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Anosognosia for motor impairments as a delusion: Anomalies of experience and belief evaluation

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Abstract
We put forward a two-factor account of anosognosia for hemiplegia—more generally, anosognosia for motor impairments—considered as a delusion. Anosognosia is a patient's lack of knowledge of their illness or impairment, and patients who lack knowledge of their motor impairments believe that they can still move limbs that are, in reality, paralysed. This belief fits the *DSM-5* definition of delusion.

In our two-factor account of anosognosia as a continued-belief delusion, the first factor—an impairment of the motor control system—results in an anomaly of experience. When patients try, but fail, to move their left arm there is an anomalous absence of immediate bodily experience of movement failure—perhaps accompanied by an illusory experience of successful movement. However, even without immediate experience of movement failure, other evidence of the motor impairments would be available—including evidence from everyday mishaps consequent on the motor impairments. The second factor in our two-factor account results in patients being unable to use this evidence to evaluate and reject the delusional belief and achieve knowledge of their motor impairments. Cognitive impairments of memory, error detection, executive function or working memory are candidate second factors that could result in this failure of belief evaluation.

Anosognosia is a patient's lack of knowledge of their illness or impairment: *a-* (without); *-noso-* (disease); *-gnosia* (knowledge). The French neurologist Joseph Babinski (1914) introduced the term as applying to patients' lack of knowledge of their hemiplegia but the term has a more inclusive use (as the etymology would suggest). Patients may be described as having anosognosia for their visual impairments, memory impairments, cognitive impairments, and so on. In this chapter, we shall focus on "the anosognosia of Babinski" (see Langer, 2009, p. 390); that is, anosognosia for hemiplegia or, more generally, for motor impairments. Anosognosia for motor impairments may follow a left-hemispheric or right-hemispheric stroke but we shall mainly discuss anosognosia for left-side motor impairments—specifically, impairment of the left arm—following a right-hemispheric stroke.

Anosognosia is usually assessed by a structured interview.[1] Interviews typically begin with general questions about the patient's health and why the patient is in hospital, and then move on to more specific questions about the motor impairment, and whether the patient is able to move the left arm. A patient who claims to be able to move the paralysed left arm may be asked to raise the left arm.

Patients are classified as having no anosognosia, or mild, moderate, or severe anosognosia, using scoring criteria such as the Bisiach Anosognosia Scale (Bisiach, Vallar, Perani et al., 1986, pp. 472–473):

> 0 The disorder is spontaneously reported or mentioned by the patient following a general question about his complaints [No anosognosia].
> 1 The disorder is reported only following a specific question about the strength of the patient's left limbs [Mild anosognosia].
> 2 The disorder is acknowledged only after its demonstration through routine techniques of neurological examination [Moderate anosognosia].
> 3 No acknowledgement of the disorder can be obtained [Severe anosognosia].

Many studies classify patients as having anosognosia only if they have a score of 2 or 3 on the Bisiach Anosognosia Scale. If a patient's left-side motor impairment is not total and some movement of the arm is possible then an affirmative answer to the question, "Can you move your left arm?", is actually *true*. For this reason, some studies include only patients whose motor impairment is total. An alternative approach is to modify the interview questions to assess whether the patient is, despite having some movement, underestimating the degree of impairment (see Vocat, Staub, Stroppini, & Vuilleumier, 2010).

In the interview used by Berti, Làdavas and Della Corte (1996), the first group of questions includes both general questions and questions specifically about the motor impairment: "Where are we? Why are you in hospital? How is your left arm? Can you move it?" (p. 429). If patients claim to be able to move the left arm then they are asked: "Please, touch my hand with your left hand. … Have you done it? … Are you sure? It is very strange because I have not seen your hand touching my hand" (pp. 429–432). Patients are classified as having no anosognosia, or mild or severe anosognosia, using the following criteria (p. 432):

[1] See Aimola Davies, White and Davies (2010, Table 23.2) for a review of the questions used in nine structured interviews for anosognosia for motor impairments published between 1952 and 2009; also see Nurmi and Jehkonen (2014, Table 2) for a review of assessment methods published between 1978 and 2013.

0 The patient answered correctly to the first group of questions [No anosognosia].
1 The patient acknowledged being in the hospital and/or being affected by a stroke, but denied his or her upper limb impairment. However, the patient acknowledged that the left arm did not reach the examiner's hand (Mild anosognosia).
2 The patient claimed that he or she had reached the examiner's hand (Severe anosognosia).

Reported rates of occurrence of anosognosia for motor impairments vary quite widely.[2] In one study of fifty-eight patients with left-arm motor impairment (Vocat et al., 2010), fifty patients were assessed within five days following a right-hemispheric stroke and sixteen (32%) had a score of 2 or 3 on the Bisiach Anosognosia Scale. Eight (18%) of forty-four patients assessed in the second week following stroke had a score of 2 or 3, and one (5%) of nineteen patients examined six months after stroke still had a score of 3 (none had a score of 2).

In this chapter, a distinction is drawn between three kinds of knowledge that may be lacking in patients with anosognosia for motor impairments: knowledge of movement failure when it occurs; more lasting knowledge of motor impairments; and knowledge of the consequences of those motor impairments for everyday activities (Section 2).

Patients who lack knowledge of their motor impairments believe that they can still move limbs that are, in reality, paralysed. This belief fits the definition of delusions, and anosognosia for motor impairments can be considered against the background of a two-factor theory of monothematic delusions. According to the two-factor theory, a first factor results in an anomalous experience that prompts the delusional idea or hypothesis and a second factor explains why the idea or hypothesis is adopted and maintained as a belief rather than being rejected on the basis of available evidence and background knowledge that counts against it (Section 3).

In our two-factor account of anosognosia for motor impairments as a delusion, a first factor—such as an impairment of the motor control system—results in the anomalous *absence* of immediate bodily experience of movement failure. However, even without immediate experience of movement failure, other evidence of the motor impairments would be available (including evidence from everyday mishaps consequent on the motor impairments). A second factor results in patients being unable to use this evidence to reject the delusional belief, "I can move my left arm", and achieve knowledge of their true condition. A patient might be unable to remember the evidence for long enough to make use of it, or unable to recognise that the evidence is incongruent with current beliefs, or unable to carry out the cognitively demanding task of belief evaluation. Consequently, cognitive impairments—of memory, error detection, executive function or working memory—are candidate second factors in anosognosia for motor impairments (Sections 4 and 5).

[2] See Aimola Davies et al. (2010, Table 23.3) for occurrence rates in 21 studies published between 1952 and 2009.

The chapter begins (Section 1) with phenomenology—patients' experience, or absence of experience, of trying but failing to move a paralysed limb—and its underpinnings in the motor control system.

1. Experience of Movement Failure

Definitions of anosognosia often mention *unawareness* of impairment and, indeed, the translation of Babinski's original paper uses that term: "the patients are *unaware* of … the existence of the paralysis which affects them" (1914/2014, p. 6; emphasis added).[3] The term "unawareness" is certainly used to express a lack of knowledge but "awareness" and "unawareness" also suggest the presence or absence of conscious experience (sensation and perception). We regard anosognosia as a failure or pathology at the level of knowledge and belief. Patients fail to know about their left-arm motor impairment and believe, instead, that they do not have the impairment and *can* still move the left arm. Patients may also sometimes believe that they *are moving* the left arm—for example, in response to a request to reach and touch the examiner's hand. It is important to distinguish between failure to *know* about one's own motor impairments and failure to *experience* one's own movement failures. (Throughout this chapter, we shall use the terms "awareness" and "unawareness" only for the presence or absence of conscious experience, and not for the presence or absence of knowledge.)

1.1 A model of motor control

The experience of movement failure (and the absence of such experience) can be understood in terms of a well-established model of motor control (Frith, Blakemore, & Wolpert, 2000; Wolpert, 1997; Wolpert & Flanagan, 2001). The key components in the model of motor control are *predictors* and *comparators*. Predictors or forward models capture the relationship between actions and their consequences. They use information about the current state of the system and about motor commands to predict the future state of the system and the resulting sensory feedback. Comparators detect any mismatch between the predicted sensory feedback and the actual feedback, and generate *prediction-error signals*.

