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Cognitive and Motivational Factors in Anosognosia

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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 paralyzed 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, paralyzed or numb?" (questions from Cutting, 1978; see also Nathanson, Bergman, & Gordon, 1952), and they 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.

Terminology and Distinctions

Before proceeding, we set out some terminological matters and some conceptual distinctions. The Oxford English Dictionary definition of anosognosia is "unawareness of or failure to acknowledge one's hemiplegia or other disability." 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 & Tranell, 1989).

Because 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; 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 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 paralyzed 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 paralyzed 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, & 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 postperformance 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 defense mechanism.

Classification of patients as having anosognosia may be complicated or even subverted by their denial that the affected part of their body (a paralyzed limb, for example) even belongs to them (Bisiach, Rusconi, & Vallar, 1991; Halligan, Marshall, & Wade, 1995). This denial of ownership of a body part is an extreme form of somatoparaphrenia (Gerstmann, 1942). Denial of ownership of a paralyzed limb may sometimes occur in conjunction with misoplegia, which is severe dislike or even hatred of 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”; however, 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 6 months after her stroke, she acknowledged that her left arm was weak—weaker than her left leg. When it was demonstrated to her that her left arm was completely paralyzed and her left leg nearly completely paralyzed, 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 paralyzed yet overestimated their ability to carry out bimanual 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 & 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, Ládavas, Straccieri, Giannarelli, & Össola, 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 among concurrent unawareness of an impairment, failure to acknowledge the impairment itself, and failure to appreciate the consequences of the impairment.

**Anosognosia as a Delusion**

A patient with anosognosia believes that he or she is able to move a limb that is, in reality, paralyzed. In its severe form, anosognosia constitutes a delusion according to
the definition offered by the Diagnostic and Statistical Manual of Mental Disorders (DSM-IV-TR) (American Psychiatric Association, 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 findings might suggest that motivation is also playing a role in some cases?

The next section provides a summary account of the two-factor theory of delusions so that, in the third section (“Anosognosia in the Two-Factor Framework”), we can address the question of how explanations of anosognosia might fit into this framework. In the “Motivation in Anosognosia” section, we consider the role of motivation in anosognosia, particularly in the light of an influential argument against interpreting anosognosia as motivated denial. In the fifth section (“A Study of Cognitive Impairments in Patients With Anosognosia”), we present an empirical study of the role of cognitive impairments in anosognosia, and in “Signs of Motivation?” we return briefly to motivation.

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 favor the true proposition, not-P. In the Capgras delusion (Capgras & Reboul-Lachaux, 1923; Ellis & Young, 1990; Stone & 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, & Coltheart, 2001; Breen, Caine, Coltheart, Hendy, & Roberts, 2000; Foley & Breslau, 1982), it is that the person that the subject sees in the mirror is not him; in delusional jealousy (Othello syndrome; Todd & 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 etiology 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.

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. 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 etiology 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.

Because 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 of 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.

The Second Factor

The argument for a second factor in the etiology 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 because it does not explain why the belief is tenaciously maintained—“firmly sustained despite...incontrovertible and obvious proof or evidence to the contrary” (American Psychiatric Association, 2000, p. 821). We need a second factor to answer the question of 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 paralyzed left-side limbs; 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.4 Thus, Coltheart (p. 1047) describes the second deficit in this way: "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" (Coltheart, 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 mechanisms involved in reasoning and also in judgment 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. 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 etiology of delusions by investigations of working memory and executive processes in patients with delusions.

**Two Factors but Three Stages**

A delusion is a pathology of belief, 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 of where
the delusion came from. 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 etiology 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, although 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.

*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 paralyzed 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 defense against depressive overwhelm. Adopting the belief that he was enjoying a fulfilling marriage, “seemed to go some way toward reconferring a sense of meaning to [BX’s] life experience and reintegrating his shattered sense of self” (Butler, 2000, p. 89).

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. 318–319). 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. 58; 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; 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).

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, paralyzed. 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.
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, & Adair, 1998; see also Frith, Blakemore, & Wolpert, 2000). In the two-factor framework, all these theorists can be regarded as proposing candidate first factors in the etiology 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 Anjan Chatterjee and Mark Mennemeier's report 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 paralyzed 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" (Chatterjee & Mennemeier, 1996, p. 229).

