Another perspective on anosognosia: Self-observation in video replay improves motor awareness

Sahba Besharati[1,2], Michael Kopelman[1], Renato Avesani [3], Valentina Moro[4*] & Aikaterini Fotopoulou[1,5*]

[1] Institute of Psychiatry, King's College London, UK.
[2] Department of Psychology, University of Cape Town, South Africa.
[3] Department of Rehabilitation, Sacro Cuore Hospital, Negrar, Verona, Italy
[4] Department of Philosophy, Pedagogy and Psychology University of Verona, Italy
[5] Research Department of Clinical, Educational and Heath Psychology, University College London, UK.
Correspondence concerning this article should be addressed to:

Aikaterini (Katerina) Fotopoulou
CEHP Research Department
University College London
1-19 Torrington Place
London WC1E 7HJ
Tel: (+44) 020 7679 3079
Fax: (+44) 020 7916 8502
Email: a.fotopoulou@ucl.ac.uk.

*Valentina Moro and Aikaterini Fotopoulou had equal, senior author contributions to this article.

Abstract

Anosognosia for hemiplegia (AHP), or unawareness of motor deficits contralateral to a brain lesion, has lasting negative implications for the management and rehabilitation of patients. A recent, bedside psychophysical intervention, namely self-observation by video replay, lead to a lasting remission of severe AHP in an acute stroke patient (Fotopoulou, et al., 2009). This procedure has been adjusted and applied here, as the basis of two intervention protocols administered independently to two patients with severe AHP. The first study used multiple, successive sessions of video-based self-observation in an acute patient, targeting first the awareness of upper limb and subsequently lower limb paralysis. The second study used a single session of video-based, self- and other- observation in a patient at the chronic stage following onset. Both protocols also involved elements of rapport building and emotional support. The results revealed that video-based self-observation had dramatic, immediate effects on awareness in both acute and chronic stages and it seemed to act as an initial trigger for eventual symptom remission. Nevertheless, these effects did not automatically generalise to all functional domains. This study provides provisional support that video-based self-observation may be included in wider rehabilitation programmes for the management and restoration of anosognosia.

Keywords: anosognosia; motor awareness; perspective; video replay; neurorehabilitation

Abbreviation: AHP= anosognosia for hemiplegia

INTRODUCTION

Unawareness of neurological deficits, i.e. the apparent inability of patients to perceive or acknowledge their impairments, is commonly referred to as anosognosia (Babinski, 1914). Anosognosia occurs in the context of several neuropathologies, including stroke, Alzheimer's (Reed, Jagust & Coulter, 1993), schizophrenia (Mohamed, Fleming, Penn & Spaulding, 1999), and traumatic brain injury (Prigatano, 1998). This study will focus on anosognosia for hemiplegia (AHP) following stroke, i.e. the denial of contralesional paralysis (Babinski, 1914). The incidence of AHP is poorly specified, with reported rates ranging from 7% to 77% of stroke patients (Orfei et al., 2007). AHP is most frequently caused by right perisylvian lesions, but is also reported in left-sided brain damaged patients (Cocchini, Beschin, Cameron, Fotopoulou & Della Sala, 2009). Although lesions of the right posterior insula (Karnath, Baier & Nagele, 2005) and premotor cortex (Berti et al., 2005) have been selectively associated with AHP, more recent results point to additional, critical lesion sites including subcortical structures (basal ganglia, hippocampus, amygdala) and deep white matter tracts (Fotopoulou, Pernigo, Maeda, Rudd & Kopelman, 2010; Vocat, Staub, Stroppini & Vuilleumier, 2010; Moro, Pernigo, Zapparoli, Cordioli & Aglioti, 2011).

The clinical characteristics of AHP may vary between and within patients in several ways (Marcel, Tegnel & Nimmo-Smith, 2004; Orfei et al., 2007). For instance, AHP can vary in severity, ranging from a mere indifference (anosodiaphoria; Babinski, 1914) to blatant denial of limb paralysis even in the face of undeniable clinical demonstrations. It can also vary in specificity (i.e. exclusively involving the upper or the lower limb) and extension (i.e. ability to recognise a deficit but not its functional consequences) (see Marcel et al., 2004 and Fotopoulou, 2012). Furthermore, clinical and experimental studies have demonstrated that certain patients have tacit or implicit awareness into their deficits, while being explicitly unaware of them (Nardone, Ward, Fotopoulou & Turnbull, 2008; Fotopoulou et al., 2010) and vice versa (Cocchini et al., 2009; Moro et al., 2011).

Many theories have been proposed to explain the causes and clinical variability of AHP (for review see Vuillleumier, 2004; Orfei et al., 2007; Heilman & Harciarek, 2010; Fotopoulou, 2013). AHP had traditionally been regarded as a secondary consequence of concomitant sensorimotor deficits or, hemispatial neglect (Cutting, 1978; Vallar & Ronchi, 2006) or a combination of sensory and other higher-order deficits, such as amnesia, general cognitive impairment and confabulation (Levine Calvanio & Rinn, 1991; Berti, Ladavas & Della Corte, 1996; Mattioli, Gialanella, Stampatori & Scarpazza, 2012). However, double dissociations have been found between AHP and such deficits (Bisiach, Vallar, Perani, Papagno & Berti, 1986; see Heilman & Harciarek, 2010 for review), pointing to the role of other factors in the pathogenesis of the syndrome. Initially some authors had argued that AHP acts as a psychological defence against depression and anxiety (Weinsein & Kahn, 1950, 1955), while more recently others have proposed that AHP is caused by a selective deficit in motor planning (Heilman, Barrett, Adair, 1998; Vallar & Ronchi, 2006), or motor monitoring (Frith, Blakemore & Wolpert, 2000; Berti et al., 2005; Fotopoulou, Tsakiris, Haggard , Rudd & Kopelman, 2008; Gabardini et al., 2012). Given that none of these factors seems sufficient to explain the multifaceted nature of AHP, there is currently some consensus regarding its heterogeneous nature (Vuilleumier, 2004; Cocchini, Beschin, Fotopoulou & Della Sala, 2010; Vocat et al., 2010; Moro, Pernigo, Zapparoli, Cordiolo & Aglioti, 2011; Fotopoulou, 2012), including a potential combination of cognitive and motivational factors (Solms, 1995; Vuilleumier, 2004; Turnbull, Evans & Owen, 2005; Feinberg Roane & Ali, 2000; Fotopoulou et al., 2010). However, the precise combinations of deficits capable of causing the syndrome and the dynamic relation between the various critical factors remains unknown.

Importantly for the present purposes, although AHP is often a transient phenomenon with spontaneous recovery occurring within days or weeks from onset (Vocat et al., 2010), the presence of AHP in the acute stage may significantly obstruct rehabilitation efforts and consequently impede long-term functional outcomes (Gialanella, Monguzzi, Santoro & Rocchi, 2005; Jehkonen, Laihosalo & Kettunen, 2006). Specifically, patients with unawareness symptoms may refuse treatments that considerably improve prognosis (Di Legge, Fang, Saposnik & Hachinski, 2005; Cherney, 2006), may not take appropriate safety measures (Hartman-Maier, Soroker & Katz, 2001; Hartman-Maier, Soroker, Ring & Katz, 2002) and may not be realistic about their rehabilitation, housing, social and financial needs (Orfei et al., 2007; Prigatano & Morrone-Strupinsky, 2010). Thus, unawareness at the acute stage is linked to longer hospital stays (Maeshima et al., 1997), less likelihood of independent living (Pedersen, Jorgensen, Nakayama, Raaschou & Olsen, 1996), lower scores on measures of functional recovery (Gialanella et al., 2005; Maeshima et al., 1997) and activities of daily living (ADL) (Maeshima et al., 1997). In fact, the impact of unawareness on ADL and functional outcomes is significant even when controlling for the extent of other cognitive deficits (Hartman-Maier et al., 2001). Thus, motor unawareness in acute stages is a specific, negative prognostic sign, compromising the course of recovery and rehabilitation and rendering the reintegration of these patients labour-intensive and costly. Moreover, it has been suggested that approximately 30% of AHP patients remain unaware of their motor deficits beyond the acute (<4 months) stage (Pia, Nepi-M'odona, Ricci & Berti, 2004; Orfei et al., 2007). Thus, the rehabilitation of AHP in the acute stage can be of long-term therapeutic significance.

Recently some progress has been made in the management and rehabilitation of AHP (Kortte & Hillis, 2011; see Prigatano & Morrene-Stupinsky, 2010 and Jenkinson, Preston & Ellis, 2011, for review). Nevertheless, to date no evidence-based treatment exists. Remission of AHP has been long reported using vestibular stimulation but unfortunately the effects of the stimulation are only temporary (Rubens, 1985; Cappa, Sterzi, Vallar & Bisiach, 1987). Transient improvement of awareness and neglect has also more recently been noted using a combination of treatments (Beschin, Cocchini, Allen & Della Sala, 2012). Beschin and colleagues investigated the effect of three treatment techniques that have traditionally been shown to temporarily improve neglect (optokinetic stimulation, prism adaptation and transcutaneous electric nerve stimulation) in five patients with both anosognosia and neglect. Both left and right hemisphere damaged patients were included and were recruited 50 to 70 days post onset. The results indicated that patients responded differently to the same treatment, with anosognosia and not neglect temporarily improving with one patient, and only neglect improving with two other patients. Furthermore, in an extensive review of rehabilitation efforts in AHP, Prigatano and Morrene-Stupinsky (2010) outlined some practical guidelines for the management of unaware patients. Firstly, the severity, 'types' of AHP and the associated neurological and neuropsychological deficits should be clearly determined. Good rapport with both the patient and the family should then be established. Lastly, they suggest that a detailed and individualized rehabilitation plan should be developed (also see Jenkinson et al., 2011).

