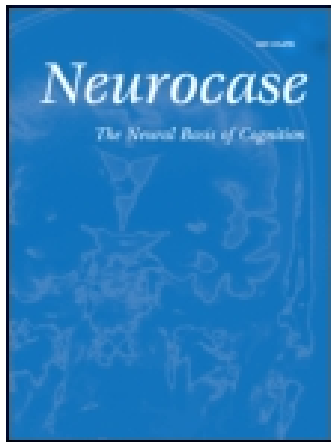


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### Controlling the alien hand through the mirror box. A single case study of Alien Hand Syndrome

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Published online: 05 Apr 2013.

To cite this article: D. Romano, A. Sedda, R. Dell'Aquila, D. Dalla Costa, G. Beretta, A. Maravita & G. Bottini (2014) Controlling the alien hand through the mirror box. A single case study of Alien Hand Syndrome, *Neurocase: The Neural Basis of Cognition*, 20:3, 307-316, DOI: [10.1080/13554794.2013.770882](https://doi.org/10.1080/13554794.2013.770882)

To link to this article: <http://dx.doi.org/10.1080/13554794.2013.770882>

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# Controlling the alien hand through the mirror box. A single case study of Alien Hand Syndrome

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Disruption of motor control in the alien hand syndrome might result from a dissociation between intentions and sensory information. We hypothesized that voluntary motor control in this condition could improve by restoring the congruency between motor intentions and visual feedback. The present study shows that, in one patient with right alien hand syndrome, the use of a mirror box paradigm improved motor speed. We speculate that the visual feedback provided by the mirror increases the sense of congruence between intention and sensory feedback, leading to motor improvement.

**Keywords:** Alien Hand Syndrome; Mirror box; Body representation; Rehabilitation; Stroke.

Alien hand syndrome (AHS) is a rare condition, generally associated with brain lesions (Ay, Buonanno, Price, Le, & Koroshetz, 1998; Bartolo et al., 2011; Feinberg, Schindler, Flanagan, & Haber, 1992; Kikkert, Ribbers, & Koudstaal, 2006; Litvan et al., 2003), that induces involuntary and unwilling goal-directed movements of the contralateral hand. Patients reach and grasp for objects without volition, touch or scratch their face and head, or even act aggressively against themselves (Spector, Freeman, & Cheshire, 2009). Finally, the unaffected hand may often oppose the action of the alien hand (Assal, Schwartz, & Vuilleumier, 2007).

Even though the term AHS has been adopted to indicate different behaviors related to the contralateral hand and different nomenclatures have been used as equivalent, recent studies are beginning to identify and define different subtypes of AHS (Aboitiz et al., 2003; Hertza, Davis, Barisa, &

Lemann, 2012; Pappalardo, Ciancio, Reggio, & Patti, 2004; Scepkowski & Cronin-Golomb, 2003). Four different subtypes of AHS have been suggested, according to the clinical characteristics and anatomical lesions of patients:

- (a) Alien Hand Sign: typically associated with lesions of the posterior corpus callosum, this subtype addresses for the subjective feeling that one's own hand is perceived as extraneous.
- (b) Intermanual conflict (diagnostic dyspraxia): usually associated with posterior corpus callosum lesions, it refers to the autonomous movement of one hand against the intended action of the other one.
- (c) Anarchic Hand: associated with lesions of the anterior corpus callosum, the left medial prefrontal area, the anterior cingulate gyrus and the supplementary motor area. This symptom concerns the denial of one's own intentions

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for actions of the hand and may present with reflexive grasping and utilization behavior.

- (d) **Supernumerary Hands:** associated with corpus callosum and right hemisphere lesions and refers to the patient's feeling of one or more extra arms.

Other authors have suggested partially different anatomical correlations, including the corpus callosum and the medial frontal lobe (Brainin, Seiser, & Matz, 2008), or the parietal lobe and the thalamic nuclei (Assal et al., 2007; Spector et al., 2009). Even though AHS mostly affects the left hand, some cases have also been reported for the right hand (Bartolo et al., 2011; Della Sala, Marchetti, & Spinnler, 1991; McNabb, Carroll, & Mastaglia, 1988; Pappalardo, et al., 2004).

