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# Action-monitoring impairment in anosognosia for hemiplegia



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## ABSTRACT

Every movement begins with action programming, and ends with a produced effect. Anosognosia for hemiplegia (AH), involving unawareness of motor deficits after brain damage, is a striking but also poorly understood symptom in clinical neurology. It has been suggested that it may result from a combination of cognitive and sensorimotor dysfunctions, including impairments in monitoring motor action and detecting the mismatch between intention and outcome. Here we investigated the relationship between motor action awareness and monitoring of self-produced movements by using a motor imaginary task, which was performed with either the intact or the affected limb. We tested 10 right brain-damaged patients, including 5 with AH, in comparison with 5 healthy controls. In a first phase, participants were asked to either realize or imagine a movement with their right or left arm. In a subsequent recognition phase, the participants had to recall whether the movement was a realized or imagined and which arm was used. AH patients performed significantly worse relative to no-AH patients and healthy controls for the left movements. Specifically, we found that AH patients believed they had realized movements with their (paralyzed) left arm even when they failed in the left execution condition. However, they also made more errors for movements actually realized with the right hand. These findings confirm that impaired action monitoring may contribute to AHP. Furthermore, our results support the notion of an action control system integrating “feedforward” signals through a comparison process between the intention and execution of movement, but also indicate that monitoring deficits in AHP are not strictly unilateral. Combined together, dysfunction of motor comparator processes and more general monitoring deficits may add up to lead to unawareness of paralysis.

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## 1. Introduction

Babinski (1914; see Langer & Levine, 2014) described for the first time anosognosia as a neurological disorder that is characterized by a lack of interest and/or ignorance of brain-damaged patients for their deficits, such as hemiplegia after stroke (the most common presentation of anosognosia). These patients fail to recognize a severe motor loss despite direct confrontation during the neurological examination or everyday activities. This unawareness of paralysis may manifest itself in inappropriate behaviors or incorrect subjective reports (e.g., the patient claims that he/she is able to move the paralyzed left arm or that he/she has just moved it), but also in unrealistic judgments about the consequences of the deficit for actions (e.g., writing, dressing) and/or unrealistic decisions (e.g., the patients may want to get out of bed or go home). Hence, anosognosia for hemiplegia (AHP) has obviously important implications for clinical management and rehabilitation (Jenkinson, Preston, & Ellis, 2011). It is generally associated with poorer recovery and greater burden for caregivers. However, the cognitive and neural mechanisms of these phenomena are still poorly understood. Moreover, they are difficult to study systematically because of the prevalence of symptoms in the acute phase and the heterogeneity of clinical manifestations (Vocat, Staub, Stroppini, & Vuilleumier, 2010).

Many theories have been proposed since the initial description of this syndrome by Babinski (1994), but none appears definitive or sufficient to explain all aspects and variations of anosognosia (Jenkinson et al., 2011; Orfei et al., 2007; Vuilleumier, 2000). Babinski and others (Langer & Levine, 2014) have insisted on the role of proprioceptive deficits, but although these are frequent and contribute to AHP, double dissociations are commonly observed (Cocchini, Beschin, Fotopoulou, & Della Sala, 2010; Vocat et al., 2010). The same applies to spatial neglect including personal and body-centered forms (Vocat et al., 2010). Several scholars including Heilman and colleagues (Gold, Adair, Jacobs, & Heilman, 1994; Heilman, Barrett, & Adair, 1998) as well as Berti and colleagues (Pia, Neppi-Modona, Ricci, & Berti, 2004; Spinazzola, Pia, Folegatti, Marchetti, & Berti, 2008) highlighted the role of deficits altering brain mechanisms for the control of movement, notably based on current theories of movement control in the normal state (Frith, Blakemore, & Wolpert, 2000). According to these viewpoints, unawareness of motor paralysis might result from a loss of motor intention (Heilman, 2014) or a destruction of monitoring processes that normally allow the brain to compare the actual motor outputs with motor commands (Wolpert, Ghahramani, & Jordan, 1995). Such deficits may lead to an inability to detect a failure in moving the affected limbs. This view accords with current theories of motor control and motor awareness (Wolpert, et al., 1995) and helps explain a number of intriguing clinical phenomena (Jenkinson & Fotopoulou, 2010). For instance, patients with AHP appear unable to distinguish between the “real” production of movements and purely internal representations of movements (Fotopoulou et al., 2008; Jenkinson, Edelstyn, Drakeford, & Ellis, 2009; Pia et al., 2013),

Moreover, motor imagery can still operate for the paralyzed limb despite AHP (Garbani et al., 2012; for review Vuilleumier, 2000).

In parallel, however, there is evidence that AHP is not a unitary phenomenon but varies according to the test procedures or patients. For instance, Marcel, Tegner, and Nimmo-Smith (2004) showed that judgment of motor performance is particularly overestimated by patients with AHP in self-related conditions (first person perspective), rather than when the same judgments are made by imagining another person in the same conditions (third-person perspective), which may reflect a more general weakening overall cognitive functioning or more extensive monitoring deficits. In addition, patients with AHP may show deficits in monitoring processes not directly related to movement but related to perception of visual events (Feinberg, 2007; Jenkinson, Edelstyn, Drakeford, Roffe, & Ellis, 2010) or to performance in other cognitive tasks (Marcel et al., 2004; Vocat, Saj, Vuilleumier, 2013), which might also contribute to the pathogenesis of AHP.

Given the difficulty to pinpoint a single mechanism responsible for AHP, several recent hypotheses (e.g., Davies, Davies, & Coltheart, 2005; Fotopoulou, 2012, 2014; Moro, 2013; Orfei et al., 2007; Prigatano, 2009; Starkstein, Jorge, & Robinson, 2010; Vuilleumier, 2004) have proposed that this phenomenon may emerge from a combination of deficits (see Vocat & Vuilleumier, 2010). For instance, Davies et al. (2005) suggested that AHP results from both an illusion (leading to delusional beliefs) and impaired evaluation of the belief process (leading to uncritical acceptance of the illusion). Similarly, Vuilleumier (2004) proposed a general “ABC model” of anosognosia, where a combination of deficits affecting at least three main domains (assessment, belief, and control operations) may interact in order to give rise to awareness of motor losses, or instead cause anosognosia or denial (see also Vocat & Vuilleumier, 2010; Vuilleumier, Vocat, & Saj, 2013). Hence, besides impairment in motor control (Bottini et al., 2009; Heilman, 2014; Pia et al., 2004), additional deficits in cognitive and motivational/emotional processes related to performance monitoring and belief shifting could also contribute to anosognosia (i.e., Check operations in ABC model). This hypothesis is supported by recent findings in a “riddle test” (Vocat et al., 2013). In this task, patients have to find a target word, initially unknown to them, based on successive verbal cues, and indicate their level of confidence for each response. Patients with AHP were found to exhibit abnormally high confidence in their guesses, unlike hemiplegic patients without AHP, reflecting selective difficulties to appraise uncertainty and to experience doubt about uncertain beliefs, even when contradicted by external evidence (e.g., additional riddle cues incompatible with initial guess). In a detailed case study, Venneri and Shanks (2004) also showed that persistent anosognosia is associated with a failure to assess the veracity of mental contents about current abilities beyond motor control. More recently, Fotopoulou (2012) proposed a more general model based on computations of ‘prior beliefs’ and ‘prediction errors’. According to this view, the ability to change beliefs and the ability to monitor performance under uncertainty could play a critical role in AHP in combination with other primary deficits in sensory, motor and/or attentional functions.

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