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# Anosognosia for apraxia: Experimental evidence for defective awareness of one's own bucco-facial gestures



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## ABSTRACT

Anosognosia is a multifaceted, neuro-psychiatric syndrome characterized by defective awareness of a variety of perceptuo-motor, cognitive or emotional deficits. The syndrome is also characterized by modularity, i.e., deficits of awareness in one domain (e.g., spatial perception) co-existing with spared functions in another domain (e.g., memory). Anosognosia has mainly been reported after right hemisphere lesions. It is however somewhat surprising that no studies have thus far specifically explored the possibility that lack of awareness involves apraxia, i.e., a deficit in the ability to perform gestures caused by an impaired higher-order motor control and not by low-level motor deficits, sensory loss, or failure to comprehend simple commands. We explored this issue by testing fifteen patients with vascular lesions who were assigned to one of three groups depending on their neuropsychological profile and brain lesion. The patients were asked to execute various actions involving the upper limb or bucco-facial body parts. In addition they were also asked to judge the accuracy of these actions, either performed by them or by other individuals. The judgment of the patients was compared to that of two external observers.

Results show that our bucco-facial apraxic patients manifest a specific deficit in detecting their own gestural errors. Moreover they were less aware of their defective performance in bucco-facial as compared to limb actions. Our results hint at the existence of a new form of anosognosia specifically involving apraxic deficits.

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Abbreviations: AHP, Anosognosia for Hemiplegia; AA, Anosognosia for Apraxia; BFA, bucco-facial apraxia; A+, apraxic patients; A-, left hemisphere brain damaged non apraxic patients; RBD, right hemisphere brain damaged patients.

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## 1. Introduction

The term *anosognosia*, initially coined to indicate the denial of motor deficits contralateral to a brain lesion (Anosognosia for Hemiplegia – AHP, Babinski, 1914), refers to a multifaceted syndrome where patients who have suffered strokes, traumatic brain injury, degenerative diseases or neuropsychiatric disorders show complete or partial lack of awareness of a variety of neurological and cognitive deficits (e.g., hemianopia, blindness, hemianesthesia, neglect, aphasia, amnesia) (Prigatano, 2010).

Clinical reports and experimental studies on AHP (Cocchini, Beschini, Fotopoulou, & Della Sala, 2010; Moro, 2013; Moro, Pernigo, Zapparoli, Cordioli, & Aglioti, 2011; Ramachandran, 1994) and Alzheimer's Disease (Mograbli & Morris, 2013) show distinct types of anosognosia where implicit and emergent residual forms of awareness are present. Moreover, patients may exclusively deny their own paralysis but recognize deficits in other patients (1st person deficit) or fail to recognize motor impairment both in themselves and in other subjects (1st and 3rd person deficit) (Marcel, Tegnèr, & Nimmo-Smith, 2004; Moro et al., 2011).

Despite the steady increase of interest in anosognosia, no studies have thus far specifically set out to investigate the possible existence of anosognosia for apraxia (AA). The term apraxia refers to a wide spectrum of disorders with in common an inability to perform skilled or learned purposeful gestures. Although sometimes co-existing with motor or sensory deficits or language disorders, apraxia is not explained by any of these (Zadikoff & Lang, 2005). Conceptual and production components of gestural organization may be differentially affected, leading to ideational (i.e., defective action and object-use knowledge) or ideomotor apraxia (i.e., defective action execution in gesture pantomime and imitation) (Leiguarda & Marsden, 2000). In limb apraxia, imitation of transitive gestures (e.g., hammering a nail) is more impaired than the imitation of intransitive gestures (e.g., waving goodbye) (Buxbaum, Kyle, & Menon, 2005). Among the body-part defined subtypes of apraxia, bucco-facial apraxia (BFA) refers to an inability to voluntarily control facial, lingual, pharyngeal and masticatory actions (e.g., protruding tongue, blinking eyes) on purpose but not in ecological situations, when the movements are automatically performed. Neuropsychological and neuroanatomical results indicate that BFA and limb apraxia are at least partially independent (Raade, Rothi, & Heilman, 1991). While limb apraxia appears to be more commonly associated with left frontal and parietal brain damage (Pazzaglia, Smania, Corato, & Aglioti, 2008), BFA follows lesions in left prefrontal areas, the central operculum, the insula, the centrum semiovale, their subcortical projections and the basal ganglia (Pramstaller & Marsden, 1996).

In this study we investigated the existence of a specific form of AA related to the possibility that subjects presenting with apraxia also show defective awareness of their difficulties. With this aim we asked patients with or without BFA to judge the correctness of bucco-facial and limb related gestures performed by themselves or by a gender-matched model. The patients' responses were compared with the evaluations provided by their therapist and caregiver. The comparison between discrepancies in judgment of actions

performed by themselves or others allowed us to distinguish deficits in awareness from non-specific difficulties in action recognition.

## 2. Methods

### 2.1. Participants

Fifteen brain damaged patients recruited from the Neuro-rehabilitation Units at the IRCCS Santa Lucia (Rome) and the Sacro Cuore Hospital (Negrar, Verona) gave their informed consent for their participation in the study. The procedures were approved by the two local Ethics Committees and the study was carried out in accordance with the guidelines of the Declaration of Helsinki. All the patients were right-handed (Briggs & Nebes, 1975) and had suffered from vascular lesions at times varying between 1 and 27 months before the assessment. They were divided into three groups depending on their symptoms and side of lesion: i) patients presenting with BFA (A+); ii) left brain damaged non-apraxic patients (A-); iii) right brain damaged non-apraxic patients (RBD). The groups were comparable in terms of age and education but differed in onset-assessment intervals with A- and RBD in more acute phases with respect to A+.