In summary, a complete agreement on the anatomo-clinical correlations of the different clinical features of the AHS is still lacking (Hertza et al., 2012; Pappalardo et al., 2004).

The sense of ownership refers to the implicit feeling that a body part belongs to one's own body, while the sense of agency refers to the feeling of being able to move a body part and control it (Longo, Schüür, Kammers, Tsakiris, & Haggard, 2008). The right parietal and frontal lobes, critically involved in the brain representation of the body, have been suggested as the possible anatomical underpinnings of such complex abilities (Tsakiris, 2010), suggesting further that body representation underpinnings and AHS anatomical localization share, at least in part, some brain areas. From a conceptual point of view, cognitive models account for AHS in terms of a disorder of agency (Schaefer, 2002; Spence, Heinze, & Galazky, 2012) given that the core feature of this deficit consists of hand movements without volitional control. However, taking into account the different subtypes of AHS, it seems reasonable to hypothesize a dissociation between a disorder of agency, due to the involvement of frontal components, such as in the case of anarchic hand and utilization behavior, and a disruption in the sense of ownership (Synofzik, Vosgerau, & Newen, 2008), more likely in the case of the alien hand behavior. Nevertheless, the two concepts of body ownership and sense of agency are closely entangled (Longo et al., 2008; Tsakiris, Prabhu, & Haggard, 2006) and both contribute to the emergent sense of self (Longo et al., 2008). Consequently, it is possible to hypothesize that both systems might be disrupted in each subtype of AHS (Spence, 2002; Synofzik et al., 2008), and

their balance account for the different features of the syndrome.

It is interesting to note that neuroimaging studies have shown similar activations in the primary motor cortex while movements were performed by both the normal and the alien hand, suggesting that conscious and unwilling AHS actions may share common neural correlates in the motor cortex (Assal et al., 2007). However the same study highlights a dissociation between intended and unintended movements in supplementary motor areas and parietal cortex, supporting the idea of an high level dysfunction in AHS, possibly due to the reduced inhibition of actions and to an impaired attentive component. The authors also highlighted that the distributed network implied during voluntary movements compared to alien movements showed stronger activation for the left dominant hemisphere especially for the prefrontal cortex, supporting the idea that the left hemisphere may play a crucial role in guiding willed action (Assal et al., 2007; Frith, Friston, Liddle, & Frackowiak, 1991). Models of motor learning and control try to explain AHS in terms of a dysfunction in the relationship between motor commands and sensory feedback, that are controlled by the motor system. According to the "forward model of motor control", in which the efference copy of the movement is used to predict the sensory consequences of a motor command (Frith, Blakemore, & Wolpert, 2000), actions of the anarchic hand occur because the effects of the visual environment are no longer inhibited by the currently intended action. Notably, the patient is aware of the discrepancies between intention and action, differently from other diseases that involve the motor system but are characterized by unawareness of the impairments, such as anosognosia for hemiplegia (Frith et al., 2000).

Independently from the theoretical point of view adopted to explain the AHS, and from its rarity, this syndrome is debilitating for the patient and requires attention when it becomes chronic. Importantly, AHS affects most everyday life activities, preventing a complete recovery to normal life in patients. Rehabilitation for AHS has been recently reported (Cantagallo, Spinazzola, Rabuffetti, & Della Sala, 2010; Pappalardo et al., 2004; Pooyania, Mohr, & Gray, 2011). Nevertheless, most of the available treatments are tailored to the individual patients and thus are quite difficult to propose as standard rehabilitative approaches.

The mirror box (Ramachandran & Altschuler, 2009) is a well known rehabilitative paradigm.

