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Rehabilitation of Face-Processing Skills in an Adolescent with Prosopagnosia:

Evaluation of an Online Perceptual Training Programme

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Abstract

In this paper we describe the case of EM: a female adolescent who acquired prosopagnosia following encephalitis at the age of eight. Initial neuropsychological and eye-movement investigations indicated that EM had profound difficulties in face perception as well as face recognition. EM underwent 14 weeks of perceptual training in an online programme that attempted to improve her ability to make fine-grained discriminations between faces. Following training, EM's face perception skills had improved, and the effect generalized to untrained faces. Eye-movement analyses also indicated that EM spent more time viewing the inner facial features post-training. Examination of EM's face recognition skills revealed an improvement in her recognition of personally-known faces when presented in a laboratory-based test, although the same gains were not noted in her everyday experiences with these faces. In addition, EM did not improve on a test assessing the recognition of newly encoded faces. One month after training, EM had maintained the improvement on the eye-tracking test, and to a lesser extent, her performance on the familiar faces test. This pattern of findings is interpreted as promising evidence that the programme can improve face perception skills, and with some adjustments, may at least partially improve face recognition skills.

Keywords: Prosopagnosia, face recognition, rehabilitation, training, encephalitis.

Rehabilitation of Face-Processing Skills in an Adolescent with Prosopagnosia:

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Prosopagnosia is a cognitive condition characterised by a relatively selective deficit in face recognition. Traditionally, the disorder has been described in a small number of individuals who acquire face recognition difficulties following neurological injury or illness, typically affecting occipitotemporal regions (de Renzi, Perani, Carlesimo, Silveri, & Fazio, 1994; Gainotti & Marra, 2011). Although acquired prosopagnosia in its purest form is a rare condition (Gloning, Gloning, & Hoff, 1967; Zihl & von Cramon, 1986), many more individuals with brain damage are believed to experience moderate-to-severe face-processing deficits alongside other cognitive impairments (Hécaen & Angelergues, 1962; Valentine, Powell, Davidoff, Letson, & Greenwood, 2006). Further, as many as 2.9% (Bowles et al., 2009) of the population may experience a developmental form of the disorder (e.g. Bate & Cook, 2012; Duchaine & Nakayama, 2005). Hence, exploration of the remediation of prosopagnosia is an urgent clinical issue that, unfortunately, has received little attention to date.

However, this issue is complicated by findings that prosopagnosia is a heterogeneous condition that not only differs in severity (e.g. Valentine et al., 2006), but also presents with a variable pattern of cognitive symptoms (Barton, 2008; Dalrymple et al., 2011; Le Grand et al., 2006; Susilo & Duchaine, 2013). For instance, individuals with acquired prosopagnosia can sometimes present with deficits in object and shape processing, topographical disorientation, visual field defects or achromatopsia (Bouvier & Engel, 2006; Landis, Cummings, Benson, & Palmer, 1986). Further, the face-processing system itself can be disrupted at a variety of stages, resulting not only in impaired identity recognition, but also in deficits in the perception of other types of facial information, such as expression, gender or

age. These patterns of impairment can be accommodated within Bruce and Young's (1986)

functional model of face-processing, which posits that face recognition is a sequential and

hierarchical process composed of several sub-stages (see Figure 1). Specifically, the model

proposes that an initial stage of early visual analysis is followed by "structural encoding",

where view-centred representations are converted into viewpoint-independent

representations. While view-centred representations can be used to independently process

some aspects of facial information (e.g. expression or lip speech), the viewpoint-independent

representations are required for identity recognition. These representations are compared to

all stored representations of familiar faces in the face recognition units (FRUs), and, if a

match is achieved, the relevant person identity node (PIN) provides access to semantic and

biographical information that is known about that person. Finally, the name of the person is

obtained.

< Insert Figure 1 >

Bruce and Young's theoretical framework was proposed in response to experimental

work (e.g. Bruce, 1986) and diary studies using typical participants, which examined the

order of these functional stages and the types of errors that typically are (or are not) made in

face recognition (Young, Hay & Ellis, 1985). Importantly, the model has also been used to

localise face-processing impairments in individuals with acquired prosopagnosia (e.g. Young,

Hellawell, & de Haan, 1988). For instance, one might conclude that a patient presenting with

deficits in both the perception and recognition of faces has a deficit at the level of structural

encoding; whereas a patient with apparently normal face perception yet deficits in identity

recognition has a deficit at latter stages of the framework (e.g. affecting the FRUs or the

PINs). In fact, these two types of impairment are commonly seen in individuals with

prosopagnosia (e.g. de Haan, Young, & Newcombe, 1987; Young et al., 1988), and are

thought to represent functional sub-types of the condition, termed "apperceptive" and

"associative" prosopagnosia, respectively (de Renzi, Faglioni, Grossi, & Nichelli, 1991). In addition, some individuals with prosopagnosia appear to have cross-modal impairments at the level of semantics (e.g. Gainotti, Barbier, & Marra, 2003), whereby the face-processing system itself is largely unaffected but familiar face recognition is disrupted by a person-specific or more generalized semantic deficit. Although there has been some debate concerning these subtypes (and particularly whether associative prosopagnosics present with perceptual impairments: Farah, 1990), Bruce and Young's model is nevertheless an important theoretical framework that has frequently been used to localize face-processing impairments, and, less often, to subsequently inform intervention strategy (e.g. De Haan, Young, & Newcombe, 1991; Ellis & Young, 1988).

Rehabilitation of prosopagnosia

A few early studies attempted to remedy acquired cases of prosopagnosia, but met with little success. First, Ellis and Young (1988) studied an eight year-old child, KD, for a period of 18 months. KD acquired severe prosopagnosia after anaesthetic complications at three years of age. She also had object agnosia, and the underlying deficit seemed to be an inability to construct adequate representations of visual stimuli. The researchers asked KD to complete four training programmes that required (a) simultaneous matching of photographs of familiar and unfamiliar faces, (b) paired discriminations of computer-generated schematic faces, (c) paired discriminations of digitized images of real faces, and (d) the learning of face-name associations. Unfortunately, none of the programmes brought about an improvement in KD's face-processing skills.

In another study, De Haan et al. (1991) attempted to rehabilitate an adult with severe acquired prosopagnosia, PH. This individual had profound face recognition impairments, but was found to display some covert recognition on several behavioural tasks, indicating he had

a higher-level impairment that was more representative of the associative sub-type. Based on this knowledge, the authors used a category-presentation method to try to improve the patient's face-processing skills. Specifically, PH was presented with the faces of famous people who were all involved in the same occupation. The researchers informed PH about the occupation performed by that set of people, and then asked him to identify the faces. Unfortunately, PH was only successful in recognizing faces from one of the six occupational categories that was used in the study, and the improvement was not maintained in a follow-up test two months later.

