Cognitive and Motivational Factors in Anosognosia

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Abstract: Anosognosia is a patient’s denial of his or her impairment or illness. The starting point for this chapter is that anosognosia can be considered as a delusion. We sketch the two-factor framework for understanding delusions and how anosognosia would fit into this framework. We set out competing views on the role of motivation in anosognosia and evaluate the prospects for an account of anosognosia at least partly in terms of motivation. We describe a study of cognitive impairments in patients with anosognosia and propose that the role of impairments of working memory and executive processes should be investigated further; and we consider how the results of a study in which motivation is not explicitly investigated might, in principle, suggest that motivation is also playing a role in some cases.

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1. Introduction

Patients with anosognosia fail to acknowledge, or even outright deny, their impairment or illness (see Orfei et al., 2007 for a recent review). Anosognosia is usually assessed by means of a structured interview beginning with questions about general health and moving to specific questions about the patient’s impairment. In this chapter, we shall be concerned with anosognosia for hemiplegia (paralysis of one side of the body) or, more generally, for motor impairments. A patient whose arm or leg is paralysed or weak following a stroke may deny the weakness in response to questions like, ‘Is there anything wrong with your arm or leg? Is it weak, paralysed or numb?’ (questions from Cutting, 1978), and may continue to deny the impairment even when it has been demonstrated. For example, the examiner may ask the patient to raise both arms and then demonstrate to the patient that one arm is not raised as high as the other. Our aim is to explore the role of cognitive impairments in anosognosia, and also the role of motivation.

1.1 Terminology and distinctions

Before proceeding, we set out some terminological matters and some conceptual distinctions. The dictionary definition of ‘anosognosia’ is ‘unawareness of or failure to acknowledge one’s hemiplegia or other disability’ (OED). As this indicates, the term can be used in a more restricted or a more inclusive way. The French neurologist Joseph Babinski (1914, 1918) introduced the term as applying only to anosognosia for hemiplegia. Etymology would suggest the more inclusive meaning – ‘lack (a-) of knowledge (-gnosia) of disease (-noso-)’. Some researchers follow Babinski in restricting ‘anosognosia’ to anosognosia for hemiplegia and then use ‘unawareness’ as a more general term. They speak of unawareness of visual impairments, unawareness of memory impairments, and unawareness of other cognitive impairments (Anderson and Tranel, 1989).

Since there is some variation in usage of the terms ‘anosognosia’ and ‘unawareness’ (and several other terms in this area) we need to be explicit about the way we use them. On the question of restricted or inclusive use, we use ‘anosognosia’ in the inclusive way. Patients may have anosognosia for visual impairments, for memory impairments, for cognitive impairments, and so on. In this chapter, we shall be concerned with anosognosia for motor impairments and, if this reference is clear from the context, we shall use the term ‘anosognosia’ without qualification.

The dictionary definition of ‘anosognosia’ mentions both ‘unawareness’ and ‘failure to acknowledge’. But the term ‘unawareness’ suggests a failure in sensation and perception while ‘failure to acknowledge’ suggests a failure in thought and speech. There is an important conceptual distinction here and it is obscured if ‘unawareness’ is used as a near synonym for ‘anosognosia’. We regard anosognosia as a failure or pathology at the level of belief. There is a mismatch between the patient’s estimate of his or her abilities and the reality of the impairment and, in a severe case of anosognosia, this mismatch is substantial. The patient believes that he or she does not have the impairment despite the fact that it is clearly present. This incorrect belief will normally be manifested in the

1 Anderson and Tranel (1989); Berti, Ládavas and Della Corte (1996); Cutting (1978); Feinberg, Roane and Ali (2000); Marcel, Tegnér and Nimmo-Smith (2004); Nathanson, Bergman and Gordon (1952); Starkstein, Fedoroff, Price, Leiguarda, and Robinson (1992).
patient’s verbal denial of the impairment in response to questions (provided, of course, that the patient answers sincerely).

A failure at the level of belief is clearly conceptually distinct from a failure at the level of sensory or perceptual experience. In principle, a patient with impaired proprioception might have no immediate bodily experience of failure to move a paralysed limb yet, on the basis of other evidence, the patient might still reach the correct belief about his or her paralysis (failure of experience without failure of belief). Conversely, a patient with intact proprioception but impaired memory might have vivid bodily experiences of failure to move a paralysed limb but, because the experiences are quickly forgotten, might fail to reach the correct belief about his or her paralysis (failure of belief without failure of experience). Furthermore, this distinction is not merely a conceptual or ‘in principle’ one. In a study of left- and right-hemisphere stroke patients, Anthony Marcel and colleagues (Marcel, Tegnér and Nimmo-Smith, 2004) assessed anosognosia in a structured interview and also asked patients to evaluate their own motor performance immediately after being asked to raise each limb with vision precluded. Some patients overestimated their motor abilities in the immediate post-performance evaluation (in which the patients had to rely on proprioception) but acknowledged their impairments in response to interview questions. Other patients showed the reverse pattern.

In order to mark this distinction we shall use the term ‘unawareness’ for the failure of concurrent sensory or perceptual experience of impairment and reserve the term ‘anosognosia’ for the failure of belief that is normally manifested in verbal denial of an impairment in response to interview questions. Sometimes, as in the first sentence of this chapter, we use the term ‘deny’ to indicate denial in thought as well as speech. We do not assume that denial in thought or speech is a psychological defence mechanism.

Classification of patients as having anosognosia may be complicated, or even subverted, by their denial that the affected part of their body (a paralysed limb, for example) even belongs to them (Bisiach, Rusconi and Vallar, 1991; Halligan, Marshall and Wade, 1995). This denial of ownership of a body part is an extreme form of somatoparaphrenia (Gerstmann, 1942). Denial of ownership of a paralysed limb may sometimes occur in conjunction with misoplegia, which is severe dislike or even hatred towards the affected limb (Critchley, 1974; see also 1955).

Anosognosia strictly so-called can be distinguished from a second condition described by Babinski (1914; quoted in Critchley, 1953, p. 230): ‘I have seen hemiplegics who, without being ignorant of the existence of their paralysis, seem to attach no importance to it. Such a state might be called anosodiaphoria.’ The patient may adopt a laissez-faire attitude and show a lack of interest in, a lack of appropriate emotion about, or an unrealistically optimistic attitude towards the consequences of, the impairment. In the literature, there is some variation in the use of the term ‘anosodiaphoria’ but, independently of the terminological issue, there is a second important conceptual distinction here. Having an incorrect belief about the severity of an impairment itself is distinct from having an incorrect belief about the seriousness of the consequences of the impairment.

Allan House and John Hodges (1988) describe an 89-year-old woman who suffered left-side paralysis following a right-hemisphere stroke. When she was examined six months after her stroke, she acknowledged that her left arm was weak, and weaker than her left leg. When it was demonstrated to her that her left arm was completely paralysed and her left leg nearly completely paralysed, she rated the strength of her left hand and wrist zero out of ten and her left hip, knee, ankle and foot two out of ten. But even while
she acknowledged her motor impairments she failed to appreciate their consequences, claiming that she could look after herself unaided and even walk upstairs (whereas, in reality, she was restricted to a wheelchair). Marcel and colleagues (2004) also report several patients who acknowledged that their left arm was paralysed yet overestimated their ability to carry out bi-manual tasks such as tying a knot, clapping hands, or shuffling cards. We might describe such patients as having anosognosia for the consequences of their motor impairment – ‘denial of handicap’ (House and Hodges, 1988) – but not anosognosia for the impairment itself. Something like the reverse dissociation is seen in patients who explicitly deny their impairment and yet show some implicit appreciation of its consequences (Berti, Làdavas, Stracciari, Giannarelli and Ossola, 1998).

Putting this conceptual distinction together with the earlier one between failure of experience and failure of belief, we need, in the end, a threefold distinction between concurrent unawareness of an impairment, failure to acknowledge the impairment itself, and failure to appreciate the consequences of the impairment.

1.2 Anosognosia as a delusion

A patient with anosognosia believes that he or she is able to move a limb that is, in reality, paralysed. In its severe form, anosognosia constitutes a delusion according to the definition offered by the American Psychiatric Association’s Diagnostic and Statistical Manual of Mental Disorders (DSM-IV-TR, 2000, p. 821): ‘A false belief . . . that is firmly sustained despite what almost everyone else believes and despite what constitutes incontrovertible and obvious proof or evidence to the contrary.’

In this chapter, we shall address four questions about anosognosia considered as a delusion.

- How would explanations of anosognosia fit into the two-factor framework for understanding delusions?
- What are the prospects for an account of anosognosia at least partly in terms of motivation?
- How should we investigate the role of cognitive impairments in anosognosia?
- In an assessment of cognitive impairments, what kinds of finding might suggest that motivation is also playing a role in some cases?

Section 2 provides a summary account of the two-factor theory of delusions so that, in section 3, we can address the question how explanations of anosognosia might fit into this framework. In section 4, we consider the role of motivation in anosognosia, particularly in the light of an influential argument against interpreting anosognosia as motivated denial. In section 5 we present an empirical study of the role of cognitive impairments in anosognosia and in section 6 we return briefly to motivation.

2. The Two-Factor Theory of Delusions

In a case of delusion, the subject believes a false proposition, \( P \), and maintains this belief despite the fact that evidence and plausibility decisively favour the true proposition not-\( P \). In the Capgras delusion (Capgras and Reboul-Lachaux, 1923; Ellis and Young, 1990; Stone and Young, 1997), the false proposition is that a close relative of the subject has been replaced by an impostor; in mirrored-self misidentification (Breen, Caine and Coltheart, 2001; Breen, Caine, Coltheart, Hendy and Roberts, 2000; Foley and Breslau, 1982), it is that the person that the subject sees in the mirror is not him; in delusional jealousy (Othello syndrome; Todd and Dewhurst, 1955), it is that the subject’s partner is being unfaithful.
Given any delusion, we can ask two questions (Coltheart, 2007, p. 1044). First, where did the delusion come from? Second, why does the patient not reject the belief? In slightly more detail, the questions are these. First, how did the subject come to regard the false proposition as a salient and serious hypothesis, a credible candidate for belief? Second, even allowing that the false proposition somehow achieved a measure of initial credibility as a candidate for belief, or even allowing that the proposition was initially adopted as a belief (Gilbert, 1991), why does the subject not subsequently reject the proposition on the grounds of its implausibility and its incompatibility with a mass of available evidence?