Specific Memory Impairment as a First Factor in Anosognosia

Earlier, 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 etiology 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: 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; 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...defects 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" (p. 115). Five minutes after it was demonstrated to her that her left arm was completely paralyzed and her left leg nearly completely paralyzed, 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: "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" (1988, p. 115).

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; the result is that one hemisphere of the brain is selectively anaesthetized. During this procedure, patients 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 31 epilepsy patients by Katherine Carpenter and colleagues (1995), 27 patients
failed to recall having had left-arm weakness when questioned 10–15 minutes after a right-side barbiturate injection, and 12 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 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: "In 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" (p. 249). 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 patient'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.

The Second Factor in Anosognosia

If a first factor is present, then a subject may be concurrently unaware of failures to move a paralyzed limb—or else may be concurrently aware of these failures but unable to integrate this information into beliefs.7 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 moturally. 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. Thus, something more—a second factor—is needed to explain why patients with anosognosia do not make appropriate use of this evidence.8

It remains to consider the neural basis and functional nature of the second factor in anosognosia.9 If Coltheart is right to assume that the second factor "is the same in all people with monotheistic delusion" (2005b, p. 154), then earlier suggestions and speculations ("The Second Factor" section) 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.

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, & 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 cutoff on this test; Marcel and colleagues (2004) reported that patients with anosognosia were no more likely to score below cutoff 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 judgment” (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 & Hamsher, 1976). The assessment included a test of 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 yields little information about which cognitive impairments best discriminate patients with somatosensory loss and anosognosia from patients with somatosensory loss but without anosognosia and thus 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.

**MOTIVATION IN ANOSOGNOSIA**

Motivation might, in principle, figure in the first factor or the second factor (or both) in the etiology of a delusion (“Motivation in the Two-Factor Framework” section). 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 & Kahn, 1950, 1951, 1953; Weinstein, Kahn, Malitz, & Rozanski, 1954) and an influential book (Weinstein & Kahn, 1955) putting forward a motivational account of anosognosia. But, as we shall see (“The Case Against Motivation” section), recent work has often been severely critical of the motivational approach.

**Patients with Brain Injury**

At the end of a study of anosognosia in 22 patients with brain tumor, Weinstein and Kahn (1950) proposed 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. 789–791). As a result of this reorganization, 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" (1950, 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 said that the manifestations of disorientation are also "symbolic expressions of the drive to be well" (1954, p. 57).

In a study of 100 patients with hemiplegia (95 following a cerebrovascular accident), Morton Nathanson and colleagues (Nathanson et al., 1952) found that all of the 26 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 regarded disorientation as a sign of a psychological defense 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, & Evans, 2004).

**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. In patients with these conditions, denial of illness has been credited with both negative and positive outcomes (see Korte & 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 hospitalization, it has been associated with delay in seeking treatment and, in the posthospitalization 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 noncerebral etiology, have been taken to support the proposal that anosognosia may have a motivational basis.

**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 & 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 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 etiology 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 & Eysenck, 1987) demonstrated that denial of hemiplegia cannot be attributed to extroverted, neurotic, or psychotic postmorbid personality styles or 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 perfectionists significantly more often than patients with hemiplegia and visuospatial neglect in the absence of anosognosia 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 & 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” (p. 362).

The Case Against Motivation

In an influential book chapter, Edoardo Bisiach and Giuliano Geminiani (1991, pp. 25–26) listed “eight important facts concerning anosognosia related to neurological disorders that affect one side of the body.” They argued 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” (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 defense against depressive overwhelm. It would surely be remarkable if motivation were sometimes a factor in delusions such as reverse Othello syndrome yet never a factor in the delusion of anosognosia.
Because our concern is with explanations of anosognosia within the two-factor framework, 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; and
- **hemispheric differences**—anosognosia is more common following right-hemisphere damage than following left-hemisphere damage.\(^2\)

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.

**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, and 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 etiology resolves. This approach would be consistent with the fact that most cases of unilateral neglect, which is a candidate first factor in anosognosia, improve rapidly over the first 10 days following stroke (Stone, Patel, Greenwood, & 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 "Premorbid Personality Styles") 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.