More recently, Powell, Letson, Davidoff, Valentine, and Greenwood (2008) investigated the rehabilitation of face recognition deficits in 20 adults who presented with a broad range of cognitive impairments following brain injury. The participants completed three training programmes targeted at the recognition of unfamiliar faces. One programme used a semantic association technique that provided additional verbal information about faces, whereas a second programme presented caricatured versions of target faces for recognition. Finally, a part-recognition technique drew participants' attention towards distinctive facial features. The authors observed small improvements in each of the three training conditions compared to a control condition where participants were simply exposed to faces. However, when the investigation was applied to a single case of profound acquired prosopagnosia, it is of note that little or no improvement was observed following the semantic association and caricaturing programmes, but the part-recognition technique yielded 25% greater accuracy than the control condition. While this study provides promising evidence that acquired deficits in face recognition may be responsive to training, the generalizability and maintenance of the improvements were not investigated.

Although there have also been few attempts to improve face-processing in developmental prosopagnosia, those studies that have been reported have met with some

success. First, Brunsdon and colleagues attempted to improve face recognition in an eight year-old child, AL, who appeared to have a problem in face perception (Brundson, Coltheart, Nickels, & Joy, 2006). The researchers gave AL a set of 17 personally-known faces to learn on stimuli cards. Six pieces of information were written on the back of each card: the person's name, age, gender, and three defining facial features. AL was shown each card and asked to identify the face. He was given feedback about his response, and discussed how he might use the distinguishing features to identify that person. AL continued the training at home until he reached 100 per cent accuracy in recognizing the faces in four consecutive sessions. This occurred after 14 sessions, within a one-month period. Encouragingly, AL's improvement was generalized to the recognition of a new set of faces, and his family reported anecdotal evidence of improvements in everyday face recognition. Further, the improvement was maintained at a three-month follow-up.

A similar study investigated K, a four year-old girl with developmental prosopagnosia who also had a problem with face perception (Schamlzl, Palermo, Green, Brundson, & Coltheart, 2008). Before training began, the researchers monitored K's eye movements while she viewed facial stimuli, and found that she tended to focus on the external rather than the internal features of the face, and particularly avoided the eye region. Following the same programme as AL, K met the criteria to stop training at session nine. Although she still could not recognize the trained faces from novel viewpoints immediately after training ceased, an improvement was observed at the one-month follow-up. Further, eye-movement recording performed after training showed an increase in the percentage of fixations directed to the internal features of the face, and particularly the eye region. However, although this training method may improve the ability to identify trained faces and may even result in a general improvement in face perception, it remains unclear whether the programme improves the encoding and recognition of untrained faces.

One study that attempted to address this issue was carried out in an adult with developmental prosopagnosia, MZ, using a training programme that aimed to improve configural processing (DeGutis, Bentin, Robertson, & D'Espositio, 2007). Prior to training MZ demonstrated profound face recognition impairments, and, using event-related potential (ERP) measures, the authors found that the face-selective N170 response was absent. During training MZ was asked to repeatedly classify facial stimuli into one of two categories, characterised by eyebrow and mouth height. She completed two one-week training periods in which she was asked to complete 4000 trials per day. Following training, the authors reported an improvement in MZ's ability to both perceive and recognize faces, and remarkably found evidence of a selective N170 response to facial stimuli. Further, following training, the authors observed increased functional connectivity between key neural areas thought to be implicated in the face-processing system. Hence, this study provides initial evidence that intervention can actually modify the neural mechanisms that are thought to underpin face-processing, at least in developmental cases of prosopagnosia.

The current study

Although few studies have examined the rehabilitation of prosopagnosia to date, the available findings raise the possibility that it is easier to remedy face-processing deficits in the developmental compared to the acquired form of the disorder. If this is the case, it is perhaps unsurprising given the additional complexities that tend to accompany neurological trauma. Alternatively, it may be that the techniques used with the developmental cases were more successful because (a) they were more effective (or better matched to the patient's particular pattern of deficits), or (b) training was carried out over a longer period of time. Indeed, evidence from other fields suggests that improvements gained during initial rehabilitation training improve compensatory strategies, whereas improvements gained from longer-term

programmes bring about neural changes that reflect a shift to a more "normal" pattern of processing (Rijntjes & Weiller, 2002; Thirumala, Hier, & Patel, 2002).

In the current paper, we report the case of EM: a female adolescent with acquired prosopagnosia, who appears to have a deficit in face perception (i.e. at the level of structural encoding). We attempted to address the issues described above by evaluating the effectiveness of a more substantial training programme in a case of acquired prosopagnosia, using a technique that was directly targeted to the presumed locus of her impairment.

CASE REPORT

Case history

EM is a right-handed female who contracted herpes simplex encephalitis at the age of eight years and three months. EM's parents contacted our laboratory when she was 14 years of age, and described her experiences of severe prosopagnosia dating back to the onset of her illness. Prior to contracting encephalitis, EM had normal face recognition skills and no history of cognitive or developmental delays or neurological disorders. Immediately following her illness, EM experienced severe problems with both face and object recognition. For instance, her mother described how EM could not find the door or sink in her hospital room. However, her object recognition skills have now largely recovered, although she still struggles with the recognition of food and the interpretation of drawings and pictures. When EM was ready to continue her education, she had to learn to read and write again. As a result of her own determination and hard-work, EM is now only one academic year behind her correct year group, and has recently started studying towards nine General Certificates in Secondary Education (GCSEs).

We tested EM over the period of one year when she was aged 14-15 years. Her main complaint at this time was with face recognition, and she experiences a profound prosopagnosia affecting the recognition of all familiar faces (she struggles to recognize any face at all, including her own face) and the encoding of new faces. However, she reported no person-specific or more generalized semantic difficulties, and immediately recalls biographical information about a person when presented with their name. When we first met EM, she was reluctant to be left alone in public places for fear of getting separated from her family or friends, although her confidence has substantially increased since this time. A recent structural MRI scan revealed lesions in the inferior and medial right temporal lobe, extending back into the inferior, lateral and posterior right occipital lobe. Additional lesions were noted in the left fusiform gyrus, inferior left temporal lobe and inferolateral left occipital lobe (see Figure 2).

< Insert Figure 2 >

Informed consent for this study was obtained from both EM and her parents, and ethical approval for the study was granted by the departmental ethics committee at Bournemouth University. EM and 14 control participants (matched according to gender, age, and IQ, see Table 1) underwent a comprehensive battery of neuropsychological tests to assess their general intelligence, memory, visuospatial, and face-processing abilities. One control participant was excluded due to a history of psychological disorders, one was excluded for failing to pass basic visual screening, and one was excluded because her face recognition scores were exceptionally high (she scored 6.5 SDs above the control mean on the Cambridge Face Memory Test (CFMT), see below), and she is suspected of being a "super-recogniser" (Russell, Duchaine, & Nakayama, 2009). The remaining 11 participants completed the majority of the tasks, but time constraints and some computer errors meant that not all participants contributed data to all tasks (see Table 1 and descriptions of individual tasks for

details). EM's performance in the assessment tasks was compared to the control participants using modified t-tests for single case comparisons (Crawford & Garthwaite, 2002). EM's performance on the assessment tasks; means, standard deviations, and N for control participants; and results of the statistical tests are presented in Table 1.