Aside from these more general intervention programmes, the first, specific, psychophysical intervention able to lead to a lasting remission of AHP was reported in a recent single case study (Fotopoulou, Rudd, Holmes, & Kopelman, 2009). Fotopoulou and colleagues used video replay to provide an AHP patient with visual feedback of her paralysis from a 'third-person perspective' (from the outside) and 'off-line' (at a time different than the one in which she initiated the movement). Patients with AHP, including the patient described in the Fotopoulou and colleagues' paper, typically remain anosognosic when their paralysed arm is brought to their ipsilateral visual field and their paralysis is demonstrated. By contrast, by providing a video-based feedback the authors noted a dramatic increase of motor awareness in this patient. Crucially, the effect was recorded immediately after the intervention and lasted at one-month follow-up.

As some patients with AHP may show greater awareness in 3rd than 1st person perspective tasks (Marcel et al., 2004), the authors hypothesised that the 3rd-person perspective video-viewing may have facilitated the updating of the patient's motor awareness. In addition, given the off-line nature of video replays, the fact that the patients motor intentions were not relevant at the time of video observation, may have facilitated awareness according to motor monitoring explanations (Berti et al., 2005; Fotopoulou et al., 2008). The aim of the current study was not to investigate these potential explanations of the effect but rather to examine the feasibility, effectiveness and optimisation of this video intervention protocol in further patients of different demographic, medical characteristics, and at different stages post-onset.

Thus, two variations of the original video-based methodology were developed and applied to the rehabilitation of two patients with severe AHP. Specifically, in the first case (an elderly woman at the acute stage of recovery following stroke), a video replay and emotional support protocol was used in multiple intervention sessions targeting unawareness of both upper and lower limb hemiplegia and their functional consequences. On the basis of the aforementioned case report (Fotopoulou et al., 2009), the video intervention initially targeted only the LUL and it was expected that viewing oneself unable to move one's arm would have led to a more generalised awareness of paralysis for both her left arm and leg. However, unlike the 2009 case study, the awareness recovery did not generalise to increased awareness for left leg movements. Therefore, both the LUL and the LLL were targeted in separate, successive sessions, covering a period of 58 days.

In the second case, a younger man at the chronic stage of recovery following stroke participated to a single session of the video intervention, which in this case included two different conditions, a self-referent and an other-referent condition, both including questions that targeted awareness about both upper and lower limbs. The 'self-referent' video clip showed the patient himself in a video replay, while the 'other-referent' video clip showed a video of another hemiplegic patient, age and gender matched, but without anosognosia. Both self-and-other videos were shown on the same day, 89 days post stroke, and the effects of this single session video intervention were monitored at different time intervals, with specialised awareness interviews targeting both upper and lower limb motor awareness. The two case studies are presented in turn below.

GENERAL METHODS

The two video interventions were an extension of the original aforementioned methodology (Fotopoulou et al., 2009), but they were conducted independently of each other by two separate research groups, one in the United Kingdom and one in Italy. The results of the two independent case studies were later communicated between the two groups and are presented successively in this paper and discussed together. Both the patients presented with severe AHP (see below for formal assessments) and were recruited as part of wider studies on AHP conducted independently by the two teams. Their common, inclusion criteria were: (1) right fronto-parietal lesion, as detected by CT investigations and confirmed by neuropsychological assessments (e.g. presence of neglect); (2) contralesional paralysis; (3) no known previous brain damage, psychiatric or neurological problems; and (4) clinical indications of anosognosia for motor impairments. Difference in inclusion criteria being: the UK group recruiting acute and sub-acute patients (less than 4 months since stroke), and the Italian group recruiting both acute and chronic (greater than 4 months) patients. Informed written consent was obtained and the study was approved by the two local ethics committees in Italy (CEP, Verona) and in U.K. (local NHS ethical committee), respectively, and carried out in accordance with the guidelines of the Declaration of Helsinki. The general method of the video intervention protocol used in both the case studies included five specific research phases (for a schematic representation see Figure 1). We aimed to extend the original Fotopoulou et al. (2009) approach, by examining the feasibility, optimisation and effectiveness of video-replay by: (1) applying this rehabilitation approach to two patients, in the sub-acute and chronic phases respectively, showing that the improvement was specifically due to the intervention; (2) introducing two new elements that are experimentally investigated, firstly the possibility to have specific and separate interventions for unawareness of upper and lower limb motor weakness; and secondly, the visual comparison with another hemiplegic patient as a possible instrument to enhance awareness of their motor weakness.

[Please insert Figure 1 here]

CASE STUDY 1

ED was an 88 year-old right-handed woman, a retired factory worker, with 10 years of education. She was previously mobile and independent, but with a medical history of ischemic heart disease and hypertension. ED was found collapsed with severe left-sided weakness and admitted to hospital. Radiology reports confirmed a dense right embolic middle cerebral artery (MCA) stroke. Figure 2 (ED) illustrates the lesion of the patient as documented in the clinical CT scan. The patient's lesion mainly involved the territory of the right middle cerebral artery, in particular the parietal and temporal cortex, insula cortex, and right precentral and rolandic operculum. The damage also extended to white matter tracts particularly the superior longitudinal fasciculus.

[Please insert Figure 2 here]

On admission, neurological examination confirmed left-sided hemiplegia (0/5 power in left limbs on Medical Research Council-MRC-Scale) and tactile loss. She also presented with dysarthria, left hemispatial neglect and hemianopsia. On initial assessment, 7 days following her stroke, ED was oriented to person, place and year, with some confusion as to the exact month. She was initially very fatigued but still cooperative. Mild decline in working memory was tested by her digit span. Formal neuropsychological assessment in the second week following onset (see Table 1 for full details; scores below the cut-off are in bold) revealed impairment in executive functions (lack of fluency & concrete thinking), but showed only small motor perseveration and mild disinhibition. She also showed personal and extrapersonal neglect. Verbal and visual recall was poor to average, but consistent with age-appropriate memory decline. ED scored in normal range for depression and anxiety (Hospital Depression and Anxiety Scale).

[Please insert Table 1 here]

Overview of video intervention and timeline

The patient participated in a standard rehabilitation program of an acute stroke rehabilitation ward that mainly targeted the patient's mobility, daily living, housing and occupational needs. Video awareness intervention was the only neuropsychological intervention the patient received. The video intervention schedule was determined by the patient's hospital schedule, medical condition, emotional state and her willingness to participate in each session. Two neuropsychologists, either together or independently, conducted the neuropsychological and awareness assessments (see below), as well as administered the video intervention. Following the same, basic video-viewing procedures as in the original study (Fotopoulou et al., 2009), we conducted two video intervention sessions for awareness of upper limb paralysis (see below for details) in sessions scheduled 15 and 20 days post stroke. The awareness improvement about the upper limb did not generalised to awareness about the left leg paralysis. Thus, motor unawareness for the left leg paralysis was subsequently specifically targeted with a modified protocol (see below for details) in sessions scheduled 41, 42 and 57 days post stroke. Monitoring of AHP throughout the intervention period was achieved by stand-alone, awareness assessments (see below for details) carried out in sessions scheduled before, in-between and following the video intervention sessions at regular intervals.

Awareness assessment (Phase I, Figure 1)

Measures. AHP was assessed by two widely used measures: (a) an awareness interview asking a number of general questions, (e.g. 'Can you move your left arm?') and several confrontational questions (e.g. 'Please, touch my hand with your left hand. Have you done it?') (Berti, Ladavas & Della Corte, 1996). The interview is scored on a 2-point scale (2-points for denial of motor impairment and failure to reach the examiners hand; 1- point for denial of motor impairment, but admits to failure to reach examiner hand; and 0 - points for no acknowledgment of motor deficits); and (b) a second awareness interview containing a total of 10 questions asking both general questions, (e.g. 'Is your left arm causing you any trouble?') and confrontational questions (e.g. 'Please, try and move your left leg for me. Did you moved it?') (Feinberg, Roane & Ali, 2000). Each question in this interview is scored as 0 = completely aware, 0.5 = partially unaware, and 1 = complete unawareness. Thus, the overall score on the interview can range from zero to ten (0= complete awareness, 10= complete unawareness of motor paralysis). Feinberg et al.'s interview was also used as the pre-and-post measure during each video intervention session, as explained below. As the original Feinberg et al.'s interview addresses only the upper limb hemiplegia, a lower limb version was created as an additional measure for the second part of video intervention protocol (see below) by substituting 'left arm' for 'left leg' as needed, e.g. "Is your leg causing you any trouble?", "Please try and move your left leg for me. Did you move it?". As there are no norms for the Feinberg et al.'s interview, we used comparison data from a previous group study on AHP. Fotopoulou, Pernigo, Maeda, Rudd and Kopelman (2010) tested a group of 7 patients with right-hemisphere damage, complete left sided hemiplegia and AHP (AHP group), and a control group of 7 patients with right-hemisphere damage, complete left side hemiplegia, but without AHP (HP group). The mean score of the AHP group was 5.64 (SD=1.38) and of the HP group 0.98 (SD=0.84).