Consider, first, a hypothetical case of hemiplegia without anosognosia. The patient wants to raise their left arm (the arm being in the raised position is the *desired state*); and they try to raise their arm. A motor command is issued. A predictor within the motor control system uses an efference copy of the motor command to generate a representation of the predicted position of the arm when the attempted movement is complete (*predicted state*) and a representation of the predicted proprioceptive feedback. The paralysed arm does not move but remains by the patient's side (*actual state*). A comparator detects the substantial disparity between the predicted position of the arm, as represented by the predicted proprioceptive feedback, and the actual position of the arm, as represented by proprioception, and a prediction-error signal is generated. Consequently, the patient has an immediate bodily experience of trying, but failing, to raise their arm—"concurrent awareness" of the specific movement failure (Marcel, Tegnér, & Nimmo-Smith, 2004, p. 34).

[3] In the original French: "les malades ignorent … l'existence de Ia paralysie dont ils sont atteints" (Babinski, 1914, p. 845).

Now consider, in contrast, a hypothetical case of hemiplegia in which *additional* impairments of the motor control system are present. As a result of these impairments, no disparity between a predicted state and an actual state is detected and no prediction-error signal is generated. Consequently, the patient does not have an immediate experience of failing to raise their arm—“concurrent unawareness” of the specific movement failure. Several proposals have been made about impairments of the motor control system that would have this consequence.

1.2 Explaining the absence of experience of movement failure

Heilman (1991, 2014; Heilman, Barrett, & Adair, 1998; Heilman & Harciarek, 2010) has proposed that, in at least some cases of anosognosia for motor impairments, there is a *motor intention* deficit. The patient desires to move their left arm but no motor command is issued. Without an efference copy of a motor command, even an intact predictor will generate neither a representation of a predicted raised position of the arm, nor a representation of predicted proprioceptive feedback consequent on that new position. The comparator may have access to information from proprioception about the actual position of the immobile left arm, but it has no information about a predicted different position. Without two represented positions to compare, the comparator detects no disparity, and no prediction-error signal is generated. Consequently, the patient does not have an immediate experience of failing to move their left arm.

This motor intention theory may account for some cases of absence of experience of movement failure, but an increasing body of evidence supports the claim that at least some patients with anosognosia are able to generate motor intentions (e.g., Berti, Spinazzola, Pia, & Rabuffetti, 2007; Garbarini, Rabuffetti, Piedimonte et al., 2012; Jenkinson & Fotopoulou, 2010; Pia, Spinazzola, Rabuffetti et al., 2013; Piedimonte, Garbarini, Rabuffetti et al., 2015). In some cases, the patient has an illusory bodily experience of successful movement—a kinaesthetic hallucination or illusory limb movement (Feinberg, Roane, & Ali, 2000; Marcel et al., 2004; Vocat et al., 2010). These illusory movements of the paralysed limb are not well accounted for by Heilman’s (1991) motor intention theory (also see Heilman, 2014) because the experience of moving the arm depends primarily on the motor commands and consequent predictions of movement, and on the match between the desired state and the predicted state (Blakemore & Frith, 2003; Frith et al., 2000).

Suppose now that motor intentions are intact and that the comparator receives information about the predicted position of the left arm. There are at least two circumstances in which the comparator would not generate a prediction-error signal and so the patient would not have an immediate experience of movement failure. One possibility is that, because of impaired proprioceptive feedback, the comparator may not receive information about the actual position of the left arm. In such a case, the comparator would not have two represented positions—predicted and actual—to compare. A second possibility is that no prediction-error signal is generated, despite the fact that information about the predicted and actual positions of the left arm is available. There may be damage to the comparator itself (Berti, Bottini, Gandola et al., 2005, p. 490; Berti & Pia, 2006, p. 247; Berti et al., 2007, p. 169; also see Garbarini, Cecchetti, Bruno et al., 2019) or the comparator’s threshold for generating a (conscious) prediction-error signal might be pathologically raised as a result of increased inherent noise in the motor system (Preston, Jenkinson, & Newport, 2010; Preston & Newport, 2014). Preston et al. (2010) suggested that, in anosognosia:

> the raising of the comparator thresholds is such that all movements are treated as both self-produced and accurate. Thus, when a movement is intended, and, importantly, a motor programme produced, the movement is treated as self-produced, even though no actual movement has taken place. (p. 3449).

Proprioceptive loss often co-occurs with anosognosia for motor impairments, and so it might not be straightforward to provide clear cases of the second possibility described in the previous paragraph. However, predictors in the motor control system predict, not only the proprioceptive feedback, but also the visual feedback that will result from the predicted state; and this predicted visual feedback is compared with actual visual feedback. Thus, there is an opportunity to test the hypothesis that, in at least some patients with anosognosia, motor intentions and predictions of movement are generated and visual feedback is available, yet no disparity between predicted and actual state is detected. In a seminal experiment, Fotopoulou, Tsakiris, Haggard et al. (2008) provided evidence that, in anosognosia for motor impairments, predictions flowing from motor intentions "dominate over sensory feedback" (p. 3433).

In this study, four patients with anosognosia for a motor impairment of the left arm following a right-hemispheric stroke and four control patients (with the left arm paralysed, but without anosognosia) were provided with false visual feedback from a realistic prosthetic hand that could be moved (unbeknown to the patient) by an assisting experimenter. The prosthetic hand was positioned (aligned with body midline) on a table in front of the patient, and was accepted by the patient as being their own hand.[4] In the critical condition (12 trials), patients were asked to raise their left hand. On six trials, the prosthetic hand moved as if the patient had succeeded in raising it, and on the other six trials, the prosthetic hand did not move. Patients were asked whether their hand moved. Patients with anosognosia reported that the hand moved, not only when it did move, but also on a substantial majority of trials when it did not move. In contrast, patients without anosognosia were able to discriminate between movement and no movement of the prosthetic hand. In two other conditions, patients were instructed not to move their left hand. In one condition (12 trials), patients were told that the assisting experimenter would passively lift their left hand for them; in the other condition (12 trials), they were told that the assisting experimenter would not attempt to lift their left hand. In each of these two conditions, the prosthetic hand moved on six of the 12 trials and both groups of patients were reliably able to discriminate between movement and no movement. Thus, anosognosia patients failed to detect a substantial disparity between predicted and actual visual feedback from the prosthetic left hand, but only when they themselves attempted to move their left hand.

[4] A suitable rubber hand was selected for each patient, so that the rubber hand resembled the patient's own hand. The patients' belief that the rubber hand was their own hand was confirmed before, during, and after the experiment, and patients "did not doubt the rubber hand was theirs" (Fotopoulou et al., 2008, p. 3437).

1.3 From experience to knowledge of movement failure

Experience, or absence of experience, of movement failure is likely to have consequences for a patient's *knowledge of movement failure at the time it occurs*, or false beliefs about successful movement. First, an immediate experience of trying, but failing, to raise the left arm will normally provide a patient with knowledge of the movement failure when it occurs. The patient will know that they have *just now* failed to move their left arm. Conversely, a patient's knowledge that they just now failed to move their arm is naturally interpreted as evidence that they had an experience of movement failure. Nevertheless, *experience* of movement failure and *knowledge* of movement failure are conceptually dissociable in both directions. Patients having a bodily experience of movement failure might be sceptical about the veridicality of this experience and might still believe that their arm moved (*experience of movement failure without knowledge of movement failure*). Equally, patients who were well informed about their condition but did not have a bodily experience of movement failure might still know that their arm did not move (*knowledge of movement failure without experience of movement failure*).

Second, a patient who does not have an immediate experience of failing to move the left arm when they try may have a concurrent *belief in successful movement*, especially if they experience an illusory movement of the paralysed limb. The patient may believe that they are *at this moment* moving their left arm.[5] Conversely, a patient's belief that they are at this moment moving their arm is naturally interpreted as evidence that they are experiencing an illusory limb movement. Nevertheless, the *experience* of successful movement and the *belief* in successful movement are conceptually dissociable in both directions. Well-informed patients experiencing an illusory limb movement might still know that their arm was not really moving (*experience of successful movement without belief in successful movement*). Equally, patients with anosognosia might conceivably believe, not only that they can move their left arm, but also that they are actually moving their arm at this very moment, even without an illusory limb movement (*belief in successful movement without experience of successful movement*).

2. Knowledge: A Threefold Distinction

In this section, we introduce a distinction between three kinds of knowledge that may be lacking in anosognosia for motor impairments. The *knowledge of movement failure* at the time it occurs (concurrent knowledge), which is normally provided by an immediate experience of trying but failing to raise the left arm, is distinguished from more lasting *knowledge of motor impairments* themselves, and *knowledge of the consequences* of motor impairments for everyday activities.