< Insert Table 1 >

General cognitive assessment and basic visual screening

Throughout the testing, EM was alert and fully cooperative and motivated. She was fluent and had normal verbal comprehension. No ideomotor, ideative or constructional apraxia was observed. All participants, including EM, were found to have normal intellectual abilities, as indicated by performance on the two subtests version of the Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler, 1999). One control participant's IQ could not be tested on the WAIS, but she performed in the average range on the equivalent subtests taken from the WAIS-IV (Wechsler, 2008). Because autism spectrum disorder is sometimes associated with poor face-processing skills (e.g. Klin et al., 1999), social functioning was assessed in all participants using the Autism Quotient (Adolescent) (Baron-Cohen, Hoekstra, Knickmeyer, & Wheelwright, 2006), a 50-item parental questionnaire. EM and all control participants scored below published cut-offs (Baron-Cohen et al., 2006), indicating normal social functioning. EM was also administered the Children's Memory Scale (CMS; Cohen, 1997), to screen for general memory deficits. She performed within the low-average to average range on all non-face subscales.

Basic visual acuity was assessed using a standard Snellen letter chart (3m) and the Hamilton-Veale contrast sensitivity test. All participants except one demonstrated normal visual acuity and contrast sensitivity. As mentioned above, this participant's data was subsequently excluded from analysis. In addition to the basic visual screening, EM was

tested on Ishihara's tests for colour deficiency (38 plates; Ishihara, 1996), and the Fly stereo-acuity test (Vision Assessment Corporation, 2007). EM showed normal colour perception, and excellent stereo-acuity (25 seconds of arc).

Lower-level vision was assessed using four sub-tests from the Birmingham Object Recognition Battery (BORB: Humphreys & Riddoch, 1993). In the Length Match test, participants are required to judge whether two lines are of the same length; in the Size Match test they judge whether two circles are of the same size; in the Orientation Match test they decide whether two lines are parallel or not; and in the Position of the Gap Match test they decide whether the position of the gap in two circles is in the same place or not. Basic object recognition was tested using the Object Decision test from the BORB. In this test, the participant is presented with a series of line drawings that depict animals or tools. In some trials the drawings represent "unreal" objects (i.e. the picture shows half of one object combined with half of another object), and the participant is asked to decide whether each of 32 drawings represents a real or unreal object. EM performed within the normal range for the length, size, orientation, and position of the gap tests. However, her performance on the object decision subtest was marginally lower than controls (see Table 1), confirming her self-reported difficulties in the recognition and categorization of objects.

Face-processing assessment

EM and controls completed a series of face processing tests to (a) confirm EM's prosopagnosia, and (b) to attempt to localise her impairments within Bruce and Young's (1986) theoretical framework:

View-dependent face perception (structural encoding): Participants completed two tests of emotional expression recognition: the Reading the Mind in the Eyes Test (RMITE: Baron-Cohen, Wheelwright, Hill, Raste, & Plumb, 2001) and the Ekman 60 faces test

(Young, Perrett, Calder, Sprengelmeyer, & Ekman, 2002). In the 36 trials of the RMITE test, participants are shown the eye region of a face, and asked to choose which of four thoughts or feelings is being expressed. In the Ekman 60 faces test, participants are shown a face and asked to choose which of six basic emotions is being expressed. The Philadelphia Face Perception Battery (PFPB: Thomas, Lawler, Olson, & Aguirre, 2008) was used to assess age and gender perception. In the 75 trials of the age subtest of the PFPB, participants are presented with two faces and asked to select which one appears older. In the gender subtest, participants are presented with 75 single faces and asked to indicate whether the person is male or female. EM was marginally less accurate than age-matched controls when asked to identify emotional expressions from the eye region (RMITE), age, and gender. She was significantly less accurate than controls when asked to discriminate emotional expression from the whole face (Ekman 60) (see Table 1).

View-independent face perception (structural encoding): The Cambridge Face Perception Test (CFPT; Duchaine, Germine, & Nakayama, 2007) is a standardised test of face perception. In each trial, participants are asked to order a set of six comparison faces based on their similarity to a single test face, where each of the comparison faces has been morphed towards the test face by different degrees. In another face matching test created within our laboratory (Bate et al., 2014) participants were asked to match a test face to one of three simultaneously presented comparison faces. The test contained 40 trials, with faces selected from the Bosphorus Face Database (Savran, Sankur, & Bilge, 2012), and cropped to remove any external features. The size, luminance, and head position of the test face differed from the comparison faces, to prevent matching based on low-level image properties. The images remained onscreen until participants responded. EM performed significantly worse than control participants on the CFPT and the face matching test (see Table 1). This confirms

that EM has a significant perceptual deficit that is most likely driving her difficulties with

face recognition.

   Face recognition and identification (FRUs, PINs and semantics): Face memory was

assessed using the Cambridge Face Memory Test (CFMT: Duchaine & Nakayama, 2006) and

a familiar faces test. In the CFMT, participants are asked to learn six faces, and then choose

which of three simultaneously presented faces shows a learnt face over 72 test items. Some

test items depict the faces from novel viewpoints, under novel lighting conditions, with added

noise, or with a combination of these manipulations. Notably, EM performed significantly

worse on the CFMT than control participants (see Table 1). In the familiar faces test, EM

was asked to recognise and name a number of personally familiar people. Fourteen familiar

face photographs were collected by EM's family, and were taken under similar lighting

conditions and using the same camera. The test also included photographs of 14 people

unfamiliar to EM, matched for age and gender, to act as distractors. All pictures were edited

to exclude hair and ears. EM was asked to indicate whether each face was familiar

(familiarity), and if so, to name the person (identification). To assess whether EM's

difficulties were face-specific or a more general person-identification deficit, she also

completed a name identification test. In this test, the name of each of the familiar people was

read to EM, who was asked to explain her relationship to that person. As it was inappropriate

to use the same tests for age-matched control participants (who were not familiar with EM

and her family), the familiar faces and names tests were administered to EM's sibling

(female, age 6 years and 9 months) for comparison. EM performed poorly on both the face

recognition and face identification components of the test – she correctly identified 8 of the

14 pictures as familiar (performance was at chance), but could only name 3 of these faces. In

contrast, EM's sibling was able to recognise and name all 14 pictures. Both EM and her

sibling could correctly state their relationship to all 14 familiar people, and provide extensive biographical information about each individual.

Summary

Overall, the results from the face processing battery suggest that EM's impairment in face recognition lies at the perceptual, rather than mnemonic level. Indeed, her poor performance on both view-dependent and view-independent face perception tests suggest that EM is impaired at the level of structural encoding – that is, she has difficulty extracting information from faces, which consequently leads to further impairments affecting facial identity recognition. However, we sought further confirmation of EM's difficulties with face perception using eye-movement technology – a technique that provides a more direct means to examine an individual's perceptual processing skills (Bate, Haslam, Jansari, & Hodgson, 2009). Indeed, if EM's face-processing deficit has perceptual underpinnings, we would expect to see abnormalities in the manner in which she looks at faces under free-view conditions.

