Research report

The riddle of anosognosia: Does unawareness of hemiplegia involve a failure to update beliefs?

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1. Introduction

Anosognosia is a striking neurological symptom that manifests as unawareness and lack of concern of for their deficit (hemiplegia, hemianopsia, neglect, etc.) in brain-damaged patients, and that cannot be explained by global cognitive dysfunction. As initially described by Babinski (1914), the most common presentation is anosognosia for hemiplegia (AHP), where patients fail to recognize a severe motor deficit despite direct confrontation during neurological examination.

This lack of awareness can affect the motor deficit itself (e.g., “I’m able to move my left arm”) but also its consequences...
for a particular action (e.g., “I can open a bottle”) and/or its general implications in everyday life (e.g., “I can go back to work”). The cognitive and neural mechanisms of this phenomenon are poorly understood, and remain difficult to study in a systematic way due the predominance of symptoms in acute stages and their heterogeneity (Vocat et al., 2010).

Several theoretical accounts of AHP have been proposed (Berti et al., 2005; Davies et al., 2005; Heilman, 1991; Levine et al., 1991; Ramachandran, 1995; Weinstein and Kahn, 1955) but none of them appears sufficient to explain all the manifestations of anosognosia, and the frequent dissociations with other disorders or between different domains of anosognosia itself (Vocat and Vuilleumier, 2010; Vuilleumier, 2000). Babinski (1914) initially emphasized that AHP was often associated with hemianesthesia and proprioceptive loss, possibly preventing direct sensory experience of weakness, whereas Bisioah et al. (1986) insisted on a particular relation between AHP and spatial neglect which might disrupt conscious awareness of left hemibody. The discovery theory of Levine et al. (1991) proposed that AHP could emerge due to a conjunction of global cognitive impairment and impaired proprioception. A role for motor neglect was also highlighted as a possible causative factor by Heilman (1991), but related to a suppression of motor intention. Likewise, a deficit in comparator mechanisms that match the predicted movements (based on intention) and the actual movement (based on sensory feedback) was also recently implicated (Bottini et al., 2010; Fotopoulou et al., 2008; Frith et al., 2000). Finally, for Weinstein and Kahn (1955) and Turnbull et al. (2002), motivational or psychological defense mechanisms could not be excluded.

More recent hypotheses (Davies et al., 2005; Vuilleumier, 2004) have suggested multiple causative factors. According to this view, AHP might emerge through various combinations of 2 (or more) deficits. Davies et al. (2005) proposed that AHP could resemble delusions, requiring the presence of a first impairment that prompts delusional beliefs and a second impairment that interferes with processes of belief evaluation which would otherwise allow rejecting the delusional belief. Similarly, Vuilleumier (2004) put forward a general “ABC model” of anosognosia, where a combination of processes involving appreciation, belief, and check operations might subserve awareness for motor losses, or cause anosognosia or denial when damaged in different ways in different patients. According to this view, the ability to modify one’s beliefs together with the aptitude to monitor performance and outcomes in case of uncertainty might play a critical role in AHP, in combination with other primary deficits in sensory, motor, and/or attentional functions (Venneri and Shanks, 2004). A recent elegant study (Jenkinson et al., 2009) also showed that patients with AHP are impaired in reality monitoring processes not strictly related to movement, but also extending to visual perception. In keeping with multifactorial causes, a recent prospective study (Vocat et al., 2010) showed that different combinations of deficits and lesions have a different impact on the presence of AHP, with proprioceptive loss being most strongly correlated with the severity of AHP in the acute stage after the stroke, but other cognitive impairments being more crucial for its persistence in chronic stages.