2.1 Concurrent knowledge of movement failure and knowledge of motor impairments

Knowledge of movement failure at the time it occurs may naturally lead to revision of long-held—but now false—beliefs, so that patients come to know that they are no longer able to move their left arm. But there is a clear distinction between concurrent knowledge of a movement failure and the relatively stable state of knowing about one's motor impairment. Patients who sometimes experience movement failure and concurrently know that they have

[5] See Babinski (1914) patient 2; Berti et al. (2007) patient CR; Berti, Làdavas, Stracciari et al. (1998) patient CC; Levine, Calvanio, & Rinn (1991) patient 6; Ramachandran (1995) patient FD.

failed to move their left arm might not be able to consolidate that information and might not achieve a lasting state of knowledge of their impairment. Equally, patients who never concurrently know that they have tried, but failed, to move their left arm may, on the basis of other available evidence, achieve a stable state of knowledge that they can no longer move their left arm.

This distinction is not a merely conceptual one. Marcel et al. (2004) assessed concurrent *knowledge of movement failure* by asking patients to raise each limb with vision precluded and, immediately afterwards, to evaluate their own motor performance. They also assessed *knowledge of motor impairments* by asking questions such as "Can you move your arms normally?" and "Is either of your arms weak?" (2004, p. 40), without asking patients actually to attempt any movement. More patients "overevaluated" their left-arm motor performance (demonstrating lack of concurrent knowledge about their failure to move the arm with vision precluded) than failed to acknowledge their motor impairment of the left arm (p. 26). In fact, Marcel et al. reported a double dissociation between lack of concurrent knowledge of movement failure and lack of knowledge of motor impairments, although the two conditions were highly associated (p. 32).

2.2 Knowledge of motor impairments and knowledge of the consequences

There is also an important distinction between *knowledge of motor impairments* themselves and *knowledge of the consequences* of those motor impairments for activities of daily living, such as washing, dressing and eating, and for other everyday tasks requiring two hands, such as tying a knot or carrying a large tray of glasses. Here, we consider the failure to appreciate the consequences of motor impairments as a further failure at the level of knowledge and belief, which can be assessed by asking patients to rate their abilities to perform everyday tasks.[6]

Marcel et al. (2004) asked patients questions about activities of daily living (e.g., "In your current state do you have any problems with dressing?"; 2004, p. 40) and about bimanual tasks (e.g., "In your present state how well, compared with your normal ability, can you tie a knot?"; p. 24). While twelve of forty-two patients with a left-arm motor impairment following right-hemispheric stroke failed to acknowledge their motor impairment itself, many more overestimated their ability to engage in activities of daily living and to carry out everyday bimanual tasks. Twenty-two patients failed to acknowledge their problems with two or more of four activities of daily living and twenty-four overestimated their ability for at least five of eight bimanual tasks. Thus, a substantial number of patients who explicitly acknowledged their motor impairment failed to appreciate the consequences of the impairment; they explicitly overestimated their ability to engage in activities of daily living and to perform everyday bimanual tasks (p. 27). These patients had anosognosia for the consequences of their motor impairment persisting longer than anosognosia for the impairment itself.

In the interview used by Berti et al. (1996), patients are asked to assess their ability to perform ten bimanual tasks (e.g., tie a knot, open a bottle), and five unimanual tasks (e.g., eat with a

[6] We do not equate this anosognosia for the consequences of motor impairments with the indifference, or lack of concern, for which Babinski (1914/2014) introduced the term "anosodiaphoria" (p. 7): "I have also observed some hemiplegics who, without being unaware of the existence of their paralysis, seemed not to attach any importance to it, as if it were a matter of an insignificant discomfort" (also see Langer, 2009, p. 391).

fork) assessed separately for the left and the right hand (p. 432).[7] Berti et al. found a double dissociation between anosognosia for the upper-limb motor impairment itself and anosognosia for the consequences of this motor impairment. Two patients overestimated their ability to perform bimanual tasks and unimanual tasks with the left hand—anosognosia for the consequences of their left-arm motor impairment—but had no anosognosia for the impairment itself (pp. 434–435, patients L.O. and A.P.). While Marcel et al. did not describe patients with the reverse dissociation, Berti et al. reported two patients with anosognosia for their upper-limb motor impairment itself, who gave realistic estimates of their ability to perform bimanual tasks and unimanual tasks with the left hand—no anosognosia for the consequences of the impairment (ibid., patients M.A. and M.E.).

2.3 The threefold distinction and assessment of anosognosia

Aimola Davies, White and Davies (2010, Table 23.4) presented an anosognosia interview that is structured in accordance with this threefold distinction. In the assessment of concurrent *knowledge of movement failure*, patients are requested, with vision precluded, to raise each arm, and then both arms, to shoulder level and are first asked questions such as, "Did it *feel* to you as if your arm was rising?". This initial stage of the assessment provides information about patients' immediate experience of movement failure, or illusory experience of successful movement, as they tried to move the affected limb. If a patient reports an illusory limb movement then the examiner continues with questions such as, "Do you believe that, when it felt as if it [this arm] was moving, it really did move?" (see Table 23.4, Q5, Step 2 'Experience' and Step 3 'Post-Performance Evaluation').

In the assessment of *anosognosia for motor impairments*, patients are asked to perform the same actions again—that is, to raise each arm, and then both arms—but now with vision permitted, so that evidence of success or failure is maximally available. Patients are asked to rate their ability to perform the action both before (prior belief) and after (posterior belief) each attempt (see Table 23.4, Q6 and Q7). An unrealistic prior belief but a realistic posterior belief is similar to a score of 2 (moderate anosognosia) on the Bisiach Anosognosia Scale; an unrealistic prior belief *and* posterior belief is similar to a score of 3 (severe anosognosia).

In the assessment of *anosognosia for the consequences* of motor impairments, patients are asked to rate their ability to carry out everyday tasks, including bimanual tasks such as tying a knot. In some cases, patients are asked to describe how they would perform the task. Patients are asked actually to perform some tasks for which clear evidence of success or failure will be available (e.g., attaching a handkerchief to a ring by tying a knot; using the affected foot to push a ball toward the examiner) and to provide ratings of their ability both before (prior belief) and after (posterior belief) their attempt (see Table 23.4, Q8 and Q9).

[7] Berti et al.'s (1996) procedure for assessing anosognosia for the consequences of motor impairments was attributed to a 1994 poster presentation by Marcel and Tégner. Della Sala, Cocchini, Beschin, and Cameron (2009) subsequently developed the *Visual-Analogue Test for Anosognosia for motor impairment* (VATA-m), which assesses anosognosia for the consequences of a motor impairment and provides normative data for diagnosis, as well as being suitable for patients with language impairments.

3. Delusion: A Two-Factor Explanatory Framework

In this chapter, we conceptualise anosognosia as a delusion, in accordance with the *DSM-5* definition: "*Delusions* are fixed beliefs that are not amenable to change in light of conflicting evidence" (APA, 2013, p. 87). Our strategy (Aimola Davies, Davies, Ogden et al., 2009; Davies, Aimola Davies, & Coltheart, 2005) will be to consider anosognosia against the background of a two-factor theory that was offered, in the first instance, as a schema for explanations of monothematic delusions of neuropsychological origin. We begin with a brief account of the two-factor theory of delusion.[8]

3.1 The two-factor theory of delusion

The starting point for the two-factor theory is that a case of delusion can be explained by answering two questions:

> **First question:** What initially prompted the delusional idea or hypothesis? What brought it to mind?
> **Second question:** Why was the delusional idea or hypothesis adopted and maintained as a belief rather than being rejected—as it should have been—on the basis of available evidence and background knowledge that counted against it?

The answers to these questions indicate two factors in the explanation of the case of delusion.

Maher (1974, 1999) proposed that a delusional idea or hypothesis arises from an attempt to explain an anomalous experience which, in turn, resulted from a neuropsychological deficit or anomaly. According to the two-factor theory, this is broadly correct. The first factor, which may be neuropsychological in nature, results in an anomalous experience—or, more generally, results in observation of a surprising fact or event—which prompts the delusional idea or hypothesis as a possible explanation. Different delusional hypotheses are usually prompted by observation of different surprising facts or events, resulting from different first factors.

An answer to the first question would not, by itself, explain a case of delusion because a delusional hypothesis is not yet a delusion. It is not even a belief, though it is a candidate for belief. When a delusional hypothesis comes to mind, it should be critically evaluated and rejected on the basis of available evidence and background knowledge but, in a case of delusion, this does not happen. Instead, the delusional hypothesis is adopted and maintained as a belief. The role of the second factor is to explain this failure of hypothesis evaluation.