Initial Assessment Results. On initial assessment, 7 days following the stroke, ED showed complete unawareness of her deficit, scoring 2 (maximum score) on the Berti et al.'s interview both for her left upper limb (LUL) and left lower limb (LLL) paralysis. In addition, she had a score of 7/10 on the original, upper limb Feinberg et al.'s interview. ED showed intractable false beliefs about her ability to move but did not produce any florid confabulations or imaginary excuses when asked if she could move her limbs. Instead, she generally remained silent or avoided direct questioning during confrontational questions. For example, when asked if she could touch the examiners right hand with her left hand, she stated: "I can move", but failed to execute the movement. When the examiner asked "Did you move?", ED remained silent, then replied "it is weak, but I can move it". When asked about her LLL weakness she was adamant that "she has no problems" walking. ED consistently reported that she "feels alright", but she acknowledged medical opinion, saying "The doctors tell me I have had a stroke, they must be right. I am not so sure, but the doctors are the experts, so I must have had a stroke", and consistently reported that she "feels alright". Neurological reports, the physiotherapist and the nurse responsible for her care additionally confirmed the patients "lack of insight".

Materials (Phase II, Figure1)

On days 13 and 14 post stroke, ED consented to the Berti et al.'s interview to be repeated and filmed by the bedside on a portable digital video camera (Sony Handycam, DCR-SR57). We subsequently edited the film content to create a 120 sec video clip showing the examiner standing on the left side of the screen, and the patient lying on the bed on the right side of the screen (her left side was on the right), with her torso being at a distance of approximately 1.5m from the camera. There was approximately 1m distance between patient and examiner. The patient was seen in front view with her upper body visible, including shoulders, arms, head and face. The edited clip contained awareness questions taken from the Berti et al.'s interview, including: a general question (e.g. "Why are you here?"), and two specific questions about the patient's upper limbs (e.g. "Can you move your left arm?") and two direct confrontations (e.g. "Please try reaching my hand with you left hand. Have you done it?"). The same edited video clip was used for both upper and lower limb video intervention, with an additional 50 secs added to the end of the clip when targeting left leg unawareness (i.e. total 170 sec clip). This later extra 50 sec clip was created by editing a filmed clip recorded on day 23 post stroke. This footage showed a direct view of ED sitting on her wheelchair, with her face, torso, upper and in this case also lower limbs fully visible. The same questions from the Berti et al.'s interview were used, this time referring to the left leg. Questions also relating to activities of daily living (ADL's) (e.g. "Can you walk on our own/without help?" or "Can you get out of bed without help?" or "Can you go to the shops without help?") were also asked (Marcel et al., 2004).

Video intervention procedures (Phase III, Figure 1)

Pre-video viewing procedures. In each of the video intervention sessions the Feinberg et al.'s interview was first administered to ED as the pre-intervention awareness measure, including also the above mentioned questions relating to ADL's (Marcel et al., 2004). In the lower limb session the modified Feinberg et al.'s interview was used.

Subsequently, a laptop computer (screen size 13) was placed on a hospital table directly in front of the patient, 50 cm from her and, to exclude possible effects of neglect deficits, 20 cm right from the centre of her visual fields. In the first video intervention session, the patient was first shown the paused frame of the above-described video clip and was asked whether she was willing to view a video of herself trying to perform an action and discuss it with the examiner. She was informed of the procedure, the possibly upsetting content of the video and was given the opportunity to ask questions, discuss any aspects of the video intervention and choose to continue. In subsequent video interventions, in addition to these procedures, the patient was reminded of the previous assessments and video replays, and again asked whether she wanted to ask anything and whether she wanted to watch the video clip again. Although as in the original study (Fotopoulou, 2009) the patient and the examiner had already established good relationship during previous assessments, it was considered important to ensure in each session that the patient felt emotionally safe and comfortable in undergoing the assessment with the examiner (also see Fotopoulou, 2008). In addition, in each video intervention session, ED was first asked to describe what she saw in the paused frame and to answer questions regarding the recognition of the identity of her own image, body-parts discrimination and location and left-right distinction.

Video-viewing procedures. The examiner made sure the patient's attention was drawn to the video and the aforementioned120s edited video clip was played back. If the patient was distracted during video observation, the video clip was paused and her attention was redirected to the video. Halfway though the video, the clip was paused and the patient was asked the following two questions: "Can you see yourself trying to move in the video?" and "Did you move?". The same procedure was followed for left lower limb paralysis (day 41, 42 and 57), with the exception of the additional 50sec clip as described above.

Post-video Viewing Procedures. In each session, immediately following the video clip viewing, ED was asked the following set of questions: "Did you see yourself in the interview?", "What did you see?", and was given the opportunity to discuss her observations and feelings with the examiner. Subsequently, the Feinberg et al.'s interview was administered as the post-intervention awareness measure, including also the aforementioned questions relating to ADL's (Marcel et al., 2004). In the lower limb session the modified Feinberg et al.'s interview was used.

Follow-up Assessment (Phase IV Figure 1)

One month after the last awareness intervention, the Berti et al.'s interview and the Feinberg et al.'s interview (see above for normative data) were administered.

Results

Left Upper Limb Video Intervention. Prior to any video intervention sessions ED's anosognosia was severe and stable. In fact, in the four initial pre-intervention assessments (including the pre-intervention assessment of the first video session), her scores were one standard deviation (SD) or more above the mean of the aforementioned, comparison AHP group (Fotopoulou et al., 2010). Following the first video intervention, her awareness for her left arm paralysis improved (Feinberg et al.'s interview scores decreased from 8/10 to 4.5/10, see Figure 3). For the first time from her stroke, the patient commented that "it is hard to move, and sometimes it is upsetting". ED acknowledged her motor weakness, agreeing with the examiner that she had a stroke, and explained that she was fearful of losing the motor ability in her arms. ED appeared upset at this point and when the examiner asked why she was upset, ED responded: "You need your arms, don't you?". ED then started to avoid further questions, making various excuses about everyday tasks she needed to complete. The examiner thus discontinued questioning and instead provided emotional support and eventually used distraction by discussion of positive topics from ED's lifetime.

[Please insert Figure 3 near here]

Five days later, ED's anosognosia had returned to pre-intervention levels with her scores falling within one SD above the comparison AHP patients' mean scores (Fotopoulou et al., 2010). Nevertheless, following the video intervention, ED showed a dramatic reinstatement of motor awareness on the Feinberg et al.'s interview (score 1/10) and began to cry at the end of it. The examiner inquired as to why she was crying, ED replied “I feel sad because I can’t move it”. The examiner then asked: “What did you see in the video?”, ED: “That I can’t move my arm”. Examiner: “What else did you see?”, ED: “That I couldn’t move my arm, I feel sad because I couldn’t move it”. The patient went on to explain: “I can’t move on my own. I wish I could, but I can’t.” Emotional support was then provided to ED, including initially understanding her negative emotions and then reflecting on some of the positive possibilities her increased insight may allow in her general rehabilitation.

The following day an increase in her anosognosia scores was noted again (5/10 on the Feinberg at al.’s interview), but this score fell within one SD below the mean of the comparison AHP group (Fotopoulou et al., 2010). Moreover, in an informal conversation ED remembered the previous day’s session and was able to describe her memories of gaining insight and having related emotions.

Left Lower Limb video intervention. Eighteen days later (41 days after her stroke) ED was less fatigued and more alert. Her anosognosia for upper limb paralysis had recovered (Feinberg et al.’s interview score 0/10). However, her anosognosia for lower limb hemiplegia seemed unaffected by both the video interventions and spontaneous recovery, scoring consistently more than one SD above the mean of the AHP comparison group (Fotopoulou et al., 2010) (see Figure 3). Moreover, she claimed that “even with the stroke, I can still walk”. Accordingly, she rated herself as fully able in questions about bipedal tasks, especially walking (Marcel et al., 2004). Interestingly, upon confrontation ED did not present with illusionary movements (Feinberg et al., 2000; Fotopoulou et al., 2008) and attributed her failures (e.g. not being able to walk at the moment) to third parties: “I can get out of this bed, but they put bars on to stop me" or "Yes I can walk, if they let me go out, I could walk" or "There is nothing wrong with my body, it's the chair". ED was rather resistant and avoidant to questions relating to her LLL paralysis, frequently making excuses, apparently in order to stop related conversations.

Immediately following the first LLL video intervention at 41days post-onset, ED's anosognosia score on the modified Feinberg et al.'s interview decreased to 4/10. Towards the end of the assessment, when asked if she could get out of bed without help, ED replied "No I can't, yet I think I can". At this point, she appeared distressed and appealed to the examiner, remarking that if it was the examiner who could not walk, she would feel sadness for the examiner ("It would make me feel sad if you couldn't walk, couldn't go anywhere on your own"). The discussion continued in this emotional tone from both parties ensuring the patient's distress had been understood and contained, and the formal assessment was concluded.