Any account of the aetiology of delusions must answer these two questions. A two-factor account offers distinct answers to the two questions in terms of two departures from normality. The first factor explains why the false proposition seemed a somewhat salient and credible hypothesis, or why it was initially adopted as a belief. The second factor explains why the proposition is not subsequently rejected.

2.1 The first factor
Max Coltheart and colleagues have put forward a two-factor theory that was tailored, in the first instance, to monothematic delusions of neuropsychological origin.² An early formulation of the two-factor theory (Davies, Coltheart, Langdon, & Breen, 2001) started from Brendan Maher’s (1974, 1988, 1992, 1999) claim that delusions are false beliefs that arise as normal responses to unusual experiences. This formulation agreed in part, but also disagreed in part, with Maher’s claim. The point of agreement was that unusual experiences figure in the aetiology of delusions. The point of disagreement was that, while Maher claimed that the unusual experience is normally sufficient to produce the delusion, the two-factor theory said that normal responses to unusual experiences do not provide an answer to the second question about delusions.

Since the two-factor theory was initially offered as a theory of delusions of neuropsychological origin, it is reasonable to expect that a neuropsychological deficit will provide an answer to the question where the delusion came from. The neuropsychological version of the two-factor theory – the two-deficit theory – does not claim that the first deficit always gives rise to an unusual conscious experience. Coltheart describes the first deficit in this way (2007, p. 1047):

The patient has a neuropsychological deficit of a kind that could plausibly be related to the content of the patient’s particular delusion – that is, a deficit that could plausibly be viewed as having prompted the initial thought that turned into a delusional belief.

It is assumed that the first deficit varies from delusion to delusion and may also vary from patient to patient with the same delusion.

2.2 The second factor
The argument for a second factor in the aetiology of delusions is that, both normally and normatively, the first factor is not sufficient to explain the delusion. The first factor prompts an apparently salient and somewhat credible hypothesis or candidate belief but the hypothesis or candidate belief normally could be, and normatively should be,
rejected. Even if the first factor explains why the hypothesis is initially adopted as a belief, it does not explain the delusion since it does not explain why the belief is tenaciously maintained – ‘firmly sustained despite . . . incontrovertible and obvious proof or evidence to the contrary’ (DSM-IV-TR, 2000, p. 821). We need a second factor to answer the question why the patient does not reject the belief.

According to the neuropsychological version of the two-factor theory, the second factor – which does its work after the generation of the delusional hypothesis, candidate belief, or initially adopted belief – is a deficit in the cognitive mechanisms responsible for belief evaluation and revision. No very detailed account of this second deficit has yet been provided but Coltheart proposes that, whatever it is, it ‘is the same in all people with monothematic delusion’ (2005b, p. 154).

Although the second deficit is poorly specified in terms of cognitive function, there are some suggestions about its neural basis. For example, following a right-hemisphere stroke, patients may deny ownership of their paralysed left-side limbs and Coltheart (2007) notes that their somatoparaphrenia is a delusion. The fact that patients with somatoparaphrenia generally have intact left hemispheres suggests that the second deficit results from right-hemisphere damage, and other evidence supports this suggestion. Thus Coltheart describes the second deficit in this way (ibid., p. 1047):

The patient has right-hemisphere damage (i.e., damage to the putative belief evaluation system located in that hemisphere).

He goes on to review evidence from group and single case studies suggesting that ‘it is specifically frontal right-hemisphere damage that is the neural correlate of the impairment of belief evaluation’ (ibid., p. 1052).

It is useful to speculate about the cognitive nature of the mechanisms of belief evaluation in terms of dual-process accounts of reasoning (Evans, 2003). Dual-process accounts propose that there are two quite different kinds of cognitive mechanism involved in reasoning – and also in judgement and decision-making. System 1 mechanisms are of types that are shared by humans and other animals, and they are typically rapid, parallel, and automatic. System 2 mechanisms, in contrast, are evolutionarily recent and perhaps distinctively human, and their operation is slow and sequential. Importantly, the operation of System 2 mechanisms is constrained by working memory capacity and depends on inhibitory executive processes to suppress default responses emanating from System 1. (Working memory involves both the temporary maintenance and the manipulation of information. Executive processes are involved in deliberate, goal-directed thought and action; they include flexible or abstract thinking, planning or decision making, and initiating or inhibiting responses.)

In terms of this dichotomy between System 1 and System 2 reasoning mechanisms, it seems natural to suppose that the mechanisms of belief evaluation and revision that are impaired in patients with delusions would belong in System 2. For these mechanisms seem more plausibly slow, sequential, and distinctively human, rather than rapid, parallel, automatic, and shared with other animals. Consequently, we might hope that some light would be shed on the cognitive nature of the second factor in the aetiology of delusions by investigations of working memory and executive processes in patients with delusions.

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3 We note, however, that there is at least one published report of a patient with somatoparaphrenia following a left-hemisphere stroke (Miura et al., 1996).

4 Here, as at many other points, we are indebted to Tony Stone.
2.3 Two factors but three stages

A delusion is a pathology of belief and so the two-factor theory focuses on explanatory factors that are departures from normality. But neither an unusual experience nor a neuropsychological deficit provides a complete answer to the question where the delusion came from. For it does not explain how the false proposition came to be regarded as a salient and serious hypothesis, why it seemed a credible candidate for belief, or how it came to be initially adopted as a belief. In the aetiology of a delusion, there is a processing stage leading from the unusual experience or neuropsychological deficit to the initial adoption of the false proposition as a belief. The processing at this stage may be quite normal although we do not rule out the possibility that it departs from normality, perhaps as the result of a neuropsychological deficit. Indeed, in principle, the first factor in a delusion might be an abnormality in this processing stage so that it leads to the initial adoption of the false belief, even without an unusual experience.

If the first factor is an unusual experience then this processing stage leads from the experience, by personal-level processes of explanation or endorsement, to the false proposition being initially adopted as a belief (Davies, this volume). If the first factor is a neuropsychological deficit that does not itself surface in consciousness as an unusual experience then there would be two possible routes to the initial adoption of the false belief. One possibility would be that, while the first deficit does not itself surface in consciousness, subsequent unconscious processing gives rise to an unusual experience and personal-level processes lead from that downstream experience to the false belief. The other possibility would be that the belief arises as the result of wholly unconscious processes, including processes of unconscious hypothesis generation and confirmation.

2.4 Motivation in the two-factor framework

The neuropsychological (two-deficit) version of the two-factor theory of delusions offers no place to motivational factors. But it seems plausible that, in some cases of delusion, motivation may play a role. Peter Butler (2000) describes a persuasive example.

Patient BX suffered a severe closed head injury in a motor vehicle accident and, even after a year of intensive inpatient rehabilitation, was still paralysed and confined to a wheelchair, unable to eat (nil-by-mouth status) or to speak without the aid of a voicing electronic communicator. He developed the delusion that he had recently married his former partner – who, in reality, had broken off all contact with him a few months after his accident (reverse Othello syndrome: a delusional belief in the fidelity of a romantic partner). This delusion persisted for some months before BX began to accept that he and his former partner were not married and that their separation was final. Butler’s summary of the case is as follows (2000, p. 89):

BX’s catastrophic TBI [traumatic brain injury] and subsequent realization of impairment seem likely to have occasioned multiple damage to his self-concept.

... In response to his demoralization and loneliness, he seems to have ... retreated into elaborate delusions concerning [his former partner] as a final defense against depressive overwhelm.

Although BX suffered severe brain injury, the case report does not suggest any specific neuropsychological anomaly that would play a key role in explaining why the proposition that he had married his former partner seemed salient and credible to BX. It does not suggest any candidate first deficit but it does suggest a first factor. The proposed explanation for the fact that BX came to regard the false proposition as a salient and
serious hypothesis, a credible candidate for belief, is that believing this proposition was a
defence against depressive overwhelm. Adopting the belief that he was enjoying a
fulfilling marriage, ‘seemed to go some way toward reconfering a sense of meaning to
[BX’s] life experience and reintegrating his shattered sense of self’ (ibid.).

On the basis of cases like that of patient BX, Ryan McKay, Robyn Langdon and Max
Coltheart (2005; see also this volume) suggest that the two-factor framework should
allow motivational factors as possible first factors. They also suggest that motivational
factors may ‘play a role in the second-factor evaluation of [doxastic] input’ (2005,
pp. 318–9). These two suggestions – that motivation may figure in the first or the second
factor – can be distinguished in terms of the two questions that we ask in any case of
delusion, ‘Where did the delusion come from?’ and ‘Why does the patient not reject the
belief?’

The first suggestion is that motivation may figure in the explanation of why the false
proposition (such as the proposition that BX had married his former partner) seemed to
the subject to be somewhat salient and credible or why the subject initially adopted that
proposition as a belief. As Alfred Mele notes (this volume, p. 000; see also 2001, pp. 29–30),
‘desires can influence which hypotheses occur to one and affect the salience of
available hypotheses’. The second suggestion is that motivation may figure in the
explanation of why the subject did not subsequently reject the false proposition on the
grounds of its implausibility and its incompatibility with a mass of available evidence. As
Mele observes, desires can surely influence the way that someone gathers, attends to, and
interprets evidence (this volume, pp. 000–00; 2001, pp. 26–30). It is plausible that cases
of delusion where motivation figures in the second factor will also be examples of self-
deception according to Mele’s (1997, 2001) account of that phenomenon (Mele, this
volume; Davies, this volume).

3. Anosognosia in the Two-Factor Framework

With this much by way of summary description of the two-factor framework, we are now
in a position to address the first of our four questions about anosognosia considered as a
delusion.

How would explanations of anosognosia fit into the two-factor framework for
understanding delusions?