The contralesional upper limb motor deficit was assessed by means of the Medical Research Council Scale (Florence et al., 1992). Clinical and demographical data are reported in Table 1.

### 2.2. Preliminary neuropsychological screening

A battery of standardized tests was used for neuropsychological screening. This involved general cognitive abilities (Raven, Court, & Raven, 1988), Verbal and Visual Memory (Spinnler & Tognoni, 1987), executive functions (non-verbal subtests of the Frontal Assessment Battery – Appollonio et al., 2005) and spatial attention (Line Bisection – Wilson, Cockburn, & Halligan, 1987). Verbal comprehension and denomination subtests of the Aachen Aphasia Test (Luzzatti, Willmes, & De Bleser, 1996) were used to assess language deficits.

As shown in Table 1, the patients' scores rule out the possibility that cognitive disorders play any major role in the experimental results. Verbal Comprehension was spared in all the patients.

### 2.3. Assessment of apraxia

The presence of BFA was ascertained by means of the Upper and Lower Face Apraxia test (Bizzozero et al., 2000). In this test 29 and 9 actions are selected to evaluate lower and upper face gestures respectively, according to the territory of the cranial nerves involved. Each action is scored 1 (correct) or 0 if there are errors in execution (i.e., amorphous movements, protracted pauses, additional movements, conduits d'approche or incomplete action). These scores are then weighted taking into account relative difficulty (cut-off: lower face = 400.04, upper face = 38.43).

The test for Upper Limb Ideomotor Apraxia (TULIA – Vanbellingen et al., 2010) consists of 48 items, including

**Table 1 – Patients' clinical data.**

Subjects	A+1	A+2	A+3	A+4	A+5	A-1	A-2	A-3	A-4	A-5	RBD-1	RBD-2	RBD-3	RBD-4	RBD-5
Age	38	69	69	60	69	69	78	75	59	68	47	59	68	70	47
Education (years)	17	13	13	5	5	17	5	17	6	13	8	17	5	13	13
Lesion	lFTI	lFP-BG	lFI-BG	lF-rP	lFTP	lTO-BG	lFI-rCer	lF	lPI	lFI	rFT	rT	rFT	rIC	rF
Interval from lesion (m.)	18	27	16	1	1	1	1	5	1	3	4	4	4	1	2
Medical Res. Council	4	1	1	5	5	5	3	1	2	1	1	1	1	1	0
<b>Neuropsychological screening</b>															
Raven 36	30	31	28	28	23	36	27	23	20	28	26	28	18	27	24
Verbal span	0	5	4	3	4	6	imp	5	3	4	4	6	4	5	4
Word Comprehension	24	30	28	30	28	28	28	30	26	24	30	30	30	30	30
Phrase Comprehension	25	27	27	28	30	27	30	27	26	27	30	30	30	25	30
Object Denomination	11	30	16	30	29	23	17	28	30	28	30	30	30	30	30
Colour Denomination	12	30	21	30	30	29	11	30	30	30	30	30	30	29	30
Compound Word Denom.	3	27	8	30	24	21	0	30	27	25	30	30	30	30	30
Story Recall Immediate	5.5	5.5	6.3	6.3	5.5	5	imp	5.6	4.7	5.1	5.5	5.5	1.1	6.6	6.6
Story Recall Delayed	5.8	5.8	6	6	5.6	2	imp	5.6	3.6	3.1	4.7	7.7	1.1	6.6	6.9
FAB (subtests 3–6)	11	12	9	9	11	12	11	10	10	12	7	8	8	11	15
Corsi Span	5	4	6	5	5	5	4	5	4	5	4	5	4	5	3
Corsi Supraspan	15.16	25.49	5.93	2.47	28.46	13.7	7.2	15.15	6.71	8.33	14.29	19	12.3	22.25	imp
Line Bisection	9	9	9	9	9	9	9	9	9	9	9	3	6	9	8
<b>Apraxia</b>															
Lower Face Apraxia	299.5	404	398.5	424	392.25	432.25	400.7	424.5	425	425.5	420.25	431.25	414.5	429.5	421.25
Upper Face Apraxia	44.5	37.25	45.5	33	45.25	45.25	44	44	40	45.25	44.75	42	43.75	41.25	43.25
Limb Apraxia	209	232	273	236	234	240	215	237	219	195	240	240	204	235	240
Use Apraxia	14	14	14	14	14	14	14	14	14	14	14	14	14	14	14
<b>Anosognosia</b>															
VATA-M	9	16	2	0	5	-5	-1	9	-5	2	0	6	4	-3	-1
VATA-L	9	13	-6	-1	3	-6	1	-1	3	6	0	2	-2	0	0

For each subject demographical and clinical data, their scores in neuropsychological screening and in the assessment of Apraxia and Anosognosia for Hemiplegia and Aphasia are reported. A+ = apraxic patients; A- = left brain damaged non apraxics; RBD = right brain damaged non apraxics; l = left; r = right; F = frontal; T = temporal; I = insular; P = parietal; BG = basal ganglia; O = occipital; Cer = cerebellum; IC = internal capsule; imp = impossible. Pathological scores are in bold.

imitation and pantomiming meaningless and meaningful gestures. A 6-point scoring method (0 = totally incorrect action, 5 = perfect performance) means that performances can be evaluated in terms of scores ranging from 0 to 240 (pathological scores  $\leq 194$ ).