The next day her scores concerning left lower limb paralysis awareness increased to 6/10 (modified Feinberg et al.'s interview). However, immediately following a second video intervention her scores declined again (4/10, corresponding to more than one SD below the mean of the AHP comparison group, Fotopoulou et al., 2010). Immediately after the interview the patient began to cry and asked the examiner: "If I can't get out of this chair, what am I going to do?". Again, the above described emotional support protocol was followed.

On the next follow-up assessment, the patient remembered the previous sessions but became increasingly distressed during recall. Accordingly, emotional support was offered to the patient, any sort of further confrontation was avoided and a two-week interval was left before the next video intervention. During this interval the patient's emotional state was monitored and supported in brief, informal visits.

Two weeks later, the third and last video intervention was administered. The patient did not show any evidence of anosognosia for upper limb paralysis, but her scores on the modified Feinberg et al.'s interview for lower limb paralysis was 5/10 (within one SD below the mean of the AHP comparison group, Fotopoulou et al., 2010). ED commented: "I can stand, but I can't get out of this chair and I don't know why". Following the video intervention, the interview showed a dramatic improvement in awareness (1/10 the modified Feinberg et al.'s interview). When the examiner asked: "Can you walk on your own?" ED remained silent for a while and hesitantly replied "Maybe a little, but not without a walking stick". The patient was mildly agitated but not tearful as in previous sessions. When asked, "Can you move your left leg?" ED replied, "I don't think so". The examiner then asked, "Can you walk?", ED said she was not able to and became upset. The above emotional support protocol was administered and the patient and examiner discussed the possibilities and opportunities for future, practical support with everyday activities.

At another assessment the next day, 58 days post stoke, ED showed no evidence of anosognosia for both left upper and lower limb paralysis (0/10 LUL and 1/10 LLL, respectively). Furthermore, ED remembered the video interventions and was able to reflect on her general disabilities and the related negative emotions. The same results were noted in a final, follow-up assessment 82 days post stroke. These findings were further confirmed by administering the Berti et al.'s interview, in which the patient showed no sign of AHP (score =0/2).

CASE STUDY 2

FG was a 70 year-old right-handed man, a retired builder with 5 years of education. He suffered a large haemorrhagic stroke in the region of the right middle cerebral artery. Figure 2 (bottom part-FG) illustrates the lesion of the patient as presented in the clinical CT scan. Lesioned areas mainly involved the basal ganglia structures and subcortical white matter (internal capsule, anterior, superior and posterior corona radiate, external capsule, superior longitudinal fasciculum and fronto-occipital fasciculum). Parts of the temporal, insular and frontal cortices were also damaged.

[Please insert Table 2 near here]

On initial examination, 74 days following the stroke, FG presented with complete left-sided hemiplegia (0/5 power in left upper and lower limb on MRC scale) and left hemispatial extrapersonal and personal neglect (see Table 2; scores below the cut-off are in bold). The patient was alert and oriented to person and place, but showed some confusion in time. Neuropsychological assessment further revealed important deficits in executive functions (Frontal Assessment Battery, FAB), but no impairments in verbal memory (i.e. story recall), language or general cognitive abilities (Mini-Mental State Examination, MMSE). There was no evidence of depression or anxiety in the Becks Depression Inventory, STAI and Affective Story Recall.

FG presented with severe AHP, reporting that he had no motor deficits in his left arm and leg. Although FG did confirm that he was in the hospital for rehabilitation, he was adamant that he could move his left arm and leg with "no problems". His only complaint was of pain in his back, neck and shoulders. He claimed to be in the hospital for further assessment of his back-pain, and maintained that the doctor had decided he should be hospitalised for rehabilitation training. When asked why he was sitting in a wheelchair, he responded that it was to help the nurses taking him to medical visits.

Overview of intervention and timeline

The patient participated in a tailored rehabilitation program in an in-patient rehabilitation Department. That program focused on his motor and neglect deficits. The video intervention, which was conducted by two neuropsychologists, was the first neuropsychological intervention the patient received for his AHP. In this patient, only one session of video intervention took place on day 89 post stroke. However, this was a "double" video session, in the sense that the viewing of a self-referent video clip as the one described in the previous case study, was followed by the viewing of a similar clip referring to another hemiplegic patient (see methods for details). Moreover, a specific "experimental" awareness task (see below) was applied as a pre-and-post assessment measure on the day of the intervention. In addition, the same measure was applied on two other dates prior to the intervention day (72 and 82 post onset) and three other dates post intervention (90, 103 and 113 post stroke). In all post intervention assessments this experimental awareness task was administered twice, one with reference to the *"self-clip"* and one with reference to the *"other-clip"*. The intervention and assessment dates were determined by the patient's hospital schedule, medical condition, emotional state and willingness to participate in each session.

Clinical awareness assessment (Phase I, Figure1)

FG's AHP was measured using three different methods (see Table 2): (1) the 4-point "Bisiach" interview (Bisiach, Vallar, Perani, Papagno & Berti, 1986); (2) the Visual-Analogue Test for Anosognosia for motor impairment (VATA-m, Della Sala, Cocchini, Beschin & Cameron, 2009); (3) the modified "Marcel" structured interview (Marcel et al., 2004; Moro et al., 2011). The Bisiach et al. interview uses a 4-point scale to evaluate the severity of the patient's unawareness identified as mild, moderate or severe (0= aware, 1= mild unawareness, 2= moderate unawareness, 3= severe unawareness). The VATA-m is a measure of unawareness of motor deficits combining both the patient's and caregiver's evaluation of the patient's motor abilities in a series of specific motor tasks (e.g. walking or drinking from a glass). A discrepancy score is then calculated with a maximum score of 36 (scores between 6.8 and 12.0 indicate mild anosognosia, scores between 12.1 and 24 indicate moderate unawareness, and scores between 24.1 and 36 indicate severe unawareness). In addition, a modified version of the Marcel structured interview (Marcel-Moro's interview, Moro et al., 2011) was used. The interview consists of 23 questions (each of them scored as 0 = aware and 1 = unaware). The questions address four different aspects of awareness, namely: (i) general awareness of illness; (ii) awareness of sensory-motor abilities for upper limb; (iii) awareness of sensory-motor abilities for lower limb; and (iv) awareness of abilities in ADL's.

In FG, no acknowledgement of the disorder could be obtained even after clear demonstrations of paralysis (scoring 3 on the Bisiach et al. interview). He also scored below the cut-off in the VATA-m, scoring 22 for bimanual and 10 for bipedal actions (both corresponding to severe AHP). During the modified Marcel-Moro's interview, FG rejected any evidence of his motor deficits and again claimed complete autonomy in his ADL's, scoring 20/23. In one instance during assessments, FG did admit to a certain degree of left upper limb weakness, but still continued to claim that he was "not paralysed".

Materials (Phase II, Figure1)

On day 82 post stroke, FG consented to his awareness testing to be filmed using a portable digital video camera (JVC GR-DVL 150 E). The patient was recorded while being seated, with the examiner standing on his left side. There was approximately 1m distance between the patient and the examiner. A 120 sec video clip was subsequently edited from this filming session. The video clip showed the patient in front view, with FG's face and body, and both right and left upper and lower limb visible in the video. The edited clip contained general questions (e.g. "Why are you here?"), specific questions about the patient's limbs ("Can you move your left arm?"), and direct confrontation questions (e.g. "Please try reaching my hand with your left hand? Have you done it?") (as described in Fotopoulou et al., 2009). In the edited clip, when the patient was asked: "Can you move your left arm?", he replied that he was able to do so and then spontaneously moved only his right arm. At this point, the patient was asked to use his left hand to reach the examiner's hand. The patient claimed that he had performed this action and that he was confident in his reply. When asked to indicate which one was his left arm the patient used his right arm to reach across and indicate his left arm, suggesting he was not suffering from left-right confusion.

An identical interview to the one described above was conducted with another hemiplegic patient, matched for age and gender, who was however fully aware of his paralysis. This interview was recorded and edited resulting in a video clip as similar as possible in content and length to FG's edited clip described above. Of course, the patient's answers to the same questions were different, in the sense that he was aware of his paralysis. For example, he reported to be at hospital because of his paralysis; he agreed that he was not able to move his left arm and leg, and, after the confrontation task, he recognised that he did not successfully reach the examiner hand.

Video Intervention Procedures (Phase III, Figure 1)

Pre-video Viewing Procedures. On two sessions prior to the day of the video intervention session, as well as on that day (89 days post stroke), an experimental measure of anosognosia, the Judgment of Actions Test (JAT, Moro et al., 2011), was used as a pre- and post- intervention awareness measure (see also below). The JAT tests the awareness of motor deficits in 23 separate and specific actions. The patient is asked to judge his or another person's ability to execute a series of unimanual, bimanual and bipedal actions (e.g. kicking a ball, climbing upstairs, picking up a glass). There are 23 questions, 10 of which refer to unimanual actions (U, 5 for each hand), 8 to bimanual (BM) and 5 to bipedal (BP) actions (Moro et al., 2011). Right unimanual actions are considered as control questions for subject's comprehension and consistency. The patient is asked to judge his or another persons' motor abilities on each action, using an 11-point scale (from 0 = "I cannot do this action at all" to 10 = "I am completely able to do this action").

