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# Atypical holistic processing of facial identity and expression in a case of acquired prosopagnosia

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## ABSTRACT

Typical face perception is mediated by holistic processing (i.e., the simultaneous integration of face parts into a whole representation). People with Acquired Prosopagnosia (AP), who have lost the ability to recognise faces after a brain lesion, should thus show atypical holistic coding. Our aim is to use the composite-face effect (CFE) as a measure of holistic processing in ST, a 48-year-old woman with AP but normal recognition of facial expressions of emotions, and matched healthy control participants. Two experiments examining the CFE for identity (Experiment 1) and for expression of emotions (Experiment 2) were conducted. Contrary to controls, in both experiments, ST showed an atypical (i.e., reversed) CFE, thus suggesting altered holistic mechanisms affecting both components of perceptual judgement. Results also suggest that normal facial expression recognition is achievable even with holistic processing difficulties, possibly through compensatory, part-based, mechanisms.

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Face; identity; expression; emotions; holistic; prosopagnosia

## 1. Introduction

Cognitive and neural evidence strongly indicates that faces represent a special category for the visual system. Human face recognition mostly relies on the synchronous activity of dedicated neurons in the (mostly right) occipito-temporal cortex (Haxby, Hoffman, & Gobbini, 2000; Kanwisher et al., 1997; Rivolta, Castellanos et al., 2014a; Rivolta, Woolgar et al., 2014b). Lesions of occipito-temporal regions cause acquired prosopagnosia (AP), a neuropsychological condition characterized by the impairment in recognizing previously known faces, and the inability to form memories of new faces despite the absence of low-level visual defect or intellectual disorders (Bodamer, 1947; Damasio, Damasio, & Van Hoesen, 1982; Grüsser & Landis, 1991; Mayer & Rossion, 2013; Rondot & Tzavaras, 1969). The recognition of familiar people in AP mainly relies on extra-facial cues, such as hair, accessories, gait, clothes and voices (see Benton, 1990 for a review).

Studies from cognitive psychology demonstrate that typical face recognition relies on dedicated perceptual mechanisms known as *holistic*, which refer to the simultaneous integration of face parts into a whole

configuration (Mckone & Yovel, 2009). The idea that holistic coding may allow for efficient face representation (Maurer, Le Grand, & Mondloch, 2002; Piepers & Robbins, 2013) was supported by studies demonstrating correlations between measures of holistic processing (see below) and face memory skills in the typical population (see DeGutis, Wilmer, Mercado, & Cohan, 2013 for correlations between holistic measures and face memory; and Richler et al., 2011 for the role of holistic processing in predicting face-recognition abilities), and by the aberrations of holistic mechanisms in patients with AP (e.g., Busigny, Joubert, Felician, Ceccaldi, & Rossion, 2010, 2014; Ramon, Busigny, & Rossion, 2010; Van Belle, de Graef, Verfaillie, Busigny, & Rossion, 2010). One of the most reliable measures of holistic processing is the *composite-face effect* (CFE), a visual illusion where the top half of one (upright) face appears to fuse perceptually with the bottom half of another, in the condition where the two halves are *aligned* (as opposed to when they are *misaligned*) (Hole, 1994; Young, Hellawell, & Hay, 1987). Albeit atypical CFE in AP has been documented in most cases (Busigny et al., 2010, 2014; Ramon et al., 2010), the existence of APs showing the typical CFE (Finzi, Susilo,

Barton, & Duchaine, 2016; Rezlescu, Pitcher, & Duchaine, 2012) indicates that face recognition deficits can co-occur with intact holistic processing (which might be due to the extent and site of the brain lesion). Note, however, that different types of holistic mechanisms exist (Mckone & Yovel, 2009; Negrini et al., 2017), which are sensitive to different manipulations (e.g., the *part-whole effect*, Tanaka & Farah, 1993; the *face-inversion effect*, Yin, 1969), and that could indicate atypical holistic mechanisms where the CFE fails to find group differences.

Albeit face identification tasks and the CFE often show anomalies in APs, more variability is seen in the recognition of expressions of emotions. According to standard models (Bruce & Young, 1986) these two abilities can dissociate; there are, in fact, APs with preserved emotion recognition (Riddoch, Johnston, Bracewell, Boutsen, & Humphreys, 2008; Takahashi et al., 1995; McNeil and Warrington, 1993; Tranel, Damasio, & Damasio, 1988) (but see Fox et al., 2011). The patient FB (Riddoch et al., 2008) presented a severe acquired prosopagnosia yet scored within the typical range on an expression recognition test (i.e., The Emotional Hexagon Test, Ekman & Friesen, 1976, which implemented faces from the Ekman and Friesen set that have been manipulated to produce images of graded difficulty; FB score: 99/120; cut-off: 94). The Authors suggested that FB relied on part-based information to a greater extent than controls and this allowed the patient to correctly solve the task. This is also seen in the congenital (or developmental) form of prosopagnosia (CP), which refers to the lifelong impairments in recognizing people by their faces (Duchaine, Parker, & Nakayama, 2003; Rivalta, Schmalzl, Coltheart, & Palermo, 2010; 2017). Despite face recognition difficulties CPs often (Humphreys, Avidan, & Behrmann, 2007; Lee, Duchaine, Wilson, & Nakayama, 2010; Palermo et al., 2011; Rivalta et al., 2010), albeit not always (Biotti & Cook, 2016), show typical emotion recognition. Thus, AP and CP cases suggest that holistic coding is necessary for identification but not for expression, and that emotion recognition might rely on featural, rather than holistic, coding (Tanaka, Kaiser, Butler, & Le Grand, 2012). From the theoretical point of view, a direct way to test this hypothesis is to check whether APs show typical or atypical CFE for emotions; atypical CFE for emotions would suggest that (atypical) holistic mechanisms for facial expressions of emotions are not sufficient to cause

facial emotion recognition deficits (Palermo et al., 2011). Unfortunately, this evidence is lacking in AP.

Thus, we here describe the case of ST, a 48-year-old Italian woman who, following right occipital brain damage, shows serious everyday-life difficulties in identifying familiar faces, albeit with typical ability to recognize facial expressions of emotions. The first aim was to investigate whether the impairment in face identification was associated with deficits in holistic processing (i.e., CFE for identity) (Experiment 1). In addition, to test whether facial expressions of emotions are processed holistically or via a part-based processing, we adopted the CFE for emotions (Experiment 2) (Calder & Jansen, 2005; Calder, Keane, Young, & Dean, 2000).

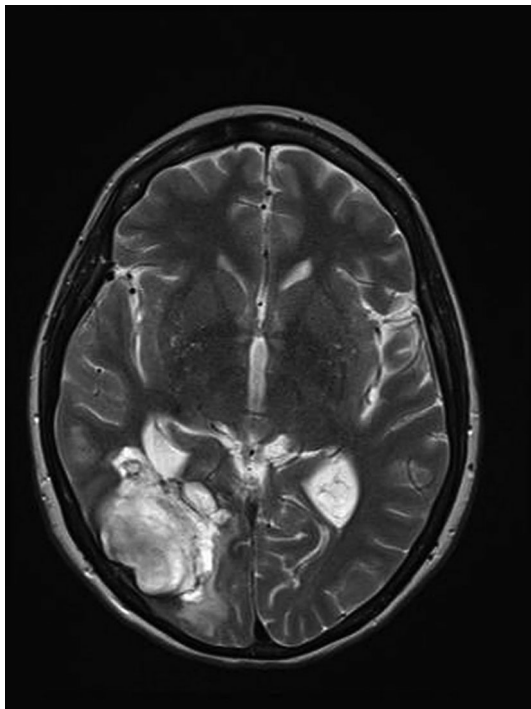
According to her neuropsychological profile (see below), we hypothesized that ST was sensitive to holistic interference (i.e., CFE) when she had to judge emotional expression, but not when the task required judging facial identity. This pattern would suggest a dissociation between the two cognitive processes, which arises earlier than holistic perceptual processing. However, considering the evidence of an interaction of emotion and identity mechanisms at low-level stages of processing in healthy subjects (Fisher, Towler, & Eimer, 2016), an alternative hypothesis would be that ST had impaired holistic processing for both emotion *and* identity judgements. If this was the case, her spared ability to categorize emotions from facial information despite severe difficulties in identity recognition could be attributed to the use of compensatory strategies (feature-by-feature processing), or, more broadly, it might suggest that holistic perception is not essential for expressions recognition, which primarily relies on part-based processing (Tanaka et al., 2012).

## 2. Methods and results

### 2.1. ST: case description

ST is a right-handed woman with 17 years of education (Master's degree in economy). She was 48 years old at the time of the assessment. In September 2014, she sustained a right occipital haemorrhage associated to thrombosis of the transverse and sigmoid sinuses. TC scan obtained in October 2014 revealed the presence of a haemorrhagic infarct in subacute phase in the occipito-temporo-parietal region of the right hemisphere. Following, a structural

MRI showed the result of the intraparenchymal hematoma in the right occipito-parietal regions (Figure 1). Consecutively, ST suffered from left homonymous hemianopia and prosopagnosia. During her hospital stay, ST complained of severe difficulties in recognition of familiar faces, such as relatives (e.g., her husband, sons) and friends. ST did not report any difficulty with face expression recognition. She lamented severe problems in recognizing familiar people, which she described as even more evident in condition of no context cues and expectations (e.g., work colleagues met outside the work place). She reported to rely on features (eye colour, earrings, beard, etc.) to overcome her face identification difficulties. ST also complained to sometimes see faces as distorted, a condition known as *prosopomorphopsia* (PM) (i.e., a rare face-selective disorder in which faces appear distorted as they were drooping, floating or bulging, Hecaen & Angelergues, 1962). Specifically, she reported that known faces appeared uglier and darker than she remembered. In the weeks immediately following the acute event, such misperceptions were persistent but not stable in terms of intensity, since they were reported as variant according to environmental conditions (i.e., brightness). In a second assessment 5 months after



**Figure 1.** Structural MRI showing ST' lesion affecting the right occipito-parietal regions (axial plane).

brain damage, ST noticed relevant improvements reporting that faces appear less swollen and ugly than earlier. At the 1-year follow up, the prosopomorphopsia has been completely recovered.

## 2.2. The diagnosis of prosopagnosia

Immediately after the acute event ST complained to experience severe everyday life difficulties in the recognition of previously known (e.g., familiars, relatives and friends) and new (e.g., professionals met at the hospital) faces. Nevertheless, she was able to recognize familiar people by the voice, clothes, and non-identity facial features (i.e., beard and glasses). Both ST and her relatives reported she never had difficulties in recognizing faces before the acute event. ST reported such condition as particularly disabling due to mistakes in recognition of people who were even highly familiar to her, such as her husband and sons. She was completely aware and worried for such difficulties, which dramatically affected her everyday life.

With the aim to formally assess ST's face recognition abilities, and to "diagnose" prosopagnosia, we administered three tasks: (1) The *recognition of familiar faces* was examined by means of an experimental task requiring identification of ST's relatives, friends and colleagues; (2) the ability to *learn new faces* has been assessed with the Cambridge Face Memory Test (CFMT; Duchaine & Nakayama, 2006), which is one the most adopted and valid test to diagnose prosopagnosia (Bowles et al., 2009); and (3) *face perception* has been assessed with the computerized version of the Glasgow Face Matching Test (GFMT, Burton, White, & McNeill, 2010), a perceptual task that requires matching of same-identity, but perceptually dissimilar, faces. Concurrently, an extensive neuropsychological assessment was conducted in order to exclude the presence of perceptual or high-order cognitive deficits. ST's performance was compared to that of typical subjects by means of a *t*-test for small samples (SINGLIMS *t*-test; Crawford, Garthwaite, & Porter, 2010).

