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Developmental prosopagnosia: A case analysis and treatment study

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This paper reports a treatment case study focused on face perception impairments designed for AL, an 8-year-old child with prosopagnosia. AL's prosopagnosia was characterized by deficits at the level of structural encoding—that is, he was unable to achieve normal basic perception of faces. This impairment then impacted on all subsequent aspects of familiar- and unfamiliar-face processing. Detailed assessment of feature processing revealed impairments in perception of facial features with a dissociation between relatively good perception of the mouth feature and poor perception of eye and nose features. Interestingly, results also suggested at least partial internal representation of facial features despite long-standing deficits in perception of these features. A treatment programme focused on training in perception, and analysis of facial features and familiar-face naming was conducted. Treatment resulted in excellent face naming for familiar faces, a decreased reliance on nonfacial cues and a reduction in AL's tendency to misidentify unfamiliar faces as family members.

Acquired prosopagnosia in adults has attracted considerable research interest from cognitive neuropsychologists. Theories of skilled adult face processing are now well established and are founded on a considerable body of empirical research as well as numerous detailed case studies of acquired prosopagnosia. In contrast relatively few cases of developmental prosopagnosia or prosopagnosia acquired during childhood have been reported in the literature (e.g., Ariel & Sadeh,

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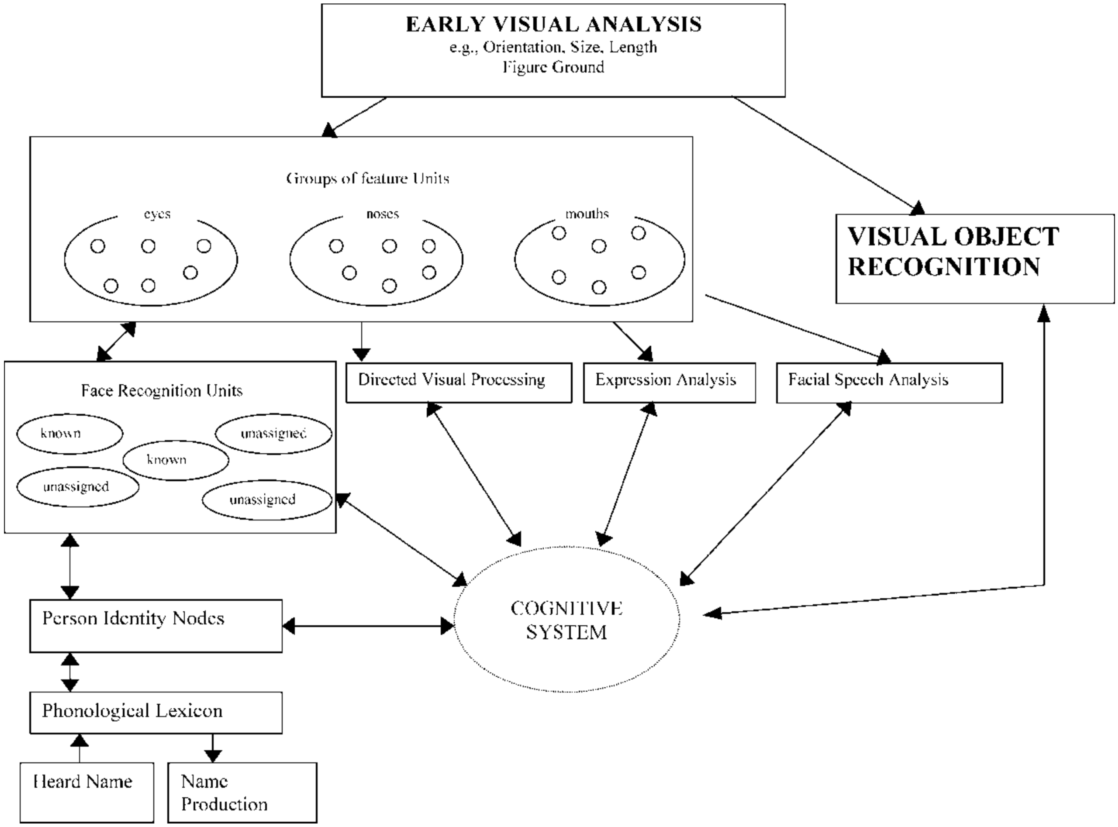

Figure 1. *Model of face recognition (adapted from Bruce & Young, 1986, and Burton, 1994).*

1996; Behrmann & Avidan, 2005, review paper; Campbell, 1992; De Haan, 1999; De Haan & Campbell, 1991; Duchaine, 2000; Jones & Tranel, 2001; Kress & Daum, 2003, review paper; Nunn, Postma, & Pearson, 2001; Schiavetto, Decarie, Flessas, Geoffroy, & Lassonde, 1997; Temple, 1992; Young & Ellis, 1989). Moreover, many of these reports focus on visual object processing and thus fail to analyse face processing in sufficient detail and/or within a cognitive neuropsychological framework (e.g., Bentin, Deouell, & Soroker, 1999; Jambaque, Mottron, Ponsot, & Chiron, 1998; Kracke, 1994; McConachie, 1976; Mottron et al., 1997; O'Hare, Dutton, Green, & Coull, 1998; Vargha-Khadem, Isaacs, & Mishkin, 1994). In addition, thus far, research investigating prosopagnosia has focused almost entirely on case descriptions and theoretical implications with only a handful of studies investigating rehabilitation strategies.

The current study includes a cognitive neuropsychological assessment of face recognition deficits in AL, an 8-year-old child with prosopagnosia, and a description of a treatment programme focused on perception of facial features and familiar-face recognition and naming.

Theoretical framework

The current study was based on the theoretical framework shown in Figure 1. The framework (see Figure 1) is briefly described here (though readers are also referred to Bruce & Young, 1986, and Burton, 1994, for further detail).