The two-factor theory has been applied to a wide range of monothematic delusions including:

- Capgras delusion—"This person I am looking at [e.g., the patient's mother] is a stranger, not my mother. My mother has been replaced by an impostor" (Capgras & Reboul-Lachaux, 1923; Ellis & Young, 1990);
- Cotard delusion—"I am dead" (Cotard, 1882; Young & Leafhead, 1996);

[8] See Coltheart, 2007, 2010; Coltheart & Davies, 2021; Coltheart, Langdon & McKay, 2011; Davies & Coltheart, 2000, in press; Davies, Coltheart, Langdon, & Breen, 2001; Langdon & Coltheart, 2000.

- Mirrored-self misidentification—“The person I see when I look in the mirror is not me” (Breen, Caine, & Coltheart, 2001; Breen, Caine, Coltheart et al., 2000);
- Fregoli delusion—“People with whom I am familiar are present in my environment, disguised” (Courbon & Fail, 1927; Langdon, Connaughton, & Coltheart, 2014);
- Somatoparaphrenia—“This [body part; e.g., the patient’s left arm] is not mine, it is someone else’s” (Halligan, Marshall, & Wade, 1995; Vallar & Ronchi, 2009); and
- The delusion of alien control—“Someone else is able to control the movements of my body” (Frith, 1992; Stirling, Hellewell, & Quraishi, 1998).

For these and other delusions, plausible first factors have been identified (Coltheart & Davies, 2021, 2022; Davies & Coltheart, 2022, in press) and there is reason to propose that, in neuropsychological cases of delusion, the second factor—resulting in failure of hypothesis evaluation—has its neural basis in damage to, or hypoactivation of, right dorsolateral prefrontal cortex (Coltheart, 2007, 2010; Coltheart, Cox, Sowman et al., 2018).

3.2 The cognitive nature of the second factor in delusions

A proposal about belief formation made by Stone and Young (1997) provides a starting point for further reflection on the cognitive nature of the second factor in delusions:

> The belief formation system contains within it a permanent tension between *two* principles that can come into conflict: a tension between forming beliefs that require little readjustment to the web of belief (conservatism) and forming beliefs that do justice to the deliverances of one’s perceptual systems [observational adequacy]. (p. 349)

A balance needs to be struck between these two cognitive imperatives: *conservatism*, that is, minimising adjustment of the pre-existing web of belief; and *observational adequacy*, that is, doing justice to one’s own perceptual experience. In delusions, the balance tips too far toward observational adequacy, at the expense of conservatism. (The Bayesian analogue of the balance tipping too far toward observational adequacy is giving too much weight to likelihoods at the expense of prior probabilities. The predictive coding analogue is giving too much weight to prediction errors at the expense of prior beliefs, understood as internal predictive models of the world.) The delusional hypothesis may have arisen as an urgently needed explanation of a surprising fact or event. But the imperative to do justice to one’s own perceptual experience needs to be inhibited, so that more conservative considerations of plausibility given one’s existing web of background knowledge and beliefs can be taken into account.

The critical evaluation that would allow a person to reject a delusional hypothesis seems to require two kinds of resources. First, it requires taking control of the balance between competing cognitive imperatives; it requires “suspending automatic biases in order to critically evaluate different hypotheses, ‘re-initialized’ as having equal priority” (Langdon & Coltheart, 2000, p. 204). Second, critical evaluation requires assessing a hypothesis in the light of evidence and plausibility—weighing up evidence and plausibility considerations and working out what to believe. This weighing up and working out should take account of the surprising fact or event that prompted the hypothesis, a mass of other recent evidence, the person's pre-existing background knowledge and beliefs, and knowledge available from family, friends and other sources.

Thus, on general theoretical grounds, it is plausible that the critical evaluation that would allow a person to reject a delusional hypothesis requires executive processes, including some inhibitory processes, and also working memory resources for the maintenance and manipulation of information. In short, hypothesis evaluation seems to be a good example of an *executive working memory* task (Engle, 2002; see also Smith & Kosslyn, 2007, p. 259: "The central executive is what does the 'work' in working memory."). It is for this reason that we have proposed (Aimola Davies & Davies, 2009) that the second factor in the two-factor theory of delusion may involve impairments of executive function or working memory.

4. Explaining Anosognosia as a Continued-Belief Delusion: A Two-Factor Account

Anosognosia for motor impairments, especially in its severe form, fits the *DSM-5* definition of delusions as "fixed beliefs that are not amenable to change in light of conflicting evidence" (APA, 2013, p. 87). However, there is an important difference between anosognosia and other monothematic delusions such as Capgras delusion, Cotard delusion, mirrored-self misidentification, Fregoli delusion, somatoparaphrenia, and the delusion of alien control. In those familiar examples of monothematic delusion, the delusional belief is newly adopted and somewhat exotic but, in anosognosia, the delusional belief is long held and commonplace. Patients with anosognosia have believed for many decades that they can move their left arm, and that they can tie a knot or carry a large tray of glasses. Following a right-hemispheric stroke, however, those beliefs are no longer true.

Anosognosia for motor impairments is a *continued-belief delusion*; Capgras delusion and the other examples are, in contrast, *new-belief delusions*. Anosognosia is not the only continued-belief delusion; another example is "a delusional belief in the fidelity of a romantic partner" (reverse Othello syndrome; Butler, 2000, p. 85). In cases of continued-belief delusion, reality has changed in a way that would normally lead to substantial revisions to long-held beliefs but, instead, the beliefs persist.[9]

When we say that we conceptualise anosognosia as a delusion, our point is not that anosognosia is sometimes accompanied by new-belief delusions. It is, indeed, true that anosognosia may be accompanied by a new-belief delusion such as somatoparaphrenia (Moro, Pernigo, Tsakiris et al., 2016). But our point is that the core belief in anosognosia, "I can move my left arm", is itself a delusion—a continued-belief delusion.

Now consider three hypothetical patients: patient A with Capgras delusion; patient B with motor impairments but not anosognosia; and patient C with anosognosia for their motor impairments. Patient A has set aside an old, commonplace, and still true belief ("The woman

[9] According to *DSM-5*, "Delusions are deemed *bizarre* if they are clearly implausible and not understandable to same-culture peers and do not derive from ordinary life experiences" (APA, 2013, p. 87). Many familiar examples of monothematic delusion are bizarre new-belief delusions, whereas anosognosia and reverse Othello syndrome are nonbizarre continued-belief delusions. The distinction between new-belief delusions and continued-belief delusions does not, however, coincide with the distinction between bizarre and nonbizarre delusions. There can be nonbizarre new-belief delusions; an example would be delusional jealousy (Othello syndrome).

who looks just like my mother and says that she is my mother is, indeed, my mother") and has adopted a new, exotic, and false belief ("My mother has been replaced by an impostor"). Patient A has adopted this new belief in response to an *anomalous experience* when looking at the woman who is, in fact, their mother. Because patient A's face processing system has been disconnected from the limbic system, the predicted autonomic response to a highly familiar face is absent (Ellis & Young, 1990). A normally functioning comparator detects this mismatch, a prediction-error signal is generated, and a new hypothesis comes to mind, "The person I am looking at is a stranger". Patient B, with motor impairments but not anosognosia, has replaced an old, commonplace, but now false belief ("I can move my left arm") with a new, unwelcome, but true belief ("I cannot move my left arm"). Patient B has adopted this new belief in response to an immediate bodily experience of movement failure when trying to move their left arm (Section 1.1). Because of patient B's motor impairment, the predicted movement of the left arm is absent. A normally functioning comparator detects this mismatch, a prediction-error signal is generated, and a new hypothesis comes to mind, "I cannot move my left arm". In contrast to patient B, patient C, with anosognosia for their motor impairments, maintains the old, commonplace, but now false belief ("I can move my left arm"). Patient C has not set aside this old belief because they have no immediate bodily experience of movement failure when they try to move their left arm (Section 1.2). Because of patient C's motor impairment, the predicted movement of the left arm is absent but, in addition, the comparator is not functioning normally. No prediction-error signal is generated, and no new hypothesis comes to mind.

Both patient A and patient C have made errors in their beliefs and, in both cases, the error is the result of an *anomaly of experience*—an anomalous experience in one case (A), and an anomalous absence of experience in the other (C). Patient A, with Capgras delusion, has made an *error of commission*—adopting a new and false belief about their mother although reality has not changed (patient A's mother has not been replaced by an impostor). Patient C, with anosognosia, has made an *error of omission*—failing to reject an old and false belief and adopt a new and true belief about their motor abilities although reality has changed (patient C's motor abilities are now severely impaired).