A patient with anosognosia believes that he or she can move an arm or leg that is, in
reality, paralysed. The patient’s belief is false but, unlike the false beliefs in some other
delusions, it is not bizarre or exotic. The vast majority of us believe that we can move our
arms and legs but, in the case of the patient with anosognosia, this long-held belief is
newly false as a result of his or her recent paralysis. Our proposal is that, despite this
difference between anosognosia and other delusions, anosognosia fits the two-factor
framework.

3.1 The first factor in anosognosia

In most delusions, the role of the first factor is to explain how a bizarrely false
proposition came to be regarded as a credible candidate for belief. The first factor may be
an unusual experience or neuropsychological deficit that prompts a new and bizarrely
false belief. In anosognosia, the role of the first factor is to explain how a familiar but
now false proposition continued to seem credible in the patient’s changed circumstances.
The first factor may be a neuropsychological deficit that prevents the patient from having
the unusual experiences of motoric failure that would have prompted revision of the patient’s long-held belief. The first factor in anosognosia impairs the patient’s concurrent awareness of paralysis.

Some theorists have proposed that somatosensory loss – particularly, proprioceptive loss – is a factor in anosognosia for hemiplegia (Levine, 1990). Some have proposed that unilateral neglect is ‘a notable suspect in anosognosia’ (Vuilleumier, 2004, p. 10). Some have proposed that, as the result of an impairment to the intentional-preparatory systems involved in motor control, paralysis is not detected (Heilman, 1991; Heilman, Barrett and Adair, 1998; see also Frith, Blakemore and Wolpert, 2000). In the two-factor framework, all these theorists can be regarded as proposing candidate first factors in the aetiology of anosognosia. If the patient’s paralysis were vividly experienced as such then the long-held belief would no longer seem credible. It continues to seem credible because a first factor impairs the patient’s sensory or perceptual experience of motoric failure.

We can see evidence of the first factor at work in a report by Anjan Chatterjee and Mark Mennemeier (1996) of retrospective observations by three patients who had recovered from their anosognosia (which had lasted from a few hours to about a week) (1996, p. 227):

E: What was the consequence of the stroke?
HS: The left hand here is dead and the left leg was pretty much.
HS: (later): I still feel as if when I am in a room and I have to get up and go walking . . . I just feel like I should be able to.
E: You have a belief that you could actually do that?
HS: I do not have a belief, just the exact opposite. I just have the feeling that sometimes I feel like I can get up and do something and I have to tell myself ‘no I can’t’.

For patient HS, the idea that he can move his paralysed limbs is still powerfully credible even though he is now, without a second factor, able to reject it. Another patient, EM, when asked, ‘Can you raise the left [arm]?’ responds: ‘It feels like it’s rising, but, it’s not’ (ibid., p. 229).

3.2 Specific memory impairment as a first factor in anosognosia

Earlier (section 2.3), we said that an unusual experience or neuropsychological deficit does not, by itself, explain how a false proposition came to be initially adopted as a belief. In the aetiology of a delusion, there is a processing stage leading from the experience or deficit to the initial adoption of the belief. We also said that, in principle,
the first factor in a delusion might be an abnormality in this processing stage so that it leads to the initial adoption of the false belief, even without an unusual experience.

Now, in the case of anosognosia, we have mentioned somatosensory loss, unilateral neglect, and impaired intentional-preparatory systems as candidate first factors. These neuropsychological deficits would give rise to the absence of the kind of unusual experience that would have prompted revision of the patient’s long-held belief. But there is also another kind of first factor that we should consider. For even a patient who did have concurrent sensory or perceptual experiences of his motoric failure might not be prompted to revise his long-held belief if the processes that would normally lead from experience to belief were to be impaired.

Marcel and colleagues discuss the case of patients who are concurrently aware of motoric failure yet deny their motor impairments in response to interview questions and they suggest that it is important to distinguish between ‘immediate episodic experience’ and ‘long-term generic memory’ (2004, pp. 32, 34):

Thus we might say that many anosognosic patients are conscious of their motor . . . deficits when instances of them occur, but that they fail to remember them in any long-term or generic way. In order to learn from experiences we may have to integrate episodic memories . . . into generic . . . representations. Acquaintance must be transformed into knowledge.

House and Hodges (1988) discuss similar issues. Their patient’s failure to acknowledge her motor impairment and her ‘obstinate denial of handicap’ (1988, p. 115) – that is, her consistent failure to appreciate the consequences of her motor impairment – persisted despite the fact that ‘the paralysis of the limbs was brought to conscious awareness by examination’ (ibid.). Five minutes after it was demonstrated to her that her left arm was completely paralysed and her left leg nearly completely paralysed, she returned to an unrealistically high rating (six out of ten) for the strength of both limbs. This patient did not show any signs of somatosensory loss or unilateral neglect and House and Hodges suggest that her anosognosia is best explained in terms of a specific impairment of the systems that would normally allow information from the limbs to be integrated into beliefs (ibid.):

Thus although primary sensations may be intact they are not assimilated to lead to a modification of central schemata, and their meaning for functioning of the limbs is lost to the individual.

The suggestion that a specific memory impairment of this kind might play a role in some cases of anosognosia receives support from a study of epilepsy patients undergoing the Wada procedure in preparation for temporal-lobe surgery. The procedure involves injection of a barbiturate into one or the other carotid artery with the result that one hemisphere of the brain is selectively anaesthetised. During this procedure, subjects suffer weakness of the side of the body opposite to the injection. Because left-side barbiturate injections produce language impairments as well as right-side weakness, many studies proceed by asking the patients questions after the effects of the barbiturate have resolved. A typical finding from such studies is that many patients fail to acknowledge their earlier weakness. In a study of thirty-one epilepsy patients by Katherine Carpenter and colleagues (1995), twenty-seven patients failed to recall having had left-arm weakness when questioned ten-to-fifteen minutes after a right-side barbiturate injection and twelve patients denied having had right-arm weakness when questioned after a left-side injection.
Nine of the patients were also questioned about their left-arm weakness early after right-side injection. Five of these nine patients denied their left-arm weakness early, while the effects of the barbiturate were still present, and also later, when the effects of the injection had resolved. But the most interesting finding for present purposes is that three of the four patients who acknowledged their left-arm paralysis at the time when it occurred failed to recall it when questioned later, even though the barbiturate did not induce a general memory impairment in these patients. Carpenter and colleagues conclude (p. 249): ‘[I]n some patients failure to recall left arm weakness can be attributed to unawareness at the time. In others it seems to be due to a specific memory deficit.’ They also suggest that the relevant memory function may be subserved by structures in the right temporal lobe of the brain.

It is important to stress that, in this discussion of an additional candidate first factor in anosognosia, we are considering impairments of memory or integration that are relatively specific – perhaps specific to information about the movements or positions of parts of the subject’s body (see Carpenter et al., 1995, p. 250, for discussion). In terms of its consequences for belief, a specific impairment of this kind – roughly, a failure to remember the experience of paralysis – would be similar to impaired concurrent awareness of the paralysis.

3.3 The second factor in anosognosia

If a first factor is present then a subject may be concurrently unaware of failures to move a paralysed limb – or else may be concurrently aware of these failures but unable to integrate this information into beliefs. But a first factor is not sufficient to account for anosognosia. Marcel and colleagues give vivid expression to the argument for a second factor (2004, p. 35):

... it is not just that they fail motorically. The consequence of such [motoric] 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, ... or that they rapidly forget them, or that they hallucinate the success of the intended action (as opposed to the movement).

In short, despite the first factor, a patient is likely to be presented with a mass of other evidence of his or her paralysis. So something more – a second factor – is needed to explain why patients with anosognosia do not make appropriate use of this evidence.

It remains to consider the neural basis and functional nature of the second factor in anosognosia. If Coltheart is right to assume that the second factor ‘is the same in all

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6 See Karnath, Baier and Nägele (2005) for the finding that anosognosia is associated with damage to a structure, the right posterior insula, that may be involved in ‘integrating input signals related to self-awareness and to one’s beliefs about the functioning of contralateral body parts’ (p. 7137).

7 This argument for a second factor might not apply if the paralysis were to last only for a very short time, during which time the patient was lying in bed and was not engaged in any everyday activities (as in the Wada procedure, for example). It also might not apply if, for some reason such as sedation or lack of arousal, evidence of paralysis was not available to the patient. In such a case, anosognosia for hemiplegia might occur even in the absence of a second factor and might not constitute a delusion.

8 In a single-case study with both structural (MRI) and functional (SPECT) imaging of the brain, Venneri and Shanks (2004) describe a patient, EN. She had anosognosia for hemiplegia persisting twenty-six months after a right-hemisphere stroke and other delusions including misidentification of place,
people with monothematic delusion’ (2005b, p. 154) then earlier suggestions and speculations (section 2.2) would lead us to investigate the following hypothesis:

**Second Factor Hypothesis:** The second factor in anosognosia is an impairment of working memory or executive processes with a neural basis in the right frontal region of the brain.

### 3.4 The discovery theory of anosognosia

David Levine’s (1990) discovery theory is an example of an explanation of anosognosia that fits the two-factor framework. Levine argues that, given a somatosensory loss, paralysis is not phenomenally immediate. In the presence of a first factor, knowledge of paralysis requires a process of discovery that is not especially demanding for cognitively intact individuals. But anosognosia for hemiplegia arises when the first factor is accompanied by additional impairments that impact negatively on observation and inference.

Levine’s proposal that cognitive impairments are involved in anosognosia has been disputed. However, the claim that anosognosia can occur without cognitive impairments is often made without a full neuropsychological assessment of the patients. For example, several studies have employed the Mini-Mental State Examination (MMSE; Folstein, Folstein and McHugh, 1975) to assess cognitive function (orientation, memory, attention and calculation, language, and visual construction). Marian Small and Simon Ellis (1996) found that only nine out of twenty patients with anosognosia for their hemiplegia scored below cut-off on this test; Marcel and colleagues (2004) reported that patients with anosognosia were no more likely to score below cut-off on the MMSE than those who acknowledged their impairments. But we need to notice that the MMSE, a general test of cognition, does not provide any detailed assessment of cognitive functions such as working memory or executive processes that seem especially relevant in cases where an individual is, as Levine puts it ‘unable to assimilate information from a variety of sources to form a consistent and accurate judgement’ (1990, p. 254).