Visual analysis, the first stage of face processing, is common to visual object, face, and word processing domains. It involves perception of basic visual properties such as size, orientation, and figure–ground relationships (akin to Riddoch & Humphreys, 1993). Basic structural encoding of the face's appearance occurs next and includes formation of an abstract representation of the face that integrates the global configuration and individual features (Bruce & Young, 1986). In Figure 1, this level of face processing is represented by groups of feature units and their connections to face recognition units as proposed by Burton (1994). Each group of features (e.g., group of "noses" or "mouths") contains feature units that represent a different "feature value" (e.g., a particular kind of mouth or nose). Burton (1994) stresses that labels employed for feature groups such as "mouth" and "nose" are simply employed for convenience and most likely also represent measurements of more complex patterns involving the interrelationship between a number facial features. Akin to Burton (1994) we conceive that feature units contain information about the visual appearance of a variety of face characteristics such as face shape, facial features, skin tone, and the spatial relationships between features. Thus, in this paper the term "facial features" does not refer only to the nose, mouth, and eye features, but incorporates a broad range of facial characteristics.

In Figure 1, recognition of a visual face stimulus as familiar occurs when the corresponding known face recognition unit is activated (akin to Bruce & Young, 1986),[1] which occurs when it receives excitatory input from the corresponding feature unit in each of the feature groups (Burton, 1994). In normal processing, this in turn activates the corresponding person identity node, which contains personal characteristics and facts about known people. Person identity nodes can also be accessed from nonfacial cues such as voice, clothing, or mannerisms. Activation of a person identity node then triggers the representation of the corresponding name in the phonological lexicon (Bruce & Young, 1986). Other specific information is also retrieved from face stimuli, in parallel with face recognition: information about facial expression, mood, and affect; analyses of facial speech movements employed in lip reading; and, finally, directed visual processing, which is proposed to represent the more strategic and selective processing needed to match and distinguish faces from one another (Bruce & Young, 1986).

Treatment of prosopagnosia

To our knowledge, there is only one published case study describing treatment of prosopagnosia in a child (Ellis & Young, 1988) and no successful studies. Treatment of prosopagnosia in adults is also rarely attempted (De Haan, Young, & Newcombe, 1991; Francis, Riddoch, & Humphreys, 2002; Wilson, 1987).[2]

Ellis and Young (1988, also see Ellis, 1989, and Young & Ellis, 1989) describe KD, a child who acquired a very severe prosopagnosia after anaesthetic complications at age 3. KD (who was studied over a period of 3 years, age 8–11) had a range of visuoperceptual difficulties including an object agnosia and a disproportionately profound prosopagnosia. Ellis and Young (1988) considered her underlying deficit to be an inability to form three-dimensional visual representations of objects, resulting in an inability to construct adequate representations of visual stimuli. KD was trained in matching/discrimination of unfamiliar and familiar-face photographs, schematic faces (simple line drawings), and digitized images

[1] In this paper we use the term face recognition to refer to recognition of a visually presented face stimulus as familiar. This is not to be confused with recognition of a person as familiar. To this end we argue that face recognition occurs at the level of face recognition units (like Bruce & Young, 1986).

[2] Polster and Rapcsak's (1996) report on unfamiliar-face learning in RJ, a densely prosopagnosic adult, is not included in this review. Their study investigates the effects of different encoding instructions on recognition memory for unfamiliar faces, but does not represent a true rehabilitation study.

of unfamiliar real faces. The final stage of training involved learning of name–face associations. No stage of treatment resulted in improvement.

Wilson (1987) describes an unsuccessful attempt to improve face–name association skills in OE, an amnesic man with prosopagnosia. The technique involved associating pictorial or motor mnemonics with the name of a familiar person depicted in the target photograph. OE failed to learn to associate photographs with either type of mnemonic.

De Haan et al. (1991) were also unsuccessful in their rehabilitation attempts with case PH, an adult with severe acquired prosopagnosia. PH showed no overt face recognition skills (no conscious recognition of familiar faces) but displayed covert (nonconscious) face recognition on a number of tasks specifically designed to measure familiarity in an incidental manner (e.g., forced-choice name decision, picture–name learning tasks). De Haan et al.'s (1991) rehabilitation aim was to "convert preserved covert face recognition into overt recognition" (p. 2585). They employed a "category presentation" method (Sergent & Poncet, 1990). PH was presented with a group of photos and was informed that all of the photographs belonged to the same occupation. He was then told the occupation category (e.g., politicians, comedians, etc). The category presentation method resulted in improved performance for only one category (out of six) with PH spontaneously naming the occupation and recognizing six out of eight exemplar photographs. This improvement was maintained on immediate posttest (when photos were again presented in random order) but not on delayed posttest (2 months later).[3]

Successful remediation of a person-based semantic disorder in an adult has been reported more recently (Francis et al., 2002). NE, a young adult with acquired brain injury, had two underlying impairments: a prosopagnosia (difficulty with recognizing faces); and a person-based semantic disorder (loss of person-based knowledge). Therapy focused on training NE to learn to identify unfamiliar faces and to relearn familiar faces. For unfamiliar faces, the most successful technique involved use of a complex mnemonic for each target photograph. NE was required to visualize an image incorporating three aspects of the target photograph: a prominent facial feature, name, and occupation. For relearning of familiar faces, the most successful technique involved rehearsal of semantic information about each person while viewing their photograph. Interestingly, there was no generalization of treatment effects to untreated faces. However, of those photos successfully treated, improvements were person specific not stimulus specific (i.e., they generalized to different photos of the target people).

In summary, few cases of childhood prosopagnosia have been reported that include a detailed assessment of face processing based on cognitive neuropsychological theories. In addition, treatment case studies are extremely rare, and to our knowledge successful treatment of prosopagnosia has only been documented once (Francis et al., 2002). Nevertheless, treatment studies have raised interesting questions about the relationship between treatment success and treatment generalization and the level of diagnosed impairment.