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, & 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, & Sterzi, 1993). In some cases, vestibular stimulation even results in improved motor performance. Gilles Rode and colleagues (Rode, Perennin, Honoré, & 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 15 or 20 minutes after vestibular stimulation. The authors conclude that there is a "motor neglect component"—"one of the many manifestations of the unilateral neglect syndrome" (Rode et al., 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 etiology of anosognosia (see Vallar, Bottini, & Sterzi, 2003, for a review).

The Selectivity of Anosognosia  Anosognosia may be selective. Patients may fervently deny their paralysis but frankly acknowledge their heart attack or stroke ("Premorbid Personality Styles" section). Some patients who have both hemianopia and hemiplegia deny their visual field deficit while acknowledging their motor impairments (Bisiach, Vallar, Perani, Papagno, & Berti, 1986). Some patients show anosognosia for paralysis of their left leg while acknowledging that they cannot move their left arm (Bisiach et al., 1986).
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, “the patient denies whatever he feels is seriously wrong with him, whether it is a hemiplegia, a craniotomy or a sense of inadequacy” (p. 789). 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.

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; 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, Lahtosalo, & Kettunen, 2006, for a recent review) and persisting anosognosia is almost invariably accompanied by neglect (Cocchini, Beschin, & 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” (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. Therefore, detection of left-side weakness following right-hemisphere damage might be more difficult than detection of 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 lateralized to the right hemisphere are those involved in somatosensory attention tasks (Coglobin, Gilron, & 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." However, 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. 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 etiology 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 of whether hemispheric differences in emotion may help to explain the hemispheric differences in anosognosia.

**Hemispheric Differences in Emotion**

In an influential paper, Guido Gainotti (1972; see also 1969) reported that catastrophic or anxious-depressive reactions are associated with left-hemisphere damage and 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) remarked 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, among 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 "the right, non-verbal hemisphere [should be considered] as more important from the 'emotional' point of view" (1972, p. 52). 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 & Irwin, 1999) supports a version of the valence hypothesis that is conceptualized 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, & Kinsbourne, 2006). Meanwhile, Gainotti (2000, 2003; see also Hagemann, Hewig, Seifert, Naumann, & Bartussek, 2005) has put forward a different refinement of the right-hemisphere hypothesis. He proposes that hemispheric differences are not related to valence—negative (right) versus positive (left)—but rather 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” (Gainotti, 2000, p. 226), but the left hemisphere may play a critical role in “functions of control and of modulation of spontaneous emotional expression” (p. 226).

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 lateralization 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.

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 (“Motivation in the Two-Factor Framework” section). 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 postacute 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 3 months after a stroke in the absence of unilateral neglect.\footnote{Therefore, one way to 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 males, M1–M6, and three females, F1–F3; ages 41–63 years) with neglect persisting at least 3 months following a unilateral stroke (Table 10.1). The patients were assessed on a large battery of neglect tests (12 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 program. 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 10.1 for the results of two assessments of motor impairments: the test described by Bisiach and colleagues (1986) and the eight functional tests of hemiplegia (Gianella & Mattioli, 1992).

<table>
<thead>
<tr>
<th>TABLE 10.1 Nine Patients With Persisting Unilateral Neglect</th>
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<td>Patients</td>
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<td>M5</td>
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<td>M6</td>
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Note: Severity scores (0–3): 0 = none, 1 = mild, 2 = moderate, or 3 = severe.
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 & 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); and
- 3 = no acknowledgement of the disorder can be obtained (severe anosognosia).

Bisiach and colleagues (1986) and also Anna Berti and colleagues (Berti, Làdavas, & Della Corte, 1996) have suggested that assessing anosognosia in patients without severe motor impairments is problematic because 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.”

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 & 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, as well as frequent contact with their families and staff. Scores for anosognosia for motor impairments and their consequences take account of her clinical judgment.

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 10.1). 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—even overambitious—in arranging excursions for himself from the hospital to the nearby casino. But he never tried to get out of his wheelchair or 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 ("Neuropsychological Assessment" section and Table 10.2), 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 3-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).