Levine, Calvanio and Rinn (1991) compared two groups of patients following a right-hemisphere stroke, six with severe anosognosia for their hemiplegia lasting for at least a month and seven for whom anosognosia had, if present at all, lasted for a few days at most. All the patients with anosognosia had severe somatosensory deficits, as did two patients without anosognosia. Levine and colleagues conducted a detailed neuropsychological assessment of all the patients and found that the patients with anosognosia, considered as a group, performed significantly worse than the patients without anosognosia on a number of tests including the Orientation, Digit Span Forward, Story Recall, and Mental Control subtests of the Wechsler Memory Scale (WMS; Wechsler, 1945), the Arithmetic Reasoning and Block Design subtests of the Wechsler Adult Intelligence Scale–Revised (WAIS-R; Wechsler, 1981), and a test of controlled word association (Benton and Hamsher, 1976). The assessment included a test of somatoparaphrenia, and persecutory delusions. Venneri and Shanks propose that an explanatory account of anosognosia including cases such as EN might draw support from ‘findings made in studies assessing the role of the right frontal lobe in the retrieval and monitoring of self related memories, as well as those evaluating the function of the right hemisphere in verifying the truthfulness of recollections’ (2004, p. 236). They suggest that, in the case of EN, ‘there may be a barrier to the natural awareness of hemiparesis as part of a wider syndrome of reality monitoring failures which also have allowed the development of other abnormal beliefs’ (ibid., p. 237).
working memory, Digit Span Backward (WMS), and a test of executive processes, a modified version of the Wisconsin Card Sorting Test (Berg, 1948). On both these tests, the group with anosognosia performed worse than the group without anosognosia although, in the case of Digit Span Backward, the trend did not reach statistical significance.

The results of the neuropsychological assessment are consistent with Levine’s two-factor claim that both somatosensory loss and cognitive impairments are required for anosognosia for hemiplegia. Indeed, Levine and colleagues claim that ‘[t]here has been no report of a patient with persistent [anosognosia] whose mental status was carefully examined and found to be normal’ (1991, p. 1777). But the study has some limitations. The patients were elderly and not well oriented to time and place and several of them did not complete all the tests. Overall, the neuropsychological assessment provides little information about which cognitive impairments best discriminate patients with somatosensory loss and anosognosia from patients with somatosensory loss but without anosognosia and so which cognitive impairments might be most important for the second factor in anosognosia.

Despite these limitations, however, this is an important study and Levine and colleagues offer a vivid qualitative description of the role of cognitive impairments in these patients with anosognosia (1991, p. 1779):

The mental disorganization and poor mental control of patients with persistent [anosognosia] prevent their developing the hypothesis that they are paralyzed and preclude their taking the necessary steps to verify it. Their mental inflexibility prevents them from rejecting the long-held belief ... that they have four fully mobile limbs, even though contradictory evidence is overwhelming.

4. Motivation in Anosognosia

Motivation might, in principle, figure in the first factor or the second factor (or both) in the aetiology of a delusion (section 2.4). Having seen how anosognosia for hemiplegia fits into the two-factor framework we turn to our second question about anosognosia considered as a delusion.

What are the prospects for an account of anosognosia at least partly in terms of motivation?

Over the past century, motivational explanations for anosognosia have been received with varying degrees of enthusiasm. During the 1950s, Edwin Weinstein, Robert Kahn and colleagues wrote a series of papers (Weinstein and Kahn, 1950, 1951, 1953; Weinstein, Kahn, Malitz and Rozanski, 1954) and an influential book (Weinstein and Kahn, 1955) putting forward a motivational account of anosognosia. But, as we shall see (section 4.4), recent work has often been severely critical of the motivational approach.

4.1 Patients with brain injury

At the end of a study of anosognosia in twenty-two patients with brain tumour, Weinstein and Kahn (1950) propose that anosognosia results from a need to be well that is ‘present in all people’ but which appears ‘in a distorted fashion’ or ‘in a new pattern of organization’ following brain injury (1950, pp. 789–91). As a result of this reorganisation, the patients deny whatever they feel to be seriously wrong with them. For the patients in Weinstein and Kahn’s study, anosognosia always involved more than one impairment or illness and, in the various types of anosognosia, ‘the same kinds of
confabulation and the same evasions, euphemisms, displacements and projections were used’ (ibid., p. 788). All the patients were temporally disoriented, particularly for time of day. Most were also disoriented for place. They claimed to be at home rather than in hospital or else accepted that they were in hospital but gave the hospital an incorrect location, usually closer to their home. Weinstein, Kahn and colleagues say that the manifestations of disorientation are also ‘symbolic expressions of the drive to be well’ (Weinstein et al., 1954, p. 57).

In a study of 100 patients with hemiplegia (ninety-five following a cerebrovascular accident), Morton Nathanson and colleagues (Nathanson, Bergman and Gordon, 1952) found that all of the twenty-eight patients with anosognosia showed some degree of disorientation. Spatial disorientation was in the direction of home or of some other location less suggestive of illness than a hospital; temporal disorientation was in the direction of a time of better health – actual or hoped for. Nathanson and colleagues regard disorientation as a sign of a psychological defence mechanism of motivated denial – partly because of the slips that the patients made, revealing some implicit appreciation of the fact that they were unwell and in hospital even while they explicitly and repeatedly maintained that they were well and, for example, at home (see also Turnbull, Berry and Evans, 2004).

4.2 Patients without brain injury

Denial of illness has been observed alongside a wide range of medical conditions, including heart disease and myocardial infarction, cancer, diabetes, and spinal cord injury. Because these medical conditions do not involve brain injury, motivational explanations of anosognosia for these illnesses have been widely espoused.9

In patients with these conditions, denial of illness has been credited with both negative and positive outcomes (see Kortte and Wegener, 2004, for a review). In the case of heart disease, the impact of denial has been shown to vary depending on the illness stage at which it is adopted. During the early stages of illness and at the point of hospitalisation, it has been associated with delay in seeking treatment and, in the post-hospitalisation phase, it has been linked with poorer compliance with treatment regimes and compromised avoidance of risk factors. However, in the hospital recovery period, denial of illness has been linked with more positive effects including protection from negative emotional states, reduced medical complications, and lower levels of anxiety and depression. These positive outcomes, coupled with the fact that denial can occur in conditions of non-cerebral aetiology, have been taken to support the proposal that anosognosia may have a motivational basis.

4.3 Premorbid personality styles

Weinstein and Kahn (1950) state that ‘it appears that the occurrence of anosognosia is related to the pattern of the premorbid personality’ (p. 780; see also Weinstein and Kahn, 1953). They associate anosognosia with a strong premorbid drive towards perfection and with the view that illness constitutes an imperfection or disgrace. When patients with this

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9 For denial of heart disease or myocardial infarction, see Levine et al. (1987), Stenstrom et al. (2005); for denial of cancer, see McKenna, Zevon, Corn and Rounds (1999), Rousseau (2000); for denial of diabetes, see Lo and MacLean (2001); for denial of spinal cord injury, see Livneh and Martz (2003), Martz, Livneh, Priebe, Wuermser and Ottomanelli (2005).
premorbid personality style are disoriented for place and time, perhaps with reduced spontaneity and initiative and with disturbed affect, they may disregard the constraints of reality and deny their impairment. However, the claim that premorbid personality has an important role in the aetiology of anosognosia has been challenged on the grounds that patients with anosognosia for their hemiplegia may frankly acknowledge other medical conditions such as heart attack or stroke (Cutting, 1978, p. 553).

Small and Ellis (1996) conducted a comprehensive analysis of anosognosia for hemiplegia, investigating proprioceptive, cognitive and personality factors. The results of the Eysenck Personality Questionnaire (Eysenck and Eysenck, 1987) demonstrated that denial of hemiplegia cannot be attributed to extroverted, neurotic, or psychotic post-morbid personality styles, nor to dissimulation or ‘faking good’. Moreover, investigations of premorbid personality (as assessed by the patient and by a close friend or relative) revealed that patients with anosognosia were not classified as perfectionist significantly more often than patients with hemiplegia and visuospatial neglect in the absence of denial or patients with acute spinal cord lesions. However, patients with anosognosia were twice as likely as patients in the other two groups to rate themselves as ‘finding it quite/very hard to admit illness’ (Small and Ellis, 1996, p. 358). Although this trend towards finding it hard to admit illness did not reach statistical significance, the authors allow that ‘personality factors may contribute to denial in some patients’ (ibid., p. 362).

4.4 The case against motivation

In an influential book chapter, Edoardo Bisiach and Giuliano Geminiani (1991, pp. 25–6) list ‘eight important facts concerning anosognosia related to neurological disorders that affect one side of the body’ and argue that these facts constitute problems for interpretations of anosognosia as ‘a defensive adaptation against the stress caused by the illness’ (1991, p. 24).

We should begin by noting that Bisiach and Geminiani’s concerns are rather different from ours. They argue against explanations of anosognosia that are cast wholly in terms of motivated denial: ‘[the motivational] explanation per se fails to account for [the] eight important facts concerning anosognosia’ (ibid., p. 25; emphasis added). They also state explicitly that two of their facts are problematic for another class of explanations, namely, ‘interpretation[s] of anosognosia in terms of general confusion or intellectual impairment’ (p. 24). Bisiach and Geminiani would have achieved their aim if they could show that there is no explanation cast wholly in terms of motivated denial – or wholly in terms of general cognitive impairments – that would cover all cases of anosognosia. But, even if they achieved their aim, it would remain plausible that motivation is a factor in some cases of anosognosia. In the case of patient BX (Butler, 2000), it is immensely plausible that one factor in his delusion was motivated denial of his separation from his partner as a defence against depressive overwhelm. It would surely be remarkable if

Levine and colleagues (1991) administered a questionnaire designed to reveal attitudes towards illness and also the Minnesota Multiphasic Personality Inventory (MMPI; Dahlstrom, Welsh and Dahlstrom, 1975). The results from the questionnaire did not provide any evidence that the patients with anosognosia had a different attitude towards illness from the patients without anosognosia. The results on the MMPI did reveal some differences between the two groups, but the authors suggest that these are best explained in terms of the mental inflexibility of the patients with anosognosia.
motivation were sometimes a factor in delusions such as reverse Othello syndrome yet never a factor in the delusion of anosognosia.