In the typical mirror box paradigm, participants move one hand in front of a parasagittal mirror while the other hand is kept behind the mirror, hidden from direct view. In this situation the mirror reflection of the moved hand is consistent with the posture of the contralateral hand behind the mirror. The vision of a moving hand congruent with the impaired one behind the mirror was found to induce motor facilitation in post-stroke patients (Altschuler et al., 1999; Yavuzer et al., 2008) or to alleviate pain in phantom limb or chronic regional pain syndrome patients (Chan et al., 2007; McCabe et al., 2003; Ramachandran & Rogers-Ramachandran, 1996; Rossi et al., 2002; Selles, Schreuders, & Stam, 2008).

We hypothesized that the congruency between motor intention and visual feedback elicited by the vision of the intact hand moving, through the mirror, would improve the voluntary control of the alien hand. From a different theoretical perspective, this paradigm would improve putatively reduced sense of agency and ownership through the embodiment of the reflected image of the functional arm that would partially restore the representation of the impaired hand. If this hypothesis was true, the mirror box would prove to be an effective and easy-to-use technique to treat the AHS.

## METHODS

### Case report

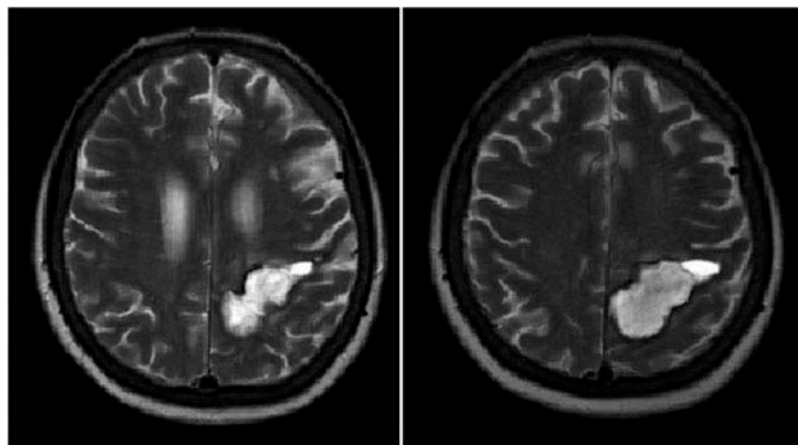
DT is a 62 years old right-handed, highly educated woman (master degree in law), who suffered from

an intracerebral haemorrhage in the left frontoparietal region, as confirmed by the MR examination performed at the time of her admission to the hospital (Figure 1).

The patient was collaborative and oriented. A general neuropsychological evaluation showed a multicomponetial cognitive deficit including number processing, verbal working and visuo-spatial short term memory, attention and executive functions (i.e. working memory, sensitivity to interference, conceptualization and programming motor sequences), finger agnosia, perceptual abilities and ideative apraxia (Table 1). The patient was diagnosed with a Gerstmann syndrome (Gerstmann, 1930; Roeltgen, Sevush, & Heilman, 1983; Strub & Geschwind, 1974). Furthermore, her standardized neurological examination (Bisiach, Vallar, Perani, Papagno, & Berti, 1986) showed extinction on double tactile stimulation (1/3) and mild personal neglect (1/3), both for the right side. A mild motor deficit was also found (1/3) for the right upper limb, specifically she presented hypostenia without hemiplegia.

The patient also presented with AHS for her right hand. This hand moved without voluntary control, reaching both personal and peripersonal space, without any aggressive behavior and she presented no signs of heminattention except for a right side extinction during a bilateral tactile stimulation and a mild difficulty of finding her right arm with the left hand while she was blindfolded. Nevertheless, DT was aware of the uncontrolled movement of the contralesional hand.