Moreover, besides a general predominance of right hemispheric dysfunction in AHP (Adair et al., 1995; Breier et al., 1995; Gilmore et al., 1992; Lu et al., 2000, 1997), neuroanatomical studies have pointed to an involvement of many different brain regions including the parietal lobe (Bisiach et al., 1986; Heilman, 1991), insula (Karnath et al., 2005), pre-motor, motor and sensory areas (Berti et al., 2005), thalamus (Starkstein et al., 1992), or more complex cortico-subcortical circuits of motor control (Pia et al., 2004). Our own work (Vocat et al., 2010) also highlighted a role for distributed lesions in areas associated with motor function, spatial attention, as well as interoceptive and affective functions, consistent with a multi-componential model of AHP.

However, although the ABC model (Vuilleumier, 2004) or 2-factor delusional theory (Davies et al., 2005) made the hypothesis that AHP might be critically linked to impairments in the generation and adjustment of one’s beliefs in face of uncertainty, no study has directly tested the ability of patients to check their belief accuracy and/or change their current beliefs. If anosognosics still rely on their past beliefs about their health and are not able to modify them according to the new condition (e.g., due to losses in sensory feedback, intentions, or bodily representations), they might indeed fail to correctly acknowledge their current handicap. Here, we therefore designed a new neuropsychological test allowing us to address this issue. Further, we aimed at using a task simple and brief enough to be administered to patients at the bedside, even in the acute stage. Our test required participants to make guesses in conditions where they should feel uncertain about their responses and could not be confident about their beliefs. Our main aim was to observe how patients with AHP could change their beliefs when confronted with incongruent information contradicting these beliefs.

2. Method

2.1. Population

We tested 11 control participants (mean age 62.8 ± 9.6, 8 women, all right-handed), with no history of neurological or psychiatric diseases, and a group of 9 consecutive patients with a first right hemispheric stroke and a full left hemiplegia. The severity of AHP in these patients was measured using the classical scale of Bisiach et al. (1986). Among them, 5 patients (mean age 56.4 ± 16.8, 3 women, all right-handed) were able to give a good spontaneous description of their plegia (Bisiach scale rating = 0) and constituted the nosognosic group; whereas 4 other patients (mean age 60 ± 10.2, 2 women, all right-handed) had clear and stable anosognosia for their hemiplegia (Bisiach score = 3 in all of them) and were therefore included in the anosognosic group. We did not test patients with intermediate Bisiach scores (ratings = 1 or 2) because it has been shown that measures of AHP may be unreliable in these cases and represent only partial or fluctuating deficits in awareness (Baier and Karnath, 2005).

2.2. Riddle test

The task required participants to guess 10 target words. For each of these words, a succession of 5 verbal clues was provided in turn. These clues were brief sentences presented...
visually on a laptop in front of the participants and were concurrently read aloud by the experimenter to ensure correct comprehension. The clues gave information about semantic or physical attributes of the targeted word that did not contain any explicit motor imagery or gestural component. Each clue appeared on the screen one after the other. They remained displayed until the end of the riddle for a given word target. Following the presentation of each clue (see example in Fig. 1, and Appendix for a complete list of the riddles), participants had to propose verbally a word that could possibly correspond to the current clue(s). If they could not find any word, they had the possibility to advance to the next clue but were encouraged to search for a possible word (all participants were generally able to do so and only rarely refused to guess a word). After a guess was uttered, the experimenter typed the proposed word on the laptop and validated it. Then, participants had to rate their level of confidence in their guess. This rating ranged from “0” (completely unsure) to “8” (completely sure) on a vertical scale presented visually. Patients gave their level of confidence by reporting the number and a verbal label associated with it (e.g., “0, completely unsure”). If no word was proposed (10.7% of trials on average), the confidence rating was skipped. This was then followed by the next clue (see example in Fig. 1). After the fifth and last clue, the rating of confidence for the last given guess was followed by the display of the “correct” word target corresponding to the definition of all preceding clues. No other feedback on accuracy was given until this final clue.

Importantly, the 5 successive clues were chosen to have a progressive informative content. The first was always ambiguous enough to have multiple potential answers, while the next successive clues gave more and more precise information on the target, in such a way that the possible answers were less and less numerous. The fifth and last clues were chosen to leave no doubt about the correct answer to the riddle. Fig. 1 shows an example of the whole succession of clues for 1 of the 10 riddles. The riddles were presented in a random order counterbalanced across participants. The test was administrated approximately one week after the stroke.