Our concern is with explanations of anosognosia within the two-factor framework. So it is important to consider whether Bisiach and Geminiani’s arguments extend to two-factor accounts. We shall focus particularly on the possibility that, in some cases of anosognosia, motivationally biased handling of the available evidence may figure in the second factor. If motivation were to play a major biasing role then such cases would plausibly be examples of self-deception (Mele, this volume; Davies, this volume). We do not suggest that motivation is a factor in all cases of anosognosia and we allow that, if motivation is sometimes a factor, it may do its work in conjunction with cognitive impairments.

We shall not discuss all eight of Bisiach and Geminiani’s putatively problematic facts, but four of the more important ones are these: time course – anosognosia is usually present only during the acute stage of the illness; remission – in some patients, vestibular stimulation (by cold water poured into the ear) results in remission of anosognosia for hemiplegia; selectivity – patients may show anosognosia for some impairments but not others; hemispheric differences – anosognosia is more common following right-hemisphere damage than following left-hemisphere damage.11 (Of these four, remission and selectivity are said also to be problematic for accounts of anosognosia in terms of general cognitive impairments.)

We shall argue that these putatively problematic facts do not pose any problem for two-factor accounts of anosognosia. In each case, the fact can be explained in terms of the first factor quite independently of whether the second factor is cognitive or motivational or both. In some cases, the fact can also be explained in terms of a partly or wholly motivational second factor.

4.4.1 The time course of anosognosia

Anosognosia is usually present only during the acute stage of the illness whereas, Bisiach and Geminiani suggest, ‘[a] goal-directed denial of illness should be characterized by an evolution opposite to that commonly observed by the clinician’ (1991, p. 25). One would expect that neurological disorders (including cognitive impairments) might improve in the days following a stroke while motivated denial might take some time to develop as a strategy for coping with illness and impairment. But what is typically found is that the time course of anosognosia patterns with neurological disorders.

In the two-factor framework, we can allow that anosognosia may be present in the acute stage following a stroke, but may then resolve because the first factor in its aetiology resolves. This approach would be consistent with the fact that most cases of

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11 The four problems mentioned in the text are numbers 1, 8, 4 and 3 on Bisiach and Geminiani’s list. The other four problems are as follows (1991, pp. 25–6): 2. Patients seldom display anosognosia for neurological disorders that do not involve neural structures responsible for higher cognitive functions. 5. Anosognosia may be manifest in verbal but not nonverbal behaviour, or vice versa. 6. Some patients with hemiplegia display misoplegia, rather than anosognosia (for discussion, see Turnbull, Jones and Reed-Screen, 2002). 7. Anosognosia may create ‘serious impediments and even danger’ rather than having positive outcomes. We must leave it to the reader to judge whether any of the four problems that we do not discuss in the text present problems for a two-factor account of anosognosia with motivation as a candidate second factor.
unilateral neglect, which is a candidate first factor in anosognosia, improve rapidly over the first ten days following stroke (Stone, Patel, Greenwood and Halligan, 1992).

We might also account for the time course of anosognosia in terms of the second factor. Bisiach and Geminiani note that, in the early stage of illness, when anosognosia is most often present, ‘the patient’s vigilance may be clouded and his evaluation of the pathological event is still incomplete’ (1991, p. 25). Their suggestion appears to be that incomplete information, and confusion or clouded vigilance, may be factors in anosognosia in the first hours or days following brain injury. In that case, anosognosia that is present in the acute stage may resolve as the neurologically produced confusion or clouded vigilance resolves. This suggestion can be adopted by an advocate of a partly motivational second factor in some cases of anosognosia. Indeed, Weinstein and Kahn’s proposal (discussed in section 4.3) was that a premorbid drive towards perfection might manifest itself in anosognosia when, as a result of other disturbances caused by brain injury, the patient disregards the constraints of reality.

4.4.2 Remission of anosognosia following vestibular stimulation

In some patients, vestibular stimulation (by cold water poured into the left ear) results in remission of anosognosia for hemiplegia (Cappa, Sterzi, Vallar and Bisiach, 1987). This fact seems to be problematic for accounts of anosognosia wholly in terms of motivated denial because it is not clear why vestibular stimulation should change a patient’s motivation or coping strategies. It also seems to be problematic for accounts of anosognosia wholly in terms of general cognitive impairments. But the facts about vestibular stimulation will not pose a problem for two-factor accounts if remission of anosognosia can be accounted for in terms of remission of the first factor.

Vestibular stimulation reduces the classical symptoms of unilateral neglect (Cappa et al., 1987; Rubens, 1985) and may also produce improvements in conditions that are associated with neglect, such as somatosensory deficits (Vallar, Bottini, Rusconi and Sterzi, 1993). In some cases, vestibular stimulation even results in improved motor performance. Gilles Rode and colleagues (Rode, Perenin, Honoré and Boisson, 1998; see also Rode et al., 1992) found that seven out of nine right-hemisphere stroke patients with unilateral neglect and hemiplegia showed improved limb movement or strength, lasting up to fifteen or twenty minutes after vestibular stimulation. The authors conclude that there is a ‘motor neglect component’ – ‘one of the many manifestations of the unilateral neglect syndrome’ (1998, p. 260) – in the motor impairments shown by these seven patients. In this case, an apparently puzzling fact about improved motor performance following vestibular stimulation is explained by postulating that unilateral neglect is a factor in the impaired motor performance before vestibular stimulation. The apparently puzzling fact about remission of anosognosia following vestibular stimulation might be explained in a similar way if neglect is sometimes a first factor in the aetiology of anosognosia (see Vallar, Bottini and Sterzi, 2003, for a review).

4.4.3 The selectivity of anosognosia

Anosognosia may be selective. Patients may fervently deny their paralysis but frankly acknowledge their heart attack or stroke (section 4.3). Some patients who have both hemianopia and hemiplegia deny their visual field deficit while acknowledging their motor impairments (Bisiach, Vallar, Perani, Papagno and Berti, 1986). Some patients show anosognosia for paralysis of their left leg while acknowledging that they cannot move their left arm (ibid.).
Selectivity poses no problem for two-factor accounts of anosognosia because it can be explained in terms of the first factor. Let us stipulate that a hypothetical patient shows a fixed degree of motivational bias, or of cognitive impairment, in handling evidence – whether it concerns the functioning of the arm or of the leg. Suppose, now, that the patient is concurrently aware of motoric failures of his left arm but concurrently unaware of motoric failures of his left leg. Then, it is surely more likely that the patient would deny paralysis of the left leg than of the left arm.

It may also be possible to explain selectivity in terms of a partly or wholly motivational second factor. Weinstein and Kahn (1950, p. 774) note that ‘the patient might deny the major disability but lay stress on some trivial aspect of his condition, a form of displacement’. More generally (ibid., p. 789): ‘the patient denies whatever he feels is seriously wrong with him, whether it is a hemiplegia, a craniotomy or a sense of inadequacy’. It is possible to imagine, for example, that a patient might feel paralysis to be more serious than a heart attack or impairment of the left leg to be a more serious loss than impairment of the left arm. The resources available for explanations of selectivity in terms of motivated denial might be further extended by appeal to a patient’s ranking of impairments, not only in terms of seriousness, but also in terms of other variables such as social acceptability.

4.4.4 Hemispheric differences in anosognosia

Anosognosia is more common following right-hemisphere damage than following left-hemisphere damage. This fact, like the previous three, might be accounted for in terms of the first factor in the two-factor framework. Candidate first factors in anosognosia include somatosensory loss, unilateral neglect, impaired intentional-preparatory systems, or specific memory impairment, and it is plausible that these candidate first factors, with the exception of somatosensory loss, are predominantly associated with right-hemisphere damage.

First, unilateral neglect is quite strongly associated with anosognosia (see Jehkonen, Laihosalo and Kettunen, 2006, for a recent review) and persisting anosognosia is almost invariably accompanied by neglect (Cocchini, Beschin and Della Sala, 2002). Neglect, like anosognosia, is reported more frequently after right-hemisphere than after left-hemisphere damage, although one study has found similar rates of occurrence of neglect in the first few days following right- and left-hemisphere damage (Ogden, 1985). Second, Kenneth Heilman and colleagues propose that, ‘in normal subjects the right-hemisphere intentional systems can help activate the motor systems for both right and left hands [but] the left hemisphere’s intentional system primarily activates the right hand’ (Heilman, Barrett and Adair, 1998, p. 1908). If this is correct then the motor-intentional deficit for the left hand following right-hemisphere damage might be more severe than the deficit for the right hand following left-hemisphere damage. So detection of left-side weakness following right-hemisphere damage might be more difficult than detection of

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12 Recent studies of patients in the first ten days following a stroke suggest a rate of occurrence for anosognosia of 17%–21% (Appelros, Karlsson and Hennerdal, 2007; Appelros, Karlsson, Seiger and Nydevik, 2002; Pedersen, Jørgensen, Nakayama, Raaschou and Olsen, 1996) and 21%–42% for right-hemisphere patients (Appelros, Karlsson, Seiger and Nydevik, 2003; Jehkonen, Ahonen, Dastidar, Laippala and Vilkki, 2000). Studies also suggest a rate of occurrence for unilateral neglect of 23% (Appelros et al., 2002; Pedersen, Jørgensen, Nakayama, Raaschou and Olsen, 1997), and 32%–42% amongst right-hemisphere patients (Appelros et al., 2003; Jehkonen et al., 2000; Pedersen et al., 1997).
right-side weakness following left-hemisphere damage. Third, the specific memory impairment proposed by Carpenter and colleagues (1995) is associated with right-temporal-lobe damage.