EYE MOVEMENT INVESTIGATION

Previous work examining eye movements in cases of acquired prosopagnosia have focused on the proportion of dwell time that is allocated to the inner features, and particularly the eyes. The majority of previous investigations have noted decreased attention to these facial regions (e.g. Lê, Raufaste, & Demonet, 2003; Stephen & Caine, 2009), with some reports of increased attention on the mouth (e.g. Orban de Xivry, Ramon, Lefèvre, & Rossion, 2008; Caldara et al., 2005; see also Bukach, Le Grand, Kaiser, Bub, & Tanaka, 2008).

Interestingly, one report failed to observe any differences in the regional distribution of fixations in two cases of acquired prosopagnosia (Rizzo, Hurtig, & Damasio, 1987), and a similar finding was found in a case of developmental prosopagnosia (Bate, Haslam, Tree, & Hodgson, 2008). All three of these cases displayed evidence of covert face recognition – a phenomenon that is traditionally associated with higher-order rather than perceptual impairments (see Bate et al., 2008). Hence, abnormalities in the regional distribution of facial fixations may be more indicative of the apperceptive rather than associative subtype of prosopagnosia.

In typical perceivers, the proportion of dwell time spent on the eye and mouth regions has also been found to vary according to emotional expression, such that participants are drawn to the eye region for sad faces and the mouth region for happy faces (Eisenbarth & Alpers, 2011). A novel application of this finding is to examine whether the same biases can be observed in EM, in order to investigate whether any abnormalities in regional scanning are absolute or still follow typical trends, albeit at a reduced level. Hence, in the current investigation, we examined the proportion of dwell time that EM spent on the eyes and mouth of faces displaying happy, sad and neutral expressions. We also measured the proportion of dwell time that she spent fixating on the nose, given that work from typical perceivers has associated this region with holistic processing (Hsiao & Cottrell, 2008; Peterson & Eckstein, 2012) – a skill that is typically disrupted in prosopagnosia (e.g. Levine & Calvanio, 1989; Sergent & Villemure, 1989).

Method

The eleven control participants described above and EM participated in this study. Participants were seated in a quiet room approximately 60cm from the screen, and were asked to place their head within a chin rest. Eye movements were recorded using the Eyelink

1000 system (SR Research Ltd, Canada), a video-based pupil/corneal reflex tracking device

sampled at 2000 Hz with spatial accuracy of between 0.25 and 0.5 degree of visual angle. A

calibration of eye fixation position was conducted prior to the experiment. Participants then

viewed 48 facial images from the Karolinska Directed Emotional Faces (KDEF) database

(Lundqvist, Flykt, & Őhman, 1998) in a random order, with an exposure time of 5 seconds

per face. The set consisted of three images of each of 16 individuals (eight male), one

displaying a happy expression, one a neutral expression and one a sad expression. Each face

was displayed in colour and upon a grey background. Images were adjusted to 762 pixels in

height and 562 pixels in width. Faces were not cropped around the hairline because the study

aimed to measure the dwell time spent on the inner features without cueing participants

towards these parts of the face. Previous studies investigating the scanning of faces

displaying different emotional expressions have reported consistent findings regardless of the

instructions given to participants. For instance, Pelphrey et al. (2002) found similar eye

movement patterns in typical participants and those with autism spectrum disorder, regardless

of whether they were instructed to "look at the faces in any manner your wish" or to "identify

the emotions portrayed in the faces". Because we wished to examine EM's natural scanning

patterns rather than any compensatory strategies that she might use to identify particular

expressions (e.g. by purposely looking to the mouth region for the presence or absence of a

smile), participants were not required to judge the emotion of each face and were instructed

to remain attentive to the faces and to allow their eyes to freely explore the stimuli. The

initial point of retinal attention for each trial was controlled by the presentation of a centrally

positioned fixation dot before the stimulus appeared.

Eye Movement Parameters and Statistical Analyses

Eye movements were analysed using Eyelink Data Viewer software (SR Research Ltd),

which allows periods of fixation to be identified and user-defined areas of interest to be

determined within the images. Seven areas of interest were drawn onto each face, covering

the eyes, nose, mouth, forehead, cheeks, chin and hair (including the ears when visible) (see

Bate et al., 2008). Data were averaged for each facial expression and for all trials (see Table

2). First, we compared the proportion dwell time allocated to the inner (i.e. the eyes, nose and

mouth) and outer (i.e. forehead, cheeks, chin and hair) features by EM and control

participants. Second, to examine more specific patterns of feature exploration, the proportion

dwell time elicited to the eye, nose and mouth regions was calculated. For each set of

comparisons, repeated measures analyses of variance (ANOVAs) were performed on the

control data, and the performance of EM was compared to that of control participants using

Crawford and Garthwaite's (2002) modified t-tests for single-case comparisons.

Results

A 3 (expression: happy, neutral, sad) x 2 (region: inner features, outer features) repeated

measures ANOVA performed on the control data indicated that more time was spent

examining the inner (M = 83.81%, SE = 14.17) compared to the outer (M = 14.17%, SE =

2.69) features, $F(1,10) = 151.784$, $p = .001$, $\eta_p^2 = .938$. There was no main effect of

emotional expression, nor did this factor interact with region, $F(2,20) = 1.246$, $p = .309$ and

$F(2,20) = 2.892$, $p = .079$, respectively. Overall, EM spent less time on the inner features and

more time on the outer features for all faces regardless of expression. When the three

expression conditions were analysed separately these differences were significant in the

neutral condition, and approached significance in the happy and sad conditions (see Table 2).

< Insert Table 2 >

A 3 (expression: happy, neutral, sad) x 3 (feature: eyes, mouth, nose) repeated measures ANOVA performed on the control data indicated a marginal main effect of expression, $F(2,20) = 3.558$, $p = .055$, $\eta_p^2 = .262$. Examination of the means suggest the proportion dwell time spent on each feature of sad faces (M = 27.47%, SE = 1.07) was lower than that spent on either happy (M = 28.08%, SE = 1.00) or neutral (M = 28.26%, SE = .97) faces. There was a clear main effect of feature, $F(2,20) = 17.560$, $p = .001$, $\eta_p^2 = .637$, and a follow-up analysis indicated that more dwell time was elicited to the eyes (M = 53.94%, SE = 6.90) compared to either the nose (M = 15.09%, SE = 2.37) or the mouth (M = 14.79%, SE = 2.75), $F(1,10) = 18.458$, $p = .002$, $\eta_p^2 = .649$. Expression and feature also interacted, $F(4,40) = 14.536$, $p = .001$, $\eta_p^2 = .592$. This was driven by less dwell time on the eyes for happy compared to neutral or sad faces, $F(1,10) = 41.993$, $p = .001$, $\eta_p^2 = .808$, and more dwell time on the mouth for happy compared to neutral or sad faces, $F(1,10) = 54.849$, $p = .001$, $\eta_p^2 = .846$ (see Table 2). EM spent less time on each feature in each condition compared to controls, but these comparisons did not reach significance (see Table 2). Further, the proportion of dwell time she spent examining the eyes and the mouth for happy compared to neutral or sad faces did not significantly differ, $F(2,30) = 1.881$, $p = .170$.