Finally, in the De Renzi and Lucchelli ideational apraxia test (1988) patients are requested to perform seven complex actions requiring the use of objects. Scores range from 2 (perfect performance) to 0 (totally incorrect performance). Total score  $<14$  indicates apraxia.

None of our patients manifested signs of limb apraxia, while all five subjects in the A+ group presented with BFA, failing in lower face (subjects A + 1, A + 3 and A + 5) or upper face (subjects A + 2 and A + 4) actions (Table 1). Their lesional data are shown in Fig. 1.

## 2.4. Assessment of AHP and language deficits

The Visual-Analogue tests for AHP (VATA-M; Della Sala, Cocchini, Beschin, & Cameron, 2009) and for Language impairment (VATA-L; Cocchini, Gregg, Beschin, Dean, & Della Sala, 2010) combine the patient's and caregiver's evaluations of the patient's abilities, the first in a series of specific motor tasks (e.g., walking, drinking from a glass) and the second in a series of communicative situations (e.g., finding the right word). A discrepancy score is then calculated with a maximum score of 36 for the VATA-M (cut-off  $\leq 6.8$ ) and 42 for the VATA-L (cut-off  $\leq 11.9$ ).

Two subjects (A + 1, and A - 3) showed signs of mild and one (A + 2) of moderate AHP. None of our patients showed deficits in awareness of aphasia.

## 2.5. Experimental task

### 2.5.1. Stimuli

Two lists of videos were arranged for the two phases of the experiment.

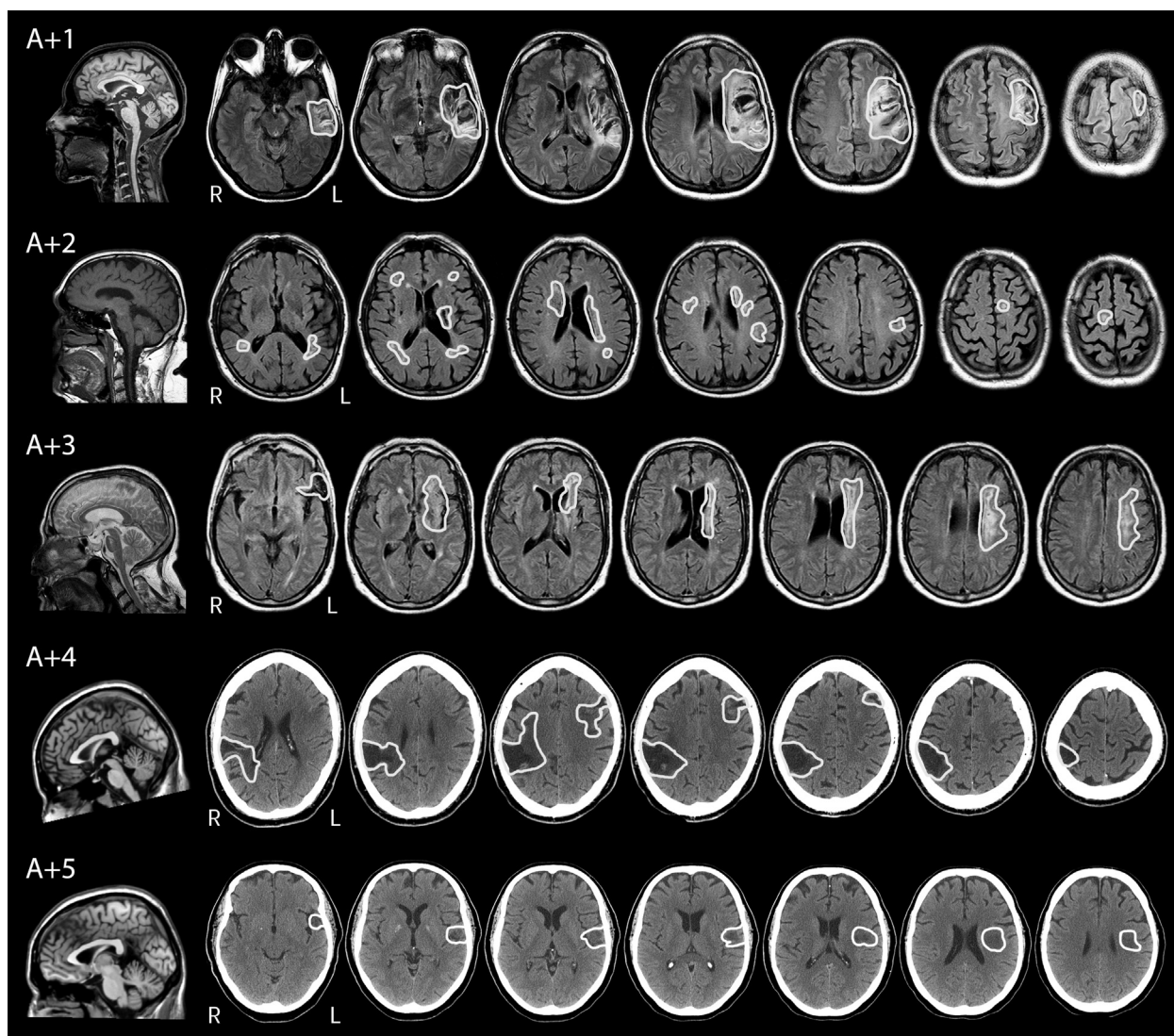
In the List 1, 28 videos were made using a Sony Handycam HDR-CX115E. These showed an actress performing bucco-facial or limb actions (14 transitive and 14 intransitive, Table 2). These videos were used as a model for patients to follow (see 2.5.2).