Video Viewing Procedures of Self Clip.

Following the pre-intervention JAT task, a laptop computer (screen size 13') was placed on a table directly in front of the patient, 50 cm from him and 20 cm right from the centre of his visual fields, to minimize possible neglect confounds. The patient was informed of the procedure, and given the opportunity to ask any questions. As in the case above and the original study (Fotopoulou, 2009) there was a good therapeutic relationship already established between the patient and the examiner.

FG was first shown the video in pause mode and asked to identify himself in the video and discriminate between left and right body-parts. The examiner confirmed that the patient was comfortable to continue and watch the video replay.

The clip was played back to the patient. He initially avoided directly looking at the video and the examiner gently guided his attention to the video clip. After the first 10 seconds the patient's attention focused on the video, although he occasionally and spontaneously shifted his attention from the video to the direct view of his left hand and back.

During playback, at the point in the video clip after the patient had answered the question "Why are you here?", the video clip was paused and the examiner asked, "Do you agree?", referring to his response in the video which acknowledged only his back and neck pains and did not report any motor difficulties. FG responded: "So, so, my arm and leg are not functioning very well, it is very bad actually". The examiner then asked, "What is?", FG: "I have to admit, the weakness is very bad". Examiner: "Can you walk?", FG: "No, I cannot". At this point the video clip was restarted. While watching the confrontation task in the video (i.e. the patient is asked to move his left arm), the patient spontaneously remarked, "I thought that I would have recovered better". The video was paused again and the examiner then asked: "How do you see yourself recovering now?", FG: "I have not recovered very much". After the end of the video playback the examiner asked: "Can you walk?", FG answering, "Not so good". Examiner: "Why?", FG: "My left leg is weak". The examiner then asked: "And can you lift it?", FG replying: "Yes, I can, but in the video the leg did not move." The JAT was subsequently administered.

Viewing Procedures of Other Video Clip. Thirty minutes after this self-referent video intervention, the video referring to the other hemiplegic patient was played back in the same manner as the self-referent video intervention. FG correctly identified that the person in the video was a hemiplegic patient ("The man in the video can not move"). During the confrontation task the video was paused and the examiner asked FG if the hemiplegic patient had managed to reach the examiners hand. He answered: "No, it is impossible." FG made no other comments during the rest of the video. The JAT (referred to the other patient) was again administered directly following the end of the video as the post-intervention awareness measure. It was repeated in the self and other referent conditions the day after (90 days in Figure 5).

Follow-up Assessment (Phase III, Figure I)

A follow-up assessment by means of the Vata-m and Marcel-Moro interview was conducted 14 days after the awareness intervention session (see Table 2). The JAT was also administered 24 days after the video session to monitor the patient's awareness (see Figure 4).

Statistical analyses

Statistical analysis was performed on the pre- and post-intervention JAT scores. Left unimanual (UM), bimanual (BM) and bipedal (BP) actions were analysed separately. Unimanual scores referring to the right hand were excluded from the analysis as the patient performed at ceiling, confirming intact attention and comprehension during these control trials. The self-referent and other-referent scores were also analysed separately, apart from one final self vs. other referent comparison applied to the post-video assessments only (there were no other-referent pre-intervention assessments). The Friedman test was used to analyse any significant differences between the JAT three baseline assessments. Since there was no difference in these scores, the average of the three baseline scores was calculated and used as a global baseline measure.

For the *self video condition*, we compared the mean baseline assessment score and the post-intervention scores. For the *other video* condition, we compared only the post-awareness assessment scores. Lastly, for the self-other video comparison we analysed the results from all JAT evaluations, excluding the baseline measures. The Friedman rank sum test was used to verify if the observed improvement in awareness was statistically significant. If this test reached a statistical significance, the Wilcoxon tests for dependent data, with False Discovery Rate correction as post-hoc test, were used.

Finally, to check if there was a stepper trend in the recovery of awareness in the self- or in the other-referred condition, we computed a simple ratio index as qualitative index. We fitted regression linear models on JAT values for right UM, left UM, BM and BP for other- and self-referent conditions on all the post-video evaluations. Then, for each typology of score, we calculated the ratio between the slope of the self-referred linear model and the slope of the other-referred linear model (self angular coefficient/other angular coefficient). If this ratio index is more than 1, it means that the self-referent awareness has a better improvement, while a ratio index lower than 1 indicates that the other-referent awareness has a better improvement. Although this index is not thought to have a strong statistical validity (mainly because of the small amount of observations), it may be useful to show the differences in the trends of responses, which could be important for discussions on clinical outcomes.

Results

Self-referred condition. Prior to any video intervention FG's anosognosia was severe and stable. As shown in Figure 4 no change in awareness was observed in the assessments prior to the video replay ($\chi^2_{(2)} = 3.07$, $p = 0.215$). All three pre-awareness assessments demonstrated severe anosognosia for left unimanual, bimanual and bipedal actions (see Figure 4). Following the video intervention, FG spontaneously remarked: "We have to work hard". When the examiner asked to what he was referring to, the patient explained that he needed to work hard on his rehabilitation, agreeing that he was not well and he was unable to move. The JAT scores immediately after the video intervention show an increase in awareness for motor deficits of unimanual left and bimanual actions (see Figure 4). Crucially, in the following assessments FG's trend of improvement seemed to continue particularly in LU and BM actions (see Figure 4).

This gradual improvement was significant in BM actions ($\chi^2_{(4)} = 13.01$, $p = 0.011$) and in LU actions ($\chi^2_{(4)} = 10.91$, $p = 0.028$), and showed a trend of significance in BP actions ($\chi^2_{(4)} = 8.74$, $p = 0.068$). Furthermore, the comparison between baseline and individual sessions shows that this gradual improvement becomes statistically significant in BM actions only in the follow-up sessions (post-hoc tests: baseline vs. 103 days: W = 36, $p = 0.028$; baseline vs. 113 days: W = 33, $p = 0.042$).

[Please insert Figure 4 near here]

Patient's improvement is additionally confirmed by clinical awareness assessment carried out 14 days after video replay, as shown by VATA-m (Della Sala et al., 2009) and Marcel-Moro's interview scores (103 days after stroke, in Table 2). Specifically bimanual VATA-m scores indicate that FG's anosognosia improved from severe to moderate following the intervention. The Marcel-Moro's interview also indicated an improvement of anosognosia for lower limb paralysis, but this improvement was only partially confirmed by the VATA-m. Results of the neuropsychological assessment demonstrated that the improvement was specific to awareness scores as neither personal, nor extrapersonal neglect, nor executive functioning scores were modified post- intervention (see Table 2).

[Please insert Figure 5 near here]

Comparison self/other referent responses. FG's scores referred to the motor ability of the other patient were significantly lower than the self-referent scores (Figure 5). This indicates greater insight into the deficits observed in the other-referent condition than the self-referent condition. In comparison to self-referent ratings, FG was better able to recognise the deficits of the 'other' hemiplegic patient in all the actions (BM: $\chi^2_{(1)} = 8.17$, $p = 0.004$; BP: $\chi^2_{(1)} = 5.4$, $p = 0.02$; LU: $\chi^2_{(1)} = 5.4$, $p = 0.02$), but not in RU action ($\chi^2_{(1)} = 0.2$, $p = 0.65$). Nevertheless, qualitatively ratio indexes between the slopes indicates better improvement in self-referent condition for left UM (1.375, self slope = -1.1, other slope = -0.8), BM (1.14, self slope = -0.4, other slope = -0.35), BP (1.20, self slope = -0.6, other slope = -0.5) but not right UM (0, self slope = 0, other slope -0.6). Thus, although FG recognised the paralysis in other patient better than in himself, the trend of recovery induced by our video rehabilitation indicates a more specific effect in self-referent condition. For clinical observations, statistical analysis and further results concerning the other-referent condition, see Supplementary Material.

DISCUSSION

We presented two patients with severe anosognosia for hemiplegia following stroke in different post-onset stages (acute versus chronic) and found that self-observation in video replay contributed to the reinstatement of motor awareness in both cases. Both patients showed stable and severe AHP in several assessments prior to the intervention and both patients showed evidence of marked improvement in motor awareness following the intervention. Although the precise intervention protocols applied, as well as the pattern of the recovery observed were different between the two patients, our results suggest that in both cases the awareness improvement cannot be accounted exclusively by spontaneous recovery. This conclusion is more obvious in the case of the chronic patient, who had showed stable and severe AHP for more than two months post-onset. We discuss these results below, in the context of the wider literature on AHP.

AHP typically remits over time as a result of spontaneous recovery, but sometimes patients remain unaware also in the chronic stage (see Pia et al., 2004 for review). Moreover, the presence of AHP in the acute stage may significantly obstruct rehabilitation efforts and consequently impede long-term functional outcomes (Gialanella et al., 2005; Jehkonen et al., 2006). As outlined in the introduction, there have been recent advances in the management of AHP by providing specific rehabilitation guidelines and strategy's (see Prigatano & Morrene-Stupinsky, 2010 and Jenkinson et al., 2011 for reviews), with temporary remission of AHP also being recently reported using a combination of techniques (see Beschin et al., 2012). Unfortunately, despite these rehabilitation efforts, there is currently no known, efficacious intervention for permanent restoration of motor awareness in patients with AHP (Kortte & Hills, 2011; Jenkinson et al., 2011).