Summary

The results of the eye-movement investigation add further weight to the behavioural findings, suggesting that EM's face recognition deficit may be underpinned by a difficulty in face perception. Critically, this abnormality is characterized by a general reduction in viewing of the inner facial features that appears absolute, and is not naturally modified by manipulations in facial expression. From these initial investigations, it was concluded that EM would benefit from an intervention programme that aimed to improve her face perception skills, rather than one that targeted higher-order processing.

REHABILITATION PROGRAMME

Previous work examining the rehabilitation of prosopagnosia is sparse, and mostly has examined the utility of short-term training programmes (e.g. Powell et al., 2008). Not only does this make it difficult to evaluate the effectiveness of a training programme (as more training may have yielded greater improvements), but there is also evidence to suggest that improvements gained during the initial stages of training recruit compensatory mechanisms, whereas later improvements bring about a normalized pattern of brain activation (Rijntjes & Weiller, 2002; Thirumala et al., 2002). Strikingly, the most successful prosopagnosia training study to date examined the utility of a longer-term programme of training, and observed neurological changes following the intervention that were interpreted as a shift towards more "normal" patterns of face-processing (DeGutis et al., 2007). It is also clear from previous work that, in order to be effective, a training programme needs to specifically target the particular mechanism that is impaired (e.g. De Haan et al., 1991; Brunsdon et al., 2006).

Hence, we created a long-term training programme for EM that was designed to improve her ability to make fine-grained discriminations between faces, targeting processing at the perceptual rather than the mnemonic level. We examined the effectiveness of the programme in improving EM's face perception skills by carrying out an eye-movement assessment post-training. Importantly, together with further behavioural tests, this data could indicate whether any gains resulted from compensatory strategies, or could be attributed to improvements within the face-processing system itself.

Method

The training programme was presented in the form of a computer game with ten levels of difficulty. EM began training at the easiest level (Level 1), and in each attempt at passing a level, was presented with 40 trials that were randomly selected from a pool of 160 trials. Each trial consisted of three simultaneously presented faces: one target face that was presented at the top of the screen, and two test faces presented below (see Figure 3). EM was required to decide which of the two test faces represented the closest match to the identity of the target face. Critically, there was a change in viewpoint between the target and test faces: in half the trials the target face was represented from a frontal viewpoint and the test faces from a 15° left or right profile, and vice versa for the remaining trials. The difficulty of the task was manipulated between levels, as stimuli were created using a series of morph continua. Specifically, 40 male faces were computer-generated using the software Facegen, and divided into 20 pairs that were morphed along continua containing 20 equidistant morph levels. One of the original images from each pair was used as the target face. The test faces were composed of faces taken from different points along the relevant morph continuum, according to the level of difficulty. For instance, Level 1 test pairs contained images displaying 100% of the identity of the target and distractor faces, Level 5 test pairs contained 80% of the relevant identities, and Level 10 test pairs contained only 55% of the identities (see Figure 3). There was no time limit within which EM was required to make her response, and feedback was given after each trial and at the end of each attempt at a level. In order to pass a level, EM was required to answer 36 of the 40 trials correctly. She then automatically proceeded to the next level.

< Insert Figure 3 >

The training programme was delivered entirely online, via a testing platform within our laboratory's website (www.prosopagnosiaresearch.org). This enabled EM to participate in training on a more flexible basis without the need to come to the laboratory, and she

sometimes completed training at home and sometimes at school. A further benefit of the programme being online was that the experimenters could monitor EM's compliance and progress remotely. Training was delivered in a flexible format (EM could take part in training as often and for as long a time as she wished), although we recommended that she completed at least half an hour of training on three occasions per week. Each time EM logged into the website to begin a new session, training would continue at the highest level she had reached during her previous session. Training stopped after 30 hours of participation, and assessment measures were taken immediately after training ceased, and again one month later.

Results

EM completed the 30 hours of training over a 14 week period, and during this time progressed to Level 8 - an indication that her perception of facial identity for trained faces improved during the programme (see Table 3 for a summary of EM's performance on the training programme). To examine whether EM's improvements on the task were face-specific, as opposed to general improvements in visuo-spatial abilities or improved compensatory strategies, we examined EM's face inversion effect (i.e. the difference between upright and inverted faces) on the training task at the beginning and end of training. These were the only times EM was tested on the training faces when they were inverted. At the beginning of training (i.e. Level 1), EM scored 34/40 for inverted faces, and 26/40 for upright faces. At the completion of training (i.e. the last level that EM successfully completed - Level 7), EM scored 25/40 for inverted faces, and 36/40 for upright faces. McNemar change tests (two-tailed) were used to assess whether EM showed significantly more correct responses to upright or inverted faces, before and after training. For inverted faces, EM performed significantly worse after training than before training, $\chi^2 (1) = 7.11$, $p = .008$. However, for

upright faces, EM performed significantly better after training than before training, $\chi^2 (1) =$ 8.1, $p = .004$. Presented another way, prior to training, EM showed a significant advantage for inverted faces over upright faces, $\chi^2 (1) = 6.12$, $p = .013$, a highly atypical pattern of results. After training, EM showed a significant advantage for upright faces over inverted faces, $\chi^2 (1) = 9.09$, $p = .003$, which mirrors the inversion effect that is typically found in studies of face recognition (see Maurer, Le Grand, & Mondloch, 2002, for a review of the inversion effect in typically developing participants).

< Insert Table 3 >

The drop in performance for inverted faces could be interpreted in two ways: first, EM may have been relying less on atypical compensatory strategies (i.e., strategies that could be used for both upright and inverted faces) after completing the training; or second, the discriminations involved in Level 7 were simply harder than those involved in Level 1, resulting in poorer performance for the untrained stimuli (inverted faces). Either way, the fact that EM's performance improved for upright but not inverted faces suggests that the training programme strengthened and improved EM's face-specific processing, rather than improving her general visuo-spatial abilities or leading to the development of new compensatory strategies.

These findings gain additional support from further eye-tracking data collected in the post-training assessment. Specifically, EM took part in the same eye-tracking study as described above, but we restricted our analyses to those that addressed her main deficit (i.e. her reduced dwell time to the inner features for all faces, regardless of expression). We found that, in the post-assessment, EM spent a greater length of time studying the inner facial features than she had prior to training. Strikingly, the mean time EM spent looking at the inner features was no longer significantly different from that observed in controls (see Table 4), and the number of trials in which EM spent less time looking at the inner features than

controls dropped dramatically (see Table 5). Pre-training, EM's dwell time spent on the inner

features was consistently lower than the mean time for controls, whereas after training she

looked at inner features for as long or longer than controls in over 43% of trials. To assess

whether this improvement in face perception also generalized to untrained faces, we asked

EM to again complete the CFPT. Although her performance remained within the impaired

range (see Table 4), her score had improved compared to her pre-training performance on this

test (see Table 5).