List 2 included 28 videos showing the actions performed by the patients during phase 1 of the experiment (see 2.5.2) and assessed by the examiner as correct or incorrect. In addition, for each incorrect action a video showing the same action (with the same category of error but performed by a gender-matched model) was selected from an archive.

Errors were divided into four categories: i) hand/arm configuration; ii) mouth configuration; iii) spatial error (measure or trajectory); iv) unrecognizable action (Buxbaum et al., 2005).

10 additional different videos in List 2 (5 correct and 5 incorrect gestures, Table S1 in Supplemental Materials) showed actions performed by the gender-matched model and these served as controls. Representative examples of stimuli are shown in Fig. 2C.





**Fig. 1 – Lesions of A+ patients.** Axial slices of the MRI/CT scans of the apraxic patients are shown (left hemisphere on the right). The tables report quantitative estimates of the damaged brain regions (sum and the percentage of lesioned voxels). The lesion analysis was performed manually by means of MRICRO software (<http://www.mccauslandcenter.sc.edu/mricro/mricron/index.html>). The plots of the lesions were drawn on the ICBM152 MRI scan template previously rotated to match the scan orientation of the patient. The definition and labels related to the anatomical maps are based on the “Automated Anatomical Labeling” atlas (AAL). Note that the A + 2 patient presented with multiple ischemic micro-lesions in both the right and left hemispheres, and the A + 4 patient shows signs of a right hemisphere lesion which occurred 2 years previously, but was asymptomatic at the onset of the left-side damage. Details concerning the analysis of each single patient’s lesions are reported in [Table S1 \(Supplemental Materials\)](#).

### 2.5.2. Procedure

The experiment was divided into two phases ([Fig. 2](#)).

**Phase 1. On-line judgment.** The subjects were seated at a distance of approximately 60 cm from a 17-inch monitor (resolution: 1024 × 768 pixels) where videos from List 1 were shown. After each video, the patients were asked to imitate, as accurately as possible, the action they had seen and their performance was video-recorded.

They were then asked to judge whether their performance was correct or incorrect (On-line judgment). When it was judged to be incorrect, the patients then categorized their

error, choosing from the four different options previously described (see [2.5.1](#)).

Their performance was also evaluated by the examiner according to the same categories of errors ([Buxbaum et al., 2005](#)).

**Phase 2. Off-line judgment.** The patients next evaluated all the actions shown in the List 2 videos which were presented in a random order (their actions, incorrect actions performed by the model and the 10 control actions) and, when incorrect, they were requested to select a category for the error.

The patients always responded verbally and the examiner recorded their responses by pressing a computer key.

**Table 2 – The list of actions used in phase 1 of the experimental task.**

Gesture	Topography	Typology of gesture	
		Transitive	Intransitive
Turning on a lighter	Limbs	x	
Sawing	Limbs	x	
Opening a zip	Limbs	x	
Fastening buttons	Limbs	x	
Writing on a keyboard	Limbs	x	
Using a kitchen whisk	Limbs	x	
Using a monkey wrench	Limbs	x	
Writing with a pen	Limbs	x	
Painting	Limbs	x	
Making the OK sign	Limbs		X
Making a “COME HERE” gesture	Limbs		X
The gesture of victory	Limbs		X
Looking through a telescope made using fingers	Limbs		X
Making the sign of wanting to drink	Limbs		X
Making the sign of wanting to eat	Limbs		X
Making the sign of phoning	Limbs		X
Making the sign of eavesdropping	Limbs		X
Indicating that a person is being crafty	Limbs		X
Inflating a balloon	Mouth	X	
Drinking with a straw	Mouth	X	
Whistling	Mouth	X	
Biting a pear	Mouth	X	
Making soap bubbles	Mouth	X	
Coughing	Mouth		X
Biting lips	Mouth		X
Making a “SHUSH” sign	Mouth		X
Sneezing	Mouth		X
Yawning	Mouth		X

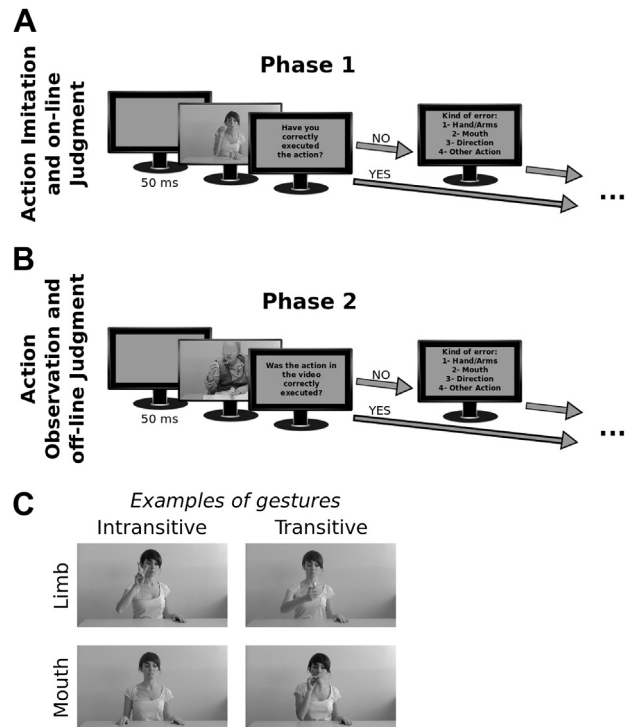
The 28 actions reported in the table constitute List 1. These were performed by an actress and shown, in random order, to the patients in 28 separate video clips. After each video, the patients were asked to imitate the action as well as they could and immediately afterwards judge their performance as either correct or incorrect.