Each response of each participant after each clue was classified as “correct response” (if it corresponded to the correct word target), “false response” (if it did not correspond), “repetition” (if the same false response was given as for the preceding step), and “no response” (if the patient could not report any word). For each participant, we also calculated a global score of correct reasoning which was obtained by summing the number of riddles that were finally resolved (with a correct answer given before or after the fifth clue). Finally, for each participant, we computed a measure of their level of confidence for the 1st, 2nd, 3rd, 4th and 5th clue, by averaging their ratings after each clue in each of the 10 riddles.

These different scores were then averaged and compared between the 3 groups of participants: healthy controls, nosognosic hemiplegic patient, and anosognosic hemiplegic patients. Statistical comparisons between the 3 groups on the different scores were performed by using the non-parametrical Mann–Whitney U-test, with a p-value threshold for significance fixed at .05. In addition, individual comparisons of response confidence level were made between the score of each anosognosic patient and both the control and the nosognosic groups using the Crawford statistical test.

2.3. Additional clinical testing

Neurological deficits, as well as executive and memory functions were also assessed in patients using several standardized clinical tests that could be administered at the bedside (see also Vocat et al., 2010). These included in particular tactile sensation [score based on 12 stimulations; 3 repetitions of 2 types of stimulation (touch or sting) on 2 sites (hand or elbow)], proprioception [score based on 9 trials; 3 joints (middle finger, wrist and elbow), 3 times each], visual field and visual extinction [scores based on 4 stimulations, with small finger movements across each hemifield], and spatial-temporal orientation. Visuo-spatial neglect was assessed using a composite score from 3 tests: star cancellation from the Behavioral Inattention Test (Wilson et al., 1987), multiple line bisection (Schenkenberg et al., 1980), and copy of the Gainotti–Ogden figure (Ogden, 1985). Several tests were used to assess general frontal functions such as verbal fluency [using names of animals and words beginning with the letter “M”, 1 min each (Thuillard and Assal, 1991)], digit span (Wechsler, 1981), and mental flexibility [category sorting task (Weigl, 1927)]. In addition, we also assessed global cognitive functioning with MMSE (Folstein et al., 1975), providing separate measures for verbal memory (word-list subtest of the MMSE), and working memory (subtraction subtest of the MMSE). The means from the 2 patient subgroups (anosognosics vs nosognosics) on each of these scores were compared by non-parametrical Mann–Whitney U-test with a threshold of significance at \( p < .05 \).

2.4. Analysis of brain lesions

We used the same procedure as previously used in larger studies on brain lesion-behavior relationships in right-hemisphere patients (Verdon et al., 2010; Vocat et al., 2010). The location and extent of brain damage was delineated in each patient, based on MRI scans obtained after the first week post-stroke (5.8 ± 1.2 days on average). MRI scans were obtained with a 3 T apparatus (Siemens Trio). The usual thickness was 6 mm (interslice gap: 1 mm). We used the T1 and T2 images, including T2-weighted fluid attenuated inversion recovery (Flair) sequences, for optimal identification of the

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1. My weights is approximatively 300 grams
2. I produce a regular sound
3. Sport makes me feel excited
4. I am usually on the left rather on the right side
5. Lovers often draw me

Fig. 1 – Example of one of the 10 riddles. Five clues were provided in successive order, until the correct response was given. All clues remained visible on the screen until resolution of the riddle, so as to minimize working memory demands.
lesion areas. We selected brain scans showing the greatest extent of lesions in all patients. The lesion area was manually reconstructed using individual images and transferred onto a standardized brain template with the MRicrO software (Rorden and Brett, 2000, www.mricro.com). This was verified by a trained neurologist who was blind to the patients’ performance. We then examined the overlap of the normalized lesion areas for the different subgroups of patients (see Fig. 2). To estimate the critical anatomical regions differentially damaged in the 2 patient subgroups, we compared the lesion overlap for the nosognosic and the anosognosic patients with a subtraction procedure and chi-squared analysis. Due to our small population, these data are mainly reported for completeness and comparison with other studies (e.g., Karnath et al., 2004; Berti et al., 2005).