< Insert Tables 4 and 5 >

A final set of tests examined whether these improvements in face perception might

also bring about an improvement in EM's face recognition skills. Indeed, we concluded from

our initial assessment of EM that her impairment was located at the stage of structural

encoding within Bruce and Young's (1986) theoretical framework. According to the

assumptions of this hierarchical model, any sub-processes that occur after the damaged

component should also be impaired, implying that EM should also experience difficulties in

face recognition even if these mechanisms are not damaged. It is therefore possible that

training in face perception may also yield improvements in face recognition. To assess this

possibility, we asked EM to repeat the familiar faces test from her initial assessment.

Strikingly, EM achieved 100% accuracy when making familiarity judgments about these

faces, and correctly identified more of the faces than she did in her pre-training assessment

(see Tables 4 and 5). Comparisons between pre-and post-training measures showed a

significant increase in correct rejection of unfamiliar faces, and a marginally significant

increase in identifying and naming familiar faces (see Table 5). However, an improvement

was not noted on the CFMT, indicating that her ability to encode novel faces for later

recognition had not changed following training (see Tables 4 and 5). When asked if she had

noticed any improvement in her everyday face recognition skills, EM reported no

improvement, although she had recognized her mother on isolated occasions. She did report an increase in confidence following training, and she had gone out without a family member on several occasions – something she had not felt able to do prior to participating in the programme.

One month after training ceased, we had the opportunity to carry out a short assessment session with EM. We decided to repeat the eye-tracking and familiar faces tests, given these assessments indicated significant gains post-training. The average length of time EM spent examining the inner facial features in the eye-tracking test had dropped slightly compared to her post-training assessment, but was still significantly greater than her pre-training performance, and within the normal range compared to controls (see Tables 5 and 6). Despite the drop in overall dwell time, EM still looked at inner features for as long or longer than controls in over 43% of trials (see Tables 5 and 6). On the familiar faces test, EM maintained her improvement to correctly reject novel faces, but her scores for familiarity judgments and correct identifications for familiar faces were no longer better than those obtained pre-training (see Tables 5 and 6).

< Insert Table 6 >

DISCUSSION

This paper aimed to improve face perception skills in an adolescent with acquired prosopagnosia, EM. Initial neuropsychological testing revealed impairments in both face perception and face recognition, in addition to object-processing deficits. An eye-movement investigation revealed a key avoidance of the inner facial features, and EM subsequently participated in an online training programme that attempted to improve her face perception

skills. After 14 weeks of training, marked improvements were noted in her perception of

trained and untrained faces, and she spent more time examining the inner facial features.

Post-training assessments also recorded an improvement in her recognition of personally

familiar faces, although these gains did not translate to EM's everyday experiences with the

same faces. She also failed to improve on the CFMT – a test of her ability to encode and

recognize unfamiliar faces. At a one-month follow-up, EM had maintained the increased

attention to the internal features, but her performance on the familiar faces test had declined.

First, EM's initial pattern of presentation merits discussion. When we first met EM,

she described profound face recognition deficits that dated back to the time she contracted

encephalitis. This was confirmed via extensive neuropsychological testing, which also

indicated severe problems with face perception and some difficulties with object-processing.

It is of note that EM's object-processing skills had improved somewhat since her illness, but

there had been no improvement in her face-processing skills. Eye-tracking evidence also

revealed profound difficulties with face perception. As observed in previous work examining

individuals with acquired prosopagnosia (Lê et al., 2003; Stephen & Caine, 2009), EM

avoided the inner features of the face, concentrating instead on the hair, ears and jawline.

However, while there is some suggestion in the literature that individuals with acquired

prosopagnosia avoid the eye region (e.g. Stephen & Caine, 2009) and overly focus on the

mouth (Caldara et al., 2005), no such pattern was observed in EM. Further, this pattern did

not vary according to emotional expression, and EM did not follow the expected biases of

spending more time on the mouth for happy faces and the eyes for sad faces (Eisenbarth &

Alpers, 2011). Hence, contrary to some previous work, it seems EM simply avoids looking

at the inner section of faces rather than avoiding or focusing on any one feature, and this

pattern is relatively absolute. This may be because she finds faces uninformative, reducing

the salience of the key facial features and encouraging her to instead treat all areas of the stimulus with equal importance.

Second, the gains made in EM's face perception skills suggest this training programme offers a promising means to treat apperceptive prosopagnosia. Indeed, there were clear improvements in EM's face perception skills, as evidenced by her progression via the training programme. Our additional investigations using inverted versions of the trained faces indicate that improvements were made to face-specific mechanisms, and did not result from the development of compensatory strategies or improvements to more generalized mechanisms. This finding was given additional weight by EM's performance on the eye-tracking test, which indicated a clear shift to a more "normal" pattern of facial scanning. Further, EM's improved performance on the CFPT also indicated that the gains could be generalized to untrained faces, although it should be noted that her performance was still impaired in comparison to age-matched control participants.

While we have presented evidence that EM's improvements can be attributed to face-specific rather than compensatory or more general mechanisms, the precise locus of improvement remains unclear. The training programme aimed to improve EM's ability to make fine-grained discriminations between faces when they are presented simultaneously – a procedure that might strengthen face perception mechanisms as the level of structural encoding (Bruce & Young, 1986). It is also possible that the training programme improved EM's configural or holistic processing skills. Although we did not take direct behavioural measures of these skills before and after training, it is generally accepted that inversion effects in face recognition are reasonable indicators of holistic processing, despite not being directly diagnostic of processing style (Crookes & McKone, 2009). The reversal of the face inversion effect that we observed after training may therefore be taken as an indicator that holistic processing mechanisms might have improved in EM. Further, previous observations

of reduced dwell time on the inner facial features in individuals with prosopagnosia have been interpreted as evidence of impaired holistic processing, despite being a blunt measure (e.g. Caladara et al., 2005; Stephan & Caine, 2009). While future research using this training programme should directly assess holistic processing (i.e. using the composite face paradigm), it can be tentatively suggested that this process was improved in EM in the current investigation.

It is less clear whether training also improved EM's face recognition skills. Her performance on the familiar-faces test indicated that she had improved in this domain, but her everyday experiences of recognizing the same faces showed little or no improvement. Further, she did not improve in her recognition of newly-encoded faces, as evidenced by her low score on the CFMT post-training. There are a number of reasons why this pattern of findings may have emerged. First, it may be that EM's improvement on the familiar-faces test resulted from practice – while this cannot be ruled out, it is of note that her performance dropped in the one-month follow-up assessment. If the improvement had resulted from practice, her performance should have been highest in this assessment. Second, it may be that EM simply did not spend enough time in training for the gains to also extend to her face recognition skills. Third, the hypothesis that EM's face recognition skills would also improve following face perception training assumes that the damage to her face-processing system is limited to the stage of structural encoding. Bruce and Young's (1986) hierarchical model posits that damage to any stage of the system prevents subsequent sub-processes from being completed, even if latter mechanisms remain undamaged. However, EM's MRI scan indicated extensive damage to the occipital and temporal lobes (see Figure 2), and it is very likely that higher-order processes are affected in addition to perceptual mechanisms (e.g. Barton et al., 2008). If this is the case, EM may have benefited more from a training programme that incorporates both perceptual and mnemonic training.