The patient was used to move the right hand without voluntary control: many times the hand unexpectedly started moving in the air, touching



**Figure 1.** Hemorrhagic stroke lesion of DT in T2 MR images; left hemisphere is on the right of the pictures.

**TABLE 1**  
Neuropsychological evaluation, tests are split by cognitive function

<i>Sub-test</i>	<i>Scores</i>
<b>Language</b>	
Phonemic word fluency (Novelli, Capitani, Vallar, & Cappa, 1986)	35
Semantic word fluency (Novelli et al., 1986)	27
Picture naming (Lansing, Ivnik, Cullum, & Randolph, 1999)	14
<b>Counting skills</b>	
AC-MT numbers dictate (Cornoldi, Lucangeli, & Bellina, 2002)	0/8*
Numerical facts (Cornoldi et al., 2002)	5/12*
Mental count (Cornoldi et al., 2002)	0/0*
<b>Verbal memory</b>	
Short Story (immediate recall) (Carlesimo et al., 2002)	0*
Short Story (later recall) (Carlesimo et al., 2002)	5
Short Story (oblivion) (Carlesimo et al., 2002)	5
Digit span (Orsini et al., 1987)	7
Backwards digit span	2*
<b>Visuo-Spatial Memory</b>	
Corsi span (Spinnler & Tognoni, 1987)	3*
<b>Attention/Executive functions</b>	
Attentional matrix (part I)	10
Attentional matrix (part II)	30
Attentional matrix (total)(Della Sala, Laiacona, Spinnler, & Ubezio, 1992)	40
Trail Making A (Giovagnoli et al., 1996)	146**
Trail Making B (Giovagnoli et al., 1996)	Ne*
Frontal Assessment Battery (Dubois, Slachevsky, Litvan, & Pillon, 2000)	11*
Verbal Judgment (Spinnler & Tognoni, 1987)	50
Letters cancellation test (Diller & Weinberg, 1977)	N-
<b>Abstract reasoning</b>	
Colored Progressive Matrix PM 47 (Spinnler & Tognoni, 1987)	17*
<b>Perceptual – gnosis functions</b>	
Benton lines (Benton, Varney, & Hamsher, 1978)	15*
Digital agnosia (Benton, Hamsher, & Varney, 1983)	13*
<b>Praxic functions</b>	
Ideomotor Apraxia (De Renzi, Motti, & Nichelli, 1980)	66/72
Ideative Apraxia	6*
<b>Neurological examination</b> (Bisiach, Cappa, & Vallar, 1983; Bisiach et al., 1986)	
Motor deficit	1/3*
Sensory deficit	1/3*(Extinction)
Visual deficit	0/3
Somatoparaphrenia	1/4*
Anosognosia for motor deficit	0/4
Anosognosia for sensory deficit	0/4
Anosognosia for visual deficit	0/4
Personal neglect	1/3*

Asterisks indicate pathological performances.

her face and head or scratching her body parts. Also uncontrolled and unwilling distal movements of the fingers were observed, such as a frequent, stereotyped opposition of the thumb to the other fingers. In order to control these arm and hand unwilling movements, the patient used to try and immobilize the whole limb by sitting on her hand. During the assessments and the experimental procedure, DT did not show utilization behaviors. Alien movements had a daily frequency, manifesting many times per day, and seemed to

increase while DT tried to do something with the other hand (i.e. having a meal, drink a glass of water, etc.). However these movements were not continuous and only rarely they clearly seemed to hinder the movements performed by the other hand.

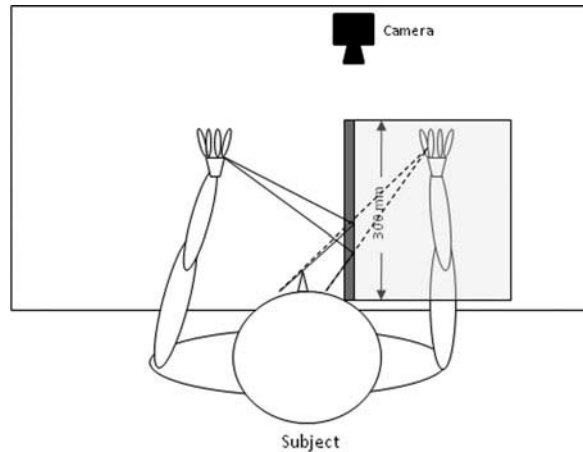
DT was deeply annoyed by these uncontrolled movements and reported the feeling that her right hand was not her own despite she “learned” that it was her own. The clinical picture of DT is compatible with a posterior form of AHS, presenting

