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Imagining the impossible: Motor representations in anosognosia for hemiplegia☆

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ABSTRACT

Anosognosia for hemiplegia (AHP) is characterised by poor insight or underestimation of hemiplegia after brain injury. Recent explanations of AHP have used an established 'forward model', which proposes that normal motor awareness involves comparing the predicted and actual sensory consequences of movements. These accounts propose that AHP patients may be able to form representations of their intended movements (i.e., motor representations), but fail to register discrepancy between intended and actual movements. A prediction arising from this proposal is that AHP patients are able to generate motor representations involving their hemiplegic limb(s). Our study provides the first direct examination of this prediction in patients with AHP. We used an existing 'grip selection task', which investigates motor representations by comparing how patients *would* grasp an object and how they *actually* grasp the same object. Eight right hemisphere stroke patients with AHP, 10 control patients (non-AHP), and 22 age-matched healthy volunteers (HVs) completed the task. Results showed that HVs outperformed both AHP and non-AHP patients in their motor representations for the hemiplegic limb; however, the performance of AHP and non-AHP patients did not differ significantly. Motor representations for the intact limb were lower than normal in AHP patients, whereas performance in non-AHP patients was midway between the AHP and HV groups. Findings suggested that the ability to form motor representations lie on a continuum, but that impaired motor representations for the paralysed limb cannot account for AHP. Distorted motor representations, in combination with other deficits, might contribute to the pathogenesis of AHP.

Anosognosia refers to a disturbance of self-awareness occurring after brain injury, in which the patient does not recognise the presence or appreciate the severity of deficits in sensory, perceptual, motor, affective or cognitive functioning (Orfei et al., 2007). The term is most frequently applied to patients with hemiplegia following right hemisphere stroke (Ellis & Small, 1997; McGlynn & Schacter, 1989), i.e., anosognosia for hemiplegia (AHP). The clinical presentation of AHP is not uniform (Ellis & Small, 1993); for example, the extent of unawareness can vary considerably. Some patients fail to recognise, appreciate the severity, or acknowledge the consequences of paralysis (Orfei et al., 2007); others deny outright any motor impairment, while some patients acknowledge the presence of a motor deficit, but explain it away (Bisiach & Geminiani, 1991). Patients with AHP may or may not also exhibit unilateral neglect (i.e., a failure to respond to stimuli presented to the contralesional side) (Berti et al., 2005; Jehkonen, Laihosalo, & Kettunen, 2006). Furthermore, AHP can occur independently at verbal and non-verbal (i.e., behavioural) levels (Jehkonen et al., 2006). That is, AHP patients may refuse to acknowledge their paralysis, but are usually content to remain in bed (Bisiach & Geminiani, 1991). In contrast, some AHP patients verbally acknowledge their paralysis, but attempt to get out of bed or perform other physical tasks that are clearly impossible (Bisiach & Geminiani, 1991). These patients are often unaware of their inability to execute bilateral tasks requiring use of the plegic limb(s) (e.g., clap hands) when asked to make self-evaluations (Berti, Làdavas, & Della Corte, 1996; Berti, Làdavas, Stracciari, Giannarelli, & Ossola, 1998; Marcel, Tegnér, & Nimmo-Smith, 2004; Nimmo-Smith, Marcel, & Tegnér, 2005).

Despite several decades of research, we are still far from a clear understanding of the cognitive processes underlying AHP. This situation may be partly attributed to limitations in the methodological and theoretical approach employed by existing studies. First, the heterogeneous presentation of AHP has resulted in a lack of consensus about how best to characterise and assess the disorder. Unfortunately, it is impossible to draw valid comparisons across studies, identify commonalities in findings, and develop a

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cohesive understanding of AHP from the results of studies failing to thoroughly characterise the disorder. Therefore, a comprehensive assessment of AHP, taking into account both verbal awareness and self-evaluations of ability/behaviour, is necessary (Marcel et al., 2004; Nimmo-Smith et al., 2005). For example, Berti et al. (1996) have adopted this approach in their procedure for assessing AHP. This method provides a robust method for characterising AHP, from which a better understanding of AHP may be developed.

A second limitation of many accounts of AHP is a failure to provide a firm theoretical framework for understanding the pathogenesis of the disorder. These accounts attempt to explain only the pathological processes involved in AHP (e.g., Bisiach, Vallar, Perani, Papagno, & Berti, 1986; Cappa, Sterzi, Vallar, & Bisiach, 1987; Cutting, 1978; Levine, Calvanio, & Rinn, 1991; McGlynn & Schacter, 1989; Weinstein & Kahn, 1950). In contrast, recent cognitive neuropsychological accounts of AHP (Berti & Pia, 2006; Frith, Blakemore, & Wolpert, 2000a) provide theoretically robust and testable explanations of the disorder, by framing their accounts within an established 'forward' model of normal motor control and awareness (Wolpert, 1997; Wolpert, Ghahramani, & Jordan, 1995), the utility of which has been demonstrated by numerous studies in normal, healthy individuals (Blakemore, 2003; Blakemore, Frith, & Wolpert, 1999; Blakemore, Frith, & Wolpert, 2001; Blakemore, Goodbody, & Wolpert, 1998; Blakemore, Rees, & Frith, 1998; Blakemore, Wolpert, & Frith, 1998) and patients with abnormal awareness of action (Blakemore, Smith, Steel, Johnstone, & Frith, 2000; Blakemore, Wolpert, & Frith, 2002; Frith, 2005; Frith, Blakemore, & Wolpert, 2000b).