One potentially promising intervention has been successfully applied in a single case study; Fotopoulou and colleagues (2009) used a single-session of self-observation in a video-replay and reported the instant and permanent reinstatement of motor awareness in a patient who was previously showing severe AHP. Unfortunately, these observations have not been as yet replicated in other patients and the intervention protocol has not been standardised. The present study aimed to replicate these findings, as well as to explore the characteristics of the intervention protocol and the feasibility of its application in acute and chronic patients with AHP. To this end, the present results provide confirmatory evidence for the restoration potential of video-based, self-observation in AHP but also suggest a number of areas that require modification and further exploration.

Both patients in the current study showed indications of a sudden and unprecedented realisation of their motor impairments immediately following the video (e.g. ED: “I can’t move on my own. I wish I could, but I can’t.”, FG: “We have to work hard!”, referring to his rehabilitation). However, unlikely in the original study (Fotopoulou et al., 2009), their awareness scores on the standardised and experimental measures used post-intervention did not reveal a corresponding, complete and generalisable recovery of awareness as compared with their pre-intervention scores. Instead, the video viewing seemed to be only the first step in a longer and more complex rehabilitation process that required additional sessions (in the case of ED), several follow-up assessments (in the case of FG) and the provision of emotional support following and in-between sessions (both patients). Moreover, the noted improvements in the awareness of upper limb paralysis following observation of the upper limb in the video did not lead to a more generalised awareness of the patients’ disabilities in uni-pedal or bi-pedal tasks. These required the passage of time and several, specific assessment sessions in the case of FG and a video intervention specifically targeting lower limb function in the case of ED. In both cases, unawareness for ADL’s, especially involving crucial lower limb abilities (e.g. walking), proved to be more resistant.

Previous studies have found AHP patients to overestimate their ability more on bipedal than bimanual actions (e.g. rowing a boat or jumping up). This difference may be explained by the fact that acute patients have less opportunities to attempt bipedal than bimanual tasks in the hospital, as well as by the fact that the practical and psychological implications of bipedal movement deficits (especially walking and driving a car), are potentially more catastrophic and thus harder to accept or acknowledge (Marcel et al., 2004). Moreover, since in the current study the effects of the video intervention on upper limb awareness did not prove to automatically generalise to other domains, it is suggested that video intervention should progressively target AHP for different modalities: (1) AHP for upper-limb weakness, (2) followed by lower-limb unawareness, (3) and finally unawareness for ADL's.

Furthermore, it should be noted that video-observation brings about strong negative emotions in patients who otherwise deny their deficits and may appear as indifferent. This observation is consistent with findings from the original study that showed that awareness improvement was not accompanied by any changes in performance on neuropsychological tests of cognitive function but rather with a large increase in depressive feelings as captured by a self-report measure. More generally, the role of negative emotions and 'catastrophic' reactions (Turnbull, Evans & Owen, 2005) needs to be taken into account in future protocols and perhaps more formal and longer sessions of emotional support need to be provided to patients. Whether these awareness-related emotions are a primary manifestation of the disorder itself, or a secondary consequence of other primary causes has long being debated in the literature (see Weinstein, 1955; Bisiach & Geminiani, 1991; Solms, 1995; Turnbull et al., 2005; Fotopoulou, 2013). This study cannot directly address such debates, but it nevertheless suggests that taking patients emotions into account may be an important component of any successful intervention for AHP (see also Prigatano, 2005), as also shown in similar syndromes such as confabulation following acute frontal damage (see Fotopoulou, 2008) and unawareness in generalised and neurodegenerative disorders such as dementia (Ownsworth, Clare & Morris, 2006).

In the case of FG, the same day as the video intervention, we showed him an other-referent video and asked him to comment on the "other" patient's disability. FG showed greater awareness for motor disorders in this other-referent condition than the self-referent condition. In comparison, Ramachandran and Rogers-Ramachandran (1996) reported three cases of anosognosic patients, two of whom extended their denial of paralysis to another "stooge" student pretending to have left hemiplegia. Similarly to FG however, the third patient immediately recognised the motor deficits of the "stooge" patient. Although our results did show a greater trend of recovery over time for the self-referent condition, the fact that he was more aware in the other-referent condition to begin with, suggest that he had less of a margin for improvement. Interestingly, "self" and "other" video replays have been recently used as an effective treatment in increasing insight in schizophrenic patients (David, Stre & Zavareri, 2012).

Finally, before firm conclusions can be drawn, it should be noted that our study has a number of limitations. Firstly, this study was limited to two single case studies. Furthermore, different intervention protocols were used in each case. The number of video observation sessions was different in ED and FG's intervention, and the results of ED's video intervention were restricted by a lack of statistical analysis. Nevertheless the basic methodology was the same for both patients, permitting us to discuss them together. The results of this study need to be replicated in a carefully controlled group study, using a standardised protocol and also comparing the results with a control group.

However, it should be noted that it was not the intention of this study to suggest a generally applicable intervention for the rehabilitation of AHP, which may indeed differ from patient to patient and hospital to hospital. Our aim was to test the feasibility of the method in two further patients, test its potential for awareness restoration in these particular patients and examine particular extensions of the original protocol. In clinical practice, the precise protocol for each individualised rehabilitation program is conditioned by personal and institutional variables. For example, it is important to consider that not all patients may be appropriate for video intervention and that it is important to individuate the times when it is possible to suggest it. Moreover, severe spatial neglect that cannot be by-passed by prompting may prevent some patients from seeing left side of the screen when watching the video replay. Severe attention and memory problems may also reduce the effectiveness of the video observation. A preserved capacity for introspection and the ability to discuss one's emotions, may also prove to be important moderators of the intervention's effects. In all cases, our results suggest that in order to establish a good clinical relationship with the patient, it is not advisable to begin the intervention too soon after first meeting the patient. In summary, this study provides further support for the finding that video-based, self-observation can reinstate motor awareness in AHP, by providing 3rd person and off-line feedback. It further highlights that while this simple, psychophysical intervention seems potent, it may not be sufficient for awareness restoration in all patients and it needs to be embedded in a wider and perhaps individualised intervention protocol, involving the targeting and training of specific facets of awareness and the management of related emotions and self-perceptions. This conclusion is consistent with recent theories of AHP that suggest it is a multifaceted syndrome entailing a dynamic interplay between neurological and psychological components (Vocat et al., 2010; Moro et al., 2011; Fotopoulou et al., 2013).

Funding: This work was funded by an European Research Council (ERC) Starting Investigator Award for the project 'The Bodily Self' N°313755 to A.F. and by the Italian Ministry of Education, University and Research (Progetti di Ricerca di Interesse Nazionale PRIN 2009), the Italian Ministry of Health (Project Code: RF-2010-2312912) to V.M., and a Commonwealth Scholarship to S.B.

Acknowledgements: We thank the patients and their relatives for their kindness and willingness to take part in the study. We are also grateful to Cristina Bulgarelli and Michele Scandola for their help in collecting data and statistical analysis. No conflicts of interest were reported.

REFERENCES

Babinski, J. (1914). Contribution e l'etude des troubles mentaux dans rhemiplegie organique cerebrale (anosognosie) [contribution to the study of mental disorders in hemiplegia (anosognosia)]. *Revue Neurologique, 27,* 845-848.

Berti, A., Ladavas, E., & Della Corte, D. (1996). Anosognosia for hemiplegia, neglect, dyslexia, and drawing neglect: Clinical findings and theoretical considerations. *Neuropsychological Society, 2,* 426-440.

Berti, A., Bottini, G., Gandola, M., Pia, L., Smania, N., Stracciiari, A. et al. (2005). Shared cortical anatomy for motor awareness and motor control. *Science, 309,* 488-491.

Beschin, N., Cocchini, G., Allen, R., & Della Sala, S. (2012). Anosognosia and neglect respond differently to the same treatments. *Neuropsychological rehabilitation, 22,* 550-562.

Bisiach, E. & Geminiani, G. (1991). Anosognosia realtes to hemiplegia and hemianopia. In G. P. Prigitano, D. L. Schacter (Eds.), *Awareness of Deficit After Brain Injurt: Clinical and Tehoretical Issues.* New York: Oxford University Press, pp. 17-39.

Bisiach, E., Vallar, G., Perani, D., Papagno, C., & Berti, A. (1986). Unawareness of disease following lesions of the right hemisphere: Anosognosia for hemiplegia and anosognosia for hemianopia. N*europsychologia*, *24*, 471-482.

Cappa, S., Sterzi, R., Vallar, G., & Bisiach, E. (1987). Remission of hemineglect and anosognosia using vestibular stimulation. *Neuropsychologia, 25,* 775-782.

Cherney, R. F. (2006). Ethical issues involving the right hemisphere stroke patient. *Stroke rehabilitation, 13,* 47-53.

Cocchini, G., Beschin, N., Cameron, A., Fotopoulou, A., & Della Sala, S. (2009). Anosognosia for motor impairment following brain damage. *Neuropsychologia, 23,* 38-63.