3. Results

All participants including patients completed the whole procedure with 10 riddles in a single session. Subjective reports during the test indicated that they enjoyed the task and were motivated in finding the correct word targets. This motivational factor was important to ensure the validity of our measure. The 3 different groups were not significantly different in term of mean age [anosognosics (58) vs nosognosics (62), p = .999; anosognosics vs controls (61), p = .503; nosognosics vs controls, p = .606], and the 2 patient groups (anosognosic vs nosognosic) were tested after a similar time delay since stroke onset (11 and 11.25 days respectively, p = .937).

A word solution was proposed and a level of confidence assigned by the participant on most trials (90% on average). We first analyzed the ratings of confidence (ranging from 0 to 8) for responses given to the 5 successive clues (averaged across all riddles). As illustrated in Fig. 3a, patients with anosognosia showed a significantly higher degree of “certainty” after the first clue than both nosognosics (p = .016) and healthy controls (p = .006). The same result persisted for the second clue (p = .016 and p = .003, compared to nosognosics and controls, respectively) and for the third clue (p = .032 and p = .001, compared to nosognosics and controls, respectively). Following the fourth clue, the certainty levels in anosognosics were still significantly different from controls (p = .006) but did not reliably differ from nosognosics (p = .190) even though the mean rating values were still numerically higher in anosognosics (see Fig. 3). By contrast, on the fifth and final clue where the riddle could generally be resolved without ambiguity, no difference was found between anosognosics and other participants, nosognosics (p = .413) or controls (p = .138). Importantly, the confidence rating values reported by nosognosics and controls did not differ for any of the 5 clues (.510 < p < .913).

This overconfidence was seen in each and every anosognosic patient as compared to other participants (see Fig. 3b), as verified by additional single-case analyses. For the 3 first clues, all 4 patients showed significant differences (Crawford test) with the control group (.001 < p < .05), with only one single comparison showing only a tendency for one patient after the first clue (p = .084). For the differences with the nosognosic group, p-value ranged from .001 to .01 for the first clue, from .001 to .09 for the second clue (3 anosognosics patients out of 4 showing a significant effect) and from .02 to .27 for the third clue (2 anosognosics out of 4 with a significant effect). These analyses confirm that each and every anosognosic patient showed a significant difference (or a strong tendency) in confidence that predominated for the 3 first clues relative to both the control and the nosognosic patients. In line with the group data, controls and nosognosic patients did not differ.

We next analyzed the quality of responses given to each clue in each riddle (50 in total). Overall, anosognosics showed significantly fewer correct responses than nosognosics (p = .032). A similar tendency was found when comparing anosognosics with controls (p = .078). Furthermore, when we examined the nature of these errors (see Fig. 4a), we observed that anosognosics repeated an incorrect response from a preceding trial much more often than both nosognosics (p = .016) and controls (p = .006), whereas the 2 latter groups made the same amount of repetitive responses (p = .51). Thus, anosognosics often failed to modify their responses when

Fig. 2 — Brain lesions in each patient group. The overlap of damaged areas is shown for (a) 5 patients with left hemiplegia but no AHP, and (b) 4 patients with left hemiplegia plus AHP. Color codes represent the number of patients with lesions affecting the same area.

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confronted with a new clue that was incongruent with their previous guess (e.g., proposing “clock” after clue No. 1 and repeating it again after clue No. 3 in the example given in Fig. 1, Table 1).

Remarkably, however, when we computed the number of riddles that were eventually resolved on the last trial (i.e., the accuracy of the final word proposed), we found no significant difference in anosognosics as compared with nosognosics ($p = .111$) or controls ($p = .412$), while the 2 latter did not differ either ($p = .583$). This final resolution outcome demonstrates that general reasoning abilities and semantic knowledge were not affected in anosognosic patients.