This latter point indicates one way in which the current training programme may be improved. Another avenue to improve the effects of training may be to increase or standardise the frequency or duration of training sessions. When EM started this programme, we recommended that she completed a minimum of three 30-minute training sessions per week. Importantly, the programme was designed to be flexible in order to fit around EM's personal and educational schedules, which often varied from week-to-week. As would be expected, there were some weeks when EM participated in more than the recommended amount of training, and in three weeks, she participated in substantially less training. When she stopped training, her performance at the beginning of the ensuing session reduced to a lower level than where it had been at the end of the previous session, which suggests that on-going engagement is important to maintain the training-based benefits. Further, the length of time that EM spent training in a single session also varied (see Table 3). When we examined EM's performance in each individual session, it became clear that she made greater progression (e.g. by actually passing levels) when she spent a longer time training, and her most successful training sessions were at least 50 minutes in duration.

It is also of note that it took EM a very long time to pass Level 1 (see Table 3), yet her progress through the other levels was more rapid. This pattern of performance suggests that EM would not have benefited from a shorter training programme, as it took her some time to develop the skills required to pass even the first level of difficulty. This finding also suggests that EM was not able to use compensatory strategies that would help her to complete the level more quickly. Hence, future attempts at remediation of prosopagnosia might recommend longer training sessions over a longer period of time.

Importantly, this study demonstrates some success in the rehabilitation of an acquired case of prosopagnosia using a remedial training strategy – a finding that has not yet been reported in the psychological literature. Indeed, the available literature only evaluates the

utility of short-term training programmes in individuals with the acquired form of the

condition, and suggests that compensatory training may be more effective that remedial

training (for a review see Bate & Bennetts, 2014). The findings reported here provide initial

evidence to suggest that remedial training may in fact be useful in acquired prosopagnosia.

Similar training programmes have also met with some success in individuals with

developmental prosopagnosia (DeGutis et al., 2007; DeGutis, Cohan, & Nakayama, 2014),

suggesting similar strategies may be useful in both forms of the condition. However, the

success of training in acquired cases is likely to be affected by more complex moderating

factors, such as the extent of the lesion, the age at which injury occurred, and the timing of

intervention.

In sum, this investigation provides encouraging evidence to suggest that a long-term

online perceptual training programme can improve face perception skills in a case of acquired

prosopagnosia. While more extensive investigation (perhaps using a modified version of the

programme) is required to fully investigate whether any gains can be achieved and

maintained in face recognition, initial evidence is nevertheless promising. Future work might

also examine whether the same training programme is effective in different subtypes of

prosopagnosia (e.g. developmental as well as acquired, and associative as well as

apperceptive), and also whether participants of different ages are able to make similar gains.

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Table 1: Results from the neuropsychological screening and face-processing assessment. Significant impairments are indicated in bold. WASI =

Wechsler Abbreviated Scale of Intelligence; CMS = Children's Memory Scale; BORB = Birmingham Object Recognition Test; CFMT =

Cambridge Face Memory Test; CFPT = Cambridge Face Perception Test; RMITE = Reading the Mind in the Eyes; PFPB = Philadelphia Face

Perception Battery.

| | | | Age-matched controls | | | Statistical tests | | | |
Task	EM	Mean	SD	n	Test statistic[a]	p	% population more extreme than EM	Estimated effect size (Z_{CC})
Age (years)	14.4-15.4	15.1	0.9	11				
WASI								
Verbal (*T* score)	52	51.44	7.79	10	0.07	.946	52.65	0.07
Performance (*T* score)	44	51.55	7.57	10	-0.95	.366	18.32	-1.00
Full-2 IQ	96	103.00	13.42	10	-0.50	.631	31.54	-0.52
CMS (Index score)								
Verbal immediate	100	-	-	-				
Verbal delayed	103	-	-	-				

Attention/concentration	97	-	-	-				
Learning	88	-	-	-				
Delayed recall	85	-	-	-				

BORB

Length match	26/30	25.63	1.80	11	0.20	.848	57.60	0.21
Size match	25/30	26.09	2.02	11	-0.52	.617	30.83	-0.54
Orientation match	22/30	25	1.79	11	-1.60	.140	6.98	-1.67
Position of gap	34/40	35.64	2.15	11	-0.73	.481	24.10	-0.76
Object decision	20/32	25.56	2.13	9	-2.48	.06	1.92	-2.61

View-dependent face perception

RMITE	13/36	23.63	3.80	11	-2.68	.06	1.16	-2.80
Ekman 60	**17/60**	45.90	6.98	10	-3.95	.008	0.17	-4.14
Age (PFPB)	43/75	60.30	7.85	10	-2.71	.06	3.25	-2.20
Gender (PFPB)	38/75	59.18	8.53	11	-1.82	.06	1.94	-2.48

View-independent face perception

CFPT[b]	**102**	39.82	5.83	11	10.21	.001	0.01	10.66

Face matching	**17/40**	30.50	3.84	10	-3.35	.002	0.04	-3.52

Face memory

CFMT	**19/72**	46.70	3.86	10	-6.84	.001	0.01	-7.18
Familiar faces: familiar hits	**8**	14[c]	-	1	4.17	.041		
Familiar faces: correct rejections	**4**	14[c]		1	8.1	.004		
Familiar faces: identity	**3**	14[c]	-	1	9.09	.003		
Familiar names	14	14[c]	-	1	-	-		

[a]All analyses except for the familiar faces tests were conducted using Crawford and Garthwaite's (2002) modified *t*-test for single-case comparisons, corrected for multiple comparisons using a sequential Bonferroni correction (Holm, 1979). Statistical comparisons for the familiar faces tests were carried out using McNemar tests, and the test statistic presented is χ^2. All results are for two-tailed tests. [b]CFPT scores measure deviation from an ideal arrangement, not accuracy. Therefore, a higher score indicates worse performance (see Duchaine et al., 2007). [c]Familiar face and name tests were administered to EM's younger sibling, but not to the age-matched control group.

Table 2: The proportion of dwell time that EM spent looking at each facial region in the eye-tracking study, compared to age-matched controls.

Cells in bold indicate a significant impairment.