**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 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 summarize 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.16
TABLE 10.2  Associations and Dissociations Between Anosognosia and Cognitive Impairments

<table>
<thead>
<tr>
<th>Patients</th>
<th>F1</th>
<th>M4</th>
<th>F2</th>
<th>M2</th>
<th>M3</th>
<th>M5</th>
<th>M1</th>
<th>M6</th>
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<tbody>
<tr>
<td>Overall anosognosia score (0–6)</td>
<td>0</td>
<td>0</td>
<td>2</td>
<td>4</td>
<td>4</td>
<td>5</td>
<td>6</td>
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<tr>
<td>Memory: Doors and People Test (DPT)</td>
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<td>Sustained attention: Lottery (TEA)</td>
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<td>Working memory: Digit Span Backward (WMS-R)</td>
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<td>Working memory: Elevator Counting With Distraction (TEA)</td>
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<td>Executive function: Wisconsin Card Sorting Test</td>
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*Note: Patient F3 is omitted because she did not complete the Doors and People Test, the Test of Everyday Attention, or the Wisconsin Card Sorting Test.*

- Impaired performance (more than one standard deviation below the normal mean) that cannot be explained in terms of unilateral neglect or language difficulties.

**Premorbid Ability, Orientation, Language and Comprehension, Visuoperceptual Function** National Adult Reading Test scores (NART II; Nelson & Willison, 1991), supplemented with background demographic information about educational and occupational accomplishments, indicated that the patients’ premorbid functioning was within the normal range (91–115). All the patients passed the Information and Orientation subtest of the Wechsler Memory Scale-Revised (WMS-R; Wechsler, 1987) without difficulty (range of 12–14; 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.

**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, & Nimmo-Smith, 1994). There 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 10.2).17

**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 among a greater number of nontargets (distractors). Sustained attention has been associated with persisting unilateral neglect (Robertson, 2001) and with poor recovery from hemiplegia (Robertson, Ridgeway,
Greenfield, & 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, & 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 10.2).

Working Memory 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-B), 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 earlier). 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 10.2).

Executive Function A computerized version of the Wisconsin Card Sorting Test (WCST; Harris, 1988) was administered using standardized instructions (Heaton, Chelune, Talley, Kay, & 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 10.2).

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 10.2, 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 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, & Davies, submitted).

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; patient F3 was excluded 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 10.1).

![Figure 10.1 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.](image-url)
Working Memory and Executive Processes in Anosognosia

The neuropsychological assessment reveals an association between anosognosia and impaired working memory as demonstrated on ECD and Digit Span Backward (Table 10.2). 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 because 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 patient M5 scored lower than the rest. Further investigation is required to understand the relationship between the two tests of working memory (because 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.

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 utilization (Lie, Specht, Marshall, & Fink, 2006), and it is acknowledged that poor performance is difficult to interpret (Cinan & Öktem Tanör, 2002; Lezak, Howieson, & 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 have a satisfactory account of the role of cognitive impairments in anosognosia. But the results from the study described here and the finding (Maguire & Ogden, 2002) that all the patients except F1 (following a left-hemisphere stroke) and M4 (following a right-basal-ganglia hemorrhage) had lesion locations that included right dorsolateral prefrontal cortex are broadly consistent with the 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.

**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). Thus, we come to our final question about anosognosia as a delusion:

In an assessment of cognitive impairments, what kinds of findings 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.\(^\text{21}\)

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 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 analyzing 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).

**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. Therefore, 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 because these might seem to be plausible candidates for motivationally biased beliefs. These expectations were not taken into account in the patients' anosognosia scores, but they are normally assessed in the context of a neurorehabilitation hospital.

Among the six male patients,\(^\text{22}\) 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. Thus, 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 anymore. 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 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. Although 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). Thus, 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. Although 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 for 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 etiology 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 stages following stroke.