Thus, it is plausible that three candidate first factors in anosognosia are predominantly associated with damage to the right hemisphere. Somatosensory loss – if it is distinguished from somatosensory neglect – can occur after damage to either hemisphere. The somatosensory processes that are lateralised to the right hemisphere are those involved in somatosensory attention (Coghill, Gilron and Iadarola, 2001; Remy et al., 1999).

The fact about hemispheric differences in anosognosia is sometimes presented as a problem for motivational accounts, such as Weinstein and Kahn’s, that appeal to premorbid coping strategies. Thus, Heilman and colleagues (1998, p. 1904) remark: ‘The coping strategy that one uses in life should not influence which side of the brain becomes damaged by stroke.’ But, Weinstein and Kahn’s account might be defended against this objection, as against the worry about the time course of anosognosia, by appeal to a partly motivational second factor. For it might be that damage to the right hemisphere is more likely to produce the disturbances in the context of which the premorbid coping strategy will be manifested.

In any case, it is not essential that motivational factors in the aetiology of anosognosia should have been present before the onset of the patient’s illness. Motivational, affective, and personality changes might result from the same brain injury that produces hemiplegia. There is a substantial body of literature on different emotional reactions following right- and left-hemisphere damage and we should briefly explore the question whether hemispheric differences in emotion may help to explain the hemispheric differences in anosognosia.

4.5 Hemispheric differences in emotion

In an influential paper, Guido Gainotti (1972; see also 1969) reports that catastrophic or anxious-depressive reactions are associated with left-hemisphere damage while indifference reactions are more frequent following right-hemisphere damage. These findings invite the view that the right hemisphere is associated with negative emotions and the left hemisphere with positive emotions. This valence hypothesis yields a suggestion about anosognosia; namely, that it results when, following damage to the negative emotional systems of the right hemisphere, only the predominantly positive left-hemisphere systems are intact. This suggestion seems initially plausible but it faces a number of challenges. Referring to Gainotti’s paper, Bisiach and Geminiani (1991, p. 25) remark that the suggestion about anosognosia is ‘contradicted by the fact that patients who deny their left hemiplegia or seem to be totally unaware of it may be intolerant of minor disorders affecting the right side of the body’.

Although Gainotti’s (1972) findings invite the valence hypothesis, Gainotti himself adopted a different interpretation. He found that, amongst left-hemisphere patients, catastrophic reactions were associated with aphasia and were ‘usually triggered by frustrating, repeated attempts at verbal expression’ (2003, p. 725). He therefore interpreted the emotional reactions of the left-hemisphere patients as being often appropriate and the reactions of the right-hemisphere patients as being less appropriate. He proposed that (1972, p. 52): ‘the right, non-verbal hemisphere [should be considered] as more important from the “emotional” point of view’. This right-hemisphere hypothesis – that the right hemisphere is dominant for the perception and expression of emotions,
irrespective of valence – yields a second natural suggestion about anosognosia, but it faces many of the same challenges as the first.

Some recent work (Davidson, 2001; Davidson and Irwin, 1999) supports a version of the valence hypothesis that is conceptualised in terms of the positive and negative responses of approach and withdrawal (Kinsbourne, 1978). This version of the valence hypothesis has been combined with a version of the right-hemisphere hypothesis for the recognition of expressions of all emotions, irrespective of valence (Root, Wong and Kinsbourne, 2006). Meanwhile, Gainotti (2000, 2003; see also Hagemann, Hewig, Seifert, Naumann and Bartussek, 2005) has put forward a different refinement of the right-hemisphere hypothesis. He proposes that hemispheric differences are related, not to valence – negative (right) versus positive (left), but to level of processing – schematic or automatic (right) versus conceptual or controlled (left). The hypothesis of right-hemisphere dominance extends to ‘automatic (expressive and autonomic) components of emotion’ (2000, p. 226) but the left hemisphere may play a critical role in ‘functions of control and of modulation of spontaneous emotional expression’ (ibid.).

None of these recent proposals about hemispheric differences in emotion immediately yields a compelling explanation of the hemispheric differences in anosognosia but, equally, they do not rule out the prospect of such an explanation. In any case, the controversy that surrounds the topic of the lateralisation of emotion makes no difference to our conclusion that hemispheric differences in anosognosia pose no problem for two-factor accounts. More generally, Bisiach and Geminiani’s (1991) influential case against motivation does not amount to a case against two-factor accounts of anosognosia that allow the possibility of motivational bias figuring as the second factor.

5. A Study of Cognitive Impairments in Patients with Anosognosia

A two-factor account of anosognosia can, in principle, allow motivation as a candidate second factor so that some cases of anosognosia may be examples of self-deception (section 2.4). Nevertheless, the dominant theoretical approaches to understanding anosognosia are not cast in terms of motivated denial. Our own view is that it is important to undertake detailed investigations of cognitive impairments in patients with anosognosia while also being alert to the possibility that motivation may sometimes be a factor. We now consider our third question about anosognosia as a delusion.

How should we investigate the role of cognitive impairments in anosognosia?

We propose that the role of cognitive impairments in anosognosia should be investigated by a detailed neuropsychological assessment of post-acute patients who are well oriented and sufficiently engaged in the activities of daily life that, even without immediate awareness of motoric failure, they would have adequate evidence of their motor impairments. The problem with this proposal is that, by comparison with anosognosia in the first few days following a stroke, persisting anosognosia is relatively rare.

When anosognosia does persist, it is likely to be accompanied by unilateral neglect. In our own review of the literature, we find only one clear case of anosognosia persisting at least three months after a stroke in the absence of unilateral neglect. 13 So one way to

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13 This is the case reported by House and Hodges (1988). In a review of patients with anosognosia reported in the literature, Cocchini and colleagues (Cocchini, Beschin and Della Sala, 2002) find only one case persisting more than one month post-CVA in the absence of unilateral neglect. This patient, who was studied by Bakchine, Crassard and Seilhan (1997), showed no sign of neglect on a test of line bisection but
investigate the role of cognitive impairments in anosognosia is to review detailed neuropsychological assessments of patients with persisting unilateral neglect.  

A study conducted by the first author (Aimola, 1999) involved nine patients (six male M1–M6, three female F1–F3; age 41 to 63 years) with neglect persisting at least three months following a unilateral stroke (Table 1). The patients were assessed on a large battery of neglect tests (twelve tests of extrapersonal neglect and two assessments of personal neglect) and all demonstrated both extrapersonal neglect and personal neglect.

Eight of the patients had right-hemisphere damage and one patient had left-hemisphere damage. All of the patients demonstrated severe motor impairments in the acute phase following their stroke. At the time of the study, all the patients had already been involved in an intensive inpatient neurorehabilitation programme. Five of the patients (F3, M1, M2, M3 and M5) were severely impaired and confined to wheelchairs; one patient (M4) was moderately impaired; and three patients (F1, F2 and M6) were only mildly impaired. See Table 1 for the results of two assessments of motor impairments: the test described by Bisiach and colleagues (1986) and the eight functional tests of hemiplegia (Gialanella and Mattioli, 1992).

5.1 Assessment of anosognosia

Anosognosia for motor impairments was assessed for upper limb and lower limb separately, on a scale proposed by Bisiach and colleagues (Bisiach and Geminiani, 1991; Bisiach et al., 1986):

0 = The disorder is spontaneously reported or mentioned by the patient in reply to a general question about his complaints (no anosognosia);
1 = The disorder is reported only following a specific question about the affected function (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).

Bisiach and colleagues (1986) and also Anna Berti and colleagues (Berti, Làdavas and Della Corte, 1996) have suggested that assessing anosognosia in patients without severe motor impairments is problematic, since a patient with only a mild impairment may quite accurately claim to be able to move the affected limb. Even for a mild impairment, we can, of course, distinguish conceptually between acknowledging it and denying it. But the problem is that failure to acknowledge a mild impairment may constitute only a minor mismatch, rather than a substantial mismatch, between the patient’s estimate of his or her ability to move the affected limb and the reality of the impairment. Such a minor departure from the truth might well fall within the range of normality and might not deserve the title of either ‘anosognosia’ or ‘delusion’.

this is not sufficient to conclude that the patient did not have neglect. The literature shows that neglect may fractionate into a variety of underlying components (Halligan, Marshall and Wade, 1989). For example, a patient may demonstrate neglect on a letter-cancellation test but not on a line-bisection test, or vice versa (Binder, Marshall, Lazar, Benjamin and Mohr, 1992). See Berti et al. (1996) and Dauriac-Le Masson et al. (2002) for cases of anosognosia without extrapersonal neglect but with personal neglect.

We note that unilateral neglect persisting more than three months after a stroke is rare. Most cases of unilateral neglect improve rapidly over the first ten days, and one study indicates that as few as ten percent of cases present in the first two or three days persist beyond three months (Stone et al., 1992).
We can respond to this problem by noting that even a mild impairment of movement or strength in a limb may present severe difficulties for the activities of daily living, such as eating, dressing, washing, and so on. Thus, in the eight functional tests of hemiplegia (Gialanella and Mattioli, 1992), even for the patients whose impairment was only mild (patients F1, F2 and M6), everyday activities were assessed as possible only with difficulty. If we consider motor impairments and their consequences for activities of daily living then we open up room for a substantial mismatch between a patient’s estimate of his or her abilities and the reality of the situation. Assessment of anosognosia for motor impairments and their consequences is a more complex matter than assessment of anosognosia for motor impairments alone. It is usually necessary to draw on information about the patients’ abilities provided by the patients’ families and neurorehabilitation staff. In the case of the study described here, it is important to note that the first author is a clinical neuropsychologist who had extended contact with each of the patients on an almost daily basis over a period of several months, and frequent contact with their families and staff. Scores for anosognosia for motor impairments and their consequences take account of her clinical judgement.

Anosognosia scores (0–3) for upper and lower limbs separately are summed to yield an overall anosognosia score for each patient (0–6; see Table 1). Two patients – F1 (the patient with left-hemisphere damage) and M4 – frankly acknowledged their motor impairments and the consequences for their everyday activities (overall anosognosia score = 0). They were distressed about the loss of their respective pastimes of playing golf and playing the guitar. Two other patients – F2 and M2 – demonstrated only mild anosognosia, acknowledging their limitations when asked specifically about them (overall anosognosia score = 2). For example, patient M2 was resourceful, and even over-ambitious, in arranging excursions for himself from the hospital to the nearby casino. But he never tried to get out of his wheelchair nor denied his limitations.