*Observers' judgments.* The patients' caregivers and/or therapists took part in the study. They were asked to judge the actions showed in the List 2 and categorize the errors.

### 3. Statistical analyses

$\chi^2$  tests were employed to compare the three groups (A+, A– and RBD, two groups at a time) for the total number of errors made during the execution of the actions, the differences between mouth and limb errors and the various categories of errors.

In order to determine the presence of anosognosia, the judgments of each patient in Phase 1 and Phase 2 were compared with the average judgments of his/her Caregiver and Therapist (R Core Team, 2013).



**Fig. 2 – Materials. Experimental Timeline of the first (A) and second (B) phases of the experiment; C) examples of the categories of gesture employed in the experiment.**

Due to the small number of patients in each group, the analyses were performed using a meta-analytic approach (Marangolo et al., 2010). This approach is in line with the philosophy of the Crawford and Howell method (1998), considering each subject as a sample of  $N = 1$  and permitting, in addition, a global measurement of the effect size for each group, in each comparison (Borenstein, Hedges, Higgins, & Rothstein, 2009).

Thus, for each patient and comparison, the odds ratio and the associated confidence intervals were computed and used to obtain the summary effect sizes and confidence intervals for each group (Viechtbauer, 2010).

### 4. Results

*Errors.* Although A+ made more errors (n.42) than A– (n.35) and RBD (n.24), only the comparison between RBD and A+ was statistically significant ( $\chi^2_1 = 5.73, p = .017, \phi = .151, p$  Bonferroni corrected = .05). Importantly, while in actions involving limbs (A+: 23, A–: 22, RBD: 18) there were no differences between the groups, in actions involving the mouth the A+ group failed in more cases than the other groups (A+ = 19, A– = 13, RBD = 6). Only the comparison between A+ versus RBD gave a statistically significant result ( $\chi^2_1 = 7.68, p = .006, \phi = .151, p$  Bonferroni corrected = .018). Separating transitive and intransitive gestures, the same difference between these two groups was present for intransitive actions ( $\chi^2_1 = 9.16, p = .0025, \phi = .27, p$  Bonferroni corrected = .007).

**Table 3 – Analysis of errors.**

		Topography	Typology of gesture		Typology of error			
			Transitive	Intransitive	Hand arm	Mouth	Spatial	Other
A+	A+1	Mouth	1	5				1.00
		Limbs	3	2	.40		.60	
	A+2	Mouth		3			.33	.67
		Limbs	1	4	.80			.20
	A+3	Mouth	2			.50	.50	
		Limbs		2	.50		.50	
	A+4	Mouth		4		.25	.25	.50
		Limbs	2	6	.63		.38	
	A+5	Mouth		4		1.00		
		Limbs		4	.50		.50	
A–	A–1	Mouth		1			1.00	
		Limbs	2	3	.80		.20	
	A–2	Mouth		2		1.00		
		Limbs		1	1.00			
	A–3	Mouth		2		.50	.50	
		Limbs	3	4	.29		.71	
	A–4	Mouth		3		1.00		
		Limbs	2	3	.40		.60	
	A–5	Mouth	2	3		1.00		
		Limbs	2	4	.50		.50	
RBD	RBD1	Mouth		1		1.00		
		Limbs						
	RBD2	Mouth		2			1.00	
		Limbs	1	2			1.00	
	RBD3	Mouth		1		1.00		
		Limbs	4	5	.11		.89	
	RBD4	Mouth		2		.50		.50
		Limbs	4	2	.17		.83	
	RBD5	Mouth			No errors			
		Limbs						

The number of errors are reported for Topography (Mouth, Limbs) and Typology of Gesture (Transitive, Intransitive). Types of errors (Hand/Arm: hand/arm configuration, Mouth: mouth configuration, Spatial: error in measure or trajectory, Other: not recognizable or totally different action) are computed as a proportion of the total number of errors.

The A+ patients frequently failed the mouth movements or executed unrecognizable actions (Table 3) while the RBD patients made spatial errors in trajectory and direction and the A– group made errors in configurations involving arms/hands and mouth but never executed totally different or unrecognizable actions.

In general, when their judgment was incorrect, both patients and observers evaluated incorrect actions as being correct. Only in a very few trials did the subjects state that a correct action was incorrect.