### 2.2.1. Familiar face recognition

From the historical perspective, prosopagnosia has been defined as the inability to recognize people that were familiar before the traumatic event (Hecaen & Angelergues, 1962). As such, we examined ST's ability to recognize 38 personally familiar faces, which included her son, daughter, husband and

relatives (three pictures of the patient's own face have also been included). All extra-face details (e.g., hair, clothing, jewellery) were cropped using the Open Source software GIMP. Each stimulus was presented on a white background until response. ST was asked to identify each individual face by trying to name it. In case of difficulty, we asked ST to describe whether faces seem familiar or not. All ST's verbalizations were registered. Overall, ST correctly recognized 50% of the shown faces. The patient showed a lot of uncertainties and a very long time to answer (over 3 sec). She was *not able to recognize her own face*, which was wrongly attributed to her father. Frequently, ST reported a failure to feel a sense of familiarity; however, when recognition was possible, naming occurred almost immediately with no hesitations. When asked, ST reported to rely on specific outstanding details, which were described as particularly salient (i.e., shape of eyes, their colour etc.). Albeit not specifically asked in the task, the patient also showed difficulties in gender and age attribution: she reported to rely on cues such as the presence of wrinkles to describe the face as depicting old or young person. As a control, ST's husband has also completed the task and was able to correctly identify all the faces. Taken together, this finding strongly confirmed the presence of prosopagnosia.

### 2.2.2. Unfamiliar face learning

The CFMT examines immediate memory for unfamiliar faces, requiring learning of target faces from different views. After a practice stage, subjects have to complete three phases: (i) introduction: recognition of six previously learned faces among distractors (test items with identical images); (ii) novel images: recognition of the target faces in which the test items are novel images (i.e., the lighting, pose or both vary); (iii) novel images with noise: recognition of the target faces presented with different levels of Gaussian noise. ST's accuracy (60%) was significantly lower ( $t = -2.02$ ,  $p = .02$ , one-tailed) than control data ( $N = 20$ ; mean: 82.7%; SD: 7.6; mean age: 45.1) published by Duchaine, Yovel, and Nakayama (2007) (Table 1). This result suggests impairment in learning and recognizing new faces.

### 2.2.3. Face matching

Face identification relies upon the fundamental ability to perceive a face as the same even when it is seen

with different lighting conditions, distance and head orientation (i.e., *within*-identity changes). Such ability refers to the structural encoding process, which allows for the construction of a view-dependent representation of the face (Bruce & Young, 1986). Here we used the Glasgow Face Matching Test (GFMT, Burton et al., 2010) in order to observe whether ST's difficulties in face recognition encompass the low-level phase of structural encoding. The GFMT (short version) measures the ability in matching two simultaneously presented faces that have the same frontal viewpoint but are subject to within-identity change (White, Rivolta, Burton, Al-Janabi, & Palermo, 2017). This introduces within-identity variations in the aspect and metric distances between images of the same individual. Each of the 40 face pairs (20 same pairs; 20 different pairs) was shown until response. On each trial, participants are required to indicate whether the two images were of the same or of different person by pressing the correspondent key on a keyboard. The experiment was run with E-Prime 2.0.

As compared with normative data (Mean = 81.2%; SD = 9.7;  $N = 194$ ; mean age: 26, age range: 18–46) provided in Burton et al., 2010, ST's accuracy performance (60%) was impaired ( $t = -2.250$ ,  $p = .01$ , one-tailed) (Table 1). RTs were not analysed since ST showed excessively long time for response (mean correct RTs: 9.3 sec). Overall, performance on the GFMT suggests early, perceptual, problems in face recognition, possibly characterizing her prosopagnosia as *apperceptive* in nature (De Renzi, Faglioni, Grossi, & Nichelli, 1991).

**Table 1.** ST's performance on standardized measures of face processing and facial expression recognition as compared to controls.

<i>Face processing</i>		
	Score	<i>p</i> value
CFMT	43/72	.02*
GFMT	24/40	.01*
<i>Facial expression recognition</i>		
Ekman 60 Faces Test		
	Score	Cut-off
Suprise	10/10	6
Happiness	10/10	9
Fear	4/10	2
Disgust	5/10	4
Anger	6/10	5
Sadness	8/10	4
Total score	43/60	

Note: CFMT = Cambridge Face Memory Test; GFMT = Glasgow Face Matching Test.

\* SINGLIMS one-tailed significant *p* values.



### 2.2.4. Facial expression recognition

ST's ability to recognize emotions from facial expressions was tested with the Ekman & Friesen 60 Faces Test (Young, Perrett, Calder, Sprengelmeyer, & Ekman, 2002), which examines the recognition of six basic facial expression (i.e., anger, disgust, fear, happiness, sadness, surprise). The test is composed of 60 trials preceded by a practice of 6 trials. Stimuli belong to the Ekman & Friesen set of Pictures of Facial Affect (1976) and appear on the screen for 5 sec each. Participants had to choose between six emotion labels, which remain on the screen for the same trial duration. Responses were verbal. ST's global score (raw score: 43/60; corrected score: 41.05/60, adjusted for age, gender and education) is within normal limit (equivalent score = 1) as compared to a sample of Italian subjects ( $N = 132$ , mean age 51.11, SD 16.5; Dodich et al., 2014) (Table 1).

### 2.2.5. General neuropsychological evaluation

A first neuropsychological assessment was conducted three weeks after injury. ST' spatial, temporal and

personal orientation were adequate. She performed within the normal range in several tasks assessing spatial cognition, praxis abilities, abstract reasoning, executive functions, verbal long-term and short-term memory and attention. Moreover, she performed well in tests evaluating colour discrimination and recognition. See Table 2 for a summary of results.

Further, a detailed evaluation of low-level visual perception and objects recognition abilities was conducted in order to investigate the presence of objects agnosia, which is frequently described in patients with AP (Clarke, Lindemann, Maeder, Borruat, & Assal, 1997). First, the Leuven Perceptual Organization Screening Test (L-POST) (Torfs, Vancleef, Lafosse, Wagemans, & de-Wit, 2014) was administered to screen for possible deficits in mid-level vision. The L-POST is composed of 15 subtests that cover a range of mid-level processes such as figure-ground segmentation, local and global processing, and shape perception (see Torfs et al., 2014, for a detailed description of each subtest). ST's overall score was 93.34% (70 out of 75 correct); the performance was

**Table 2.** Results of the general neuropsychological assessment.

Cognitive domain	Test	Raw score	Cut-off
Visuo-spatial abilities	Line bisection (horizontal) (Fortis et al., 2010)	2.09	5.75
	Cancellation task—Bells Test (Gauthier, Dehaut, & Joannette, 1989)	0	5
	Sentence reading (Zoccolotti et al., 1989)	0	1
	Vertical reading	18/18	–
	Line bisection (vertical)	6/6	–
Ideomotor apraxia	Imitation of gestures (De Renzi, Motti, & Nichelli, 1980)	71	52
Buccofacial apraxia	Imitation of orofacial movements (Spinnler & Tognoni, 1987)	20/20	–
Nonverbal executive functions	Clock Drawing Test (Mondini, Mapelli, Vestri, & Bisiacchi, 2003)	9.5	8
Numerical abilities	E.N.P.A.		
	<i>Numbers repetition</i>	10/10	8.8
	<i>Dictation</i>	10/10	6.3
	<i>Words-numbers</i>	7/7	4.2
	<i>Calculations—sum</i>	3/3	2.2
	<i>Calculations—subtraction</i>	3/3	1
	<i>Calculations—multiplication</i>	4/4	1.4
Cognitive domain	Test	Raw score	Equivalent score
Praxis abilities	Rey-Osterrieth Complex Figure—copy (Caffarra, Vezzadini, Dieci, Zonato, & Venneri, 2002)	30	0
Nonverbal executive functions	Weigl Sorting Test (Laiacina, Inzaghi, De Tanti, & Capitani, 2000)	12	3
Abstract reasoning	Standard Progressive Matrices (Spinnler & Tognoni, 1987)	29/48	1
Lexical access	Phonemic verbal fluency (Spinnler & Tognoni, 1987)	40	4
	Semantic verbal fluency (Spinnler & Tognoni, 1987)	42	3
Verbal short-term memory	Numerical span (Monaco, Costa, Caltagirone, & Carlesimo, 2013)	6	3
Verbal working memory	Reverse numerical span (Monaco et al., 2013)	5	4
Verbal long-term memory—Immediate recall	List of 15 words (Carlesimo, Caltagirone, Fadda, Marfia, & Gainotti, 1995)	67/75	4
	Prose memory (Spinnler & Tognoni, 1987)	46.5	4
Verbal long-term memory—Delayed recall	List of 15 words (Carlesimo et al., 1995)	15/15	4
Visuo-spatial long-term memory	Rey-Osterrieth Complex Figure—recall (Caffarra et al., 2002)	7	0
Attention	Attentive matrices (Spinnler & Tognoni, 1987)	48	2

Raw scores and equivalent or cut-off scores are provided. For the equivalent score: 0 = pathological performance; 1 = borderline performance; 2–4 = performance within the normal range (see Capitani & Laiacina, 1997 for psychometric details).

Abbreviations: E.N.P.A = Esame neuropsicologico per l'afasia (Capasso & Miceli, 2001).

**Table 3.** Low-level vision and object processing evaluation: results.

Cognitive domain	Test	Raw score	Cut-off
Low-level visual abilities	L-POST		
	<i>Fine shape discrimination</i>	5/5	4
	<i>Dot lattices</i>	5/5	4
	<i>RFP contour integration</i>	5/5	4
	<i>Shape ratio discrimination (Efron)</i>	5/5	4
	<i>Figure ground segmentation</i>	5/5	4
	<i>Kinetic object segmentation</i>	5/5	5
	<i>RFP texture surfaces</i>	4/5	4
	<i>RFP fragmented outline</i>	4/5*	5
	<i>Biological motion</i>	5/5	3
	<i>Recognition of missing part</i>	5/5	4
	<i>Global Motion Detection</i>	5/5	4
	<i>Dot counting</i>	2/5*	4
	<i>Recognition of objects in isolation</i>	5/5	5
	<i>Embedded figure detection</i>	5/5	3
	<i>Recognition of object in a scene</i>	5/5	5
		<i>Total score</i>	70/75
Perceptual processing and space perception	Visuospatial judgment (Benton et al., 1978)	26	19
	VOSP		
	<i>Screening test</i>	20	15
	<i>Incomplete letters</i>	20	17
	<i>Silhouettes</i>	16	16
	<i>Object decision</i>	18	15
	<i>Progressive silhouettes</i>	15	14
	<i>Dot counting</i>	9	8
	<i>Position discrimination</i>	20	18
	<i>Number location</i>	6*	7
	<i>Cube analysis</i>	7	6
	BORB		
	<i>Lenght match (version A)</i>	24/30	23,7
	<i>Lenght match (version B)</i>	21/30	
	<i>Size match (version A)</i>	23/30	22,5
	<i>Size match (version B)</i>	27/30	
	<i>Orientation match (version A)</i>	25/30	19,6
<i>Orientation match (version B)</i>	24/30		
Verbal naming	Figure naming (AAT)	40/40	
Cognitive domain	Test	Raw score	Equivalent score
Visual-perceptual analyses	Overlapping Figures (Della Sala et al., 1995)		
	<i>Concrete</i>	32/36	1
	<i>Abstract</i>	15/35	0
	<i>Total score</i>	47/71	0
Colour processing	Ishihara Test for Colour Blindness		
	<i>Numbers</i>	21/25	17
	<i>Paths</i>	9/13	
	Farnsworth-Munsell 28 HueColor Vision test (Farnsworth, 1943)		normal
	Colour naming (AAT subtest)	7/7	
	Colour semantic knowledge	6/6	

Raw scores and equivalent or cut-off scores are provided. For the equivalent score: 0 = pathological performance; 1 = borderline performance; 2–4 = performance within the normal range (see Capitani & Laiacona, 1997 for psychometric details).