Cocchini, G., Beschin, N., Fotopoulou, A., & Della Sala, S. (2010). Explicit and implicit anosognosia or upper limb motor impairment, *Neuropsychologia, 48,* 1489-1494.

Cutting, J. (1978). Study of anosognosia. *The journal of Neurology, Neurosurgery and Psychiatry, 41,* 548-555.

David, A. S., Ster, I. & Zavarei, H. (2012). Effect of video self-observation vs. observations of others on insight in psychotic disorder. *The Journal or Nervous and Mental Disease, 200,* 358-361.

Della Sala, S., Cocchini, G., Beschin, N., & Cameron, A. (2009). Vata-m: Visual-analogue test assessing anosognosia for motor impairment. *The Clinical Neuropsychologist*, *23*, 406-427.

Di Legge, S., Fang, J., Saposnik, G. & Hachinski, V. (2005). The impact of lesion side on acute stroke treatment. *Neurology, 65,* 81-86.

Dubois, B., Slachevsky, A., Litvan, I. & Pillon, B. (2000). The FAB-A Frontal Assessment Battery at beside. *Neurology, 55,* 1621–1626.

Feinberg, T. E., Roane, D. M., & Ali, J. (2000). Illusory limb movements in anosognosia for hemiplegia. *Journal of Neurology, Neurosurgery Psychiatry, 69,* 511-513.

Fotopoulou, A., Tsakiris M., Haggard, P., Rudd, A. & Kopelman, M. (2008). The role of motor intention in motor awareness: An experimental study on anosognosia for hemiplegia. *Brain, 131*, 3432-42.

Fotopoulou, A., Rudd, A., Holmes, P., & Kopelman, M. (2009). Self-observation reinstates motor awareness in anosognosia for hemiplegia. *Neuropsychologia*, *47*, 1256-1260.

Fotopoulou, A., Pernigo, S., Maeda, R., Rudd, A., & Kopelman, M. (2010). Implicit awareness in anosognosia for hemeplegia: Unconscious interference without conscious re-representations. *Brain*, *133,* 3564-3577.

Fotopoulou, A. (2012). Illusions and delusions in anosognosia for hemiplegia. *Brain, 135*, 1344-1347.

Fotopoulou, A. (2013). Time to get rid of the ‘modular’ in neuropsychology. *Journal of Neuropsychology*, 1-19.

Frith, C. D., Blakemore, S. J., & Wolpert, D. M. (2000). Abnormalities in the awareness and control of action. *Philosophical Transactions of the Royal Society of London: Biological Sciences, 355,* 1771-1788.

Gialanella, B., Monguzzi, V., Santoro, R., & Rocchi, S. (2005). Functional recovery after hemiplegia in patients with neglect: The rehabilitation role of anosognosia. *Stroke, 36*, 2687-2690.

Garbarini, F., Rabuffetti, M., Piedimonte, A., Pia, L., Ferrarin, M., Frassinetti, F., Gindri, P., Catagalllo, A., Driver, J., & Berti, A. (2012). 'Moving' a paralysed hand. *Brain, 135,* 1486-1497.

Hartman-Maeir, A., Soroker, N. and Katz, N. 2001. Anosognosia for hemiplegia in stroke rehabilitation. *Neurorehabilitation and Neural Repair*, *15,* 213–222.

Hartman-Maeir, A., Soroker, N., Ring, H. and Katz, N. 2002. Awareness of deficits in stroke rehabilitation. *Journal of Rehabilitation Medicine*, *34*, 158–164.

Heilman, K. M. Barrett, A. M. & Adair, J. C. (1998). Possible mechanisms of anosognosia: A deficit in self-awareness. *Philosophical Transactions of the Royal Society of London: Biological Sciences, 353,* 1903-1909.

Heilman, K. M. & Harciarek, M. (2010). Anosognosia and anosodiaphoria of weakness. In G.P. Prigatano GP (Ed.). *The Study of Anosognosia*. New York: Oxford University Press (pp. 89–112).

Jehkonen, M., Laihosalo, M., & Kettunen, J. (2006). Anosognosia after stroke: Assessment, occurrence, subtypes and impact on functional outcome reviewed. *Acta Neurol Scand*, *114,* 293-306.

Jenkinson, P. M., Preston, C., & Ellis, S. (2011). Unawareness after stroke: A review and practical guide to understading, assessing, and managing anosognosia for hemiplegia. *Journal of clinical and Experimental Neuropsychology, 33,* 1079-1093.

Karneth, H., O., Baier, B., & Nagele., T. (2005). Awareness of the functioning of one's own limbs mediated by the insular cortex? *The Journal of Neuroscience, 25*, 7134-7138.

Kortte, K. B. & Hills, A. E. (2011). Recent trends in rehabilitation interventions for visual neglect and anosognosia for hemiplegia following right hemisphere stroke. *Future Neurology*, *6*, 33048.

Levine, D.H., Calvanio, R. & Rinn, W. E. (1991). The pathogenesis of anosognosia for hemiplegia. *Neurology, 41,* 1770-1781.

Maeshima, S., Dohi, N., Funahashi, K., Nakai, K., Itakura, T. and Komai, N. (1997). Rehabilitation of patients with anosognosia for hemiplegia due to intracerebral haemorrhage. *Brain Injury*, *11*, 691–697.

McIntosh, R.D., Brodie, E.E., Beschin, N., & Robertson, I.H. (2000) Improving the clinical diagnosis of personal neglect. *Cortex*, *36*, 289-292.

Marcel, A. J., Tegner, R., & Nimmo-Smith, I. (2004). Anosognosia for plegia: Specificity, extension, partiality and disunity of bodily awareness. *Cortex, 40,* 19-40.

Mattioli, F., Gialanella, B., Stampatori, C. & Scarpazza, C. (2012). General intellectual impairment in chronic right hemisphere damaged patients with anosognosia: a group study. *Neuropsychological rehabilitation*, *22,* 501-515.

Mohamed, S., Fleming, S., Penn, D. L., & Spaulding, W. (1999). Insight in schizophrenia. *Journal of Nervous and Mental Disease, 187*, 525-531.

Moro, V., Pernigo, S., Zapparoli, P., Cordioli, Z., & Aglioti, S. (2011). Phenomenology and neutral correlates of implicit and emergent motor awareness with Patents with anosognosia for hemiplegia. *Behavioural Brain Research*, 225, 259-269.

Nardone I. B., Ward, R., Fotopoulou & Turnbull (2010). Attention and emotion in Anosognosia: evidence of implicit awareness and repression? *Neurocase: The Neural Base of Cognition*, *13,* 5-6.

Nasreddine, Z.S., Phillips, N.A., Bedirian, V., et al. (2005). The Montreal Cognitive Assessment, MoC. *Journal of American Geratric Societey, 53,* 695-699.

Orfei, M. D., Robinson, R. G., Prigatano, G. P., Starkstein, S., Rsch, N., Bria, P., . . . Spalletta, G. (2007). Anosognosia for hemiplegia after stroke is a multifaceted phenomenon: A systematic review of the literature. *Brain*, *130*, 3075-3090.

Ownsworth, T., Clare., L. and Morris, R. (2006). An integrated biopsychosocial approach to understanding awareness deficits in Alzheimer's disease and brain injury, Neuropsychological Rehabilitation: An International Journal, 16, 415-438,

Pia, L., Neppi-M'odona, M., Ricci, R., & Berti, A. (2004). The anatomy of anosognosia for hemiplegia: A meta-analysis. *Cortex*, *40*, 367-377.

Prigatano, G. P. (1998). Anosognosia, delusions, and altered self-awareness after brain injury. *BNI Quarterly, 4,* 40-48.

Prigatano GP. (2005). Disturbances of self awareness and rehabilitation of patients with traumatic brain injury: a 20-year perspective. *Journal of Head Trauma Rehabilitation, 20*, 19–29.

Prigatano, G. P., & Morrone-Strupinsky, J. (2010). Management and rehabilitation of persons with anosognosia and imparied self-awareness. In G. P. Prigatano (Ed.), *The study of anosognosia* (pp. 495-516). New York: Oxford University Press.

Pedersen, P. M., Jørgensen, H. S., Nakayama, H., Raaschou, H. O. & Olsen, T. S. (1996). Frequency, determinants, and consequences of anosognosia in acute stroke. *Journal of Neurological Rehabilitation, 10,* 243–250. Ramachandran, V.S. and Rogers-Ramachandran, D. (1996) Denial of disabilities in anosognosia. *Nature, 382,* 501.

Ramachandran, V.S., & Rogers-Ramachandran, D. (1996). Denial of disability in anosognosia. *Nature*, 382, 501.

Reed, B. R., Jagust, W. J., & Coulter, L. (1993). Anosognosia in alzheimer's disease. *Journal of Clinical and Experimental Neuropsychology, 15,* 231-244.

Rorden, C. & Brett, M. (2000). Stereotaxic display of brain lesions. Behavioral *Neurology, 12*, 191–200.

Rubens, A. B. (1985). Caloric stimulation and unilateral visual neglect. *Neurology, 35,* 1019-1024.

Solms, M. (1995). Is the brain more real than the mind? *PsychoanalyticPsychotherapy, 9,* 107–20.