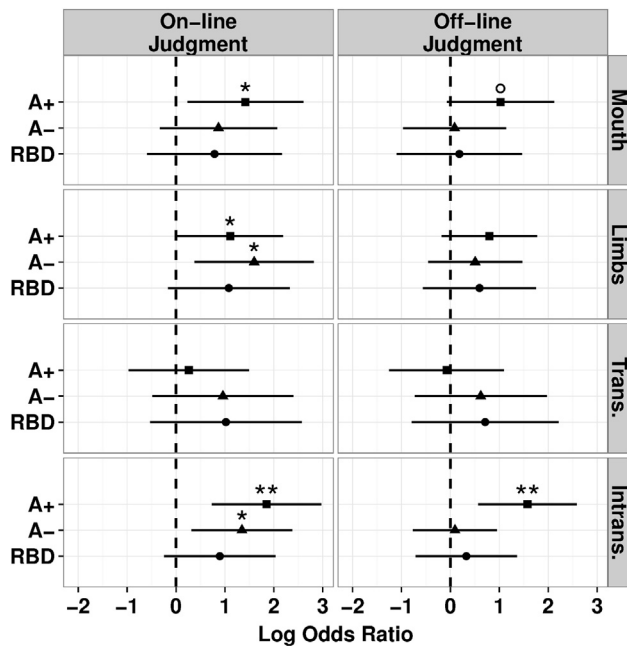
**Control videos.** In the ten actions used as controls in order to exclude any general difficulties in action recognition, all three groups performed well with no differences when they were compared with the average evaluations of the caregivers/therapists (A+:  $z = .56$ ,  $p = .58$ ,  $CI = -.58, 1.04$ ; A–:  $z = -.10$ ,  $p = .92$ ,  $CI = -.79, .79$ ; RBD:  $z = 0$ ,  $p = 1$ ,  $CI = -.84, .75$ ).

**Phase 1. On-line judgment.** The meta-analyses carried out on all 28 actions indicated that when asked to judge their own gestures directly after execution, all three groups recognized fewer errors than their observers (A+:  $z = 2.99$ ,  $p = .003$ ,  $CI = .47, 2.24$ ; A–:  $z = 2.96$ ,  $p = .003$ ,  $CI = .46, 2.28$ ; RBD:  $z = 1.96$ ,  $p = .05$ ,  $CI = .00, 2.12$ ). Crucial to our study, in the limb actions both the A+ and A– groups failed in the judgments of their own actions (A+:  $z = 2.01$ ,  $p = .04$ ,  $CI = .3, 2.18$ ; A–:  $z = 2.57$ ,  $p = .01$ ,  $CI = .38, 2.82$ ; RBD:  $z = 1.70$ ,  $p = .09$ ,  $CI = -.17, 2.32$ ),

while in the mouth actions only group A+ ( $z = 2.35$ ,  $p = .02$ ,  $CI = .23, 2.60$ ; A–:  $z = 1.42$ ,  $p = .15$ ,  $CI = -.33, 2.07$ ; RBD:  $z = 1.11$ ,  $p = .26$ ,  $CI = -.59, 2.16$ ) failed in their judgments (Fig. 3). A statistical difference was also present for intransitive actions (mouth + limb) in left damaged groups (A+:  $z = 3.24$ ,  $p = .001$ ,  $CI = .73, 2.97$ ; A–:  $z = 2.56$ ,  $p = .01$ ,  $CI = .32, 2.38$ ; RBD:  $z = 1.54$ ,  $p = .12$ ,  $CI = -.24, 2.03$ ) (Fig. 3).

In conclusion, some degree of difficulty in judging one's own action immediately after the execution was found in all the left brain damaged patients. Importantly, however, this was not specific for the facial or limb actions. Therefore, this difficulty may be considered as a generic impairment likely due to the role of the left hemisphere in motor planning rather than to apraxia.

**Phase 2. Off-line judgment.** When asked to judge their own actions shown in a video, only group A+ failed ( $z = 2.54$ ,  $p = .01$ ,  $CI = .23, 1.75$ ; A–:  $z = .80$ ,  $p = .42$ ,  $CI = -.42, 1.01$ ; RBD:  $z = .86$ ,  $p = .39$ ,  $CI = -.52, 1.34$ ). Dividing the gestures into categories depending on topography, a trend towards statistical significance was observed, with the A+ group failing exclusively in the judgment of mouth actions ( $z = 1.84$ ,  $p = .066$ ,  $CI = -.18, 1.77$ ; A–:  $z = .16$ ,  $p = .87$ ,  $CI = -.97, 1.14$ ; RBD:  $z = .28$ ,  $p = .78$ ,  $CI = -1.1, 1.48$ ). In addition, the same group failed in the evaluation of intransitive ( $z = 3.06$ ,  $p = .002$ ,  $CI = .57, 2.58$ ; A–:  $z = .21$ ,  $p = .83$ ,  $CI = -.77, .95$ ; RBD:  $z = .61$ ,  $p = .54$ ,  $CI = -.71, 1.36$ ) but not transitive gestures (Fig. 3).



**Fig. 3 – Judgment of actions executed by the patients.** Summary effects from the meta-analyses of the comparison between the patients' and the observers' judgments. Effects are separately reported for each group, phase, topography and typology of action. A 0 score indicates the correspondence between patient and observer judgments. Scores below 0 indicate that the patient's judgments are more severe than those of the observers (underestimation), and score over 0 indicates that the patient judges the actions more leniently than the observers (overestimation). ° = trend towards statistical significance ( $p = .06$ ); \* =  $p \leq .05$ ; \*\* =  $p \leq .01$ . Intervals of confidence (CI) are reported in the text.