Failure to elicit treatment generalization both to untreated items and also to alternative versions of the treated items has been common in treatment of visual recognition difficulties, for both objects and faces (see Humphreys & Riddoch, 1994). Francis et al. (2002) concur with Ellis and Young (1988) that level of impairment is an important factor in remediation outcome and particularly findings of generalization. Francis et al. (2002) propose that person-specific generalization in their study within the treated group of photos (i.e., generalization of treated photos to other photos of the same person) may have been related to the fact that NE did not exhibit perceptual deficits. They propose that failures to achieve

[3] Sergent and Poncet (1990) also failed to demonstrate lasting improvements following category presentation in their case study of PV, an adult with acquired prosopagnosia.

this type of generalization in other cases may relate to difficulties earlier in face processing and particularly at a perceptual level (Ellis & Young, 1988).

The following case description details the face-processing impairments of AL, an 8-year-old boy with developmental prosopagnosia, resulting from impairments in structural encoding. The detailed assessment was guided by the face-processing framework shown in Figure 1 and focused on the microstructure of structural encoding and particularly feature perception.

CASE REPORT

AL is an 8-year-old[4] child with prosopagnosia and visual-processing deficits of presumed developmental origin. At the time of treatment AL was in Year 2 of a mainstream school. He received individual educational assistance for 2.5 hours per day. His developmental history and visual object agnosia have been discussed in detail previously (Joy & Brunsdon, 2002) so are only covered briefly here.

Developmental history

AL was born in Sri Lanka and lived in an orphanage until adopted by Australian parents at 9 months of age. His gestational age and prenatal history are unknown. At 12 months of age he presented with marked motor delay. Initial neurological examination revealed hypotonia in the lower limbs and a left internal strabismus but intact extraocular movements and cranial nerves. Other investigations, which included spine and skull X-rays and brain magnetic resonance imaging (MRI), were normal. Hearing tests were also normal.

At age 4 years, neuropsychological assessment revealed a range of significant cognitive and visual recognition deficits. Intellectual testing indicated a borderline/mild intellectual delay; however, verbal skills were superior to nonverbal skills. AL made some striking visual errors on picture-naming tasks and was unable to recognize photos of family members. At this time, he was also described as having difficulties negotiating his physical environment with poor topographical orientation skills. Formal visual assessment at $5\frac{1}{2}$ years rated his visual acuity at 6/9 for both eyes. Visual fields and colour vision were reported as normal.

At age 7 years, full assessment of cognition and visual recognition skills was undertaken. AL's verbal intellectual skills now fell within the average range for his age. On formal assessment, his single-word reading, spelling, and basic number skills were average to above average for age, and language skills were age appropriate. In contrast, his performance on attention and executive tasks was impaired, and visuo-spatial and nonverbal abilities remained well below average. Copying, writing, and constructional skills also fell well below age expectations.

Visual agnosia

AL's visual processes were assessed (at age 7) using the BORB (Birmingham Object Recognition Battery; Riddoch & Humphreys, 1993), and results are detailed in Joy and Brunsdon (2002). AL performed outside the range of controls for perception of basic visual properties such as size, length, orientation, and position of gap (BORB Tests 2–5). Perception of size and of orientation were most impaired (at chance) with milder impairments evident in perception of length and position of gap. More marked impairments were evident in figure/ground segmentation, object constancy (i.e., perception of objects across different viewpoints), visual organization, and the processing of more complex visual material (see Joy & Brunsdon, 2002, for more detail). Semantic knowledge regarding objects was intact.

[4] AL was 7 years old when assessment commenced but turned 8 during the pretreatment assessment phase. Published age-based norms were used accordingly—that is, 7-year-old norms were used when AL was aged 7, and 8-year-old norms were used after his birthday.

Joy and Brunsdon (2002) conclude that AL's semantic, language (input and output), reading, and spelling systems were intact. Furthermore his difficulties in object recognition most likely represented inefficiencies in processing, most evident under more challenging conditions. That is, there appeared to be a cascading effect of partial impairments at earlier levels flowing through to, and impacting on, more complex recognition.

Thus, in terms of the model of face processing shown in Figure 1, AL had documented impairment in early visual analysis, particularly with perception of size, orientation, and figure–ground. His language skills were normal, indicating no impairment in phonological input or output processes.

Assessment of face-processing skills

Face-processing skills were assessed prior to treatment, and results are shown in Table 1. AL's 4-year-old adopted sister (ML) was used for comparison on many tasks as she, like AL, had extensive exposure to the same close family members. She also served as a suitable age match for AL's delayed nonverbal and spatial abilities, which could have affected his performance on some tasks. ML was adopted into the same family at 6 months of age. Her development had been normal.

Familiar-face naming

AL was presented with a set of 34 colour photographs consisting of 17 familiar faces (i.e., 15 close family members and 2 of AL's close friends) and 17 matched unfamiliar-face distractors (in random order).[5] The photographs were presented in two conditions: with hair and without (i.e., hair was cropped using photoediting software). AL was asked to name each person (i.e., "who is this person, do you know their name?"). ML, his younger sister, had no difficulty with this task.

Stimuli with hair. AL's overall response accuracy was 47% (16/34). He correctly named 6/17 of the familiar faces and misidentified 7 unfamiliar faces as familiar. Error responses (for familiar faces) included reporting that the face was not familiar or providing an incorrect name. With both familiar and unfamiliar faces he made errors in age and gender judgement and relied heavily on nonfacial cues (such as hair-style and glasses).

Stimuli without hair. AL achieved a total of 17/34 correct. He named 4/17 of the familiar faces correctly and misidentified 4 unfamiliar faces as familiar. For most of the familiar-face stimuli, AL reported that they were not familiar, so did not offer a name. All other naming-error responses (for both familiar and unfamiliar faces) involved faces with glasses, all of which AL incorrectly identified as that of his mother (who also wore glasses).

Face recognition units

Face familiarity task ("Is this person familiar?"). AL, on a different testing occasion, using the same stimuli as those above, was simply required to indicate whether each stimulus face was familiar to him. His overall accuracy for faces with hair was 74% (25/34) and without hair was 68% (23/34), both significantly above chance (binomial $p = .003$ and $p = .017$, respectively), but well below the performance of his 4-year-old sister, ML (34/34 on both tasks). AL correctly identified 12/17 of the familiar faces with hair and 10/17 of the familiar faces without. He made four false-positive errors (with unfamiliar faces) in both conditions.