Abbreviations: L-POST = Leuven Perceptual Organization Screening Test; RFP = Radial frequency pattern; VOSP = Visual Object and Space Perception battery; BORB = Birmingham Object Recognition Battery; AAT = Aachener Aphasia Test.

below the cut-off in the Dot counting and Radial frequency pattern (RFP) fragmented outline subtests. Furthermore, the Overlapping Figures Test (Della Sala, Laiacona, Trivelli, & Spinnler, 1995) was used to examine visual-perceptual analyses. ST's score was below the normal range due to selective difficulties in meaningless patterns. Nevertheless, at the Visual Object and Space Perception battery (VOSP, Warrington & James, 1991) the patient performed well within the typical range in all subtests evaluating perceptual processing and object perception. Regarding space perception, ST's scores were in the normal

range except for some difficulties in the number location task, likely to be attributed to the presence of hemianopia. Adequate visuospatial analysis was confirmed from the results at the Birmingham Object Recognition Battery (BORB, Riddoch & Humphreys, 1993) length match, size match and orientation match subtests, together with the Visuospatial Judgment test score (Benton, Varney, & deS Hamsher, 1978). Moreover, verbal naming was preserved (see Table 3 for a summary of the results).

Taken together, ST's neuropsychological performance indicates intact abilities in perceptual

organization and space perception, except for some difficulties in abstract shapes discrimination and isolated difficulties in dot counting, likely to be attributed to left hemianopia. Results from perceptual and naming tasks suggest that structural encoding and recognition of visually presented stimuli are unimpaired. ST does not show any impairment in basic-level object recognition. Moreover, a tactile recognition task revealed no impairments in recognizing objects either through non-visual modality (accuracy 10/10).

### 2.2.6. Conclusions

A formal evaluation of ST' face recognition abilities strongly indicates the presence of a severe impairment in the (i) recognition of previously known personally familiar faces, (ii) ability to learn and remember new faces (CFMT), (iii) ability to match unfamiliar faces (GFMT). Besides the presence of visual field reduction (left homonymous hemianopia), ST did not show visual-perceptual defects, achromatopsia, objects agnosia, and topographical disorientation, which are known to be frequently associated in patients with face recognition difficulties (Barton, Press, Keenan, & O'Connor, 2002; Bouvier & Engel, 2006; Clarke et al., 1997; Gauthier, Behrmann, & Tarr, 1999). Overall, ST's face-selective impairment, together with the results from the neuropsychological assessment and clinical observations indicate a diagnosis of a "pure" acquired prosopagnosia.

## 2.3. Holistic processing measures

### 2.3.1. Identity composite experiment

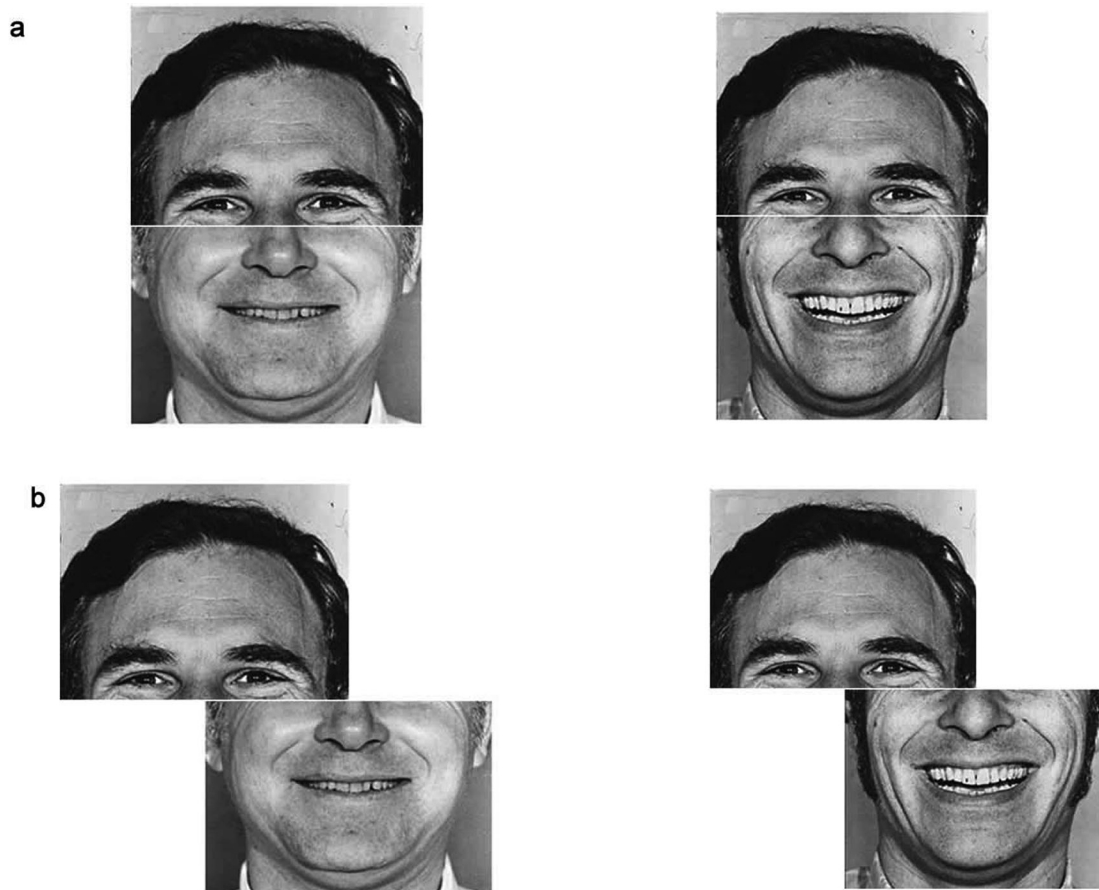
**2.3.1.1. Participants.** Eleven healthy subjects were tested (6 females; mean age:  $41 \pm 13$ ).

**2.3.1.2. Materials and methods.** Stimuli were selected from the Ekman 60 Faces Test (Young et al., 2002). Images of eight individuals were chosen (4 males; 4 females). All the faces displayed the same expression (i.e., happiness). Images were separated into top and bottom halves by cutting stimuli horizontally at the bridge of the nose; the two halves were separated by a 5 pixel gap (see Ramon & Rossion, 2012, for a similar procedure). By combining top and bottom parts we obtained a set of 32 composite stimuli. Besides eight individuals originally selected, which constituted *congruent* stimuli (i.e., top and

bottom parts with same identity), twenty-four were *incongruent* stimuli (i.e., top and bottom parts of different individuals with the same gender). The same set of aligned stimuli was then arranged to create a misaligned one. Misaligned stimuli were formed by horizontally shifting the bottom part to the left or to the right of the top one by about half of the image width. Thus, 64 stimuli were obtained (32 aligned; 16 with left bottom misalignment; 16 with right bottom misalignment). On each trial, two face stimuli (S1 – S2) were shown sequentially. S2 was maintained congruent (top and bottom belonged to the same identity) while we randomly varied the congruency of S1 (congruent S1: top and bottom of the same identity; not congruent S1: top and bottom of different identities). Thus, we obtained "same" and "different" conditions. For the "same" condition, according to the congruency of S1, the trial interval randomly showed: (i) same top halves/same bottom halves (congruent S1) or (ii) same top halves/different bottom halves (not congruent S1). Experimental variables were aligned/misaligned; same/different response; S1 congruent/ not congruent; response to the top/bottom halves (see Figure 2 for an example of stimuli).

**2.3.1.3. Procedure.** The experiment was run by using E-prime 2.0. Participants were seated at approximately 43 cm viewing distance from the monitor. Stimuli were presented on a white background. On each trial two composite faces (S1–S2) were sequentially displayed and participants were requested to judge whether either the top or the bottom halves of the two stimuli showed the same or different identities, by pressing the corresponding keys. Each trial showed two aligned or misaligned composite faces. The side of misalignment (left; right) was randomized between trials. Each aligned stimulus was 10.8 (width)  $\times$  15.2 cm (height) ( $13.1 \times 19.4^\circ$  in visual angle, viewed from a distance of 43 cm). Each misaligned stimulus was 16.1 (width)  $\times$  15 cm (height) ( $21.4 \times 19.4^\circ$  in visual angle, viewed from a distance of 43 cm). Reaction times and accuracy were recorded. At the beginning of each trial a fixation cross was presented for 1000 ms at the centre of the screen, followed by S1 (150 ms), a second fixation cross (150 ms) and S2 (150 ms). Participants had a maximum of 3500 ms for response after S2 disappearance. Trials were separated by 1000 ms interstimulus





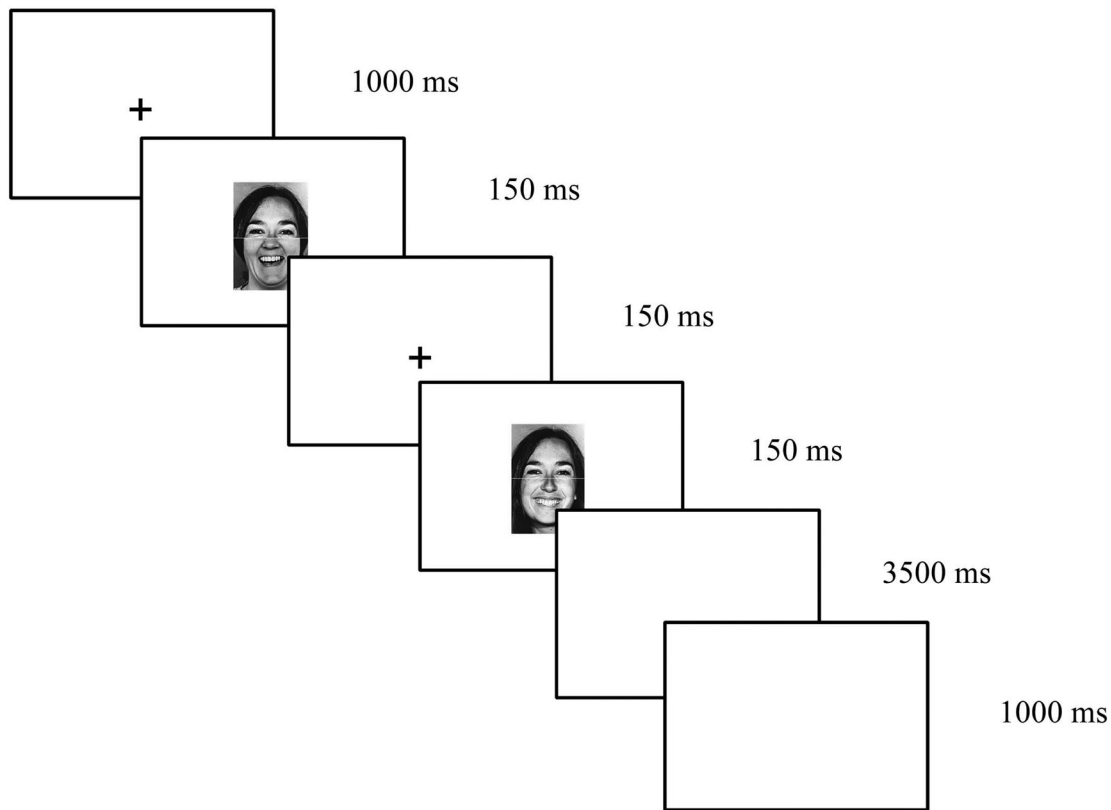
**Figure 2.** *Identity composite experiment: example of stimuli.* The figure shows an example of aligned (a) and misaligned (b) “same” trial, top target condition. The participants have to judge if the top halves of two sequentially presented faces (prime and target; left and right in the Figure) belong to the same or to different individuals.

interval (ISI) (Figure 3). Participants completed four blocks of 64 trials differing with respect to the part they have to classify, either the top or the bottom target. Blocks order was fixed for each participant (bottom, top, bottom, top). Aligned and misaligned trials were randomized within blocks.

**2.3.1.4. Analyses.** In line with Rossion (2013), analyses were conducted only considering the “same” condition. Since the trial “congruent S1—congruent S2” with same top halves actually showed two identical faces, they were not considered in the analyses. Therefore, analyses were conducted on critical condition’s trials (i.e., incongruent S1—same response trials). We performed a *group-level analysis* using mixed effects models. Mixed effects models allow accounting for both inter-subject and intra-subject variability, increasing the reliability of the model (Bates, 2015). These analyses were performed on a *single trial* basis, for both experiments (identity and emotional expressions) separately, comparing controls’

performance to ST’s (both in the initial testing and in the 18-months follow-up).