We also examined performance on several additional neurological and neuropsychological tests. Compared to non-anosognosic patients, the anosognosics showed no significant difference on any of these tests, including tactile sensation ($p = .555$), proprioception ($p = .601$), visual field loss ($p = .116$), visual extinction ($p = .345$), spatio-temporal orientation (correct in all patients), as well as visuo-spatial neglect tasks ($p = .997$), verbal memory ($p = .178$), and working memory ($p = 1.000$). Most importantly, no difference was found for tests probing frontal lobe functions, including verbal fluency (categorical: $p = .386$; phonological: $p = .965$) and digit span ($p = .720$). Remarkably, the patients did not differ either on measures of reasoning and mental flexibility in the Weigl category sorting test ($p = .539$). MMSE scores were also comparable in anosognosic and nosognosic patients (27.5 and 28.5, respectively, $p = .132$). The “normal range” of these scores indicates that both groups of patients showed no severe cognitive alteration. In addition, none of them showed a severe amnesia, profound neglect, and/or massive executive dysfunction.

Finally, we performed an exploratory anatomical analysis comparing the lesions overlap of anosognosic and nosognosic patients. Using a standard subtraction analysis (see Fig. 5a), the maximal lesion overlap in affected patients fell in the
anterior parietal-temporal cortex junction and subcortical white matter (MNI coordinates: 40, -42, 24). An additional statistical analysis with a voxel-by-voxel chi-square procedure (see Fig. 5b) showed a peak of lesion differences in the supramarginal gyrus (MNI coordinates: 49, -43, 24). Thus, both analyses pointed to the parieto-temporal junction and accord with other studies indicating that this region is frequently lesioned in AHP and related disorders (Bisiach et al., 1986; Feinberg et al., 2010; Vocat et al., 2010).

### Table 1 — Number of each type of response for each participant.
A response was classified as “Correct response” (if it corresponded to the correct word solution of the riddle), “False response” (if it did not correspond), “Repetition” (if the same false response was given as for the preceding step), and “No response” (if the patient could not report any word).

<table>
<thead>
<tr>
<th></th>
<th>Correct response</th>
<th>False response</th>
<th>Repetition</th>
<th>No response</th>
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</thead>
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<tr>
<td>Controls</td>
<td>19.64</td>
<td>20.00</td>
<td>5.00</td>
<td>5.36</td>
</tr>
<tr>
<td>Nosognosics</td>
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<td>17.00</td>
<td>3.40</td>
<td>8.00</td>
</tr>
<tr>
<td>Anosognosics</td>
<td>13.75</td>
<td>23.75</td>
<td>10.50</td>
<td>2.00</td>
</tr>
</tbody>
</table>

Fig. 4 – Different types of response given to the riddles. Repetitions correspond to false words that were proposed more than once to successive clues in the same riddle. Resolutions correspond to correct responses given after the fifth (final) clues. The number of repetitions and resolutions (mean for the 10 riddles) are plotted for each group (a; controls, nosognosics, and anosognosics) or detailed for the four anosognosic patients (b).