as an alien hand sign subtype, with a mild frontal component of the syndrome. Her symptoms are consistent with the lesion site in the rostro-caudal area. Indeed her hemorrhage, which involved the fronto-parietal region, had a major impact on the parietal lobe at the cortical and subcortical level, also partially involving the posterior corpus callosum. Only a small involvement of the left frontal lobe in the middle frontal gyrus was shown by the MR. Nevertheless, it should be noted that AHS is typically associated with right hemisphere lesions (Aboitiz et al., 2003; Hertz et al., 2012), while our patient suffered from a left hemisphere stroke, so the anatomo-clinical correlation should be considered with caution.

### Mirror box paradigm

The patient was administered with an experimental mirror box task. To test the subjective effects of the task, she also answered a question through a Visual Analogue Scale (VAS).

During the mirror box training, the patient was comfortably seated in front of a table, on which the mirror box was placed. This consisted of an opaque box with a hole on the wall facing the patient, where she could introduce her hand, and a mirror featuring its parasagittal wall. The patient placed her arms on the table, keeping the right, affected hand, inside the mirror box and the other one outside the box, in front of the mirror. Then, she was asked to perform rhythmic tapping movements with both index fingers. In this situation the patient was able to see only her intact hand moving, and producing a mirror reflection compatible with the image of the affected hand, which was instead invisible inside the mirror box. Half of the times the mirror was left uncovered, thus providing visual feedback (mirror condition) and half of the times the mirror was covered, producing no feedback (no-mirror condition) (see Figure 2 for a schematic view of the experimental setting). Each condition was presented 8 times in a counterbalanced order, for a total of 16 blocks. To assess any improvement in the patient's motor control, the number of tapping movements made in 15 seconds by the alien hand was counted. The hypothesis is that the vision of a mirror reflection compatible with the image of the affected hand would increase motor control, thus increasing the frequency of tapping of the affected hand. Therefore we expected the frequency of tapping to be higher in the mirror than in the no-mirror condition.



**Figure 2.** Mirror Box Settings and measures. The patient has her right hand inside the mirror box (in gray) while she sees in the mirror her left normal one. She performed tapping movements with both hands with a camera recording the performance. Number of tappings of the right index finger produced in 15 seconds have been calculated as a measure of fine tuned movements.