Person identity nodes

AL had no difficulty discussing or providing personal information about people he knew (e.g., their occupation, where they lived, their age, their likes and dislikes), indicating intact person identity nodes.

[5] Each unfamiliar face was matched (according to gender, age, hair-style, and presence or absence of a beard and/or glasses) as closely as possible to one of the familiar-face photos.

Table 1. Face-processing assessment results

	AL				Control data: ML	Control data: Published normative data	
Task	*N*	*Total correct*	*Proportion correct*	*Time taken*[a]	*Proportion correct*	*M*	*SD*
Face perception and structural encoding							
Unfamiliar-face matching and discrimination							
Unfamiliar faces with hair (colour photographs)	*22*		1.0				
Unfamiliar faces without hair (colour photographs)	*22*		.77				
Benton Face Recognition Test							
Items 1–6 (simple front view)	*6*	6				5.4[b]	1[b]
Items 7–28 (different views and altered lighting)	*21*	8				13[b]	1.76[b]
Discrimination of facial features							
Scrambled-faces task	*14*		1.0				
Discrimination of eyes, nose, and mouth features: *in isolation*	*24*		.88		.92		
Discrimination of eyes, nose, and mouth features: *within the face*							
Mouth	*40*[c]		.78	6.38	.83 (*N* = *24*)[c]		
Nose	*40*[c]		.50	8.03	.79 (*N* = *24*)[c]		
Eyes	*40*[c]		.52	8.88	.96 (*N* = *24*)[c]		
Total	*120*[c]		.60		.86 (*N* = 72)[c]		
Perception of facial features							
Total	*40*		.60		.85		
Skin	*9*		.56		.78		
Mouth	*8*		.88		1.0		
Nose	*4*		0		1.0		
Eyes	*7*		.57		.86		
Eyebrows	*7*		.71		.86		
Chin/face shape	*5*		.60		.60		
Face judgement tasks							
Gender[d]	*18*		.77		1.0		
Age[e]	*18*		.72		.83		
Emotional expression[f]	*24*		.58				
Face imagery tasks							
1. Familiar faces similarity judgement	*10*		.80		.80		
2. Familiar face features judgement	*50*		.70		.68		

[a]Average time taken per stimulus item, in s. [b]Published normative data from Benton, Hamsher, Varney, and Spreen (1983). [c]AL was given each set of eight facial-feature stimuli five times (giving a total of *N* = 40 for each feature and *N* = 120 overall). The control, ML, was given each feature set a total of three times (*N* = 72 overall). [d]Forced choice: male/female. [e]Forced choice: adult/child. [f]Forced choice of 6 for each item.

Therefore, in terms of the framework shown in Figure 1, AL's impairment must lie at the level of face recognition units or earlier (given intact name retrieval and person identity nodes).

Face perception (structural encoding)
Face matching and discrimination.
Unfamiliar faces with and without hair. Stimuli for this task included 22 pairs of unfamiliar colour front-view photographs (presented with hair and without hair on separate occasions). AL had to decide for each pair whether the photos were the same or different (11 pairs were the same, and 11 were different). For photos with hair cues his performance was perfect. Again, his performance was affected by the removal of hair cues (see Table 1).

Benton Face Recognition Task. The Benton Face Recognition Task (Benton, Hamsher, Varney, & Spreen, 1983) assesses face-matching skills under simple and more complex conditions, using black-and-white photographs with hair cues removed. In the simple condition subjects are required to match a front-view target face to an identical face image among five different front-view face distractors. In the complex condition the front-view target face must be matched to three same-face images shown in different angles or under altered lighting (e.g., increasing shadows), shown among three different-face distractors. AL's performance with simple comparisons was perfect. In contrast, his performance fell below controls when required to match under more complex conditions (see Table 1).

Discrimination of facial features.
Scrambled-faces task. This task required discrimination between scrambled and normal faces, where scrambled faces had facial features placed in the wrong positions. AL's performance on this task was flawless (see Table 1).

Discrimination of eyes, nose, and mouth features in isolation. The stimuli used for this task were taken from the Children's Memory Scale face memory subtest (Cohen, 1997). Using computer photoediting software, eight different noses, eight different eyes, and eight different mouths were extracted from the face stimuli (see Figure 2).[6] AL was presented with a total of 24 black-and-white stimuli: 8 pairs of eyes, 8 pairs of noses, and 8 pairs of mouths (half of the pairs for each feature were the same, and half were different). As shown in Table 1, AL and his younger sister were equally competent at this task. Both ML and AL made two errors involving

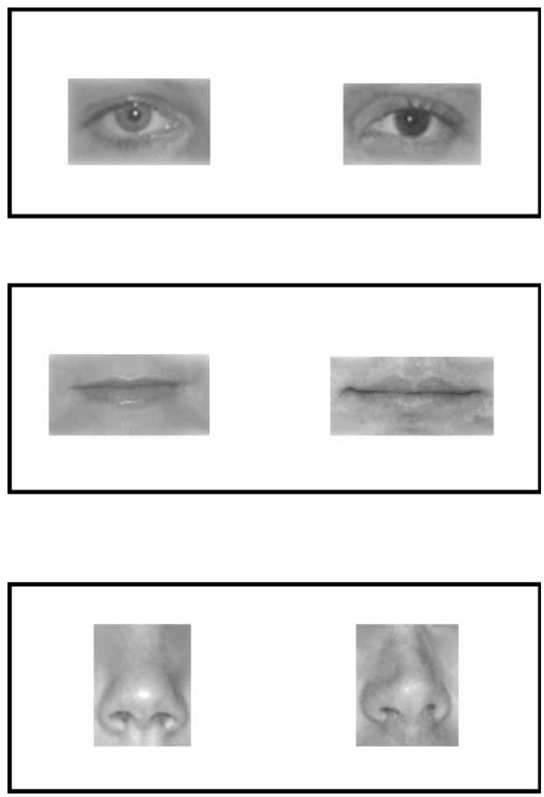

Figure 2. *Examples of stimuli used for the "discrimination of eyes, nose, and mouth features" task: "in isolation" condition.*

[6] Due to copyright restrictions the actual stimuli used in this study could not be reproduced in Figures 2 and 3, however suitably equivalent stimuli have been constructed for illustrative purposes.

nose pair stimuli, and AL made one additional error involving an eye pair stimulus.