The analyses were conducted by using generalized mixed effects models (on a binary distribution) on accuracy (coded as 0–1), and linear mixed effects models on response times (RTs). All models included *alignment* (2 levels: aligned vs. misaligned), *top–bottom* (TB) (2 levels: target in top vs. bottom of the face), and *condition* (2 levels: controls vs. ST) as independent fixed factors (in a full factorial design), and random intercepts. In models on accuracy, *p*-values were obtained by “models comparison method” (i.e., by comparing the deviance in two models—with vs. without the effect of interest—in a chi-squared test). In the model on RTs, only correct trials (accuracy = 1) were considered. Outliers exceeding 2 SD from the mean were excluded (mean and SD were computed on each subject). *P*-values, in all post-hoc multiple comparisons, were adjusted according to the Tukey HSD method. Only main effects and interaction involving the condition factor (main variable of interest) are reported below.



**Figure 3.** Identity composite experiment: trial procedure. Here is shown an example of same-top target condition: two same top are paired with different bottom halves (aligned, S1 not congruent).

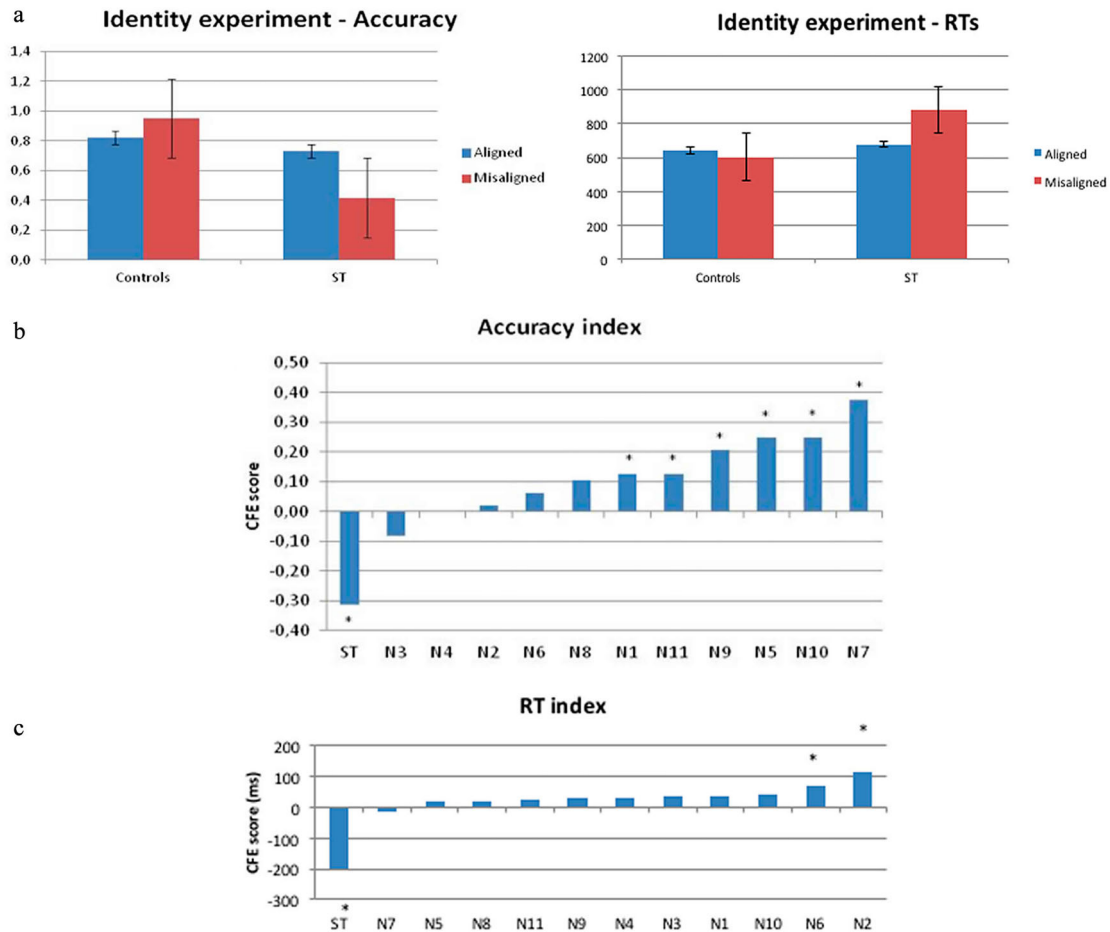
Moreover, we ran single-subject analyses by comparing aligned and misaligned trials for each control subject and ST (*Single-subject analyses*). We here used *linear models* (LM) for RTs and *generalized linear models* (GLM) on binomial distribution for accuracy. Alignment and top/bottom target were considered as independent within-subjects variables. RTs associated with correct responses were cleaned at the individual-subject level by removing trials above or below 2 SD from the subject's mean.

### 2.3.1.5. Results

**2.3.1.5.1. Single-subject analyses.** *Accuracy:* in 9 out of 11 subjects the numerical difference is in the direction expected for healthy participants (i.e., they are more accurate in misaligned rather than aligned trials); of these, 6 show a statistically significant misaligned vs. aligned difference (Figure 4b). *RTs:* in 10/11 subjects the numerical difference between conditions is in the expected direction (i.e., they are faster in misaligned than aligned trials) and 2 of these show a statistically significant misaligned vs. aligned difference (Figure 4c). ST,

instead, showed a “reversed” CFE (i.e., she is faster and more accurate in aligned, rather than misaligned, trials), which is statistically significant in terms of both accuracy ( $\chi^2(1) = 9.803, p = .002$ ) and RTs ( $F(1,48) = 8.599, p = .005$ ). See Figure 4a for a comparison between controls' average scores (accuracy and RTs) and ST performance in aligned and misaligned conditions.

**2.3.1.5.2. Group-level analyses.** *Accuracy:* the model showed a statistically significant main effect of condition:  $\chi^2(1) = 5.658, p = .017$ ; and a significant alignment \* condition interaction ( $\chi^2(1) = 36.644, p < .001$ ). The main effect of condition showed that, on average, control participants ( $m = 0.88$ ) were more accurate than ST ( $m = 0.57$ ) in the task. In the interaction effect, post-hoc comparisons based on the *alignment* showed that ST was more accurate in aligned ( $m = 0.73$ ) than misaligned ( $m = 0.42; z = 3.104, p = .002$ ) trials, while healthy participants were more accurate in misaligned ( $m = 0.95$ ) than aligned ( $m = 0.82; z = -6.099, p < .001$ ) trials (i.e., ST thus shows an opposite effect of alignment) (Figure 5a). Comparisons based on the *condition* showed that ST



**Figure 4.** Identity composite experiment: single-subject analyses results. a: average observed accuracy scores (left column) and average observed RTs (right column) in the aligned and misaligned condition for both ST and controls. b: the figure shows accuracy face composite (CFE) indexes (resulting from comparison between the aligned and misaligned condition) for each participants alongside with ST. c: the figure shows RTs face composite indexes (resulting from comparison between the aligned and misaligned condition) for each participants alongside with ST. Asterisks indicates significant differences between aligned and misaligned trials at the single-subject level. Bars represent standard errors.

presented significantly lower accuracy than healthy participants ( $z = -4.497$ ,  $p < .001$ ) in misaligned trials, while, for aligned trials, ST and healthy participants did not show any difference ( $z = -0.778$ ,  $p = .431$ ) (Figure 5a).

**RTs:** the model showed a significant interaction effect of alignment \* condition ( $F(1, 918.83) = 13.085$ ,  $p < .001$ ) (Figure 5b). Post-hoc comparisons showed that while controls were slower in aligned trials ( $m = 628$  ms) than in misaligned ones ( $m = 599$  ms) ( $t(924) = 3.155$ ,  $p = .002$ ), ST was slower in misaligned ( $m = 881$  ms) than aligned ( $m = 679$  ms) trials ( $t(924) = -3.002$ ,  $p = .003$ ). Comparisons based on the condition showed that ST presented a *trend* towards statistical significance for slower RTs compared to healthy participants ( $t(13) = 2.126$ ,  $p = .054$ ) in misaligned trials, while, for aligned trials, ST and healthy

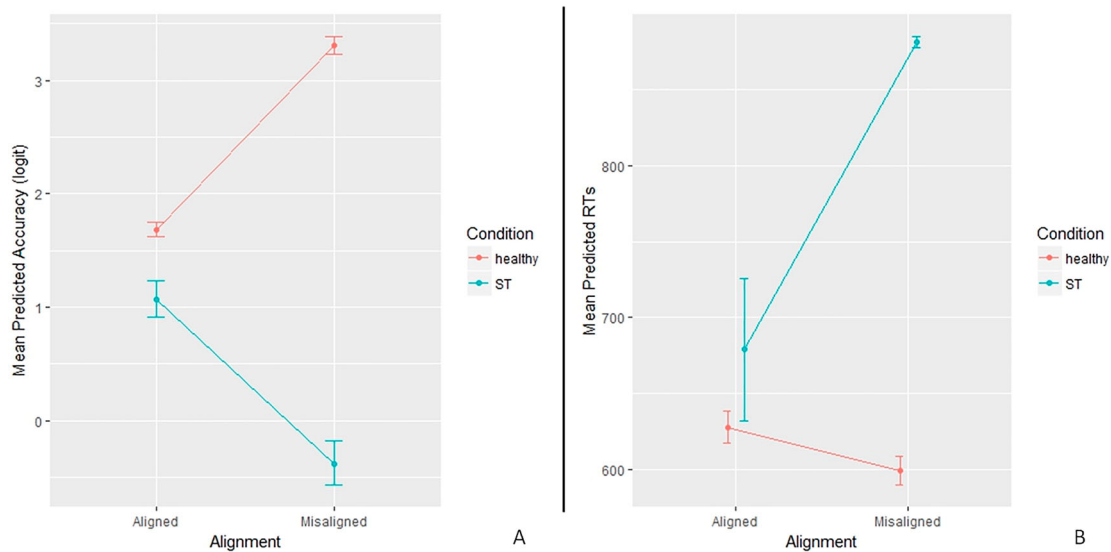
participants did not show any difference ( $t(11) = 0.443$ ,  $p = .666$ ) (Table 4 for results).

### 2.3.2. Emotional expression composite experiment

**2.3.2.1. Participants.** Thirteen healthy subjects were tested (12 females; mean age:  $39 \pm 13$ ).

**2.3.2.2. Materials and methods.** Stimuli were selected from the Ekman & Friesen 60 Faces Test (Young et al., 2002). We chose photographs depicting four different individuals (two males and two females), each one showing three distinct facial expressions. According to Baudouin and Humphreys (2006) we selected expression of happiness, anger and fear since happiness and anger are easy to be discriminated both from the top and the bottom part (see Baudouin & Humphreys, 2006, for a comprehensive description).

## IDENTITY TASK



**Figure 5.** Identity composite experiment: group-level analyses results. In this and following plots: dots represent the mean predicted value and error bars represent 95% confidence intervals; the blue line represents ST's scores, while the red line represents average control participants' scores. Panel A: Plot representing the alignment \* condition interaction effect on participants' accuracy. Panel B: Plot representing the same effect on participants' reaction times.

Moreover, such expressions are well recognized by our patient (see results from Ekman & Friesen test).

For each of the four individuals selected, all possible combinations were created, obtaining 36 composite

**Table 4.** ST's and control's performance at the Identity composite (Experiment 1) and Emotional expression composite (Experiment 2) tasks.