NOTES

1. The authors' names are listed in alphabetical order.
2. Anderson and Tranel (1989); Berti et al. (1996); Cutting (1978); Feinberg, Roane, and Ali (2000); Marcel et al. (2004); Nathanson et al. (1952); Spinazzola, Pia, Folegatti, Marchetti, and Berti (2008); Starkstein, Fedoroff, Price, Leiguarda, and Robinson (1992).
4. 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).
5. Here, as at many other points, we are indebted to Tony Stone.
6. Patients with unilateral neglect fail to respond to stimuli presented on the side opposite to their lesion—for example, on the left side following a right-hemisphere lesion. Unilateral neglect is not a unitary condition and one important distinction is between extrapersonal neglect and personal neglect. In extrapersonal neglect, patients neglect left-side stimuli external to their body—for example, neglecting to copy the left side of a picture or neglecting to cross out lines or other targets on the left side of a sheet of paper. In personal neglect, patients neglect the left side of their body—for example, combing their hair only on the right side. Research on unilateral neglect has involved both attentional and representational theories (Bisiach, Capitani, Luzzatti, & Perani, 1981; Bisiach & Luzzatti, 1978; Bisiach, Luzzatti, & Perani, 1979; Halligan & Marshall, 1994; Robertson & Marshall, 1993). The role of personal neglect in anosognosia might be conceived as a failure to direct attention towards the paralyzed side of the body or a failure to represent that side of the body. Representational theories of personal neglect have connections with research on the "body schema" (see Heilman, 1991, p. 58, for a brief discussion).
7. 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).

8. This argument for a second factor might not apply if the paralysis were to last only for a very short time and, during that time, the patient were lying in bed and 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 were 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.

9. See Berti et al. (2005) and Karnath et al. (2005) for recent proposals about the neural basis of anosognosia. 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 26 months after a right-hemisphere stroke and other delusions including misidentification of place, sonatophrenia, 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” (p. 237).

10. For denial of heart disease or myocardial infarction, see Levine et al. (1987) and Stenstrom et al. (2005); for denial of cancer, see McKenna, Zevon, Corn, and Rounds (1999) and Rousseau (2000); for denial of diabetes, see Lo and MacLean (2001); for denial of spinal cord injury, see Livneh and Martz (2003) and Martz, Livneh, Priebe, Wuermser, and Ottomanelli (2005).

11. Levine and colleagues (1991) administered a questionnaire designed to reveal attitudes towards illness and also the Minnesota Multiphasic Personality Inventory (MMPI; Dahlstrom, Welsh, & Dahlstrom, 1975). The results from the questionnaire did not provide any evidence that the patients with anosognosia had an attitude towards illness different from the attitudes of 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.

12. 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. 25–26): 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
behavior, or vice versa. 6. Some patients with hemiplegia display misplicegia, rather than anosognosia (for discussion, see Turnbull, Jones, & 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 presents problems for a two-factor account of anosognosia with motivation as a candidate second factor.

13. Recent studies of patients in the first 10 days following a stroke suggest a rate of occurrence for anosognosia of 17–21% (Appelros, Karlsson, & Hennerdal, 2007; Appelros, Karlsson, Seiger, & Nydevik, 2002; Pedersen, Jørgensen, Nakayama, Raaschou, & Olsen, 1996) and 21–42% for right-hemisphere patients (Appelros, Karlsson, Seiger, & Nydevik, 2003; Jehkonen, Ahonen, Dastidar, Laippala, & Vilki, 2000). Studies also suggest a rate of occurrence for unilateral neglect of 23% (Appelros et al., 2002; Pedersen, Jørgensen, Nakayama, Raaschou, & Olsen, 1997) and 32–42% among right-hemisphere patients (Appelros et al., 2003; Jehkonen et al., 2000; Pedersen et al., 1997).

14. This is the case reported by House and Hodges (1988). In a review of patients with anosognosia reported in the literature, Cocchini and colleagues (2002) find only one case persisting more than 1 month poststroke 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; however, 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, & 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, & 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.

15. We note that unilateral neglect persisting more than 3 months after a stroke is rare. Most cases of unilateral neglect improve rapidly over the first 10 days, and one study indicates that as few as 10% of cases present in the first 2 or 3 days persist beyond 3 months (Stone et al., 1992).

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

17. 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.

18. 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 color, 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 10 consecutive correct matches, the principle is changed without warning. The test is terminated when the
patient achieves 10 consecutive correct responses on each of six matching principles or when the 128 cards are exhausted.

19. The assumptions behind our approach involve some idealization, particularly the assumption that trials are independent. Nevertheless, we regard this as a reasonable and pragmatic method for analyzing these data.

20. 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 & Ellis, 1996).

21. Turnbull et al. (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.

22. The three female patients were not working outside the home in the months before the stroke.

REFERENCES