Discrimination of eyes, nose, and mouth features within the face. The same discrimination task as that above was administered but with the features placed within a face (see Figure 3). The stimulus pairs used in this task were exactly the same as those used in isolation. AL and ML were informed that some of the faces would differ according to their nose, eyes, or mouth, and that others would be exactly the same. No other feature cues were provided. AL's response time for each stimulus was recorded.[7] AL's overall ability to discriminate eye, nose, and mouth features "within a face" fell well below that of his younger sister (see Table 1; Generalized Estimating Equation, GEE, analysis, $z = -3.02$, $p = .003$).[8] AL's overall performance on the "within-face" condition fell well below his overall performance when required to discriminate the same features "in isolation" (60% correct and 88% correct respectively, GEE, $z = 2.22$, $p = .026$). In contrast his sister's performance on both the "in isolation" and "within-face" conditions was similar.

This suggested that although AL was able to perceive and discriminate individual facial features relatively well (though we do not claim that this processing was normal), he was significantly worse at extracting the same information from within the facial gestalt.[9]

Most interestingly, he demonstrated a significantly better performance with "mouth" stimuli, which was well above chance ($p < .001$), than with "nose" and "eye" stimuli (GEE: nose, $z = -2.08$, $p = .038$; eye, $z = -1.97$, $p = .049$), which were both at chance level (nose, $p = .125$; eye, $p = .119$). Furthermore, AL's performance with "mouth" stimuli did not significantly differ from that of ML (GEE: $z = 0.51$, $p = .613$), but performance with "nose" and "eye" stimuli was significantly worse than that of ML (nose, $z = 2.35$, $p = .019$; eye, $z = 2.91$, $p = .004$).[10] ML's performance on "eye", "mouth", and "nose" stimuli did not differ significantly (GEE: $\chi^2 = 2.60$, $p = .27$). AL's response time showed a similar pattern with a significant degree of variation across facial-feature type, $F(2, 109) = 3.968$, $p = .022$. His response time for "mouth" stimuli was significantly faster than that for "eye" stimuli ($t = 2.74$, $p = .007$) and "nose" stimuli ($t = 1.97$, $p = .050$). Response time for "eye" and "nose" stimuli did not differ significantly ($t = 0.62$, $p = .539$).

Perception of facial features. AL's perception of facial features was assessed using eight colour photos of unfamiliar faces. The task involved 40 forced-choice questions (5 per photo) about facial features. Questions were designed to explore his perception of six different categories of facial features (i.e., skin tone, lip shape/colour, nose shape, eye colour/shape, eyebrow shape, overall face shape). For example, questions included: "Does he have a long pointy nose or a small round nose?"; "Does she have thick or thin eyebrows?"; or "Does he have a long thin face or a round face?". The same task was completed by

[7] Response time was recorded using the timing function in Microsoft PowerPoint. Response time was measured from the moment of stimulus display to AL's verbal response.

[8] GEE is an approach to the analysis of correlated response data, which is particularly useful when the responses are binary. It takes account of the correlations between observations and uses them when calculating both parameter estimates and their standard errors. Interested readers can refer to Hanley, Negassa, Edwardes, and Forrester (2003) for more information.

[9] It could be argued that this finding was simply a task complexity effect (i.e., the visual complexity of looking at and comparing two faces is greater than that when just comparing two noses/eyes/mouths). We would argue against this for two reasons. First, AL was able to perform the "within-face" task relatively well with mouth stimuli (as described in the next section), which suggests that he was able to cope with the task demands. Second, he was able to perform other face discrimination tasks of a similar complexity (e.g., Benton Face Recognition Test, Items 1–6), suggesting that in general he was able to cope with complex front-view discrimination tasks.

[10] We do not argue that AL's processing of the mouth region was normal (as the current control was not age matched), just that it was relatively intact when compared with other facial regions.