	EM	Age-matched controls			Modified t-test[a]			
		Mean	SD	n	t[b]	p	% population more extreme than EM	Estimated effect size (Z_{CC})
Happy								
Eyes	36.23	49.92	23.97	11	-0.55	.596	29.82	-0.57
Mouth	7.56	19.73	10.12	11	-1.15	.276	13.81	-1.20
Nose	10.48	14.59	8.75	11	-0.45	.662	33.12	-0.47
Total inner	54.27	84.24	9.96	11	-2.88	.060	0.82	-3.01
Total outer	41.50	14.02	9.07	11	2.90	.060	0.79	3.03
Neutral								
Eyes	34.80	57.72	20.71	11	-1.06	.314	15.71	-1.12
Mouth	2.28	12.37	9.48	11	-1.02	.332	16.61	-1.06

Nose	9.10	14.70	7.63	11	-0.70	.498	24.91	-0.73
Total inner	**46.18**	84.79	9.67	11	-3.82	.021	0.17	-3.99
Total outer	**48.56**	13.46	8.27	11	4.05	.016	0.12	4.237

Sad

Eyes	28.69	54.17	24.21	11	-1.01	.337	16.87	-1.05
Mouth	7.95	12.27	8.38	11	-0.49	.632	31.61	-0.52
Nose	13.84	15.96	8.48	11	-0.24	.816	40.78	-0.25
Total inner	50.48	82.40	10.64	11	-2.87	.060	0.83	-3.00
Total outer	43.64	15.04	9.77	11	2.80	.060	0.94	2.93

Overall

Eyes	33.24	53.94	22.87	11	-0.87	.406	20.32	-0.90
Mouth	5.93	14.79	9.13	11	-0.40	.696	34.82	-0.42
Nose	11.14	15.09	7.87	11	-0.48	.641	32.06	-0.50
Total inner	**50.31**	83.81	9.94	11	-3.23	.048	0.45	-3.37
Total outer	**44.57**	14.17	8.91	11	3.267	.048	0.42	3.41

[a]All analyses were conducted using Crawford and Garthwaite's (2002) modified *t*-test for single-case comparisons, corrected for multiple

comparisons using a sequential Bonferroni correction (Holm, 1979). All results are for two-tailed tests.

Table 3: EM's progress through the training programme.

Level	Number of Attempts	Number of Sessions	Average Session Length (minutes)	Total Time (minutes)	Average Score (max. 40)
1	136	18	35.44	638	24.94
2	12	1	56	56	31.83
3	15	3	32	96	31.87
4	15	1	55	55	29.47
5	28	10	17	170	30.64
6	48	18	18.44	332	29.67
7	6	2	35	70	32.83
8	64	12	35.42	425	24.75
Totals	324	65	35.50	1842	29.50

Table 4: Post-assessment measures. Cells in bold indicate a significant impairment.

Task	EM	Age-matched controls			Statistical tests			
		Mean	SD	n	*Test statistic[a]*	*p*	% population more extreme than EM	Estimated effect size (Z_{CC})
Face perception								
CFPT	**68**	39.20	5.75	10	4.63	.002	0.05	4.83
% dwell time on inner features	84.32	83.81	9.94	11	0.05	.962	51.91	0.05
% dwell time on outer features	14.67	14.17	8.91	11	0.05	.958	52.09	0.06
Face memory								
CFMT	**22**	46.7	3.86	10	-6.101	.0006	0.009	-6.39
Familiar faces: familiar hits	14	14	-	1	-	-		
Familiar faces: correct rejections	14	14	-	1	-	-		
Familiar faces: identity	9	14	-	1	3.2	.073		

[a]All analyses except for the familiar faces tests were conducted using Crawford and Garthwaite's (2002) modified *t*-test for single-case

comparisons, corrected for multiple comparisons using a sequential Bonferroni correction (Holm, 1979). Statistical comparisons for the familiar

faces tests were carried out using McNemar tests, and the test statistic presented is χ^2. All results are for two-tailed tests.

Table 5: Comparison of EM's face perception and face memory scores at pre-training, immediately post-training, and in a one-month follow-up session. Cells in bold indicate a significant improvement following training.

Task	Pre-training	Post-training	Follow-up	Pre-training and post-training		Pre-training and follow-up	
				$\chi^{2\,a}$	p^b	$\chi^{2\,a}$	p^b
Face perception							
CFPT[c]	29.16	**52.78**	-	22.04	< .0005	-	-
Dwell time on inner features[d]	47/47	**21/48**	**21/48**	24.04	< .0005	25.04	< .0005
Dwell time on outer features[d]	47/47	**24/48**	**24/48**	22.01	<.0005	22.01	<.0005
Face memory							
CFMT	19	22	-	1.33	.248	-	-
Familiar faces: hits	8	14	12	4.17	.082	2.25	.266
Familiar faces: correct rejections	4	**14**	**14**	8.1	.032	8.1	.032
Familiar faces: identity	3	9	8	4.17	.082	3.2	.148

[a]McNemar change tests. [b]Results are for a two-tailed test, with sequential Bonferroni correction for multiple comparisons (Holm, 1979). [c]For the purposes of comparison, CFPT scores were transformed to percentage correct (see Rezlescu, Pitcher, & Duchaine, 2012). [d]For the purposes

of comparison, we calculated the number of trials in which EM's dwell time for inner features was less than the mean control dwell time for

inner features for that trial. These transformations were performed so we could conduct McNemar change tests (which require binary, rather than

continuous data) and analyse whether the changes in EM's face perception and eye movement patterns following training were statistically

significant, rather than simply showing how EM performed relative to controls before and after training (those results are presented in Tables 4

and 6).

Table 6: Follow-up measures taken one month post-training. Cells in bold indicate a significant impairment.

| Task | EM | Age-matched controls | | | Statistical tests | | | |
		Mean	SD	n	test statistic[a]	p	% population more extreme than EM	Estimated effect size (Z_{CC})
Face perception								
% dwell time on inner features	80.68	83.81	9.94	11	-0.30	.769	38.46	-0.32
% dwell time on outer features	17.68	14.17	8.91	11	0.38	.714	64.30	0.39
Face memory								
Familiar faces: familiar hits	12	14	-	1	0.5	.480		
Familiar faces: correct rejections	14	14	-	1	-	-		
Familiar faces: identity	**8**	14	-	1	4.17	.041		

[a]Crawford and Garthwaite's (2002) modified *t*-test for single-case comparisons was used to compare the face perception measure to control data.

Statistical comparisons for the familiar faces tests were carried out using McNemar tests, and the test statistic presented is χ^2. All results are for

two-tailed tests with sequential Bonferroni correction for multiple comparisons (Holm, 1979).

Figure Captions

Figure 1: An adaptation of the theoretical framework proposed by Bruce and Young (1986).

Figure 2: Images from a recent structural MRI scan. EM has lesions in the inferior and medial right temporal lobe, extending back into the inferior, lateral and posterior right occipital lobe. There are additional lesions in the left fusiform gyrus, inferior left temporal lobe and inferolateral left occipital lobe.

Figure 3: Examples of trials from Levels 1, 5 and 10 of the training programme. The target face is identical in all levels and contains 100% of one identity. The identities represented in the pairs of test faces become progressively more ambiguous in each level, and the task is to decide which face is the most similar to the target face.