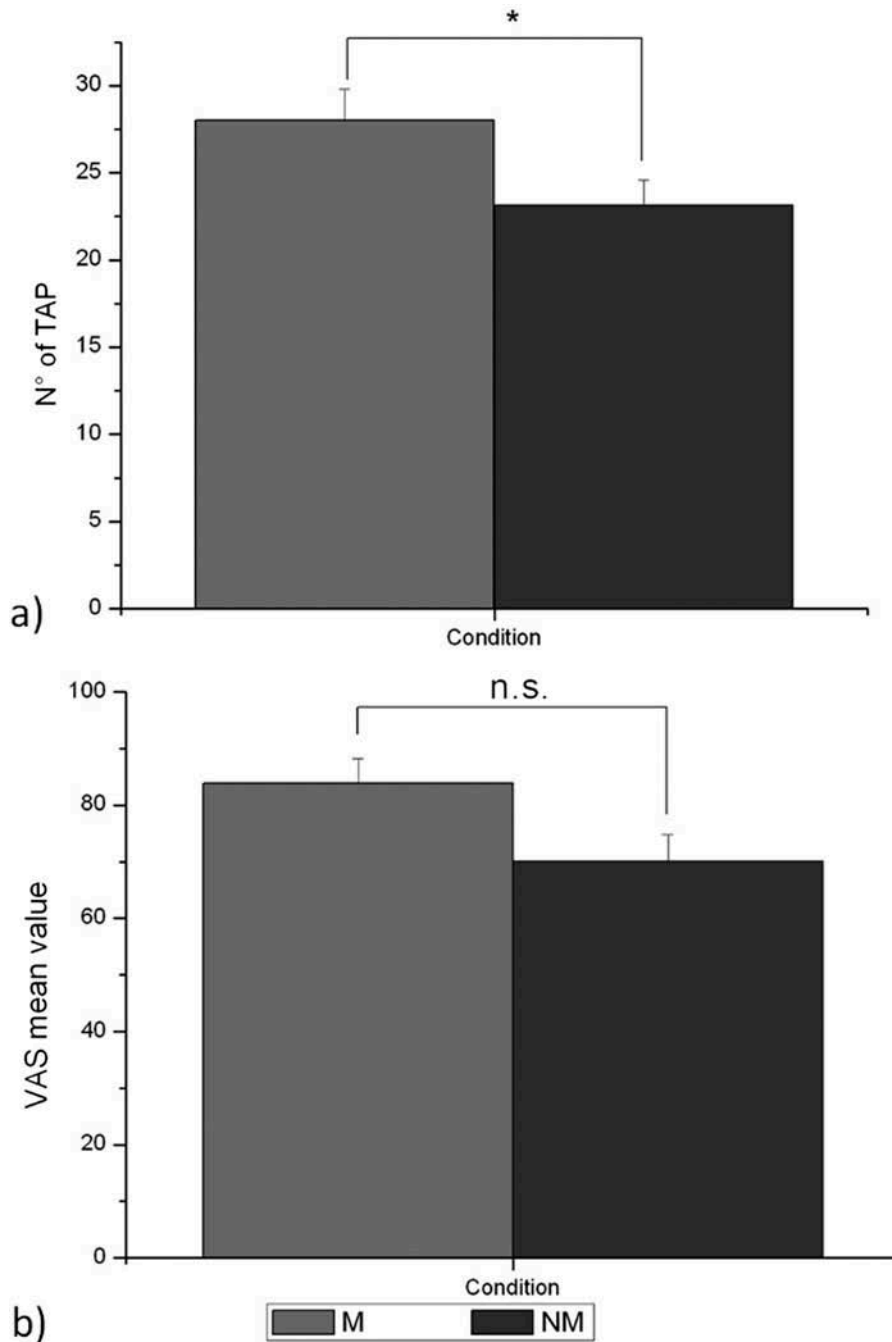
After completing each experimental condition, the patient was given a vertical VAS (100 mm length  $\times$  approximately 1 mm thickness), designed to investigate her awareness of movements accuracy. The VAS was presented vertically in order to avoid biases in response due to the eventual presence of neglect. The instructions of the VAS was: “How did you perform the movement? Put a sign on the bar near to the top if you think you moved well, otherwise put it near to the bottom.”

### RESULTS

Data were analyzed using PASW<sup>®</sup> Statistics 18 (IBM<sup>®</sup> SPSS<sup>®</sup> Statistics © 2010, Chicago, IL, USA) applying the non parametric Wilcoxon Test.

DT performed on average  $28 \pm 1.78$  (standard error) tapping movements in the mirror condition and  $23.12 \pm 1.47$  movements in the no-mirror condition (Figure 3a). At the VAS, she judged her performance to be accurate at  $83.87 \text{ mm} \pm 4.35$  in the mirror condition and  $70.12 \text{ mm} \pm 4.65$  in the no-mirror condition (Figure 3b).

Statistical comparisons between conditions in each task revealed a significant difference between the mirror and no-mirror conditions in the tapping task ( $Z = -2.205$ ;  $p = .027$ ) but not in the VAS ( $Z = 1.68$ ,  $p = .09$ ), suggesting an improvement in DT performance while tapping and seeing the mirror reflection of her movement, without a parallel change in her subjective evaluation of the condition.