### 4. Discussion

By using a novel and simple “riddle” test, our study reveals that patients with AHP present with a specific impairment in evaluating the certainty of their guesses and adjusting to new conflicting information, unlike other patients who also suffer from right-brain damage and left hemiplegia but show no anosognosia. Although our sample was relatively small, this pattern was observed in group comparisons as well as in single-case analyses, with at least a statistical tendency for
A preceding trial and new information, rather than reject their current beliefs and make a new guess. In other words, while nosognosics or controls would usually change their mind and find another word that could suit better the succession of clues, anosognosics tended to persist with their preceding response and find in the new clues any thin link that could justify the given word. Remarkably, during testing, we observed that these patients did not simply persevere on their verbal responses; but they often tended to give reasons to do so and verbally formulate ad hoc explanations. For example, for the riddle illustrated in Fig. 1, an anosognosic patient answered “bread” after the first clue, but then said, after the second clue: “it is just as I thought, it is small bread...”; and after argued: “yes, like when we cut the bread, with the knife, it makes a regular noise...” These connections were often non-obvious but their justification offered with some logic. It is therefore unlikely that the impaired ability to change belief reflected a purely cognitive deficit in reasoning. These issues echo some of the debates in the current research on error monitoring in healthy people, where the nature of error and feedback monitoring functions are still unclear and associated with both sensorimotor and motivational factors (Holroyd and Coles, 2002; Ullsperger and von Cramon, 2004), as we have discussed elsewhere (Vocat and Vuilleumier, 2010). Moreover, this behavior is reminiscent of the fanciful justifications often provided by anosognosic patients (e.g., “my hand is asleep...”) when they are confronted with their weakness during neurological examination (Assal, 1983; Nathanson et al., 1952).

Importantly, such tendency to overconfidence and persistence in current beliefs was not accompanied by a more general impairment in executive functions, vigilance, spatial attention, or memory (as measured with our clinical bedside testing). In particular, no evidence for greater deficits in frontal lobe functions was found on clinical tests of reasoning and mental flexibility (Weigl sorting test) or word fluency (both phonological and semantic), which are typically highly sensitive to prefrontal lesions of various origins (Lezak et al., 2012). Thus, although we did not use standard neuropsychological tests of high-level executive function, there is no reason to think that our riddle task was more difficult, abstract, or demanding on memory, visual-spatial, or linguistic functions than some of the other bedside tasks; if
anything, the Weigl sorting test used here (requiring the flexible formation of categories based on shape properties) would actually be anticipated to be more abstract and cognitively challenging. Moreover, the present findings accord with previous studies that reported no significant correlation between anosognosia and several frontal lobe deficits (Anderson and Tranel, 1989; Starkstein et al., 1992; Vocat et al., 2010). However, because our testing was limited by clinical constraints due the acute neurological settings, it would interesting in future studies to explore whether the deficits observed here does or does not correlate with standard frontal tests as well as more specific decision tasks and self-monitoring tests that are not routinely tested in the clinic.

Therefore, the specific impairment in evaluating and correcting current belief states, as we observed here in anosognosics, might point to some dysfunction in higher-level executive functions that depend on brain areas distinct from those concerned with standard frontal tasks. We suspect that this more specific deficit might reflect damage to prefrontal cortical areas (Coltheart, 2010; Goel et al., 2009) or fronto-striatal circuits (Samejima and Doya, 2007) that are known to be recruited by self-monitoring, error detection, or set shifting conditions, and also frequently found to be damaged in patients with AHP (Pia et al., 2004; Vocat et al., 2010). Damage to the right insula is also common in AHP (Vocat et al., 2010; Baier and Karnath, 2008) and might also contribute to abolish predictive affect signals related to uncertainty and error risk (Preuschloff et al., 2008; Singer et al., 2009). Further research is needed to better understand the exact cognitive mechanisms underlying belief formation and verification, as well as their neural substrates (Harris et al., 2008). Interestingly, functional neuroimaging work in healthy volunteers suggests that judgments of disbelief (“false” vs “true” responses) made in response to statements from various domains (e.g., semantic “devious means friendly” or personal “you were born in New York”) correlate with activation of insula and anterior cingulate regions associated with error monitoring (Harris et al., 2008; Kapogiannis et al., 2009), as well as areas in the right temporoparietal junction recruited by perspective taking and shifting (Sommer et al., 2007), all commonly damage in patients with AHP than without AHP (Vocat et al., 2010), particularly when AHP persists. Interestingly, our own overlap analysis (see Fig. 5) showed that this region, the right temporoparietal junction, was more frequently damaged in anosognosic patients (although these results are difficult to interpret since they are based on a small sample of patients).