Despite the difference between anosognosia and more familiar monothematic delusions, such as Capgras delusion, it has proved theoretically illuminating to explain anosognosia by appeal to two factors.[10] A first factor results in an anomaly of *experience* and a second factor results in a failure of hypothesis evaluation—or better, because the delusional idea is already believed, the second factor results in a failure of *belief evaluation*.

4.1 Candidate first factors in the two-factor account of anosognosia for motor impairments

In our two-factor account of anosognosia for motor impairments, the first factor results in an anomalous absence of immediate bodily experience of movement failure—perhaps accompanied by an illusory experience of successful movement. As described earlier (Section 1.2), this anomalous absence of experience in anosognosia arises from one or

[10] Because anosognosia is a continued-belief delusion, our two-factor account departs from the exposition of the two-factor theory of delusion (Section 3.1) in one way. Patients with anosognosia have believed "I can move my left arm" for many decades, and so the first factor in anosognosia is not indicated by the answer to the first of the two questions: "What initially prompted the delusional idea or hypothesis? What brought it to mind?".

another of several possible impairments of the motor control system—a motor intention deficit, impaired proprioceptive feedback, damage to, or abnormal functioning of, the comparator. Thus, these impairments are candidate first factors.

Two other candidate first factors are proprioceptive loss (now considered independently of the motor control system) and unilateral visuospatial neglect (unilateral neglect). Proprioceptive loss—long considered a possible factor in the aetiology of anosognosia (Babinski, 1914, 1918; Levine, 1990; Levine, Calvanio, & Rinn, 1991)—could give rise to an anomaly of experience that would make an independent contribution to the aetiology of anosognosia. Consider a patient who did not have immediate experience of movement failure, because no prediction-error signal was generated in the motor control system. But suppose that the patient's proprioception was intact. Such a patient could still have conscious bodily experiences of the position of their left arm, immobile at their side. Proprioceptive loss would block this experiential route to knowledge of their motor impairment.

In a similar way, a left-side attentional deficit—unilateral neglect—would obstruct the route to knowledge of motor impairments that is provided by patients' visual experience of their paralysed limbs. Vuilleumier (2004, p. 10) described unilateral neglect as "a notable suspect in anosognosia" and anosognosia persisting more than three months after stroke is almost invariably accompanied by unilateral neglect (e.g., Aimola, 1999; Aimola Davies et al., 2009; Cocchini, Beschin, & Della Sala, 2002). Interestingly, in the study by Vocat et al. (2010; see p. 3588), patients with both severe proprioceptive loss and severe unilateral neglect in the first twelve days following a right-hemispheric stroke were found to be significantly more likely to have anosognosia than patients with only one or neither of those two deficits.

4.2 Arguments for a second factor in anosognosia for motor impairments

The two-factor theory of delusion is supported by an argument for a second factor in the explanation of a case of delusion (e.g., Davies & Coltheart, in press). There are multiple examples of people who have the delusional hypothesis come to mind but are not delusional—they evaluate and reject the hypothesis. A second factor results in failure of hypothesis evaluation and thus explains why, in a case of delusion, the delusional hypothesis was not rejected. Our two-factor account of anosognosia is supported in a similar way.

First, there is an empirical argument. There are patients who have a first factor—resulting in absence of experience of movement failure, or even illusory experience of successful movement—and have the delusional idea come to mind, but do not have anosognosia. For example, patient EM (Chatterjee & Mennemeier, 1996) was asked, "Can you raise both arms? … Can you raise the left one?", and responded: "It feels like it's rising, but, it's not" (p. 229).

Second, there is a more theoretical argument for a second factor. Even without immediate experience of movement failure, other evidence of motor impairments is available:

> [I]t is not just that they fail *motorically*. The consequence of such failures is that, in trying to get out of bed to go to the toilet or to lift an object, they fall over or incur a similar accident, often lying helpless or hurting themselves. Unless such patients have some other problem, it is unlikely that they are unaware of these incidents (even if they are unaware of the reason for them), or that they rapidly forget them, or that

they hallucinate the success of the intended *action* (as opposed to the movement). (Marcel et al., 2004, p. 35)

> [P]atients with AHP [anosognosia for hemiplegia] do not express mere uncertainty regarding the perception of sensations or movement from the left limbs, nor do they just complain of movement illusions. They instead ignore the wealth of evidence that they are paralysed (e.g., their disabilities, occasional accidents, others' feedback) and adhere to the 'delusional' belief that they have functional limbs. The explanation of the latter belief … requires the postulation of another dysfunction. (Besharati, Forkel, Kopelman et al., 2016, p. 982)

So—even without immediate experience of movement failure—there is sufficient evidence for patients to reject the long-held belief that they can move their left limbs. There must be a second factor that explains why anosognosia patients are unable to make appropriate use of this evidence—including evidence from everyday mishaps consequent on the motor impairment (e.g., the glasses on the tray that the patient was trying to pick up are now on the floor).

4.3 Candidate second factors in the two-factor account of anosognosia for motor impairments

In principle, there could be at least four kinds of case in which patients who did not have immediate bodily experience of movement failure might also be unable to make use of other evidence to achieve knowledge of their motor impairments.

> **Option 0. No relevant evidence:** There might not be any relevant evidence for the patient to use. This might be the situation of a patient, perhaps sedated, lying in bed and not trying to engage in any everyday activities. Having mentioned this kind of case, we set it aside. If such a case of anosognosia were possible then it would not fit the definition of delusion and our two-factor account would not obviously be applicable.
> **Option 1. Unable to remember the evidence:** There might be relevant evidence available, but the patient might be unable to remember it for long enough to make use of it.
> **Option 2. Unable to recognise that the evidence calls for evaluation of current beliefs:** There might be relevant evidence available and remembered by the patient. But the patient might be unable to recognise that the evidence provides a reason to evaluate current beliefs.
> **Option 3. Unable to carry out the task of belief evaluation:** There might be relevant evidence available and remembered, and the patient might recognise that the evidence provides a reason to evaluate current beliefs. But the patient might be unable to carry out the cognitively demanding task of belief evaluation.

In this section, we shall discuss only Options 1 and 3. (We shall provide a substantive account of Option 2 in Section 5.1.)

Option 1 highlights the fact that a general memory impairment is a candidate second factor in anosognosia. Such an impairment could help explain why a patient is unable to use evidence and clues of many kinds to reject old beliefs that are now false and to adopt new beliefs that are more realistic. Vocat et al. (2010) found that anosognosia in the second week after stroke, was significantly associated with memory impairment (see p. 3587, Table 1) and with lesions in the hippocampus (p. 3590, Figure 4B). Klingbeil, Wawrzyniak, Stockert et al. (2020) used lesion network-symptom-mapping (based on maps of functional connectivity in the healthy brain) to investigate 49 patients with motor impairments following right-hemispheric stroke—25 with anosognosia and 24 without. They found that, in the patients with anosognosia, a region of right posterior hippocampus that was spared was previously functionally connected to regions that were now damaged. This could have led to hippocampal dysfunction as a remote effect of the lesion—*diaschisis*. The authors suggested that, even without direct hippocampal damage, "diaschisis induced memory deficits" (p. 3) might contribute to anosognosia, not only by preventing the effective use of evidence from multiple sources, but also by "perturb[ing] the stable encoding of updated beliefs" (p. 5).

Patient NS, whose anosognosia for motor impairments persisted a year after a severe closed head injury (Cocchini et al., 2002), is a good example of impaired memory preventing the use of evidence to update beliefs. Patient NS had left-side paralysis and unilateral neglect, and he also suffered from "anterograde amnesia for self-related information … he regularly failed to recall what he was doing or whom he had met a few minutes earlier" (p. 2031). Because of his unilateral neglect (a first factor) and associated *allochiric movements*,[11] patient NS did not have immediate experience of motoric failure unless his left limbs were repositioned on the right side of space and he attempted to move them. Even when patient NS did momentarily acknowledge his hemiplegia, the anterograde memory impairment meant that he was unable to retain this information for long enough to achieve more realistic beliefs about his changed circumstances. He continued to believe that he could move his left limbs, that he could go surfing, and that he could jump out of the car to buy a newspaper.