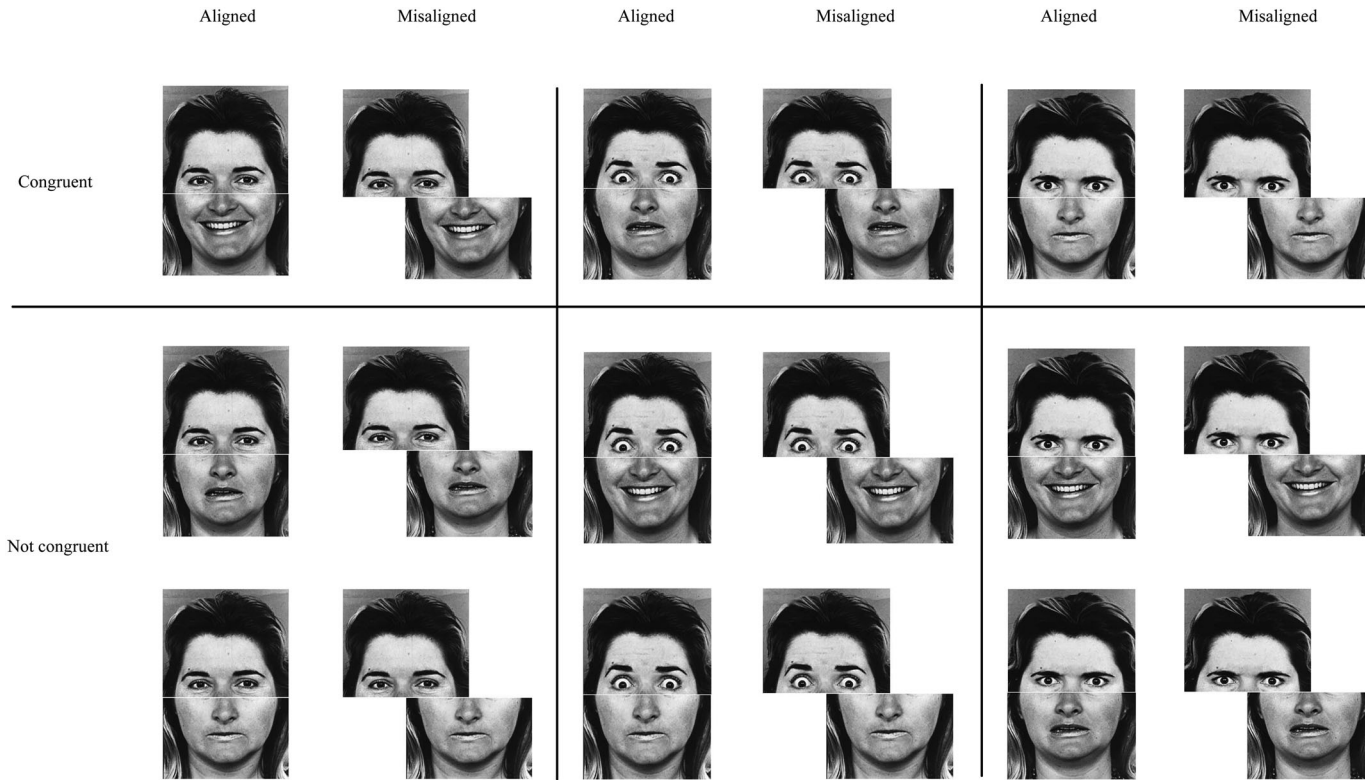
		a. Top target.					
		Testing			Follow up		
Identity composite							
Accuracy				RTs		Accuracy	
	ST	Controls	ST	Controls	ST	ST	ST
Aligned	62,5	76.1 (42.7)	836	672 (238)	50	624	
Misaligned	58,3	97.7 (14.9)	875	620 (233)	70,8	686	
Emotional expression composite							
Accuracy				RTs		Accuracy	
	ST	Controls	ST	Controls	ST	ST	ST
Aligned	75,0	76.0 (42.8)	803	790 (330)	75,0	623	
Misaligned	41,7	92.3 (26.7)	864	709 (317)	66,7	625	
		b. Bottom target.					
Identity composite							
Accuracy				RTs		Accuracy	
	ST	Controls	ST	Controls	ST	ST	ST
Aligned	83,3	87.5 (33.1)	569	593 (223)	87,5	549	
Misaligned	25,0	92 (27.1)	892	578 (202)	54,2	667	
Emotional expression composite							
Accuracy				RTs		Accuracy	
	ST	Controls	ST	Controls	ST	ST	ST
Aligned	95,8	80.4 (39.7)	672	722 (360)	91,7	583	
Misaligned	58,3	92.0 (27.2)	835	749 (296)	75,0	731	

Means and standard deviations (in brackets) are showed. Left column: testing phase; Right column: follow-up evaluation. The Table shows results from the top-target (Table 4a) and bottom-target condition (Table 4b).

stimuli (12 congruent, 24 incongruent). Each *incongruent* composite depicted a face constituted by two halves of the same individual showing different emotions: one on the top half and a different one on the bottom half (e.g., happiness on the top and fear on the bottom). *Congruent* composites were made by two halves showing a unique emotion expressed by the same individual. A correspondent misaligned set of stimuli was created through the same procedure as in Experiment 1. See Figure 6 for an example of stimuli.

**2.3.2.3. Procedure.** Experimental procedure was the same as in Experiment 1. To avoid possible perceptual interference generated from identity information, in each trial faces of the same individual were shown, which could be congruent or incongruent for emotional expression. Subjects were required to judge whether the two upper or lower halves of the faces showed the same or different emotions. We stressed that they did not have to judge the identity of the faces; rather they had to attend to the emotional expression by ignoring the identity. Participants completed four blocks of 80 trials (response to the top; bottom; top; bottom).

**2.3.2.4. Analyses.** Analyses were the same as for Experiment 1.



**Figure 6.** *Emotional expression composite experiment: example of stimuli.* A matrix example of stimuli used. Each pair displays the aligned and the correspondent misaligned version. Here are shown stimuli for the congruent (upper row) and not congruent (second and third row) conditions. Congruent stimuli depicted a face showing a unique facial expression of the same identity (from left to right: happiness; fear; anger). Not congruent stimuli are composite faces in which the upper and lower halves have same identity but differ for the expressed emotions. From left to right, second row: top happiness-bottom fear; top anger-bottom happiness; top anger-bottom happiness. From left to right, third row: top happiness-bottom anger; top fear-bottom anger; top anger-bottom fear. Four different face identities were used.

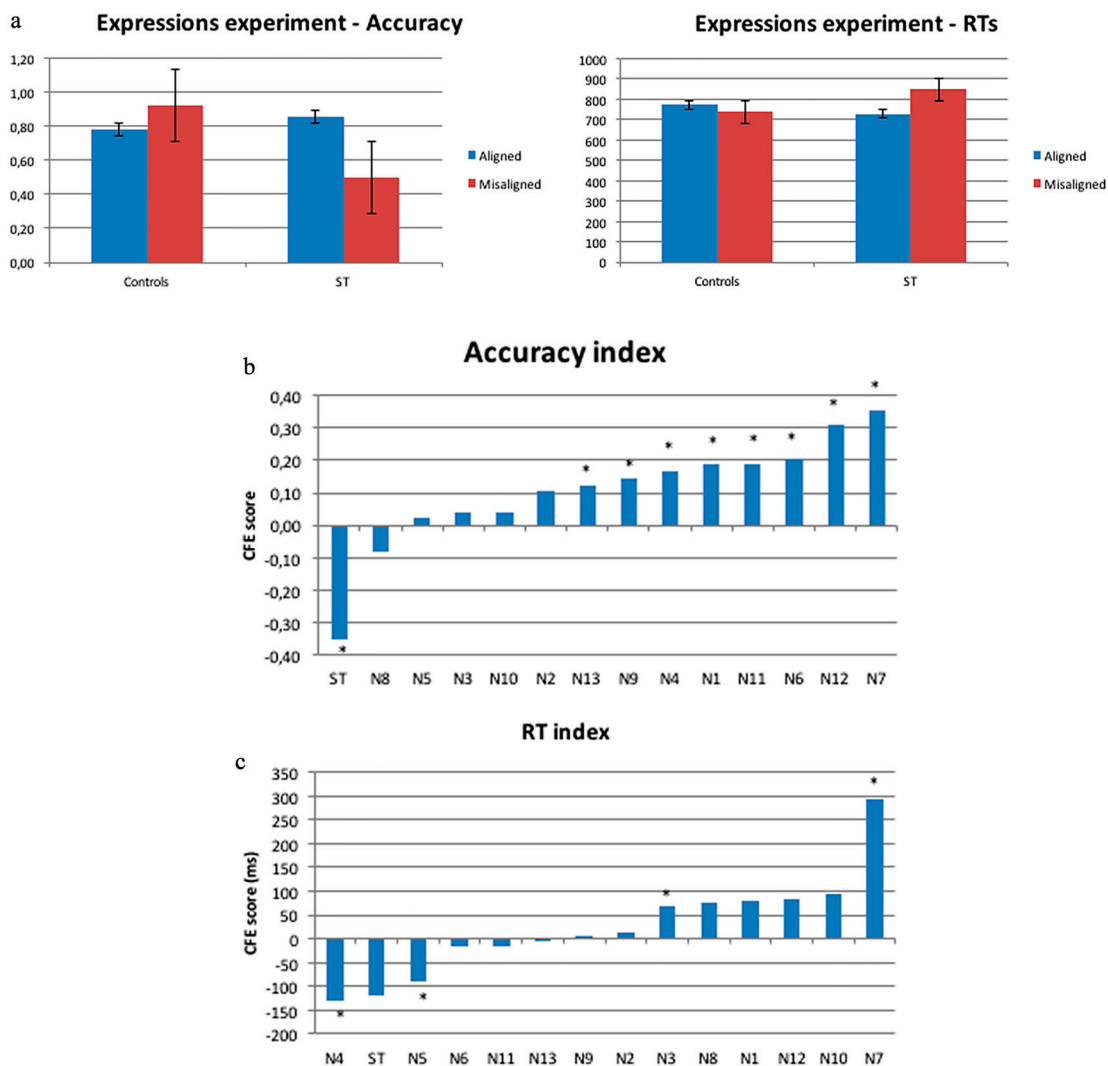


### 2.3.2.5. Results

**2.3.2.5.1. Single-subject analyses.** *Accuracy:* in 12 out of 13 subjects the numerical difference is in the expected direction (“typical” CFE); 8 out of 12 show a statistically significant misaligned vs. aligned difference. ST showed a statistically significant reversed CFE ( $\chi^2(1) = 15.033, p = .001$ ) (Figure 7b). See Figure 7a for a comparison between controls’ average scores (accuracy and RTs) and ST performance in aligned and misaligned conditions.

*RTs:* in 8/13 subjects there is a typical CFE (2 of these show a statistically significant misaligned vs. aligned difference). ST showed a reversed CFE in a trend toward significance ( $F(1,57) = 3.435, p = .07$ ) To note,

that also two controls displayed a statistically significant “reversed” CFE (Figure 7c). However, when looking at both accuracy and RTs, we note that these subjects showed a trade-off between accuracy and correct RTs (i.e., they were faster in aligned trials, but they were better in misaligned trials; subject N4, RTs: aligned 518ms, misaligned 647ms; accuracy: aligned 0.83, misaligned 1. Subject N5, RTs: aligned 455ms, misaligned 546 ms; accuracy: aligned 0.83, misaligned 0.85). It is also important to note that the CFEs for RTs and accuracy have opposite directions for subjects N4 and N5; whereas this is not evident for ST, who showed a congruent reversed-CFE for both accuracy and RTs (Figure 7b and c).



**Figure 7.** Emotional expression composite experiment: single-subject analyses results. a: average accuracy scores (left column) and average RTs (right column) in the aligned and misaligned condition for both ST and controls. b: the figure shows accuracy face composite (CFE) indexes (resulting from comparison between the aligned and misaligned condition) for each participants alongside with ST. c: the figure shows RTs face composite indexes (resulting from comparison between the aligned and misaligned condition) for each participants alongside with ST. Asterisks indicates significant differences between aligned and misaligned trials at the single-subject level. Bars represent standard errors.

To take into account the above-mentioned trade-offs, we have computed a global measure of CFE that combines accuracy rates and correct RTs (see Busigny & Rossion, 2010, for a similar procedure applied to the face inversion effect in a case of AP) and we have compared ST with the control's mean using SINGLIMS *t*-test for small samples (Crawford et al., 2010). To do this, we calculated the Inverse Efficiency score (IE) for each participant and for ST ( $IE = RTs/accuracy$ ) then we calculated the CFE index using the following formula:  $(IE_{aligned} - IE_{misaligned}) / (IE_{aligned} + IE_{misaligned})$ . Our results showed that ST has a significantly lower CFE index ( $-0.33$ ) in comparison to the control participants (mean:  $0.1$ ;  $t(12) = -265.8$ ,  $p < .001$ , two-tailed).