Importantly, we found that after receiving all 5 clues with progressively increasing informative consent, anosognosics patients were able to find the correct word solution on the last step as often as the controls or nosognosics did. This suggests that anosognosics had no general problem in reasoning but required a repeated signal of errors, or a larger incongruence between a new clue and the previous guess, in order to prompt a re-appraisal of their preceding responses and to trigger a new solution. The threshold to evoke doubt about the current belief might be pathologically increased in these patients, presumably due to their lesion site or perhaps other factors including personality or past experiences (Weinstein and Kahn, 1953). This observation is also consistent with the fact that AHP is typically transient after acute brain injury: as the patients progressively recover from their neurological deficits and receive more informative feedbacks on their current sate, their awareness of paralysis increases and their anosognosia tends to disappear. This view also supports the potential benefits of daily confrontation with the deficit to enhance awareness and facilitate rehabilitation (Fotopoulou et al., 2009; Ownsworth et al., 2006).

On a more general level, our findings accord with the proposal that some aspects of AHP after right-brain damage might reflect the role of the left hemisphere as a “story-teller” (Ramachandran, 1995) or “interpreter” (Gazzaniga, 2000) that uses any available information in a given situation and builds the most plausible “story” with it (see also Assal, 1983). In this process, relevant congruent information tends to be highlighted while incongruent elements are ignored or suppressed, as if they were pulled out of consciousness. The role of the right hemisphere (e.g., mediated by fronto-striatal or insula circuits) might constitute a “devil’s advocate” that help evaluate and reject inappropriate beliefs generated in the left hemisphere, by signaling self-relevant errors and promoting the resolution of uncertain inferences (Coltheart, 2010; Goel et al., 2009). In patients with anosognosia, the right hemispheric lesion might disrupt this “devil’s advocate” function and reduce the ability to check and change current beliefs about one’s own state despite the presence of incongruent information. Due to a lack of error signals and subsequent check operations, the patients may be abnormally confident in their beliefs and fail to integrate new information in order to modify them when confronted with incongruence. Hence, anosognosics might be unable to change their past beliefs (“my left arm is fully functional”) simply because they have no grounds to do so at both the sensory-motor and affective-motivational levels. This account of AHP partly converges with the “discovery hypothesis” put forward by Levine et al. (1991), but we surmise that the discovery failure may reflect a more specific impairment in belief monitoring abilities rather than from a general cognitive defect or dementia (as proposed by Levin et al.); and that an abnormal experience of motor weakness may not only be caused by losses in proprioception but also influenced by other deficits including spatial neglect, impaired motor intention, amnesia, etc. (Vuilleumier, 2004; Davies et al., 2005), perhaps in different combinations in different patients (see Vocat and Vuilleumier, 2010 for the description of a multifactorial model of self-monitoring and anosognosia). This variety of factors may account for differences in terms of the specificity and selectivity of AHP commonly observed in the clinic (Cocchini et al., 2010; Fotopoulou et al., 2008; Marcel et al., 2004). Importantly, like all previous studies investigating neuropsychological correlates of AHP (e.g., Vocat et al., 2010; Marcel et al., 2004), our finding of a strong association between anosognosia and a deficit in beliefs updating does not demonstrate that the latter is causal. As proposed for other deficits, such impairment is likely to be neither sufficient nor necessary to produce anosognosia but we suspect it could be crucial in some cases, particularly in combination with other deficits disrupting a normal experience of motor function (e.g., due to concomitant losses in feedback or feedforward control mechanisms). Importantly, a deficit in belief shifting alone would not be sufficient to

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cause anosognosia (for hemiplegia and/or other concomitant deficits) but likely to provoke denial only (or predominantly) for a function for which direct information (e.g., motor intention or proprioceptive signals) is degraded or unavailable for the patient (see also Vuilleumier, 2004; Levine et al., 1991).