**Figure 3.** In (a) the graph shows the mean number of tapping movements, asterisk indicate for a significant difference; while in (b) the graph displays mean value of VAS self-evaluations and n.s indicates that difference was not statistically significant. Bars represent Standard Error, M = Mirror condition (light gray); NM = No-Mirror condition (dark gray).

Considering both conditions, VAS judgments and tapping performances correlated at a significant level (Spearman's  $\rho = .56$ ;  $p = .02$ ), suggesting that the patient was, at least in part, conscious of the positive effect of the task on her performance.

## DISCUSSION

The Alien Hand Syndrome (AHS) is a rare neuropsychological disorder in which patients experience involuntary and unwilling goal-directed movements of the contralesional hand (Brion &

Jedynak, 1972; Sala et al., 1991). This syndrome is debilitating for the patient as it interferes with everyday activities. However, at present, there are no established treatments for this condition. Here we present a case of right AHS, which we attempted to modulate through the mirror box paradigm.

The most reliable explanation of the mirror box treatment effectiveness is that the intact arm, reflected in the sagittal mirror, is perceived in the felt location of the affected arm which is somehow revitalised by the attribution of sensorimotor features of the intact arm (Ramachandran & Altschuler, 2009). The mirror box paradigm has never been used in case of AHS.

According to our working hypothesis, in the present experiment we found that, in the mirror condition, we observed not only a quantitative improvement of the tapping movements, as quantified by the number of tapping the patients performed in 15 seconds, but also a qualitative improvement of the fine motor control during the task execution. During this condition we noticed that the patient was able to move only the index finger (as requested) while the other fingers and the whole hand were kept stationary in a comfortable position; in each one of the 8 mirror trials we did not observe any unexpected and unwilling movement. Interestingly, the patient sometimes spontaneously reported to feel in control of her hand. Strikingly, DT reported that: “[my hand] ameliorated, it does not fly here and there as she pleases, even though sometimes I still have difficulties in controlling it. Now I can feel the hand, even though she annoys me a bit, while before I did not even feel it, now it seems more my hand!” By contrast, in the no-mirror condition we observed that, after a few seconds, DT started to lose control of her right hand producing incoherent movements with the other fingers at first, and with the whole hand after a while. In some trials, in this no mirror condition, the patient removed the hand from the mirror box, a behavior that we interpreted as a manifestation of the syndrome, given that the patient explicitly declared that she did not want to remove it, and tried to put again the hand inside the box. Moreover, DT verbally reported to feel confident with the efficacy of her performance. Despite the lack of a statistical difference in the VAS judgments between the mirror and no-mirror conditions, considering both conditions together, we observed a significant correlation between the VAS judgments and the tapping performance, showing

the reliability of the patient self-evaluation and at least a partially conscious insight on the task efficacy.

Unfortunately, as we had the opportunity to perform this mirror box paradigm under experimental control only once, due to clinical constraints of the hospital setting, we cannot provide evidence for a long term effect of such a rehabilitation approach and the improvement found so far could be limited to the exposure to the mirror visual feedback. However, during DT’s standard rehabilitation program in the 5 days following the experimental procedure presented here, DT performed also some mirror box treatment sessions and she spontaneously reported that after a few days the unwilling movements decreased both in term of frequency and intrusiveness even if they did not disappear completely. Although anecdotal, this observation suggests that a prolonged treatment with the mirror box could be beneficial in patients with AHS.

Furthermore, DT’s neuropsychological evaluation suggested that our patient did not present AHS isolated. Gerstman syndrome symptoms may have contributed to enhance AHS, as the left/right disorientation and finger agnosia may have impacted on the perception of her own arms (Hertza et al., 2012). On the other hand, AHS symptoms might confound the patient’s performance at the tests for ideative apraxia, in which the patient obtained a pathological score. It might be possible that DT did not have a deficit in identifying the correct way of using objects, rather the unwilling movements of the right hand might have determined the poor performance when the patient was required to perform bimanual tasks. Indeed her performance was inadequate for bimanual movements such as opening a plastic bottle or sharpening a pencil, and not for unimanual actions. This pattern of results seems to suggest that the pathological score is more related to the presence of AHS than to a mere apraxic deficit.

