Questionnaire

Brief interview
Where are we? Why are you now in the hospital? Is there anything wrong with you? How is your left arm? Can you move it? How is your left leg? Can you move it?

Anosognosia for somatosensory impairments

Upper limb
Tactile modality
How is sensation in your arm? Are you able to perceive a light touch on your left hand?

Proprioceptive modality
Do you have any problem in perceiving where your hand is if I move your left wrist in different positions?

Lower limb
Tactile modality
How is sensation in your leg? Are you able to perceive a light touch on your left foot?

Proprioceptive modality
Do you have any problem in perceiving where your foot is if I move your left ankle in different positions?

Anosognosia for hemiplegia

Upper limb

Analytic movements
Can you move your arm upward? Can you put your left hand on your left shoulder? Can you open and close completely your left hand?

Functional activities

Monomodal actions
Can you drink a glass of water using your left hand? Can you open a door with your left hand? Can you use the telephone with your left hand?

Bimodal actions
Can you open a bottle using two hands? Can you tie a knot using two hands? Can you wash your face with two hands?

Lower limb

Analytic movements
Can you bend your left hip moving your foot 10 cm across the floor? Can you straighten your left knee? Can you lift your left foot from the floor?

Functional activities
Can you walk? Can you climb the stairs? Can you ride a bicycle?

B.1. Assessment of joint position senses

To assess the patients’ sense of their joint position, the examiner placed their wrist and ankle in different positions and subjects had to report the direction their wrist and ankle were facing.

B.1.1. Joint position sense of wrist

(1) Flexion
(2) Radial deviation
(3) Ulnar deviation
(4) Extension
(5) Anatomical position
(6) Radial deviation
(7) Flexion
(8) Extension
(9) Anatomical position
(10) Ulnar deviation

B.1.2. Joint position sense of ankle

(1) Pronation
(2) Supination
(3) Dorsal flexion
(4) Plantar flexion
(5) Anatomical position
(6) Supination
(7) Dorsal flexion
(8) Pronation
(9) Plantar flexion
(10) Anatomical position

References