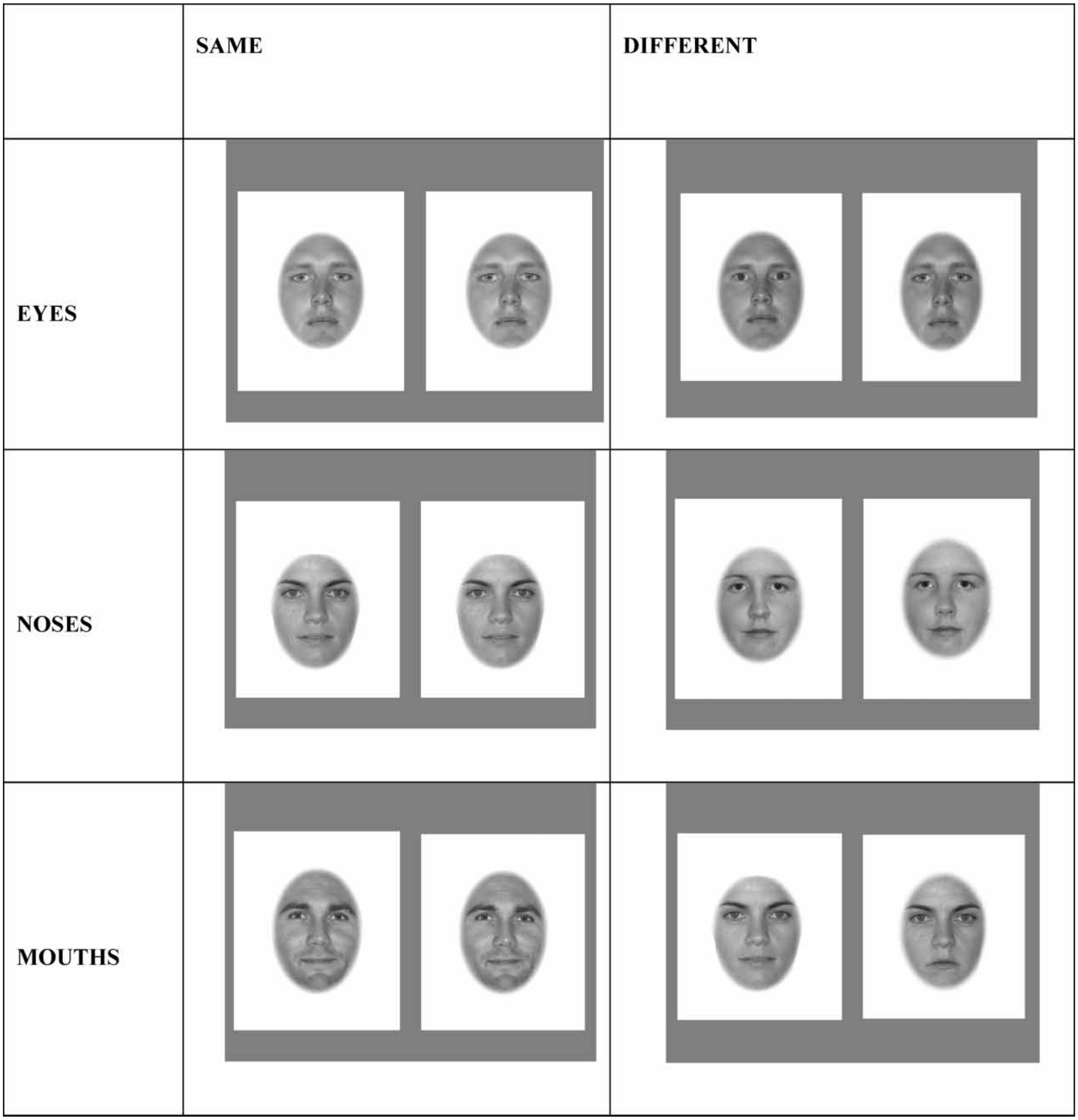

Figure 3. *Examples of stimuli used for the "discrimination of eyes, nose, and mouth features" task: "within-face" condition.*

AL's 4-year-old sister, ML. AL's overall performance (see Table 1) was not significantly different from chance ($p = .057$) and was well below that of ML (McNemar, $p = .021$). It is difficult to make comparisons at an individual facial-characteristic level due to the small number of stimuli; however, it is interesting to note that AL performed relatively better with "mouth" questions than with "nose" and "eye" questions, consistent with his performance on the eyes, nose, and mouth feature discrimination task (see Table 1).

Making judgements about unfamiliar faces. AL's ability to judge age and gender was assessed using selected stimuli ($N = 18$, with an equal number, $N = 9$, of males and females, and adults and children) from the face memory subtest of the Children's Memory Scale (Cohen, 1997), which employs coloured photographs with minimal hair cues. AL's response accuracy was significantly above chance (gender, $p = .012$; age, $p = .033$) but fell below the level of his younger sister (see Table 1).

AL was able to demonstrate and describe a range of facial expressions (e.g., happy, sad, angry) but he had difficulty judging emotional expressions from photographs. His ability to perceive and judge emotional expression was assessed using the Spence (1995) test of "perception of emotion from facial expression". AL performed well above chance ($p < .001$) but poorly compared to normal controls (see Table 1).[11]

Internal representation of facial features

This section investigates whether AL was able to extract enough information from the face stimulus to form internal representations of familiar-face features and characteristics.

Familiar faces similarity judgement. This task involved 10 forced-choice questions requiring AL to judge which two out of three familiar people look most alike (the response choices consisted of either three children or three adults). For example, he was asked, "Imagine a picture of your dad, your uncle J, and your cousin B. Which two look most like each other?" Both AL and ML answered 8 out of 10 questions correctly (see Table 1).

Familiar faces features judgement. AL and ML were asked forced-choice questions about facial features of family members (e.g., "imagine a picture of your sister; does she have big round eyes or thin narrow eyes?"). AL performed at an equivalent level to ML on this task (see Table 1). Most interestingly, he performed better on this measurement of internal representation of face features than in his perception of such features measured previously.

It cannot be definitively argued that AL and his sister completed these tasks solely by inspecting and comparing internal representations of facial features. It is possible that they were able to make some judgements based on general knowledge about the person's appearance or specific memory for isolated distinctive features. However, given AL's long-standing severe face perception impairments his performance on the imagery tasks was surprisingly good. We argue that at very least he had formed partial representations of facial features for highly familiar faces (see Table 1).

FUNCTIONAL LOCALIZATION OF IMPAIRMENT

With reference to the current framework shown in Figure 1, we propose the following pattern of processing impairment for AL:

1. Person identity nodes: AL had no difficulty discussing or providing personal information about people he knew.
2. Phonological lexicon: AL's language comprehension and expression were normal (particularly at the single-word level). Reading aloud was also normal for age (Joy & Brunsdon, 2002).
3. Visual analysis system: We propose that AL's prosopagnosia largely stemmed from core deficits in early visual analysis skills. AL performed outside the range of controls for perception of size, length, orientation, and position of gap (BORB Tests 2–5). Impairments in perception of size and orientation were most impaired (at chance) with milder impairments evident in perception of length and position of gap (Joy & Brunsdon, 2002).
4. Access to feature units: Assessment results suggest that AL's perception of facial features

[11] Spence (1995, p. 51) states "my previous research (Milne & Spence, 1987) demonstrated that most children over the age of 8 are able to decode facial expressions correctly".

(particularly those representing the eye and nose regions) and thus his access to feature units from visually presented faces was impaired. We propose that this most probably represents the follow-on effect of his early visual analysis impairments. Thus, when shown the visual stimulus of a face AL was unable to extract certain feature information from that face.