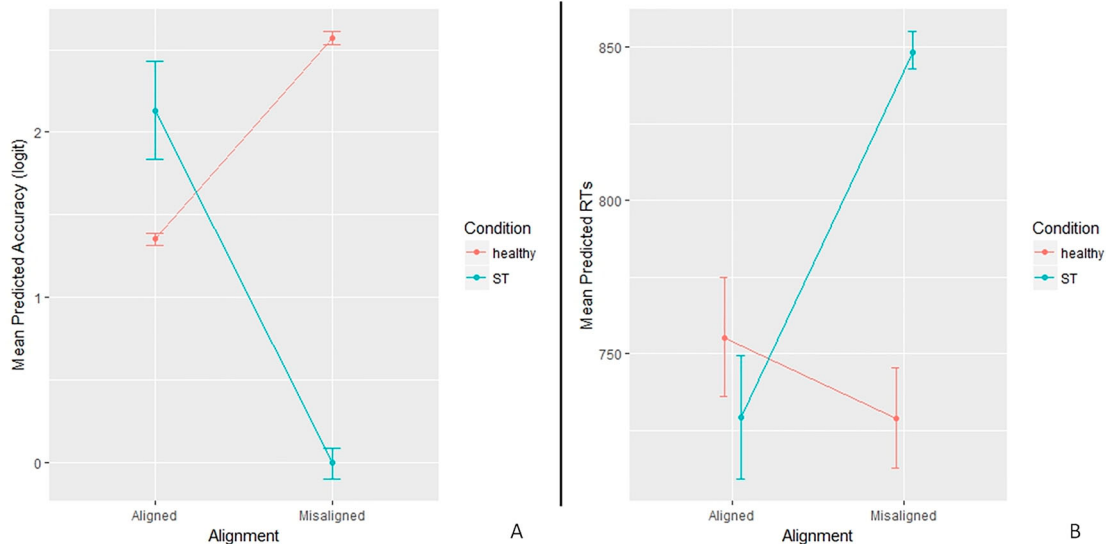
**2.3.2.5.2. Group-level analyses. Accuracy:** the model showed a statistically significant alignment \* condition interaction:  $\chi^2(1) = 38.3$ ,  $p < .001$  (Figure 8a). Post-hoc comparisons based on alignment showed that ST was more accurate in aligned ( $m = 0.85$ ) than misaligned ( $m = 0.50$ ;  $z = 3.361$ ,  $p < .001$ ) trials, while healthy participants were more accurate in misaligned ( $m = 0.92$ ) than aligned ( $m = 0.78$ ;  $z = -6.780$ ,  $p < .001$ ) trials. Comparisons based on the condition showed that ST presented significant lower accuracy than healthy participants ( $z = -3.916$ ,  $p < .001$ ) in misaligned trials, while, for aligned trials, ST and healthy participants did not show any difference ( $z = -0.944$ ,  $p = .345$ ).

**RTs:** The model on participants' RTs showed a significant alignment \* condition interaction ( $F(1, 1043.28) = 4.142$ ,  $p = .042$ ) (Figure 8b). Post-hoc comparisons based on the alignment showed that ST did not present a statistically significant difference between aligned ( $m = 729$  ms) and misaligned trials ( $m = 848$  ms) ( $t(1046) = -1.640$ ,  $p = .101$ ), while healthy participants showed a trend towards shorter RTs in misaligned ( $m = 729$  ms) than aligned trials ( $m = 755$  ms) ( $t(1046) = 1.904$ ,  $p = .057$ ). Comparisons based on the condition did not highlight any statistical significant difference between ST and healthy participants, neither in aligned ( $t(12) = -0.137$ ,  $p = .893$ ) or in misaligned trials ( $t(13) = 0.493$ ,  $p = .630$ ) (Table 4 for results).

### 2.3.3. Control analyses

ST's neuropsychological assessment highlighted left hemianopsia. It is thus possible that ST's problems shown in the CFE (identity and emotions) are due to a low-level deficit rather than holistic processing impairments. This is unlikely since the offset of the misaligned face was equally distributed between left and right. However, to exclude potential low-level effects on ST's performance we ran additional eight control analyses focusing on the effect of trials N-1. In both Experiments, aligned and misaligned trials were interleaved. Thus, on trial N participants may fixate on the top (or bottom) half of an aligned face.

## EMOTION TASK



**Figure 8.** Emotional expression composite experiment: group-level analyses results. Panel A: Plot representing the alignment \* condition interaction effect on participants' accuracy. Panel B: Plot representing the same effect on participants' reaction times.

On trial N + 1 the misaligned face may be offset to the left or to the right-hand side. Consequently, there is the possibility that the patient fixated in the same location during the two trials' presentation and, in the trials where the misaligned face was offset to the left side, the task-relevant part of the face is more likely to be in the blind visual field.

For this reason, we separately analyzed: (i) trials in which participants view an aligned trial followed by a misaligned trial (a condition in which the patient may fixate in the same location), and (ii) trials in which a misaligned trial is preceded by another misaligned trial (in this latter case the patient might be inclined to fixate on the location of the misaligned top face half). We expected that, if the hemianopia is playing a role in these results, the performance should be worse in aligned trials that are followed by misaligned trials than on misaligned trials followed by misaligned trials. On the other side, if ST fixated on the central cross throughout, we do not expect to find such a difference. Control analyses were performed for each Experiment, for both testing and follow up.

As for our other results (see Results section), the analysis was conducted on top-target only since the CFE on the bottom half is generally weaker than the CFE on the top half (Rossion, 2013). We considered trials in which the target was aligned or misaligned to the left side. Eight models were run on these data, considering accuracy and reaction times (RTs) in the Identity task (test and follow up) and in the Emotion task (testing and follow up). All models performed on RTs were linear models, while all models performed on the accuracy were general linear models on a binomial distribution, in which the statistical significance of each effect was tested by using likelihood ratio tests (LRTs). Each model included the following independent variables: Alignment (2 levels: Aligned vs. Misaligned Left), N-1 Alignment (3 levels: Aligned, Misaligned Left, Misaligned Right), and their interaction. Outliers exceeding 2 SD from the mean were excluded from RTs.

Results showed that none of the main effects of N-1 Alignment or the interactions N-1 Alignment \* Alignment resulted as statistically significant. Such findings allowed us to exclude possible effects on the performance due to ST's left hemianopia (see Supplementary Material for further details).

### 3. Follow up

At the follow up examination (18 months after the acute event) ST reported persisting difficulties in familiar face recognition, which were still evident in the clinical setting. Her prosopometamorphosis was almost completely recovered since she reported to do not experience facial misperception as before. The reduction of the left hemifield was still present at the neuroophthalmological visit. ST was retested with the CFE, for both identity and emotions, and the Ekman 60 Faces Test (Young et al., 2002). Additionally, we used the *face inversion effect* (FIE) paradigm (Yin, 1969) in order to examine ST with a further measure of holistic processing.

Moreover, four statistical analyses were run to test possible differences in ST's performance in the Identity and Expressions experiments between the first measurements and follow up (*Analyses pre-post*). Two generalized linear models (on a binomial distribution) were used to test ST's accuracy in both identity and emotional expressions tasks, while two linear models were used to test ST's RTs in the same tasks. All these models included alignment, TB and time (2 levels: pre vs. post) as independent factors, in a full factorial model. RTs from incorrect trials or exceeding 2 SD from the mean were excluded from the analyses.

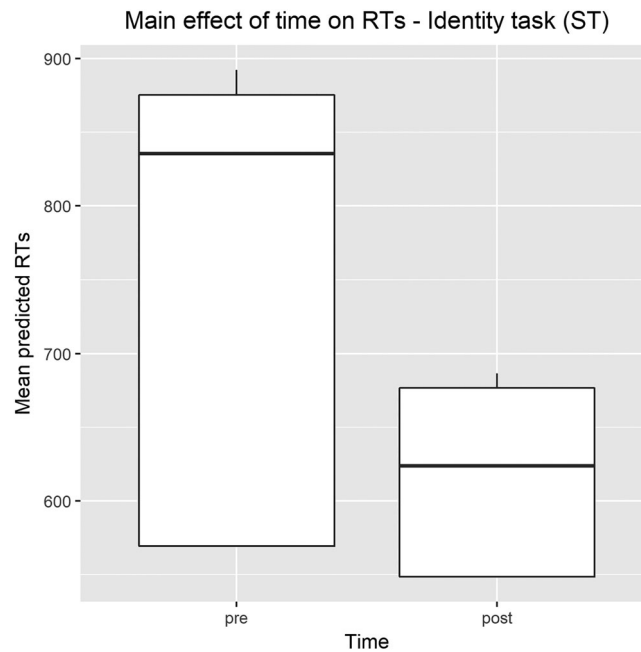
#### 3.1. Identity composite experiment

##### 3.1.1. Analyses pre-post

*Accuracy:* The analyses did not show any statistically significant effect involving the factor time (all  $\chi^2 < 2.5$ , all  $p > .1$ ). *RTs:* The analyses on RTs highlighted a statistically significant main effect of time:  $F(1, 103) = 11.24$ ,  $p = .001$  (identity task) (Figure 9). RTs in the first measurement (mean = 749 ms) were slower than in the follow up experiment (mean = 622 ms). No other interaction effects including the factor time resulted as statistically significant (all  $F_s < 2.1$ , all  $p_s > .15$ ).

##### 3.1.2. Group-level analyses

*Accuracy:* The model on participants' accuracy showed a statistically significant main effect of condition:  $\chi^2(1) = 4.0273$ ,  $p = .045$ ; and a significant alignment \* condition interaction:  $\chi^2(1) = 15.235$ ,  $p < .001$  (Figure 10a). The main effect of *condition* showed that, on average, control participants ( $m = 0.88$ ) were more accurate than ST ( $m = 0.66$ ) in the task. In the



**Figure 9.** Identity composite experiment: Boxplot representing ST's RTs during the first measurement (after the acute phase) and during the follow-up, in the identity composite experiment. This plot highlights the statistically significant main effect of the factor Time in the linear model.

interaction effect, post-hoc comparisons based on the *alignment* indicated that ST did not show a statistically significant difference between aligned ( $m = 0.69$ ) and misaligned ( $m = 0.63$ ;  $z = 0.934$ ,  $p = .350$ ) trials, while healthy participants were more accurate in misaligned ( $m = 0.95$ ) than aligned ( $m = 0.82$ ;  $z = -6.099$ ,  $p < .001$ ) trials. Comparisons based on the *condition* led to the same pattern found in the first measurement: ST presented lower accuracy than healthy participants in misaligned trials ( $z = -3.402$ ,  $p < .001$ ), while for aligned trials ST and healthy participants did not differ ( $z = -0.886$ ,  $p = .375$ ).

*RTs*: the model on participants' RTs showed a significant alignment \* condition interaction ( $F(1, 929.05) = 6.0597$ ,  $p = .014$ ) (Figure 10b). Post-hoc comparisons based on the *alignment* showed that ST presented an opposite trend effect direction compared to healthy participants: while healthy participants were slower in aligned trials ( $m = 628$  ms) than in misaligned ones ( $m = 599$  ms) ( $t(931) = 3.207$ ,  $p = .001$ ), ST was slower in misaligned trials ( $m = 678$  ms) than aligned ones ( $m = 575$  ms) ( $t(931) = -1.727$ ,  $p = .085$ ). Comparisons based on the *condition* showed no differences between ST and healthy participants, neither in misaligned ( $t(11) = 0.572$ ,  $p = .579$ ) or in aligned trials ( $t(11) = -0.470$ ,  $p = .648$ ). See Table 4 for results.