Our own proposal, in line with Option 3, is that at least some patients may have difficulty with the cognitively demanding task of belief evaluation. Although anosognosia is a continued-belief delusion, the anosognosia patient's belief evaluation task is similar in outline to the hypothesis evaluation task for a person with a new-belief delusion (Section 3.2). It requires taking control of the balance between cognitive imperatives of conservatism and observational adequacy. In anosognosia, the balance tips too far toward conservatism, at the expense of observational adequacy. (The Bayesian analogue of the balance tipping too far toward conservatism is giving too much weight to prior probabilities at the expense of likelihoods. The predictive coding analogue is giving too much weight to prior beliefs at the expense of prediction errors.) The delusional belief has been held for decades but the imperative to do justice to pre-existing background knowledge and beliefs needs to be inhibited, so that new evidence can be taken into account.

Belief evaluation requires assessment of hypotheses in the light of plausibility *and evidence*—evidence that might not all point in the same direction, especially if the patient experiences illusory limb movements. It requires weighing up these competing considerations and working out what to believe. Crosson, Barco, Velozo et al. (1989) point out that the transition,

[11] Patient NS moved his right limbs when asked to move his left limbs—allochiric movements (Cocchini et al., 2002, p. 2036). When he was asked to move the left limb, a motor command was issued for the right limb, which did, indeed, move, so that there was no mismatch between the predicted and the actual state, and no prediction-error signal was generated.

from concrete pieces of evidence about specific difficulties in particular situations, to a more abstract and general belief about having a motor impairment, may present an additional challenge. A considerable degree of understanding is needed to recognise "some common thread in the activities with which the patient has trouble" (1989, p. 47). Thus, we propose that the second factor in anosognosia for motor impairments (as in new-belief delusions) may involve impairments of executive function or working memory (Aimola Davies et al., 2009).

4.4 Impairments of executive function or working memory in patients with anosognosia

Support for the proposal that the second factor in anosognosia may involve impaired executive function or working memory comes from our study of patients with unilateral neglect persisting at least three months following stroke. Five of the patients (3 to 22 months post-stroke) had anosognosia for their motor impairments and four (4 to 14 months post-stroke) did not. A detailed neuropsychological assessment of all nine patients was conducted (Aimola, 1999; Maguire & Ogden, 2002),[12] followed by a statistical investigation of the patients who met the criterion that they had results on all the neuropsychological tests for which the score was the number of correct responses out of a fixed total. Thus, the statistical analysis included seven patients (four with anosognosia and three without) and fifteen scores from neuropsychological tests of visuoperceptual function, sustained attention, memory, executive function, and working memory. For a brief account of the neuropsychological assessment that includes the statistical analysis, see Aimola Davies et al. (2009).

In brief, the statistical analysis demonstrated that only three (of fifteen) test scores were significantly predicted by the overall anosognosia scores for upper and lower limbs. One was the Elevator Counting with Distraction subtest of the Test of Everyday Attention (Robertson, Ward, Ridgeway, & Nimmo-Smith, 1994). This subtest is described by the authors as a test of working memory, but it also has a clear inhibitory component. The other two test scores were Categories Achieved and Perseverative Errors from the computerised version of the Wisconsin Card Sorting Test (WCST), which was administered using standardised instructions (Heaton, Chelune, Talley et al., 1993). The WCST is a demanding test of executive function, involving set-shifting, complex working memory operations, error detection, and feedback utilisation (Lie, Specht, Marshall, & Fink, 2006).

5. The Two-Factor Account and the ABC Model of Anosognosia

In our two-factor account of anosognosia for motor impairments as a continued-belief delusion, the first factor results in an anomalous absence of immediate bodily experience of movement failure, which prevents the patient from gaining knowledge of movement failure when it occurs. Alternative experiential routes to concurrent knowledge of movement failure (and, thence, to more lasting knowledge of motor impairments) might be blocked by proprioceptive loss or unilateral neglect (Section 4.1). The two-factor account is supported by empirical and theoretical arguments for a second factor in the explanation of anosognosia—a factor that results in a failure of belief evaluation (Section 4.2). A second factor must explain why patients are unable to make appropriate use of available evidence to achieve knowledge of their motor impairments. We have considered impairments of anterograde memory, executive function, and working memory as candidate second factors (Section 4.3).

[12] The first author of the paper by Maguire and Ogden (2002) is the last author of the present chapter.

The two-factor account is similar in structure to Levine's (1990; Levine et al., 1991) discovery theory of anosognosia (for discussion, see Davies et al., 2005). According to Levine, proprioceptive loss prevents the patient from having immediate knowledge of whether the affected limb is moving, but discovery of paralysis would still be straightforward for cognitively healthy individuals. Thus, Levine proposed that patients with motor impairments who suffered a proprioceptive loss (a first factor) would have anosognosia for their motor impairments only if they also had a second factor, "cognitive defects impairing the ability to observe and to infer" (1990, p. 254). The structure that is shared by the two-factor account and the discovery theory is well described by Vuilleumier (2004, p. 11) "[A]ny neurological dysfunction susceptible to alter the phenomenal experience of a defect might provide the ground out of which anosognosia can develop when permissive cognitive factors are also present."

Vuilleumier's ABC (Appreciation, Belief, Check) model of anosognosia (Vocat, Saj, & Vuilleumier, 2013; Vocat & Vuilleumier, 2010; Vuilleumier, 2004; Vuilleumier, Vocat, & Saj, 2013) conforms to the same structure. Understood as the (A + BC) model, it offers a two-factor account of anosognosia. Impaired Appreciation operations (e.g., impairments of the motor control system, proprioceptive loss, or unilateral neglect) alter patients' conscious experience and prevent them from gaining "direct first-person knowledge" about their motor impairments (Vuilleumier, 2004, p. 15). But impaired Appreciation operations are not sufficient to explain anosognosia and so additional impairments are required. These are grouped under the Belief and Check headings and can be summarised as "an inability to question beliefs or knowledge [Belief] … or an inability to trigger reactions of doubt or verification in case of uncertainty [Check]" (Vuilleumier et al., 2013, pp. 204–205).

5.1 Belief updating in anosognosia for motor impairments

Vocat et al. (2013) investigated belief updating in anosognosia for motor impairments, testing patients in the second week after a right-hemispheric stroke—four patients with anosognosia and five without—and healthy control participants. They used a riddle task that required the participant to guess ten target words (AIRPLANE, TOOTH, CARROT, KEY, COW, HEART, SHADOW, GARDEN, MATCHES, BROOM). For each word, five successively more informative clues were offered and participants were asked to guess the word, and to indicate their level of confidence, after each clue. Neither the target words nor the clues were related to motor abilities or impairments.

The first clue allowed many possible answers and was sufficiently uninformative to create doubt in healthy participants. For example, the first clue for the target word HEART was "My weight is approximately 300 grams". The fifth clue was intended to leave no doubt about the correct answer; for example, "Lovers often draw me" for HEART. Anosognosia patients were no less able than participants in the other two groups to solve the riddle by giving the correct response after the fifth clue. However, patients with anosognosia gave significantly higher confidence ratings than other participants after the first three clues. Thus, it appeared "as if the anosognosics could not experience 'doubt' anymore" (Vocat et al., 2013, p. 1777). In the terms that we have used earlier (Section 3.2), it seemed that the patients with anosognosia could not suspend or inhibit judgement and consider their first guess as a hypothesis having only "equal priority [with other] possible hypotheses" (Langdon & Coltheart, 2000, p. 206).

Most interestingly, given the character of anosognosia as a continued-belief delusion, the patients with anosognosia showed clear evidence of failure to update their beliefs. They were significantly more likely than the other two groups—indeed, more than twice as likely—to produce the same incorrect guess to two consecutive clues in the same riddle. Vocat et al. (2013) proposed that the anosognosia patients' impairment of belief updating resulted from a problem with *error detection*—in this case, detection of mismatch or incongruence between the presented clue (evidence) and the patient's guess (current belief). The anosognosia patients "required a repeated signal of errors, or a larger incongruence between a new clue and the previous guess, in order to prompt a re-appraisal of their preceding responses and to trigger a new solution" (p. 1778).

This proposed connection between impaired belief updating and a difficulty in detecting mismatch, incongruence or error fits well with formulations of the ABC model in which first factors (A) disrupt "direct appreciation of motor losses" and second factors (BC) damage "the capacity to monitor and detect error in performance" (Vuilleumier et al., 2013, p. 208). Hence, Vocat et al. (2013) suggested: "anosognosics might be unable to change their past beliefs … *simply because they have no grounds to do so* at both the sensory-motor and [cognitive-]affective-motivational levels" (p. 1778; emphasis added).