5. Feature unit representations: AL's early visual analysis impairment affected his ability to form feature unit representations at a developmentally appropriate rate. However, for highly familiar faces, his internal representations of facial features (or feature units) were surprisingly well formed in comparison to his very severe face perception impairments. AL performed at an equivalent level to his younger sister on face imagery tasks and better than he performed on face perception tasks. We do not argue that his feature unit representations were normal, just that (in comparison to his perception impairments) he was able to access them relatively well via the phonological lexicon, person identity nodes, and face recognition units.
6. Unfamiliar-face processing (directed visual processing, expression analysis, and age/gender judgements): Early impairments in visual analysis and their impact on access to feature units have a cascading impact on all aspects of unfamiliar-face processing, as shown by AL's reduced performance on perception of emotion, face matching, and discrimination (with hair cues removed and under more complex conditions).

In summary, in terms of the current framework of face processing (shown in Figure 1), we argue that AL's severe developmental prosopagnosia resulted predominantly from deficits in early visual analysis, which impacted on both familiar-face recognition and unfamiliar-face processing in a cascading and cumulative manner. Despite this, feature unit representations appeared surprisingly well developed for highly familiar faces (but most probably not entirely normal).

TREATMENT STUDY

Treatment focus

AL's core impairment was in visual analysis, which affected his face processing at an early perceptual level. Thus, face perception was poor, and, more specifically, AL was unable to isolate and analyse individual features and characteristics within the face gestalt. Therefore, the current treatment programme focused on direct training in perception and analysis of facial features and was limited to 17 familiar photographs. Training specifically aimed to improve AL's ability to focus on, perceive, and analyse facial features while reducing his dependence on nonfacial cues such as hair-style or glasses.

Aims of the treatment study

The aims of the current treatment study were to:

1. Improve AL's ability to perceive and discriminate facial features and facial characteristics.
2. Decrease AL's reliance on nonfacial cues such as hair-style and glasses and thus reduce his tendency to misidentify unfamiliar faces as close family members.
3. Improve AL's ability to recognize faces of familiar family members.

Treatment design

Three baseline assessments of face naming were conducted approximately 2 weeks apart, for faces with and without hair cues. Stimuli used for baseline assessments were identical to those used for pretreatment assessment of familiar-face naming. Feedback regarding accuracy of response was not provided during any assessments.

Treatment was commenced following Baseline 3 and targeted familiar-face stimuli without hair ($N = 17$). All target photos were included in treatment, even those named consistently correctly over baselines. These photos were included in an attempt to improve AL's methods for determining their identity, to increase his reliance on facial features, and therefore hopefully to reduce his likelihood of misidentifying unfamiliar faces as immediate family members.

Target stimuli were randomly divided into two groups (Group A contained eight target photos, and Group B contained nine target photos). Group A and Group B photos were trained in turn. Target photo naming was assessed midtreatment (after treatment of Group A photos), immediately posttreatment (after treatment of Group B photos), and 3 months posttreatment.

An additional group of photos (Group C) was compiled and included 11 different photos of target familiar faces. This group of photos was included to determine whether treatment effects were photo specific or person specific—that is, improvement was not only evident for a particular photo but also generalized to alternative photos of the same person. Group C photos were not a target of treatment, but were assessed both before treatment and immediately posttreatment.

Posttreatment assessment of discrimination of eyes, nose, and mouth features within the face and perception of facial features was conducted to determine whether these skills improved when compared to pretreatment assessments.

Treatment methods

For treatment, each target photo was presented to AL on a white background. AL was first asked to identify the person depicted in the photo (he was told their name if incorrect) and was then taught to perceive and remember five defining characteristics (which were written on the reverse of each photo). For each photo, the first two characteristics were the person's age (child/adult) and the person's gender (male/female). These were included as an initial strategy to increase AL's ability to identify the person depicted in the photo—AL's ability to determine the age and gender of a person (although not perfect) was superior to his ability to name them. The final three characteristics were defining facial features (such as "long thin face", "wide nostrils", "high curved eyebrows", "wrinkles around the eyes", "freckles"). AL was required to observe, discuss, and remember the characteristics for each photo (see Appendix 1 for home practice instructions). Treatment for each photo set was discontinued when AL achieved 100% correct for four consecutive homework sessions.

Treatment study results

Stimuli with hair. There was a marked improvement in AL's ability to name familiar faces with hair between the first and third baseline assessments (McNemar, $p = .004$). Familiar-face naming for "with hair" stimuli was therefore not a focus of treatment; however, performance was monitored posttreatment.

Stimuli without hair. There was no significant change in familiar-face naming "without hair" between any baselines (McNemar all comparisons, $p > .5$; see Table 2). However, a dramatic improvement was evident immediately posttreatment when compared to all baselines (Baseline 1, McNemar, $p = .004$; Baseline 2, $p = .004$; Baseline 3, $p = .002$) with total correct improving from 6/17 to 16/17 correct. This improvement occurred over just 14 home practice sessions (conducted over 1 month) and was maintained over time with no decline evident when assessed at 3 months posttreatment.

Another treatment aim was to reduce the number of unfamiliar faces identified as familiar (i.e., the number of false-positive errors), particularly those identified as family members. As shown in Table 2 there was a general decrease in false-positive errors (unfamiliar faces without hair identified as family members) during treatment. Due to the small number of items only one comparison was significant: the decline in false-positive errors between Baseline 3 and assessment at 3 months posttreatment (binomial, $p = .031$).