## 3.2. Emotional expression composite experiment

### 3.2.1. Analyses pre-post

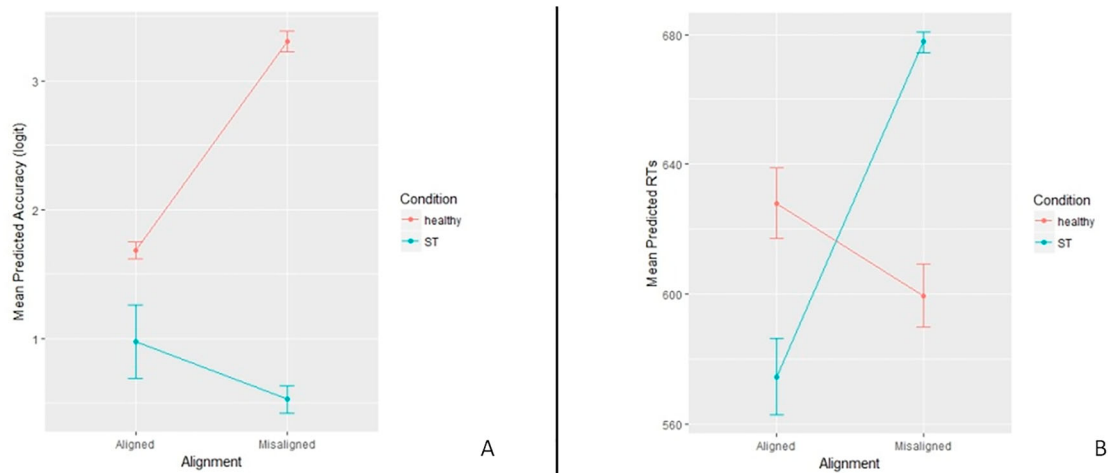
*Accuracy*: The analyses did not show any statistically significant effect involving the factor time (all  $\chi^2 < 2.5$ , all  $p > .1$ ). *RTs*: The analyses on RTs highlighted a statistically significant main effect of time:  $F(1, 123) = 14.60$ ,  $p < .001$  (Figure 11). RTs in the first measurement (mean = 772 ms) were slower than in the follow up experiment (mean = 637 ms). No other interaction effects including the factor time resulted as statistically significant (all  $F_s < 2.1$ , all  $p_s > .15$ ).

### 3.2.2. Group-level analyses

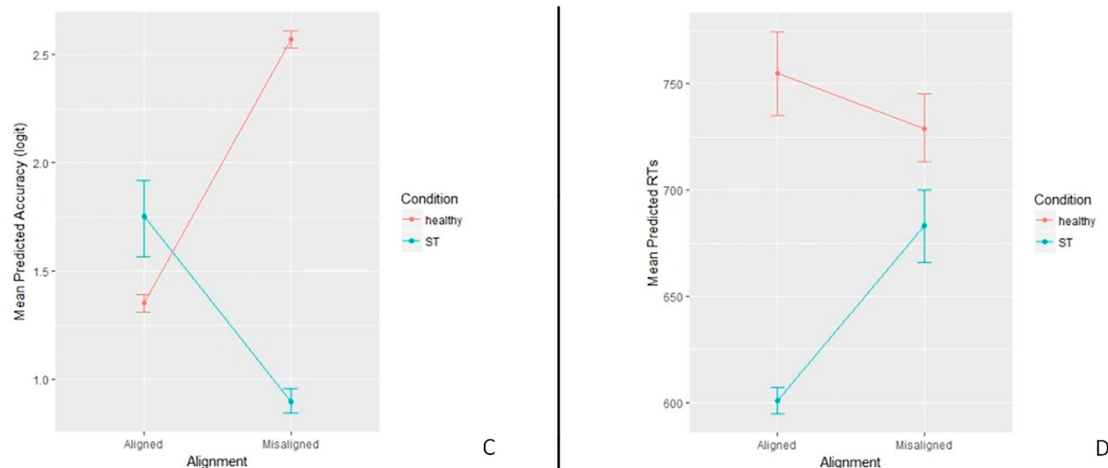
*Accuracy*: The model on participants' accuracy showed a significant alignment \* condition interaction ( $\chi^2(1) = 14.213$ ,  $p < .001$ ) (Figure 10c). Post-hoc comparisons based on the *alignment* showed the same pattern found in the identity task: ST did not present a significant difference between aligned ( $m = 0.83$ ) and misaligned ( $m = 0.71$ ;  $z = 1.581$ ,  $p = .114$ ) trials, while healthy participants were more accurate in misaligned ( $m = 0.92$ ) than aligned ( $m = 0.78$ ;  $z = -6.780$ ,  $p < .001$ ) trials. Comparisons based on the *condition* led to the same pattern found in the first measurement: ST presented significantly lower accuracy than healthy

# FOLLOW UP

## IDENTITY TASK



## EMOTION TASK



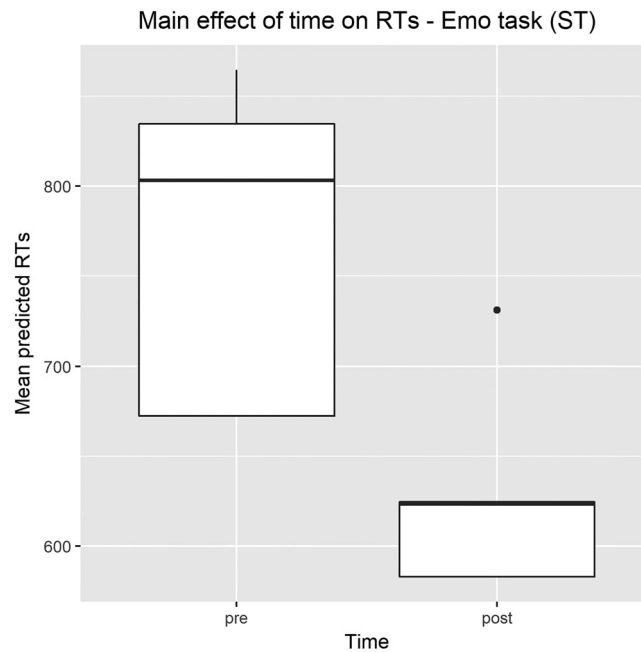
**Figure 10.** Follow up experiments: Panel A and B: Plots representing the alignment \* condition interaction effect on participants' Accuracy (A) and reaction times (B) in the follow up identity composite experiment. Panel C and D: Plots representing the same effect on participants' Accuracy (C) and reaction times (D) in the follow up emotional expression composite experiment.

participants ( $z = -2.496, p = .013$ ) in misaligned trials, while, for aligned trials, ST and healthy participants did not show any statistically significant differences ( $z = 0.543, p = .587$ ).

**RTs:** the model on participants' RTs showed a trend towards significance in the alignment \* condition interaction ( $F(1, 1055.98) = 2.809, p = .094$ ) (Figure 10d). Even though this effect is not strictly statistically significant, multiple comparisons were performed to compare this effect with the same effect in the first measurement. Post-hoc multiple comparisons on the

interaction effect did not lead to clear differences, still reflecting the same pattern found in the first measurement. Comparisons based on the alignment showed that ST did not present a statistically significant difference between aligned ( $m = 601$  ms) and misaligned trials ( $m = 683$  ms;  $t(1053) = -1.226, p = .220$ ), while healthy participants showed a trend towards shorter RTs in misaligned ( $m = 729$  ms) than aligned trials ( $m = 755$  ms) ( $t(1053) = 1.927, p = .054$ ). Comparisons based on the condition did not highlight any significant difference between ST and healthy participants, neither





**Figure 11.** Emotional expression composite experiment: Boxplot representing ST's RTs during the first measurement and during the follow-up, in the emotional expression composite experiment. This plot highlights the statistically significant main effect of the factor Time in the linear model.

in aligned ( $t(12) = -0.737$ ,  $p = .475$ ) or in misaligned trials ( $t(13) = 0.265$ ,  $p = .795$ ) (Table 4 for results).

### 3.3. Ekman 60 faces Test

ST's expression recognition ability was at normal range (raw score: 48/60; corrected score: 46.05/60, adjusted for age, gender and education; equivalent score: 3; Dodich et al., 2014).

### 3.4. Face inversion experiment

#### 3.4.1. Material and methods

Stimuli were selected from the Face Database (Minear & Park, 2004). Thirty-two stimuli were chosen (16 females; 16 males) and divided in four age range subsets (18–29; 30–49; 50–69; 70–94 years old). All stimuli were modified in greyscale and cropped to remove extra-face stimuli. For each trial a fixation cross was presented at the centre of the screen (1000 ms), followed by the target (3000 ms), a fixation cross (2000 ms) and a slide with two simultaneously presented probes (3000 ms). Participants were instructed to select one of the two faces (left or right) matching the one previously shown, by pressing the corresponding key. The experiment was divided in four blocks of 32 trials

differing for stimuli orientation, for a total of 128 trials. Blocks order was fixed for each participant (upright, inverted, upright, inverted). The experiment was run with E-Prime 2.0. A face inversion score was calculated as following: *inverted-upright* for correct RTs, and *upright-inverted* for accuracy. ST's performance was compared to that of five healthy subjects (3 females; mean age:  $44 \pm 13$ ) by means of a *t*-test for small samples (SINGLIMS *t*-test; Crawford et al., 2010).

#### 3.4.2. Results

ST's face inversion score for RTs was significantly lower as compared to the controls' average score (ST: 20.2; control's mean: 78.7, SD = 32.7;  $t(4) = -2.761$ ,  $p = .03$ , one tailed). ST's face inversion score for accuracy did not differ from the control's (ST: 3.1; control's mean: 2.2, SD = 4.1;  $t(4) = .20$ ,  $p = .42$ , one-tailed). Thus, although accurate, ST failed to show an upright superiority for RTs, as opposite to control subjects. It is known that correct RTs are a highly sensitive measure of the effects of face inversion in normal observers (Rossion, 2008).

To conclude, severe difficulties in face recognition were still present at the follow up evaluation. As for the initial assessment, ST showed reduced identity-CFE. The absence of face inversion effect further

confirmed the presence of atypical holistic processing in our patient. Moreover, she still showed atypical expressions-CFE despite spared emotion recognition ability. As compared to the initial assessment, a decrease in ST' reaction times was observed for both Identity and Expressions experiments, congruent with a general improvement which could be ascribed to a spontaneous recovery or to the adoption of perceptual strategies.

#### 4. General discussion

In the present study we examined the sensitivity to the CFE for both identity and facial expressions in a case of AP (ST) who was severely impaired in identity, albeit not in emotion recognition. First, we assessed whether the impairment in face recognition was associated with the lack of perceptual integration of face parts into a whole. To this aim, we examined ST's sensitivity to the CFE for identity processing (Experiment 1). Second, we tested the expressions-CFE to ascertain whether her spared facial expressions recognition relies on holistic mechanisms or, rather, on "compensatory" part-based strategies (Experiment 2).

Results from Experiment 1 (Identity composite) showed a "typical" composite face effect in the control group, characterized by better performance for misaligned than aligned trials. The difference in the performance between matching two same top halves when their different bottom halves are aligned Vs. misaligned is considered as an index of perceptual integration of the irrelevant part with the attended one (Rossion, 2013). This (typical) integration, where *misaligned* trials are better and faster processed than *aligned*, is seen in controls only. ST, on the other hand, showed an opposite pattern (i.e., better performance on aligned trials), thus suggesting atypical holistic coding. The reduced face-inversion effect for ST emerged in the follow-up examination provided further support to such interpretation. Congruently, performance on the GFMT (Burton et al., 2010) revealed severe difficulties when ST had to match faces varying for within-identity changes, which might be ascribed to a low-level perceptual difficulty characterized by weak holistic integration of face features. Our results are in line with evidences of weak or atypical CFE in APs (Busigny et al., 2010; Ramon et al., 2010). However, findings indicating typical sensitivity to the CFE in APs (see Finzi et al.,

2016; Rezlescu et al., 2012) and in CPs (Biotti et al., 2017), pointed out some variability in these populations which can be due to different experimental manipulations or, in the case of APs, to different location/size of brain damage.