If Vocat et al.'s (2013) suggestion is correct, it makes a difference to our understanding of the second factor in the two-factor account of anosognosia. Earlier in this chapter (at the beginning of Section 4.3), we considered ways in which patients with a first factor resulting in absence of immediate experience of movement failure might also be unable to make use of available evidence (including evidence from everyday mishaps) to achieve realistic beliefs about their motor impairments. A patient might be unable to remember the evidence for long enough to make use of it (Option 1), or unable to recognise that the evidence calls for evaluation of current beliefs (Option 2), or unable to carry out the cognitively demanding task of belief evaluation (Option 3). We discussed impairments of anterograde memory (Cocchini et al., 2002) and executive function or working memory (Aimola, 1999; Maguire & Ogden, 2002) as candidate second factors in line with Options 1 and 3.

We can now add impaired error (or mismatch) detection as a candidate second factor in line with Option 2. We can also provide a fuller account of Option 2 and some further articulation of Option 3. (*Italics* indicate changes from the earlier versions of Options 2 and 3.) Option 1 remains unchanged, but for convenience it is repeated here.

> **Option 1. Unable to remember the evidence:** There might be relevant evidence available, but the patient might be unable to remember it for long enough to make use of it.
> **Option 2. Unable to recognise that the evidence calls for evaluation of current beliefs:** There might be relevant evidence available and remembered by the patient. But the patient might be unable to recognise that the evidence provides a reason to evaluate current beliefs *because the patient does not recognise that the evidence is incongruent with current beliefs*.
> **Option 3. Unable to carry out the task of belief evaluation:** There might be relevant evidence available and remembered, and the patient might recognise that the

> evidence provides a reason to evaluate current beliefs *because the patient recognises that the evidence is incongruent with current beliefs*. But the patient might be unable to carry out the cognitively demanding task of belief evaluation.

5.2 Impaired error detection and reality monitoring in anosognosia for motor impairments

Contemporary theories of anosognosia for motor impairments appeal to a domain-specific failure of error detection. A comparator within the motor control system fails to detect a substantial disparity between predicted and actual feedback from the left arm and fails to generate a (conscious) prediction-error signal. As a result of this first factor, the patient has no immediate bodily experience of movement failure (Section 1.2). Vocat et al.'s (2013) results using the riddle task, with target words and clues that were not related to motor abilities or impairments, suggest a more general problem with detection of mismatch or error:

> In patients with anosognosia, the right hemispheric lesion might … reduce the ability to check and change current beliefs about one's own state despite the presence of incongruent information. Due to a lack of error signals and subsequent check operations, the patients may be abnormally confident in their beliefs and fail to integrate new information in order to modify them when confronted with incongruency. (2013, p. 1778)

Is there other evidence to support the proposal that an impairment of error detection *extending beyond motor control for the paralysed limbs* might play a second-factor role in the explanation of anosognosia (in line with Option 2)? Two studies (Preston et al., 2010; Ramachandran, 1995) have suggested that this problem with detection of mismatch or error extends to actions performed using the unimpaired limbs.

Patient FD (Ramachandran, 1995) had anosognosia and experienced illusory movements of her paralysed left arm. When asked to point to the examiner's nose with her left hand, she said that she could clearly see her hand pointing: "it is about two inches from your nose" (p. 32). Ramachandran investigated error detection related to patient FD's *unimpaired right hand* by using a mirror box to provide false visual feedback. Patient FD placed her right hand in the box and moved it up and down. Vision of her right hand was precluded by a mirror, in which she viewed the reflection of a stationary (left) hand positioned so as to appear to be a (right) hand at the location of her moving right hand. Patient FD apparently failed to detect the disparity between the predicted and actual visual feedback; that is, movement of the felt hand *versus* no movement of the seen hand. She reported that "she could clearly see the hand move up and down" (p. 33).

A different visuomotor error detection paradigm (cf. Fourneret & Jeannerod, 1998; Nielsen, 1963) was used by Preston et al. (2010). Subjects reached with their unimpaired right hand toward target locations directly in front of them. False visual feedback introduced an angular perturbation (leftward or rightward in the horizontal plane) from the hand's actual movement. An anosognosia patient, GG, failed to detect disparities of up to 20 degrees between the false visual feedback and the actual trajectory of the movement of his *unimpaired right hand* (while healthy control participants easily detected disparities of 8 degrees).

These two error detection studies involved discrimination between online information with an internal origin—how one is intending and trying to move the unimpaired hand—and online information with an external origin—how the unimpaired hand is observed to be moving. In contrast, *reality monitoring* is discrimination between *remembered* information that had an internal origin and *remembered* information that had an external origin. Jenkinson, Edelstyn, Drakeford and Ellis (2009, Experiment 2) investigated reality monitoring for actions. In the first (acquisition) phase of their experiment, three patients with anosognosia were presented with short phrases for actions that were to be performed, imagined, or observed (twenty action phrases in each of three conditions). In the second (test) phase (when twenty new actions were included), the anosognosia patients achieved significantly less good source attribution performance—reporting whether an action correctly identified as not new was *performed*, *imagined* or *observed*—than patients without anosognosia and healthy volunteers.

Building on this work, Saj, Vocat and Vuilleumier (2014) also investigated "whether self-monitoring deficits for movements in patients with [anosognosia for motor impairments] are selective for the affected limb … [or] would extend to the non-paralyzed limb" (p. 95). In the first (encoding) phase of the experiment, five patients with, and five without, anosognosia and five healthy control participants were instructed either to perform or to imagine actions (e.g., rub shoulder, wave goodbye) using either the left or the right arm (twenty actions in total, five in each of four conditions). In the second (recognition) phase (when five new actions were included), participants were asked, for each of the twenty-five actions, whether it was new, or had been successfully executed, only attempted, or imagined, and which arm had been used. The performance of the patients with motor impairments following a right-hemispheric stroke, but without anosognosia, was essentially the same as that of the healthy control participants, save that the patients without anosognosia acknowledged their failure to execute actions with the left arm. In contrast, patients with anosognosia never classified actions as having been only attempted. They classified actions attempted or imagined using the left arm—and even some executed using the right arm—as having been executed using the left arm (thirteen of twenty-five actions on average). They also classified some actions imagined using the right arm as having been executed. Saj et al. concluded:

> Patients with [anosognosia for their motor impairments] … often misattributed movements previously executed with the right arm to the left arm, and generally showed greater confusion between executed and imagined for both sides, rather than strictly unilateral deficits concerning the left arm. (p. 103)

Thus, in both error detection and reality monitoring paradigms, patients with anosognosia fail to distinguish between internally represented and externally perceived movements of their limbs—both the impaired and the unimpaired limbs. The results of another experiment suggested that impaired reality monitoring in patients with anosognosia extends even beyond the domain of movement and action. Jenkinson et al. (2009, Experiment 1) investigated whether patients with anosognosia are impaired in distinguishing between imagined (internally represented) and externally perceived visual stimuli that are unrelated to limb movement. In the acquisition phase, ten anosognosia patients were presented with words for visual objects and, immediately after each word, either perceived or imagined a drawing of the corresponding object (twenty words in each of two conditions). In the test phase, patients were presented with the same words (along with twenty new words) and reported whether they were *perceived*, *imagined*, or *new*. Anosognosia patients reported imagined visual stimuli as having been perceived and their source attribution performance was significantly less good than patients without anosognosia and healthy volunteers.

In a recent study, Cocchini, Scandola, Gobbetto et al. (2022) used the vicarious agency paradigm (Wegner, Sparrow, & Winerman, 2004) to investigate the sense of agency for visually observed hand gestures (e.g., wave, make a fist) in patients with left-side motor impairments following stroke—seven with anosognosia and nine without. Patients sat in front of a full-length mirror with their hands remaining still on their lap, hidden from view, and were asked to look at the mirror and not to move their arms during the experiment. The examiner, hidden by a screen, stood behind the patient's chair and reached their hands around the patient so that the examiner's arms appeared where the patient's own arms would have been. On each trial, the patient heard an instruction to perform an action (e.g., "wave") and then, while the patient watched in the mirror, the examiner performed an action—with either their left or their right hand, and either matching or incongruent with the instruction heard by the patient (four conditions in total). After 48 trials in a single condition (e.g., right hand; match), the patient was asked the question, "How much control did you feel that you had over the arm's movements?", and responded with a rating from 1 (not at all) to 7 (very much).