The forward model proposes that whenever a voluntary movement is executed two sources of information are produced: (i) somatosensory feedback reflecting the *actual* consequences of the movement, and (ii) a *prediction* of the expected sensory feedback arising from the intended movement. According to the model, these sensory predictions form the basis of motor awareness, whereas actual somatosensory feedback is not sufficient or necessary to construct knowledge of motor behaviour in normal individuals (Blakemore & Frith, 2003; Blakemore et al., 2001; Blakemore, Rees et al., 1998; Blakemore, Wolpert et al., 1998; Fourneret & Jeannerod, 1998; Haggard, 2005; Haggard, Clark, & Kalogeras, 2002; Haggard & Eimer, 1999; Wolpert & Flanagan, 2001). The forward model further proposes that normal motor awareness depends on a comparator, which checks for congruence between predicted and actual sensory feedback. When movement does not occur as planned the comparator detects a mismatch between predicted and actual sensory feedback, which produces conscious awareness of an error. This is particularly evident in situations when intended movement and sensory feedback do not match, such as when visual feedback regarding intended movements are reversed by use of a mirror (Fink et al., 1999), or when distinguishing between self-generated movements and those caused by another (Blakemore, 2003; Blakemore et al., 1999, 2001; Blakemore, Oakley, & Frith, 2003; Farrer et al., 2008).

Recent cognitive neuropsychological accounts (Berti & Pia, 2006; Frith et al., 2000a) have used the forward model to predict the pattern of intact and impaired functions that produce AHP. For example, Berti and Pia (2006) draw attention to the forward model's proposal that sensory predictions form the basis of normal motor awareness. This proposal implies that whenever a sensory prediction is created, and the comparator does not detect a mismatch with actual feedback, individuals might construct the belief that they have executed movement as intended. Accordingly, pathological awareness in AHP might occur if the ability to generate representations of intended movements (i.e., motor representations) and predict their expected sensory consequences were preserved, but patients fail to detect when these predictions are not congruent with actual sensory feedback. Under these circumstances motor awareness in AHP becomes based entirely on sensory predictions, which erroneously indicate successful execution of the intended movement. In contrast, hemiplegic patients without anosognosia (i.e., non-AHP) possess preserved awareness of their motor impairment because they are able to generate motor representations and detect when the predicted and actual sensory consequences of their movement do not match. This account is in direct contrast to an earlier 'feed-forward' hypothesis proposed by Heilman (1991), which assumed that AHP arises from a *loss* of intention to move. That is, if patients with AHP do not intend to move, the comparator is not primed to expect movement, and a subsequent lack of movement does not create a discrepancy signal or indicate an error to the AHP patient. However, this explanation is not supported by physiological studies (Berti, Spinazzola, Pia, & Rabuffetti, 2007; Hildebrandt & Zieger, 1995), which report normal muscle electrical activity (electromyography, EMG) in AHP patients instructed to move their hemiplegic limb. This activity suggests intact intention to move in AHP.

Although recent cognitive neuropsychological accounts of AHP lead to clear theoretical predictions, direct examined of these predictions is scarce. One prediction requiring examination is the ability of AHP patients to generate motor representations. Berti et al. (2005) have reported lesion analyses which suggest that AHP patients have some spared activity in pre-motor areas, which are known to be involved in planning movements and motor representations (Beltramello et al., 1998; Grèzes & Decety, 2001; Roland, 1993; Roland, Larsen, Lassen, & Skinhut, 1980); it is, therefore, possible that patients with AHP can form a distorted representation of intended movements (Berti et al., 2005). However, no existing behavioural study has directly examined motor representations in AHP.

The ability to generate motor representations has been examined in healthy individuals (Johnson, 2000b) and neurological patients (Buxbaum, Johnson-Frey, & Bartlett-Williams, 2005; Johnson, 2000a; Johnson, Sprehn, & Saykin, 2002) using motor imagery (i.e., "a dynamic state during which the representation of a given motor act is internally rehearsed within working memory without any overt motor output", Decety & Grèzes, 1999, p. 177). Johnson (2000b) developed a 'grip selection task' to assess motor representations in healthy individuals using implicit motor imagery (i.e., tasks requiring mental simulation of planned movements to solve, rather than instructing participants to explicitly imagine making particular movements; Johnson, 2000b). The task requires participants to first make a prospective motor imagery judgement (MI condition), indicating how they *would* grasp a wooden dowel/handle presented at several orientations (i.e., choosing to use either an overhand (pronated) or underhand (supinated) power grip, as one would clench the handle of a hammer). Participants are then presented with a real dowel in the same orientations as the MI condition and asked to actually grasp them using a power grip (motor control (MC) condition). Using this method, Johnson (2000b) found a near perfect correlation between MI and MC grip selections, suggesting that MI judgements are analogous to MC judgements in healthy individuals.

Motor imagery is also an ideal method for assessing motor representations in patients with hemiplegia, because it involves movement planning in the absence of overt execution. Johnson and colleagues (Johnson, 2000a; Johnson, Sprehn et al., 2002) have used a variant of the grip selection task to assess motor representations in acute and chronic hemiplegic stroke patients with preserved awareness (i.e., non-AHP), concluding that motor representations are intact in acute and chronic patients. However, a major limitation of both studies was a failure to include a healthy control group, which means it is not possible to determine whether the ability