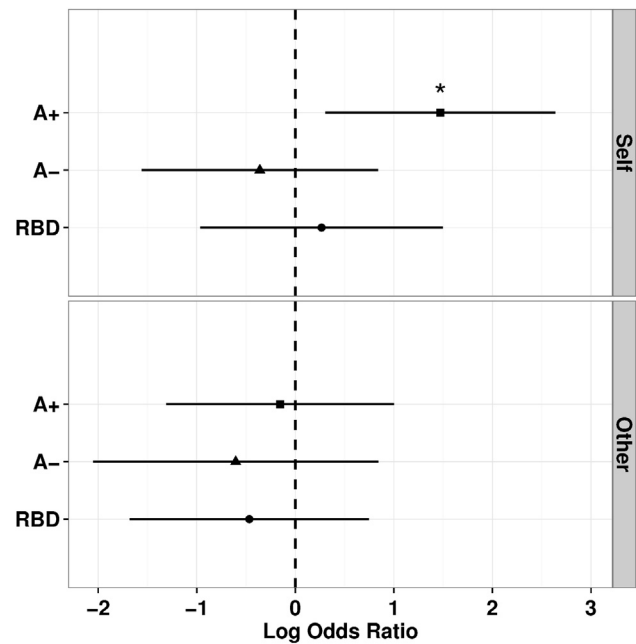
*Self versus other.* Separating judgments concerning patients' incorrect actions from judgments referred to incorrect actions performed by models, only the A+ group fail (A+:  $z = 2.47$ ,  $p = .013$ ,  $CI = .3, 2.64$ ; A-:  $z = -.59$ ,  $p = .56$ ,  $CI = -1.56, .84$ ; RBD:  $z = .42$ ,  $p = .67$ ,  $CI = -.96, 1.5$ ) and this was exclusively with reference to patients' actions (Fig. 4).

Individual data are discussed in the [Supplemental Material](#).

## 5. Discussion

Anosognosia has mainly been explored in right hemisphere damaged patients. However, [Della Sala et al. \(2009\)](#) circumvented linguistic disorders by using non-verbal tools and demonstrated that anosognosia may also be present after left hemisphere lesions ([Della Sala et al., 2009](#)).

In our study we have identified a new form of anosognosia in patients affected by BFA which is specifically related to apraxia (AA). This deficit is specific to bucco-facial gestures (characterized by impaired execution) and does not generalize to actions involving limbs. This defective awareness is present when patients evaluate their own actions rather than those of



**Fig. 4 – Judgment of actions performed by the patient themselves and those performed by others.** Summary effects from the meta-analyses of the comparison between the patients' and the observers' judgments are reported, separated for the three groups.

other people. Finally, in contrast with what has been found in cases of AHP ([Fotopoulou, Rudd, Holmes, & Kopelman, 2009](#)), self-observation using a video does not seem to impact the awareness of deficits.

### 5.1. AA

Our BFA patients, but not the non-apraxic left damaged and RBD, failed to report their errors when they were watching their action execution in a video. The total number of errors did not differ between the two left hemisphere damaged groups. Moreover they were not different depending on the category of errors between the three groups ([Buxbaum et al., 2005](#)). In contrast, while non apraxic patients are aware of their errors and judge their actions just as external observers do, the A+ group are not aware of their failures and judge incorrect gestures to be correct.

Since the patients were not affected by comprehension deficits and were able to provide "yes/no" responses as required, it is unlikely that aphasia influences the results.

The structure of the task reproduces the logic of other efficacious tests employed to detect AHP and for aphasia, the Vata-M ([Della Sala et al., 2009](#)) and the Vata-L ([Cocchini, Beschini, et al., 2010; Cocchini, Gregg, et al., 2010](#)). As in these tests, the patient's judgement concerning the correctness of his/her own actions is compared with the judgments of two external observers and any discrepancy between the evaluations is analyzed.

The results suggest that AA is independent from anosognosia for aphasia. The distinction between AHP and AA is more complex. [Della Sala et al. \(2009\)](#) found signs of AHP in



40% of their sample of left hemisphere damaged patients assessed by means of non-verbal tasks. In our groups, two BFA and one non apraxic patient show signs of AHP in the Vata-M. It is thus plausible that in some patients AHP and AA coexist.

It is worth noting that our results show a specificity in anosognosia that is selective in that it exclusively involves bucco-facial and not limb actions. This suggests that there are different mechanisms relating to AHP and AA (see 5.2).

Previous studies have demonstrated deficits in action recognition in patients affected by frontal lesion and apraxia (Pazzaglia, Smania, et al., 2008; Pazzaglia, Pizzamiglio, Pes, & Aglioti, 2008; Moro et al., 2008). Nevertheless, deficits in action recognition were excluded in our study due to the patients' spared ability to judge the actions of other.

Finally, the failure to judge intransitive actions demonstrated by all the left brain damaged patients (both apraxics and non apraxics) is an unexpected result and is something which has not been found in previous studies (Pazzaglia, Smania, et al., 2008). We can not exclude the possibility that in this experiment the transitive gestures were simpler to judge than the intransitive ones.

## 5.2. Topography in AA

Our BFA patients did not perform worse than the other groups when judging all types of gestures. Tellingly however they did exhibit a specific impairment for actions involving their mouth and face. This topography, previously also found in AHP as a distinction between upper and lower limb (Della Sala et al., 2009; Moro et al., 2011), may speak in favor of modularity in AA.