In contrast, patients F3, M1, M3 and M5, with moderate or severe motor impairments, all seriously underestimated the extent and the consequences of their impairments (overall anosognosia scores = 4–5). For example, patient M3, while sometimes acknowledging his impairments, repeatedly tried to get out of his wheelchair while at home alone and injured himself, finally having to be placed in a nursing home for his own safety.

Patient M6 showed only mild motor impairments and, on one of the tests (Bisiach et al., 1986), did not show any lower-limb weakness. Nevertheless, the eight functional tests of hemiplegia reveal, not only that everyday activities were assessed as possible only with difficulty, but also that a weakness of the left leg was evident when patient M6 was walking. Indeed, he sometimes required a cane, dragging his left leg behind him. Despite these serious limitations, patient M6 insisted that he could leave the hospital, live at home, and cook for the family – even though this proved clearly beyond him when he made short visits home. His anosognosia for motor impairments and their consequences was assessed as severe (overall anosognosia score = 6).

When we consider associations and dissociations between anosognosia and cognitive impairments (section 5.2 and Table 2), we focus on the patients with moderate or severe anosognosia (patients F3, M1, M3, M5 and M6) and group the patients with mild anosognosia (patients F2 and M2) together with the patients with no anosognosia (patients F1 and M4). In support of this grouping, we note that some studies use a three-point scale on which patients who acknowledge their impairment in response to a specific
question (Bisiach’s score of 1) are scored 0 and classified as not having anosognosia (Berti et al., 1996).

5.2 Neuropsychological assessment

We noted earlier that some theoretical claims about anosognosia have been based on studies that used only a general test of cognition, such as the Mini-Mental State Examination (MMSE), to assess cognitive impairments. It is methodologically important that the study described here includes a detailed neuropsychological assessment – as does the study of Levine and colleagues (1991). We shall summarise the assessment, beginning with tests on which all nine patients performed in the normal range and moving on to tests in four main areas of cognitive function – memory, sustained attention, working memory, and executive function – where impairments were apparent.15

5.2.1 Premorbid ability, orientation, language and comprehension, visuoperceptual function

National Adult Reading Test scores (NART II; Nelson and Willison, 1991), supplemented with background demographic information about educational and occupational accomplishments, indicated that the patients’ premorbid functioning was within the normal range (91 to 115). All the patients passed the Information and Orientation subtest of the Wechsler Memory Scale–Revised (WMS-R; Wechsler, 1987) without difficulty (range of 12 to 14; maximum = 14), demonstrating that there was no disorientation or confusion. All the patients also passed the tests of language and comprehension, although patient F1, following a left-hemisphere stroke, demonstrated mild communication problems. On the tests of visuoperceptual function, the patients demonstrated no problems with low-level perception.

5.2.2 Autobiographical, visual and verbal memory

The patients did not demonstrate difficulties with memory for people and incidents in their past and they all scored in the normal range on the Digit Span Forward and Logical Memory (Story Recall) Immediate and Delayed subtests of the WMS-R. However, four patients (F2, M2, M4 and M6) demonstrated serious problems on subtests of the Doors and People Test (DPT; Baddeley, Emslie and Nimmo-Smith, 1994). The was a double dissociation between anosognosia and impaired memory as demonstrated on the DPT. Patient M5 had anosognosia but not impaired memory; patient M4 had impaired memory but not anosognosia (Table 2).16

5.2.3 Sustained attention

Tasks that assess sustained attention or vigilance typically require the patient to monitor the presentation of intermittent sensory targets (visual or auditory) that may occur amongst a greater number of non-targets (distractors). Sustained attention has been associated with persisting unilateral neglect (Robertson, 2001) and with poor recovery

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15 For further details of the neuropsychological assessment and structural MRI, see Aimola, 1999; Maguire and Ogden, 2002.

16 Time issues prevented patient F3 from completing the full neuropsychological battery. She did not complete the Doors and People Test, the Test of Everyday Attention, or the Wisconsin Card Sorting Test.
from hemiplegia (Robertson, Ridgeway, Greenfield and Parr, 1997). Five patients (M1, M2, M4, M5 and M6) demonstrated sustained attention problems, as evidenced by their low scores on the Lottery subtest of the Test of Everyday Attention (TEA; Robertson, Ward, Ridgeway and Nimmo-Smith, 1994). There was a double dissociation between anosognosia and impaired sustained attention as demonstrated on the TEA. Patient M3 had anosognosia but not impaired sustained attention; patient M4 had impaired sustained attention but not anosognosia (Table 2).

5.2.4 Working memory
While memory involves the preservation of information, working memory involves both the temporary maintenance and the manipulation of information. In many cases, performing the correct manipulation requires the subject to retain information about serial order. A simple test of working memory is Digit Span Backward (WMS-R), in which the subject is required to reverse the order of a series of digits presented by the examiner. Two patients (F1 and M1) demonstrated problems on this test, but the performance of patient F1 could be explained by her language difficulties (noted above). The formal assessment of working memory also included the Elevator Counting with Distraction subtest of the TEA, which requires the patient to respond to two types of auditory tones by counting the low tones and ignoring the high tones. Four patients (F1, M3, M5 and M6) demonstrated problems on this test, but again the performance of patient F1 could be explained by her language difficulties. Thus, in this group of patients, anosognosia was associated with impaired working memory (Table 2).

5.2.5 Executive function
A computerised version of the Wisconsin Card Sorting Test (WCST; Harris, 1988) was administered using standardised instructions (Heaton, Chelune, Talley, Kay and Curtiss, 1993). Performance on the WCST is commonly assessed using two measures, Categories Achieved and Perseverative Errors.

Patient M4 demonstrated normal performance but all the other patients were impaired on this test. (In fact, all the patients except F1, M3 and M4 performed very poorly on both measures; that is, more than two standard deviations below the normal mean.) There was a clear one-way dissociation between impaired executive function, as demonstrated on the WCST, and anosognosia (Table 2).

5.3 Statistical analysis
The findings of the neuropsychological assessment are suggestive but one disadvantage of the method of associations and dissociations is that we need to impose boundaries between patients who are classified as impaired on a neuropsychological test and those who are not. (In Table 2, patients are classified as impaired if their performance is more than one standard deviation below the normal mean and this cannot be explained in terms of unilateral neglect or language difficulties.) In order to investigate further the

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17 In this test, the patient is required to match each of 128 test cards to one of four reference cards (one red triangle, two green stars, three yellow crosses, or four blue circles) according to the colour, shape, or number of stimuli on the cards. The matching principle is not explained to the patient and must be inferred from the response (right or wrong) to the patient’s attempted match. After ten consecutive correct matches, the principle is changed without warning. The test is terminated when the patient achieves ten consecutive correct responses on each of six matching principles or when the 128 cards are exhausted.
relationship between cognitive impairments and anosognosia for motor impairments, we
carried out a statistical analysis of the neuropsychological test scores and anosognosia
scores using a standard general linear model (GLM) approach (see Smithson,
Aimola Davies and Davies, submitted, sections 2–4).

For this purpose, we consider only those neuropsychological tests for which the score
is the number of correct responses out of a fixed total. Each such test is considered as a
fixed number of independent trials with a probability of a correct response on each trial.
The number correct on each test can be thought of as following a binomial distribution,
thereby enabling us to determine whether the anosognosia score predicts the probability
of a correct response for a given test. In the setting of a GLM we may compare a ‘null’
model against one including anosognosia status as a predictor using standard maximum
likelihood methods. Only seven of the nine patients could be included in the analysis.
Patient F1 was excluded because language difficulties affected her performance and
patient F3 because time difficulties precluded her completion of the full battery of tests.

The primary candidates for tests whose scores are significantly predicted by
anosognosia are the Elevator Counting with Distraction (ECD) subtest of the TEA, the
Logical Memory Immediate (LMI) subtest of the WMS-R, and the two WCST scores,
Categories Achieved (WCA) and Perseverative Errors (WPE). (Note that LMI does not
quite achieve a significant effect.) These predictions are all in the expected directions.
ECD, LMI, and WCA are negatively associated with anosognosia, whereas the WPE
score is positively associated.

For each of the three test scores that are significantly predicted by the anosognosia
score (ECD, WCA, WPE), the model with anosognosia as a predictor is significantly
better than the null model but does not yield an acceptable fit. It turns out that the sum
of ECD and LMI yields a better fit than ECD alone but that the best combination of tests is
the sum of three, ECD, LMI and WPE (with the WPE score reversed) (Figure 1).

5.4 Working memory and executive processes in anosognosia
The neuropsychological assessment reveals an association between anosognosia and
impaired working memory as demonstrated on Elevator Counting with Distraction (ECD)
and Digit Span Backward (Table 2). The statistical analysis shows that anosognosia is a
significant predictor of the ECD score but a predictive model including only ECD does
not yield an acceptable fit. If impaired working memory is a factor in anosognosia then
this is not surprising, since patient M1, who demonstrated working memory problems on
Digit Span Backward, scored in the normal range on ECD. Digit Span Backward could
not be included in the statistical analysis because the score is not the number of correct
responses out of a fixed total. An acceptable model fit is achieved if we add to ECD the
score on Logical Memory Immediate (LMI), a test on which all the patients scored in the
normal range, but patient M1, and also patient M5, scored lower than the rest. Further
investigation is required to understand the relationship between the two tests of working
memory (since impairments on ECD and Digit Span Backward are doubly dissociated)
and also to understand the relationship between working memory and LMI, which is a
test of the maintenance of a structured body of information.