Spalletta, G., Serra, L., Fadda, L., Ripa, A., Bria, P., & Caltagirone, C. (2007). Unawareness of motor impairment and emotions in right hemispheric stroke: A preliminary investigation. *Geriatric Psychiatry, 22,* 1214-1246.

Turnbull, O. H., Evans, C. E. Y., & Owen, V. (2005). Negative emotions and anosognosia. *Cortex, 41*, 67-75.

Vallar, G. & Ronchi, R. (2006). Anosognosia for motor and sensory deficits after unilateral brain damage: a review. *Restorative Neurology and Neuroscience, 24,* 247-257.

Vocat, R., Staub, F., Stroppini, T., & Vuilleumier, P. (2010). Anosognosia for hemiplegia: A clinical-anatomical prospective study. *Brain, 133*, 3578-3597.

Vossel, S., Weiss, P.H., Eschenbeck, P., & Fink, G. R. (2013). Anosognosia, neglect, extiniction and lesion site predict impairment of daily living after right hemisphere stroke. *Cortex, 49,* 1782-1789.

Vuilleumier, P. (2000). Anosognosia. In J. Bogousslavsky, & J. L. Cummings (Eds.), *Behavior and mood disorders in focal brain lesions* (pp. 465-519). Cambridge, UK: Cambridge University press.

Vuilleumier, P. (2004). Anosognosia: The neurology of beliefs and uncertainties. *Cortex*, *40*, 9-17.

Weinstein, E. A., & Kahn, R. L. (1950). The syndrome of anosognosia. *Neurology and Psychiatry*, *64*, 772-791.

Weinstein, E. A., & Kahn, R. L. (1955). *Denial of illness: Symbolic and physiological aspects*. Springfield, IL: Charles C. Thomas.

Wilson, B. A., Cockburn, J. & Halligan, P. W. (1987). *Behavioural Inattention Test* (BIT). Bury St Edmunds: Thames Valley Test Company.

Zigmond, A. S. & Snaith, R. P. (1983). The Hospital Anxiety and Depression Scale (HADS). *Acta Psychiat Scand*, *67*, 361–70.

TABLE 1. ED's neurological and neuropsychological profile in repeated assessments.

Function	Test (max score)		Days from onset	
			7	58 post video
Awareness[a]	Feinberg	LUL (10)	**7***	1
	Berti	LUL (-2)	**2**	0
		LLL (2)	**2**	0
Motricity	MRC[b]	Power LUL (5)	**0**	**1**
		Power LLL (5)	**0**	**0**
Mood	HADS[c]	Depression (10)	2	np
		Anxiety (10)	5	np
Memory[d]	MOCA	Verbal recall	3	np
Working Memory	Digit Span	Forwards	5	6
		Backwards	**2**	3
Personal Neglect	Comb & Razor[e]		**-0.38**	np
Extra Personal Neglect	BIT[e]	Star omissions	**50**	np
		Line bisection (3)	**0**	**1**
		Line Crossing (40)	**5**	np
		Copy (3)	**0**	**1**

Tactile Extinction	Bisiach [e]	Upper limb	**3**	1
		Lower limb	**3**	2
Visual Extinction	Bisiach[e]	Upper visual field	**3**	1
		Lower visual field	**3**	1
Executive Functioning	FAB[f]	Similarities	**1**	**1**
		Lexical fluency	**1**	np
		Motor Series	2	2
		Go-no-Go (inhibitory control)	2	np
GCF[g]	LCFS		7	7

[a]Awareness interview (Berti et al., 1996); AHP questionnaire (Feinberg et al., 2000)
[b] MRC= Medical Research Council (Guarantors of Brain, 1986)
[c]HADS=Hospital Anxiety and Depression scale, cut-off=8 points (Zigmond, A. S. & Snaith, R. P.,1983)
[d]MOCA=The Montreal Cognitive Assessment (Nasreddine, 2005)
[e]Comb/Razor test (MacIntoch, Brodie, and Beschin, 2000); BIT= Behavioural Inattention Test (Wilson, Cockborn & Halligan, 1987); tactile and visual extinction= (Bisiach, Vallar, & Perani, 1986)
[f]FAB= Frontal Assessment Battery (Dubois et al., 2000)
[g]GCF= Global Cognitive Functioning; LCFS= Levels of Cognitive Functioning Scale

TABLE 2. FG's neurological and neuropsychological profile in the pre- and post-training assessments

Function	Test	(max score)	Days from onset	
			74	103 (14 days after V)
Awareness [a]	Bisiach	LUL (3)	**3***	np
		LLL (3)	**3**	np
	VATA-m (Max=36)	Bimanual	**22**	**12**
		Bipedal	**10**	**7**
	Marcel-Moro Modified interview	Total score (23)	**20**	**8**
		General (2)	**2**	**2**
		Upper Limb (9)	**7**	**3**
		Lower Limb (8)	**7**	1
		Daily life activities (4)	**4**	**2**
Motricity	MRC [b]	Power LUL (5)	**0**	**0**
		Power LLL (5)	**0**	**0**
Mood [c]	B.D.I.		5	8
	S.T.A.I.	Y-1 State	38	40
		Y-2 Trait	51	33
	Affective Story Recall (4)		0	1

Memory [d]	Story Recall		12.08	12.07
Working Memory	Digit Span	Forwards	4	4
Personal Neglect	Comb & Razor[e]		**-0.05**	**-0.11**
Extra Personal Neglect	BIT[e]	Stars omissions (46)	**42**	np
		Line bisection (9)	**0**	**3**
		Line Crossing (40)	**9**	**11**
		Copy (4)	**0**	**0**
	Tactile extinction		**0**	**0**
	Visual extinction		**0**	**0**
Executive Functioning	FAB [f]	Similarities	**1**	3
		Lexial fluency	**1**	2
		Motor Series	2	**1**
		Go-no-Go (inhibitory control)	**1**	**0**
General Cognitive	MMSE [g] (30)		23.7	np

[a] Bisiach interview (Bisiach, Vallar, Petrani, Papagno & Berti, 1986); VATA-m= Visual-Analogue Test for Anosognosia for motor impairments (Della Sala, Coccini, Bechin & Cameron, 2009); Marcel-Moro modified interview (Moro, Perngio, Zapparoli, Cardiolo & Aglioti, 2011)
[b] MRC= medical research council (Guarantors of Brain, 1986)
[c] BDI= Becks Depression Inventory (Beck, Ward & Mendelson, (1961); STAI= State-Trait Anxiety Inventory (Speilberg, Gorsuch, Lushene, 1089); Affective story recall test (Turnbull, Evan & Owen, 2005)
[d]Story recall & Digit span (Spinnler & Tognoni, 1987)
[e] Comb/Razor test= test of personal neglect, bias is calculated according to MacIntoch, Brodie, and Beschin (2000); BIT= Behavioural Inattention Test (Wilson, Cockburn & Halligan, 1987)
[f] FAB= Frontal Assessment Battery (Apollonio at al., 2005)
[g] MMSE= Mini-Mental State Examination (Folstein, Folstein, McHugh,1975)

CAPTIONS TO FIGURES

FIGURE 1. Identifiable phases in the video replay intervention methods. Specific indications useful to employ and implement the video-intervention methodology are reported for each of the four phases of the training.

FIGURE 2. Neuroimaging examinations with white boundary lesion demarcation. *Top part:* ED's CT scan. *Bottom part:* FG's CT scan. Right hemisphere is on the right side. Lesion mapping was performed using MRIcro software (Rorden & Brett, 2000) by an examiner who was blind to the clinical feature of the patients. We superimposed the lesion of each subject on the Automatic Anatomical Level template provided with MRIcro and calculated the number of lesioned voxels in each anatomical region of interest. The tables show the percentage of lesioned tissue affected by the lesion in the grey (left tables) and white (right tables) matter.

F IGURE 3. Left upper and lower limb awareness progression after self-observation using video replay in multiple intervention sessions. The figure shows the awareness recovery for upper and lower limb paralysis over time. The original and adapted Feinberg et al.'s interview were used to acquire separate awareness scores for left upper limb (LUL) and left lower limb (LUL), respectively, before and after the intervention. Comparison data from previous right hemisphere groups with and without AHP on the original Feinberg et al.'s interview are also reported (Fotopoulou et al., 2010). The intervention led to a large decrease in unawareness in all sessions but awareness was not completely, nor permanently restored and a smaller increase of unawareness was noted between sessions, until total recovery was progressively achieved. Interventions targeting the arm had no effect on leg unawareness, which remained constant until it was specifically targeted by video-observation.

Figure 4. Judgement of Actions Test (JAT) scores for self-referent condition before and after video intervention. Median (95% Interval of Confidence, C.I.) values are shown for each condition and each observation. The three pre-video observations in the left side of the dotted vertical line were averaged to a unique baseline for statistical tests.

Figure 5. Comparison of self-referred vs. other-referred ratings on judgment of actions test (JAT). Median (95% C.I.) values are shown for each condition and each observation. Statistical significances are represented by brackets containing

the self- and the other-referent distributions and are related to their overall comparison. Brackets underlying a single other-referred distribution represent a statistical significance in the overall trend. For details of statistical results, see the Supplementary Materials.