The presence of an atypical CFE in a case of AP is consistent with the proposal that holistic processing contributes to efficient face recognition. The link between holistic coding and face recognition ability was demonstrated in a study of DeGutis et al. (2013): using a regression-based approach, authors found that both part-whole and composite measures showed robust correlations with the CFMT. As authors suggested, it should be noted that holistic processing is not the unique contributor to efficient face recognition, since holistic face processing and non-holistic processing were found to independently predict face recognition ability. Furthermore, in support to the role of holistic coding in face recognition, results from Richler, Cheung, and Gauthier (2011) indicated that holistic processing predicted face recognition abilities on the CFMT and in a task of face identification.

The second aim of the current study was to examine whether typical facial expressions recognition in our patient relies on holistic mechanisms or, rather, it might be supported by "part-based" strategies. Results from Experiment 2 (Facial expression composite) are similar to those from Experiment 1 (Identity expression composite): the control group showed a misalignment advantage (i.e., a typical CFE), whereas ST had an opposite pattern (i.e., better performance on aligned trials).

Taken together, results from the two composite tasks demonstrate that holistic processing difficulties for both identity and facial expressions can be involved in a patient with AP, and that these difficulties cannot be explained by low-level (i.e., left hemianopsia) problems (see Supplementary Material). To the best of our knowledge, there are only few studies examining CFE for facial expressions in APs. Interestingly, one study examining the sensitivity to the composite interference in a patient with AP (HJA) showed a residual ability to judge facial emotions although it was qualitatively different with respect to the controls (Baudouin & Humphreys, 2006). Specifically, HJA did not show a configural interference effect for facial emotion judgements, leading authors to suppose that he relied on the analyses of critical local features.

Nevertheless, the task involved high-order categorization abilities given that it required naming emotions. Thus, to date, little is known on how facial expressions are processed at a low-level of perceptual processing in patients with face recognition impairments. Furthermore, even though HJA seemed not to benefit from configural information for face discrimination, he was not formally assessed with a CFE paradigm for identity processing.

Moreover, our results showed that facial emotions recognition can be spared in a patient with altered holistic coding for facial expressions. Actually, results from the emotion recognition task (Ekman 60 faces test) and the Facial expressions composite task indicated that ST was able to recognize facial expressions even in the presence of atypical holistic processing for emotions. We suppose that ST's residual ability in recognizing facial expressions might be ascribed to the adoption of part-based, compensatory strategies, based on a piecemeal analysis of face parts. Considering ST's severe face recognition difficulties, our findings seem to suggest that alternative, part-based, mechanisms did not support efficient identity recognition in our case. The presence of typical emotion recognition despite reduced expressions-CFE may also indicate the possibility that emotion recognition is not necessarily mediated by holistic processing. Congruently, studies in healthy participants showed that expression and identity rely on different processing styles, with expressions judgement relying on a more piecemeal analysis of local features (e.g., Tanaka et al., 2012). Even though some studies provided data in favour of holistic processing for facial expressions in healthy individuals (e.g., Calder et al., 2000; Calder & Jansen, 2005), other studies have demonstrated that recognition of facial expressions involves analytic processing (Blais et al., 2012; Bombari et al., 2013; Eisenbarth & Alpers, 2011; Scheller, Büchel, & Gamer, 2012; Smith, Gosselin, Cottrell, & Schyns, 2005).

It is still debated the extent to which emotion and identity processing are independent or not (e.g., Calder & Young, 2005). We reported a dissociation between identity and facial expressions recognition in a patient with AP; however, such segregation was only evident at a high semantic level (i.e., recognition and naming phase), thus suggesting an initial stage of holistic integration common to both identity and emotion processing. The fact that the two

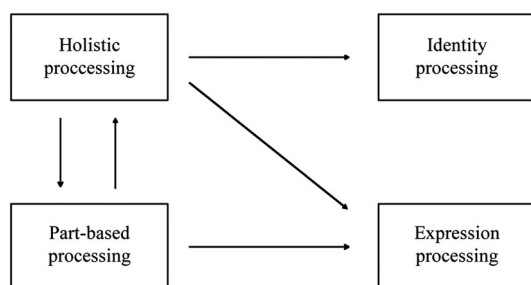
mechanisms diverge, at least at a certain level, provides support to neuropsychological case studies showing double dissociations (i.e., prosopagnosics with identification deficit and preserved emotion recognition, e.g., Riddoch et al., 2008; Tranel et al., 1988). Vs. patients with impaired emotional but not identity recognition e.g., Young, Newcombe, de Haan, Small, & Hay, 1993), and is also in line with cognitive and neural models on anatomical and functional distinct systems subserving emotions and identity processing (e.g., Bruce & Young, 1986; Haxby et al., 2000). Identity and emotions processing dissociations were also observed in CPs individuals, in which the facial expressions recognition (e.g., Duchaine et al., 2003, 2007; Palermo et al., 2011) and matching (Fisher, Towler, & Eimer, 2017) is generally unimpaired, despite their face recognition difficulties. CPs also showed atypical holistic processing measured with different tasks (e.g., Avidan, Tanzer, & Behrmann, 2011; DeGutis, Cohan, Mercado, Wilmer, & Nakayama, 2012; Duchaine et al., 2007) and they were found to adopt a part-based processing in the identity matching of external and internal features (Towler, Fisher, & Eimer, 2018).

However, what remains controversial is whether the dissociation between identity and emotion processing reflects early perceptual mechanisms or later stages requiring memory and semantic-related retrieval information. From a behavioural perspective, our findings support a model in which a common stage of emotion and identity processing exists, at early perceptual stages. Congruently, a strong interaction between the two mechanisms was found in healthy individuals (Fisher et al., 2016); here, early perceptual symmetrical interference effects between identity and emotions have been reported (i.e., the unattended identity and the unattended expression, which were varied orthogonally, both interfered with the performance in the expression and the identity matching tasks, respectively). Event-related potentials (ERPs) recording of the N250r components revealed that identity and expression processing interact at an early stage, when visual representations are matched with those stored in face memory (Fisher et al., 2016).

Our results strictly parallel findings of CPs, with impaired identity recognition but spared expression recognition in the presence of weakened holistic processing of both expression and identity (Palermo et al.,

2011). This is consistent with a model proposing an early, holistic coding stage for multiple facial attributes (Model B, see Palermo et al., 2011) in which the split between expression and identity coding occurs after a common stage of holistic processing. Authors suggested that CPs used atypical compensatory strategies for expressions recognition, such as an over-reliance on non-holistic cues. Here we propose to extend such model by integrating a stream of featural processing which can interact with holistic mechanisms (Figure 12). It has, in fact, been demonstrated that facial emotion recognition recruits dissociable, but interactive dorsal and ventral routes in the core face processing network (Meaux & Vuilleumier, 2016): the regions involved differ with respect to the sensitivity to holistic, as compared to analytic facial coding, and their engagement was modulated by task demands. In particular, holistic information activated fusiform, inferior occipital areas and amygdala, whereas the perception of local features mainly engaged STS and prefrontal areas.

These findings corroborated a *dual-code view* in which emotion processing may be neither strictly holistic nor featural, and that the relative contribution of such processes varies between different expressions (Meaux & Vuilleumier, 2016). This allows overcoming a strictly dichotomous segregation between analytic and holistic coding of facial information. It is plausible that expression coding in healthy individuals is mediated by an interaction between holistic and part-based mechanisms and that the extent to which



**Figure 12.** Model of processing for identity and expressions mechanisms (adapted from Palermo et al., 2011). An early, perceptual holistic coding is common to identity and expression processing. Holistic processing and part-based analyses interact at an early stage of analyses. However, whereas holistic processing seems to be essential to an adequate identity recognition, expression coding is likely mediated by both holistic and part-based mechanisms. When holistic processing is disrupted or altered, part-based mechanisms can help achieving expression, but not identity recognition (or in a lesser extent).

these processes are engaged depends on task or expression type (Meaux & Vuilleumier, 2016). However, by considering our findings and those observed in CPs (Palermo et al., 2011), when holistic processing is weak or damaged, featural processing compensates for emotions, but not for identity recognition. Moreover, ST's atypical holistic processing is congruent with occipito-temporal damage, whereas part-based mechanisms are subserved by STS and prefrontal regions (Meaux & Vuilleumier, 2016), which were spared in our patient. Taken together, our results are in line with other findings suggesting that holistic processing is not mandatory for accurate facial expressions recognition (Tanaka et al., 2012). Specifically, it is possible that emotion recognition relies on a continuum between featural and holistic mechanisms (Farah, 1991) depending on stimulus ambiguity and the type of emotion displayed (Tanaka et al., 2012).

It has to be noted that, although difficulties in recognizing facial expressions are described as relative uncommon in CPs (e.g., Humphreys et al., 2007; Lee et al., 2010; Palermo et al., 2011; Rivolta et al., 2014b; 2010), a recent study of Biotti and Cook (2016) found CPs (particularly those individuals with apperceptive profile) exerted difficulties in classifying facial expressions. Authors investigated emotion recognition in CPs with the use of different measures. Participants were requested to make binary classifications if whole-face expressions in morphed stimuli (Experiment 1); to judge facial emotion using only the eye-region (Experiment 2); and to classify emotions from vocal affect (Experiment 3). CPs showed face-specific expression recognition deficits. Such difficulties have not been ascribed to aberrant holistic processing given that they were evident both when participants were asked to categorize emotions using cues from the whole and from the eye region in isolation. It is possible that the ability to detect difficulties in emotion recognition may be sensitive to the procedure used.

At this regard, a potential limitation of our study is the use of a single measure of emotion recognition. The Ekman 60 Faces Test is a well-known neuropsychological test assessing facial expression recognition and it is widely used in the face processing literature (see Mestry et al., 2012; Bate et al., 2015 in acquired and Palermo et al., 2011 in congenital form of prosopagnosia). Identity and emotions measures used in

our study have long stimuli presentation durations (Ekman 60 Faces: 5s; CFMT, GFMT and familiar face recognition: until subject's response) so we can suggest that they do not differ for task difficulty. However, we cannot rule out that the adoption of more sensitive tests and the analysis of RTs (which were not collected in the current study) might have highlighted subtle deficits in emotion recognition. Future studies on individuals with prosopagnosia might examine performances in different measure of facial expressions, involving for example both static and dynamic stimuli. A further potential limitation concerns the use of stimuli showing facial expression of happiness in the Identity Composite experiment. Nevertheless, since we kept constant the emotion by varying the identity (all faces showed happy expressions), it is unlikely that unexpected "interference" might have affected the data. Moreover, our subjects and ST were carefully instructed to attend solely to the identity by ignoring the emotional expressions before running the experiment. A final point of reflection concerns the sensitivity of the CFE as a "diagnostic" tool in clinical neuropsychology. With respect to the group of controls, ST showed an "opposite" CFE (i.e., aligned more accurate and faster than misaligned trials), with clear deficits in the misaligned condition. We exclude that this atypical performance might be due to problems in understanding tasks instructions since ST did not manifest any problem in understanding instructions of the (numerous) tasks adopted in the neuropsychological assessment. Critically, even one control on the composite identity (accuracy data—Figure 4), and two controls on the composite expression (RTs data—Figure 7), showed a CFE in the direction of ST's; these participants however, by definition, did not show any problem with faces. These results might indeed suggest that the composite-face task is not always valid in clinical neuropsychology, and that multiple tests should be adopted to assess holistic processing in prosopagnosia. ST's atypical inversion effect (at follow-up) further corroborates her holistic deficits.

In conclusion, our findings demonstrated that holistic processing difficulties for both identity and facial expressions can be involved in a patient with severe AP. The atypical pattern we found for both identity and expressions-CFE suggested that her altered holistic coding affected both components of perceptual judgement. Such results provide further data in

support to a common stage for identity and emotion processing at low-perceptual level. Since our patient demonstrated difficulties for identity, but not facial expressions recognition, our results might be indicative of a segregation of the two mechanisms at a high semantic level, showing that facial expression processing is achievable even with holistic coding difficulties, possibly through compensatory, part-based mechanisms.

## Disclosure statement

No potential conflict of interest was reported by the authors.

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