Other recent theories about AHP have highlighted the importance of motor planning and motor intention disruptions (Berti et al., 2005; Heilman, 1991; Vallar et al., 2003), or anomalies in body ownership and motor agency (Baier and Karnath, 2008). These theories all agree with the fact that elementary neurological deficits (in motor or sensory function) are not sufficient to fully explain AHP, but hypothesize that some impairment in higher-level monitoring processes must be involved. In our study, however, we further suggest that the latter high-level deficits may not necessarily imply specific functions related to motor actions or attention, but can be observed in other domains when patients are confronted with partial and uncertain information — as demonstrated here in our new “riddle” test. Although this disruption alone is neither necessary nor sufficient to lead to anosognosia, we believe that it may provide a “fertile field”, for the emergence of full-blown anosognosia when co-existing with other deficits (including in motor intention and comparator processes) and that it could contribute to the severity and/or persistence of anosognosia. Likewise, using an ingenious reality monitoring task, Jenkinson et al. (2009) reported that both patients with AHP and those without AHP failed to discriminate between motor memories internally generated by intentions versus externally generated by actual execution, while only the patients with AHP additionally failed to judge the reality of past visual imagination versus experience, suggesting a more general deficit in monitoring processes beyond motor function only.

Our study is not without limitations. In particular, although we found consistent anomalies in confidence responses in every patient with AHP (with at least a strong statistical tendency), our sample was relatively small (leading to relatively low statistical power for some of our analysis). Testing acute stroke patients at the bedside with specific and quantitative experimental tasks is challenging, and AHP tends to disappear after a few days or even hours in many cases (Vocat et al., 2010). Nonetheless, future studies need to recruit larger groups with different degrees of anosognosia and different types of neurological deficits, as well as to explore the role of more specific decision making processes. Having established this phenomenon of overconfidence in a guessing task for the first time, we hope that future research will investigate and confirm similar behaviors in other neuropsychological syndromes associated with anosognosia (e.g., amnesia, blindness). Preliminary data with our “riddle task” administered to an anosognosic patient with cortical blindness suggest the exact same pattern of findings: over-confidence and difficulties to update wrong beliefs. Thus, with a different kind of anosognosia, with no relation to motor functions and a different distribution of lesions, the same higher-level cognitive processes associated with belief formation and veriﬁcation seemed also to be crucially involved. The role of such processes has certainly been underestimated and understudied until recently. Therefore, a major goal for future studies on anosognosia should be to clarify the neuro-cognitive substrates implicated in belief monitoring, and better understand their interaction with other deficits affecting motor monitoring in AHP.

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Appendix A. The 10 riddles

**Riddle 1:** Airplane
I have wings
I can fly
I can move very fast
I exist in various sizes
I have wheel

**Riddle 2:** Tooth
I’m white
I have deep roots
I need daily maintenance
I’m covered with enamel
Some children keep me for the little mouse

**Riddle 3:** Carrot
I am a food
I am very cultivated
I am a vegetable
I am usually orange
The rabbits adore me

**Riddle 4:** Key
I can be of metal
I can be carry in a hand
It’s better not to lose me
I protect against thieves
I allow people to return home

**Riddle 5:** Cow
I am an animal of the female gender
I am sometimes black and white
I can be sacred
Sometimes I wear a bell
I chew when I have nothing else to do

**Riddle 6:** Heart
My weights is approximatively 300 g
I produce a regular sound
Sports makes me feel excited
I am usually on the left rather on the right side
Lovers often draw me

**Riddle 7:** Shadow
Without the sun, I could not survive
You often takes advantage of me when it’s hot

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Riddle 8: Garden
I can tell you the time
You can’t lose me or catch me

Riddle 9: Matches
I can be in a pocket or in a box
I change color after use
I fear water
My modern competitor is the vacuum cleaner

Riddle 10: Broom
I am a current object
I have a rigid part, the other soft
The cleaners use me frequently
I change with the seasons
Sometimes, professionals take care of me

Riddle 11: Garden
I love flowers but also vegetables
I can be hanging or secret
I love flowers but also vegetables
I need sun, water and fresh air

Riddle 12: Matches
I am a current object
I have a rigid part, the other soft
The cleaners use me frequently
I am essential to witches
My modern competitor is the vacuum cleaner

References


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