18 The assumptions behind our approach involve some idealisation, particularly the assumption that trials
are independent. Nevertheless, we regard this as a reasonable and pragmatic method for analysing these
data.
All the patients except M4 showed impaired executive function on the WCST. The statistical analysis shows that anosognosia is a predictor of both measures of performance on the WCST, Categories Achieved (WCA) and Perseverative Errors (WPE), while the best combination of tests is achieved by adding WPE to the sum of ECD and LMI. The WCST is a demanding test involving cognitive functions including set-shifting, complex working memory operations, error detection, and feedback utilisation (Lie, Specht, Marshall and Fink, 2006) and it is acknowledged that poor performance is difficult to interpret (Cinan and Öktem Tanör, 2002; Lezak, Howieson and Loring, 2004). Further investigation would be required to determine which components of the test are responsible for the difficulties demonstrated by most of these patients.

Much remains to be done before we shall have a satisfactory account of the role of cognitive impairments in anosognosia. But the results from the study described here, and the finding (Maguire and Ogden, 2002) that all the patients except F1 (following a left-hemisphere stroke) and M4 (following a right-basal-ganglia haemorrhage) had lesion locations that included right dorsolateral prefrontal cortex, are broadly consistent with the Second Factor Hypothesis (section 3.3): The second factor in anosognosia is an impairment of working memory or executive processes with a neural basis in the right frontal region of the brain.

6. Signs of Motivation?
The study that we have described did not include any formal assessment of motivational, affective, or personality factors, and the same is true of the study by Marcel and colleagues (2004). So we come to our final question about anosognosia as a delusion.

In an assessment of cognitive impairments, what kinds of finding might suggest that motivation is also playing a role in some cases?

There is, of course, no way to limit in advance where relevant evidence might come from. We simply indicate some possibilities.

In principle, it might happen that a neuropsychological assessment reveals no cognitive impairments in a patient who has anosognosia. Or it might be that one patient shows more anosognosia than would be predicted by an otherwise well supported association between anosognosia and cognitive impairments. In such a case, one possible hypothesis would, of course, be that the assessment of cognitive impairments was not sufficiently discriminating. But clearly, an alternative hypothesis would be that motivational, affective, or personality factors, rather than just cognitive factors, figure in the explanation of anosognosia in this patient. Some recent studies of anosognosia focus primarily on factors of these kinds.

In the study by Marcel and colleagues (2004), many more patients failed to appreciate the consequences of their impairments for activities of daily living than failed to acknowledge the impairments themselves. The fact that some patients who acknowledge their motor impairments still overestimate their ability to carry out motor tasks could potentially be explained in several ways. For example, patients may have difficulty in

19 Some other studies have assessed personality styles or attitudes to illness but have found no theoretically significant differences between patients with and without anosognosia (Levine et al., 1991; Small and Ellis, 1996).
20 Turnbull, Jones and Reed-Screen (2002); Turnbull, Evans and Owen (2005); see also Turnbull and Solms (2007) and the Cortex Forum on Neuropsychoanalysis for which it is the target article.
inferring or working out the consequences of their impairments (Marcel et al., 2004, p. 32). Alternatively, patients might be motivated to deny their inability to carry out the activities of daily living. Marcel and colleagues explored the possible role of motivation by analysing whether the gender of patients influenced their overestimates of ability to carry out bimanual and bipedal tasks. (The reason for this analysis was that it is plausible that the significance of these abilities for self-esteem varies with gender, and varies differently for different tasks.) The only task on which a significant influence of gender on overestimation was found was driving a car: men overestimated their ability more frequently than women (2004, p. 27).

6.1 Beliefs about returning to work
In the study that we have described, anosognosia scores already reflect anosognosia for both motor impairments and their consequences for activities of daily living. So we cannot look for signs of motivation by asking whether patients who acknowledge their motor impairments may fail to appreciate the consequences of those impairments for daily activities. Nevertheless, it is of some interest to consider expectations about returning to work since these might seem to be plausible candidates for motivationally biased beliefs. These expectations were not taken into account in the patients’ anosognosia scores but are normally assessed in the context of a neurorehabilitation hospital.

Amongst the six male patients, M3 and M5 were the two most concerned about their rehabilitation outcome and the two who best appreciated that it was very unlikely that they would be able to return to work. Patients M1, M2, M4 and M6 also expressed concerns about the future but they were eager and anxious to return to work immediately. In fact, returning to work was not a realistic expectation for any of these six patients. So patients M1, M2, M4 and M6, unlike patients M3 and M5, were seriously overestimating their ability to return to work.

There was a double dissociation between anosognosia and overestimating ability to return to work. Patients M3 and M5 had moderate anosognosia for their motor impairments and the consequences for activities of daily living yet they correctly estimated that a return to work was unlikely. Patients M2 and M4 had at most mild anosognosia for their motor impairments and the consequences for activities of daily living but overestimated their ability to return to work. This double dissociation might suggest that overestimating ability to return to work is to be explained in terms of factors different from the cognitive impairments that are associated with anosognosia. Motivational factors would be one candidate.

The case of patient M4 is particularly striking. He had no anosognosia for his motor impairments and their consequences for activities of daily living. He had made a moderately good recovery from hemiplegia to the extent that he could walk with a cane, but he still had motor impairments. He acknowledged these impairments and was concerned about not being able to play the guitar any more. Patient M4 did have some cognitive impairments of memory and sustained attention but he scored in the normal range on tests of working memory and executive function – the areas of cognitive impairment that may be implicated in anosognosia in these patients. Nevertheless, patient M4 seriously overestimated his ability to return to work in his highly skilled and

21 The three female patients were not working outside the home in the months before the stroke.
potentially dangerous job in the construction industry. One natural hypothesis is that
patient M4’s false belief on this topic was motivationally biased and a case of self-
deception.

Other hypotheses are, nevertheless, possible. For example, patient M4 had severe
extrapersonal and personal neglect and these impairments played a significant role in
making it impossible for him to return to work. It is difficult to discover one’s own
unilateral neglect and, in line with Levine’s (1990) discovery theory of anosognosia,
denial of neglect can occur in the absence of cognitive impairments. While patient M4
did not have anosognosia for his motor impairments he did have severe anosognosia for
his extrapersonal and personal neglect (see Azouvi et al., 1996, for interview questions
that assess anosognosia for neglect). So a speculative alternative to the motivational
hypothesis would be that anosognosia for neglect played a part in patient M4’s
overestimating his ability to return to work.

Conclusion
This chapter has made use of the two-factor theory of delusions as a framework for
considering anosognosia as a delusion. In anosognosia, the first factor is an impairment
that prevents the patient’s paralysis or weakness from making itself known to the patient
through immediate experience of motoric failure. The second factor is an impairment that
prevents the patient from making appropriate use of other available evidence of his or her
motor impairments. Levine’s discovery theory is an example of a two-factor account of
anosognosia.

Motivation may play a role in some cases of delusion and a case in which
motivationally biased handling of the available evidence figured in the second factor
would plausibly be an example of self-deception. While accounts of anosognosia as
motivated denial were advanced during the mid-twentieth century, recent work has often
been severely critical of the motivational approach. Nevertheless, there is no good
argument against two-factor accounts of anosognosia that allow the possibility of
motivation figuring in the second factor.

In the two-factor theory of delusions, the second factor has been assumed to be an
impairment of systems of belief evaluation and revision. The functional nature and neural
basis of the second factor have not been well specified but there are good reasons to
consider the hypothesis that it might be an impairment of working memory or executive
processes with a neural basis in the right frontal region of the brain. The results of an
investigation of cognitive impairments in anosognosia are broadly consistent with that
hypothesis. Further theoretical work is needed to reach a better understanding of the role
that impaired working memory or executive function might play as a second factor in the
eaetiology of delusions. In the case of anosognosia, future empirical research should
include systematic and detailed investigation of candidate first factors, cognitive factors,
and motivational, affective and personality factors, in patients at both the acute and the
chronic stage following stroke.
References


Table 1.
Nine patients with persisting unilateral neglect: Patients’ age, time since stroke, side of stroke; motor impairments assessed by the test described by Bisiach and colleagues (1986) and by eight functional tests of hemiplegia (Gialanella and Mattioli, 1992); anosognosia scores for upper and lower limb and overall anosognosia score.

<table>
<thead>
<tr>
<th>PATIENTS</th>
<th>F1</th>
<th>F2</th>
<th>F3</th>
<th>M1</th>
<th>M2</th>
<th>M3</th>
<th>M4</th>
<th>M5</th>
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<td>Age</td>
<td>56</td>
<td>41</td>
<td>63</td>
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<td>56</td>
<td>59</td>
<td>48</td>
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<td>Time since stroke (# months)</td>
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<td>14</td>
<td>21</td>
<td>7</td>
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<td>Side of stroke</td>
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<tr>
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<td>2</td>
<td>1</td>
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<td>Motor Impairment (Bisiach): Lower Limb (0–3)</td>
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<td>2</td>
<td>3</td>
<td>1</td>
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<td>Motor Impairment Eight Functional Tests (0–3)</td>
<td>1</td>
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<td>Anosognosia for Hemiplegia Lower Limb (0–3)</td>
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<td>Overall Anosognosia Score (0–6)</td>
<td>0</td>
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<td>4</td>
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Severity scores (0–3): 0 = none, 1 = mild, 2 = moderate, or 3 = severe.
Table 2.
Associations and dissociations between anosognosia and cognitive impairments. Shaded cells indicate impaired performance (more than one standard deviation below the normal mean) that cannot be explained in terms of unilateral neglect or language difficulties. Patient F3 is omitted as she did not complete the Doors and People Test, the Test of Everyday Attention, or the Wisconsin Card Sorting Test.

<table>
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<tr>
<th>PATIENTS</th>
<th>F1</th>
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<td><strong>Working Memory:</strong></td>
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<td>Elevator Counting with Distraction (TEA)</td>
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<td><strong>Executive Function:</strong></td>
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<td>Wisconsin Card Sorting Test</td>
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Figure 1.
Unweighted Sum of WPE + ECD + LMI versus overall anosognosia score: relationship between overall anosognosia score (0–6) and sum of scores on Elevator Counting with Distraction subtest of the Test of Everyday Attention, Logical Memory Immediate Recall subtest of the Wechsler Memory Scale–Revised, and Wisconsin Card Sorting Test (Perseverative Errors). Patients F1 and F3 are not included in the analysis.