In healthy young adults, the sense of agency is stronger in the *match* conditions than in the *incongruent* conditions (Wegner et al., 2004, Experiment 2). In healthy older adults, the sense of agency is reduced and the effect of matching is attenuated, but not abolished (Cioffi, Cocchini, Banissy, & Moore, 2017). Cocchini et al. (2022) found no difference in the sense of agency between the left-hand and right-hand conditions; nor between the match and incongruent conditions. But there was a significant difference between the patients with anosognosia for their motor impairments (mean sense of agency rating = 5.43) and patients with motor impairments but without anosognosia (mean sense of agency rating = 3.38). Patients with anosognosia were more likely than patients without anosognosia to report a feeling of controlling the hand gestures viewed in the mirror.

It seems unlikely that the illusory sense of agency depended on motor intentions—both because patients were instructed not to move their hands or arms and because the sense of agency was no less strong when the viewed action was incongruent with the heard instruction. Patients (with or without anosognosia) responded to the check question, "To what degree did you feel you could anticipate the movements of the arm?", with higher ratings in the match conditions than in the incongruent conditions (4.57 *versus* 2.19). Thus, it seems that the patients with anosognosia attended to the heard instruction and were somewhat able to predict what hand gesture they would see in the match conditions; but just seeing themselves in the mirror, appearing to make a hand gesture, was sufficient to generate a sense of agency.

This illusory sense of agency—for both impaired and unimpaired limbs—seems to be broadly consistent with other findings that we have reviewed in this section.[13]

The first five experimental studies (Jenkinson et al., 2009, Experiments 1 and 2; Preston et al., 2010; Ramachandran, 1995; Saj et al., 2014) provided evidence that, in patients with anosognosia, impairments of error detection and reality monitoring extend to actions performed using unimpaired limbs, and even to domains beyond movement and action. The Cocchini et al. (2022) study added that, in the vicarious agency paradigm, patients with anosognosia have a relatively strong sense of agency for an action that appears to be theirs—even when it is incongruent with a heard instruction. Taken together, the results of these studies might indicate: a monitoring impairment "not limited to the contralesional side" (Cocchini et al, 2022, p. 11); "a more global deficit for motor awareness" (Preston et al., 2010, p. 3449); "more global monitoring difficulties" (Saj et al., 2014, p. 103); "a breakdown of reality monitoring processes for information not directly related to movement" (Jenkinson et al., 2009, p. 468); or even damage to a domain-general "anomaly detector" (Ramachandran, 1995, p. 39). A difficulty in detecting disparities between what is internally represented—intended, imagined, heard, or antecedently believed—and external reality could contribute to patients' inability to make appropriate use of available evidence to achieve knowledge of their motor impairments.

A difficulty in detecting such disparities might also help to explain the performance of some patients with anosognosia in the Berti et al. (1996) study. Patients were asked to estimate their *potential* ability to perform bimanual tasks, such as opening a bottle or a jam tin, and were then asked *actually* to perform the tasks—and to score their actions. Some patients not only claimed, hypothetically, that they *would* be able to perform these tasks well, but also gave themselves high scores *after* attempting the action with real objects—despite the fact that an unopened bottle or jam tin would provide clear evidence of failure (see Berti et al., 1996, p. 435, Table 6).

6. Conclusion

Anosognosia for motor impairments fits the *DSM-5* definition of delusions as "fixed beliefs that are not amenable to change in light of conflicting evidence" (APA, 2013, p. 87). In this chapter, we have put forward a two-factor account of anosognosia for motor impairments. Anosognosia is a continued-belief delusion, rather than a new-belief delusion, and the first factor results, not in an anomalous experience, but in the anomalous absence of immediate bodily experience of movement failure. Patients with motor impairments, but without immediate experience of their movement failure, could—if they had no additional impairments—use other available evidence to achieve knowledge of their motor impairments. So there must be at least a second factor in the aetiology of anosognosia—one or more additional impairments that prevent patients with anosognosia from making appropriate use of available evidence to update their beliefs.

[13] Cocchini et al. (2022) put forward a possible alternative interpretation of their findings in terms of interhemispheric compensation for left-side motor impairments, while also noting that this interpretation "will certainly need further investigation" (p. 9). It is beyond the scope of the present chapter to evaluate this alternative interpretation.

The structure of our two-factor account (Davies et al., 2005) is similar to Levine's discovery theory (Levine, 1990; Levine et al., 1991) and it is shared by Vuilleumier's ABC model (Vocat et al., 2013; Vocat & Vuilleumier, 2010; Vuilleumier, 2004; Vuilleumier et al., 2013). The two-factor structure is also evident in a recent structural neuroimaging study (Pacella, Foulon, Jenkinson et al., 2019):

> We thus postulate that deficits in motor monitoring, associated with a compromised premotor network [first factor], need to be combined with other salience and belief updating deficits [second factor], collectively leading to a multifaceted syndrome in which premorbid beliefs and emotions about the non-paralysed self dominate current cognition about the paralysed body" (p. 6).

We have considered three candidate second factors. We agree with Cocchini et al. (2002) that some patients might be unable to make appropriate use of available evidence because of a general memory impairment (Option 1). And we agree with Vocat et al. (2013) that some patients might have evidence available and remembered, but might still be unable to recognise the incongruence between the evidence and their current beliefs because of an error detection impairment (Option 2). We, ourselves, propose that some patients might have evidence available and remembered, and might recognise the incongruence between the evidence and their current beliefs. But some of these patients might still be unable to carry out the cognitively demanding task of belief evaluation—weighing up the evidence and working out what to believe—because of impairments of executive function or working memory (Option 3).

Patients with no general memory impairment and no error detection impairment might use the evidence from everyday mishaps, such as failures to complete bimanual tasks, to reject the current beliefs with which the evidence is most obviously incongruent. They might adopt new and more realistic beliefs about activities of daily living, such as washing, dressing and eating, and about other everyday tasks, such as cutting a steak, tying a knot or carrying a large tray of glasses. In short, they might overcome their anosognosia for the consequences of their motor impairment—but, crucially, they would still have anosognosia for the motor impairment itself. (See Section 2.2. for the double dissociation between anosognosia for the impairment itself and anosognosia for the consequences of the impairment.)

The patients would still need to evaluate and reject the core belief, "I can move my left arm", in anosognosia for their motor impairment. Belief evaluation is a cognitively demanding task (Sections 3.2. and 4.3) and there are particular challenges in the case of anosognosia. As Crosson et al. (1989) observed, it is not a straightforward matter to proceed from evidence about specific mishaps in particular situations (e.g., getting dressed, carrying a large tray of glasses) to the new general belief, "I cannot move my left arm" (Section 4.3). The task would be even less straightforward if illusory experiences of successful movement were to provide (misleading) evidence in support of the to-be-rejected belief. Thus, some patients with impairments of executive function or working memory might still fail to achieve full knowledge of their true condition (see Section 4.4).

Over the last two decades, research has advanced our understanding of many aspects of anosognosia for motor impairments, including preserved motor intentions (e.g., Garbarini et al., 2012; Jenkinson & Fotopoulou, 2010), implicit knowledge (e.g., Antoniello & Gottesman, 2020; Fotopoulou, Pernigo, Maeda et al., 2010), perspective—first-person *versus* third-person—and 'centrism'—egocentric *versus* allocentric (e.g., Besharati, Jenkinson, Kopelman et al., 2022; Marcel et al., 2004), theorising anosognosia in the predictive coding framework (e.g., Fotopoulou, 2014; Kirsch, Mathys, Papadaki et al. 2021), assessment (e.g., Della Sala et al., 2009; Moro, Besharati, Scandola et al. 2021), rehabilitation (e.g., Jenkinson, Preston, & Ellis, 2011; Moro, Scandola, Bulgarelli et al., 2015), and the neural basis of anosognosia (e.g., Berti et al., 2005; Vocat et al., 2010)—particularly, in recent years, disconnection of neural networks (e.g., Monai, Bernocchi, Bisio et al., 2020; Pacella et al., 2019). We conceptualise anosognosia as a delusion (Davies et al., 2005; Aimola Davies et al., 2009), considered against the background of a two-factor theory of delusion that was applied, in the first instance, to monothematic delusions of neuropsychological origin (e.g., Coltheart, 2010). When full neuropsychological investigations are included in studies of anosognosia for motor impairments, strengths and deficits are revealed in the areas of orientation, attention and working memory, sensation and perception, memory and learning, concept formation, reasoning and executive function. It is then possible to develop individual neuropsychological profiles for series of cases of anosognosia. We might hope that these neuropsychological profiles would shed light on the patterns of co-occurrence of cognitive impairments of memory, error detection, executive function and working memory, with anosognosia for motor impairments.

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