The hypothesis that the congruence between the motor intentions of the patient and the sensory feedback from the environment could have improved movements efficacy in DT seems to be confirmed by our clinical results. We suppose that the image of the hand observed in the mirror has been embodied in the motor control system, replacing to some extent the representation of the affected hand inside the box. This visual feedback may have thus provided a representation of a contralesional limb with better functionality with consequent improvement of motor performance triggering a



normal functioning. The rapid effect of our mirror box paradigm on behavior, without the need of any learning period, supports the hypothesis of an automatic and quick incorporation of the mirror image as a body part, not based on neural plasticity (Altschuler et al., 1999).

Interestingly, we also observed a partial, although not complete, relationship between the motor performance and the awareness of it. While there was no difference at the VAS scores between conditions, we found a correlation between the VAS scores and the performance, that suggests some degree of transfer of the behavioural effect at the conscious level.

Future studies could try to investigate further the relationship between motor improvement and explicit or conscious perception of the improvement, to shed further light on the mechanisms of the mirror box paradigm.

It is worth noting that, although AHS is usually described as a consequence of right hemispheric lesions (Assal et al., 2007; Brainin et al., 2008; Spector et al., 2009), our patient presented with this condition after a left fronto-parietal lesion. In our view this could not be attributed to an atypical hemispheric lateralization, as the patient Oldfield handedness scale (Oldfield, 1971) indicated a full right handedness and she showed a complete Gerstmann syndrome typically associated with left fronto-parietal and/or callosal lesions (Gerstmann, 1930; Roeltgen et al., 1983; Strub & Geschwind, 1974). Furthermore rare cases of right alien limb associated with left or bilateral cerebral lesions (Della Sala et al., 1991; McNabb et al., 1988) have already been reported.

Finally, our patient did not show an evident ideomotor apraxia, and our dependant variables could not discriminate between improvement of praxis and restored AHS. Thus, we cannot rule out the possibility that the mirror box treatment could have induced an improvement in voluntary motor control itself, restoring motor praxis rather than directly affecting alien hand movements. Further studies using the mirror box paradigm in groups of patients with either apraxia or the AHS could clarify this point. Nevertheless, it is interesting to note that an effect on apraxia is not mutually exclusive with the restoring of the AHS, rather it might be possible that the embodiment of a functional arm would act on both impairments, restoring the sense of agency, improving the sense of ownership and ameliorating the planning of motor sequences.

A final remark is that this paradigm could work only for the posterior type of AHS, which is supposed to depend on a disconnection between sensory-motor and intention systems, rather than for the anterior form of AHS which seems to depend on a failure to inhibit motor behavior of the non-dominant hemisphere (Hertza et al., 2012). However, the embodiment hypothesis could explain an eventual improvement also for the frontal subtypes of AHS: providing a visual feedback congruent with motor intentions could trigger the embodiment of the reflected arm, thus partially restoring the body representation of that arm. The hypothesis that the restored body representation influences action control and motor planning should be tested also for frontal forms of AHS before concluding for dissociable mechanisms and treatments.

Even though the present case may not be taken as a representative of all cases of AHS, given the anatomo-clinical peculiarities, we believe that, the rationale underpinning the use of the mirror box approach, has the theoretical basis to work also with different types of AHS and more in general with high cognitive dysfunctions of action and body representation. Given these premises, we believe the present work gives new insight that should be explored also with other forms of motor dysfunction, such as frontal AHS or apraxia, especially when considering that, so far, effective and reliable treatments for these symptoms are still lacking.

In conclusion the modulatory effect of the mirror box on motor performance of this AHS patient might be considered a novel finding suggesting that even high-level body representation deficits may benefit from such a treatment.

Original manuscript received 7 June 2012

Revised manuscript accepted 16 October 2012

First published online 5 April 2013

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