It has been suggested that anosognosia may include a number of specific deficits deriving from impairments in anatomo-functionally discrete monitoring systems, each involved in the general control and monitoring of motor, sensory, spatial, memory and language functions (Marcel et al., 2004; Spinazzola, Pia, Folegatti, Marchetti, & Berti, 2008). Moreover, the finding that premotor areas (where a lesion induces a deficit in motor planning and control) are also affected by a lesion in AHP (Berti et al., 2005) supports the hypothesis that there are multi-componential rather than single-domain monitoring systems. Studies on the multilevel component framework postulate a monitoring system specific to each domain which is anatomo-functionally related to the to-be-monitored function (Vallar & Ronchi, 2006). This is in keeping with the function-dependent specificity of various forms of anosognosia reported in the literature. In this vein, a cognitive or motor deficit might extend to lack of awareness when lesions affect specific-function monitoring components. However, in order to guarantee the consistency and continuity of a patient's experience, these monitoring systems need to be connected to a second level of awareness, probably linked to the networks involved in error processing, metacognitive processes, beliefs and construction of self (Fotopoulou, 2013; Prigatano, 2010; Vocat, Saj, & Vuilleumier, 2013).

It is worth mentioning that deficits in self-appraisal (i.e., the cognitive ability that helps people to select appropriate tasks on the basis of accurate evaluation of their abilities, Rosen et al., 2010) may play an important role in determining anosognosia and overestimation of self performance (Rosen et al., 2010). Interestingly, our anosognosic patients show

lesions involving the frontal cortical and subcortical networks that are involved in self-related processes. Thus, the suggestion may be made that awareness of deficits is normally guaranteed by the interaction between modular specific monitoring systems and more general self-related processes (Mograbli & Morris, 2013; Rosen et al., 2010). Long fronto-temporo-insulo-parietal white matter tracts are involved in the explicit, declarative awareness of self (Moro et al., 2011; Pia, Neppi-M'odona, Ricci, & Berti, 2004), while the error processing networks include the anterior cingulate cortex, the insula, the basal ganglia and their connections to the lateral frontal cortex (Karnath, Baier, & Nägele, 2005; Klein, Ullsperger, & Danielmeier, 2013).

Our patients' lesions are different in terms of site and extension and so we cannot directly correlate specific networks to the symptoms. It is worth noting, however, that in all our patients, the lesions involve the left frontal areas, together with the insula or the basal ganglia and their surrounding white matter. It is thus possible that AA patients fail to acknowledge their own errors in the ideation and planning of actions. A similar hypothesis is suggested by Sirigu, Daprati, Pradat-Diehl, Franck, and Jeannerod (1999) who found a failure in apraxic patients to compare internal and external feedback regarding movement.

## 5.3. On-line and off-line judgment

It has been found that in cases of AHP self-observation using video replays contributes towards the reinstatement of motor awareness, in both the acute and chronic stages after the onset of damage. In a single case study, Fotopoulou et al. (2009) used video replays and recorded a dramatic increase in the patient's motor awareness. Video replay forces patients to shift their perspective from that of an agent to that of an observer. In addition, in line with explanations related to the monitoring of motor awareness, given the off-line nature of video replay, the patient's motor intentions are not relevant at the moment of observing the video, and an increase in his/her motor awareness is thus facilitated (Berti et al., 2005).

Our results show a different pattern in apraxic and non apraxic subjects. Indeed, in the comparison between the judgments expressed by patients immediately after the execution of actions (phase 1, 1st person perspective) and those expressed after video observation (phase 2, 3rd person perspective), the non apraxic subjects improve their performance and judge their actions as the observers do. In contrast, the apraxic patients do not show this improvement. This suggests that in on-line/first person judgment an important role may be played by the on-line action programming, i.e., the subjects judge their actions referring to the action planning more than to the outcome of action execution. Viceversa, the apraxic failure in the recognition of their actions also when these are seen in a video (and thus not planned at the moment of the judgment) may be suggestive of the presence of deficits in action representation. Although this first versus third person dissociation, originally described in right hemisphere damaged patients, may hint at the differences between AHP and AA, further studies on larger samples are necessary.

Finally, in apraxic patients we found a dissociation regarding self versus others. Indeed, the patients normally



recognized the errors in the videos when the actions were executed by a model, but did not with their own executions. Studies with larger samples will make it possible to verify whether or not this dissociation is constant in AA.

Of course the study has some limitations. Firstly, similar investigations concerning limb apraxia are necessary in order to confirm the dissociation between AHP and AA and establish the existence of double dissociations between AA in patients with bucco-facial and limb apraxia. Nevertheless, due to the novelty of the study, which for the first time investigates a new form of anosognosia, we chose to start with a group study on a pure form of apraxia. Focusing on patients with BFA allowed us to explore whether defective awareness of gestures was topographically associated to defective execution due to the higher-order motor control impairment typical of apraxia. Moreover, the small number of subjects meant that there were limitations regarding the possibility of analyzing individual variability in depth and correlating patients' behavior with a detailed study of the lesions involved. This is due to the fact that it is necessary to exclude subjects with disorders in communication and limb apraxia. Further research will also establish whether emotional and affective variables influence patients' judgments.

### Conflict of interest statement

The authors declare that they have no financial or other relationships that could be interpreted as a conflict of interest affecting this article.

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### Supplementary data

Supplementary data related to this article can be found at <http://dx.doi.org/10.1016/j.cortex.2014.05.015>.

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