Generalization of treatment effects. Two treatment generalization effects are considered: first, treatment generalization to photos of people not included in the initial treatment group; and second, generalization to different photos of those included in the treatment group. Although

Table 2. *Treatment outcome for AL*

	Total or proportion correct				
	Baseline 1	*Baseline 2*	*Baseline 3*	*Immediately posttreatment*	*3 months posttreatment*
Familiar-face naming[a]					
With hair	.35	.65	.88	.100	.94
Without hair	.24	.35	.35	.94	.94
Unfamiliar faces identified as AL's mother[b]					
With hair	6	5	2	0	0
Without hair	4	4	6	2	0
Discrimination of eyes, nose, and mouth features: *within the face*					
Accuracy[a]					
Total	.60				.80
Mouth	.78				.81
Nose	.50				.81
Eyes	.53				.75
Response time[c]					
Total	7.76				9.6
Mouth	6.38				7.84
Nose	8.03				9.78
Eyes	8.88				11.16
Perception of facial features[a]					
Total	.60				.90
Skin	.56				1.0
Mouth	.88				1.0
Nose	0				.50
Eyes	.57				1.0
Eyebrows	.71				.86
Chin/face shape	.60				.80

[a]Proportion correct. [b]Total number. [c]Average response time per item, in s.

comparison between photo sets A and B is difficult to analyse statistically due to small numbers ($N = 8$ and $N = 9$, respectively) some observations relating to treatment generalization deserve a brief mention. Given that Set A was trained first, if treatment generalization to untrained photos did occur, we might hypothesize that total correct for Set B would improve prior to their being targeted for treatment, or that Set B would improve more quickly during treatment. However, both sets took a similar amount of training to reach criterion—Set A took a total of eight practice sessions to reach criterion (total correct each session: 3, 7, 7, 7, 8, 8, 8, 8), and Set B took a total of six to reach criterion (total correct each session: 4, 8, 9, 9, 9, 9). In addition, Set B photos did not improve prior to being targeted for treatment (following treatment of Set A). At midtreatment, assessment showed perfect naming of Set A photos but only three of Set B photos were named correctly. In sum, the limited data available suggest that treatment did not generalize to untrained target items.

In regard to generalization between different photos of the same person, the number of Group C photos named correctly also improved with treatment. Those "without hair" improved from 5/11 at pretreatment to 11/11 immediately posttreatment indicating that treatment effects were not photo specific.

Finally, there was also anecdotal evidence that AL's ability to identify family members in "real life" improved. AL's mother reported that following treatment she had noticed that AL was less likely to confuse family members, was less likely to misidentify strangers as familiar, and also seemed better at recognizing family members in family photographs.

Discrimination of eyes, nose, and mouth features (*within-face condition*). There was a significant improvement in AL's overall ability to discriminate between eyes, nose, and mouth features (within-face condition) following treatment (GEE, $z = 3.09$, $p = .002$; see Table 2). There was a significant improvement for perception and discrimination of "nose" and "eye" stimuli (GEE: nose, $z = 2.31$, $p = .021$; eye, $z = 2.20$, $p = .028$, respectively). Total correct for "mouth" stimuli did not change significantly ($z = 0.78$, $p = .434$), although this was relatively good pretreatment (i.e., did not differ significantly from the control). In fact, posttreatment AL performed at an equivalent level to the control ML not only on "mouth" (GEE, $z = -0.3$, $p = .768$) but also on "nose" ($z = 0.07$, $p = .942$) and "eye" stimuli ($z = 1.68$, $p = .093$).

Overall average response time increased with treatment, $F(1, 204) = 8.67$, $p = .004$, perhaps indicating a more careful and deliberate approach from AL (see Table 2). However, only the change for "eye" stimuli was significant ($t = -2.11$, $p = .036$).

Perception of facial features. Pre- and posttreatment results for the perception of facial features task are shown in Table 2. Treatment successfully improved AL's ability to perceive facial features overall (McNemar, $p = .002$). Small numbers make it difficult to statistically compare results according to individual face features, but AL's perception of all features improved to some extent.

Discussion

To our knowledge this study represents the first successful treatment of prosopagnosia in a child. The aim of treatment was to enable AL to reliably recognize familiar faces by improving his ability to focus on, perceive, and analyse facial features and hence reduce his dependence on nonfacial cues such as hair-style or glasses. Treatment outcome included improved perception and analysis of facial features, almost perfect recognition and naming of target familiar faces, and a reduction in the frequency of false-positive errors. Treatment effects were not specific to trained photos (i.e., were generalized to different photographs of the target group). Treatment aims were achieved surprisingly quickly, and benefits were enduring.

Treatment success

In terms of treatment effects, the current study clearly illustrates marked improvements in AL's ability to recognize photos of familiar faces. In addition, there is strong evidence that perception of (and discrimination between) different facial features within the face gestalt also improved. Significant posttreatment improvements were evident in feature discrimination involving both "eye" and "nose" regions and overall perception of facial features in general. The significant increase in processing latency for all feature regions also suggested a more deliberate and careful approach to feature processing in general. Within the theoretical framework, treatment effects may be interpreted as improved input to the feature groups and particularly those representing "nose" and "eye" regions. These improvements then enable the formation of more distinguishable feature units for a wider range of features, thus improving the system's ability to activate the correct face recognition unit using visually presented feature information.

A number of authors have argued that level of impairment in prosopagnosia is an important factor in treatment outcome (Ellis & Young, 1988; Francis et al., 2002; Wilson 1987)—in particular that prosopagnosia arising from perceptual deficits, like that of AL, is most resistant to treatment and also least likely to show treatment generalization effects. Despite this, the current treatment study was successful and provided some evidence of treatment generalization effects (treatment effects were not photo specific).

Developmental structural encoding impairments and cascading deficits

The current framework predicts that structural encoding impairments would have a cascading effect on all subsequent aspects of face processing including familiar-face recognition and also the processing of unfamiliar faces. Although many developmental cases, like AL, support the prediction (e.g., Ariel & Sadeh, 1996; De Haan & Campbell, 1991), others do not. In fact, some cases vary considerably in their pattern of cascading deficits and have surprisingly intact ability to perform specific face judgements. For example, cases KD[12] (Young & Ellis, 1989), EP (Nunn et al., 2001), and YT (Bentin et al., 1999) all had significant structural encoding deficits but were reportedly able to make good judgements about unfamiliar faces (for KD perception and naming of facial expressions were relatively good, and both EP and YT were able to make normal judgements about facial expression, age, and gender from unfamiliar photographs).

We argue that the contrasting patterns of cascading impairments in these cases most likely relates to subtle differences in exact type and severity of structural encoding impairment, which have not been explored in assessment, perhaps including differences in facial-feature perception. Structural encoding deficits in childhood are most commonly diagnosed by a failure on face discrimination tasks, and early feature perception is not usually investigated.

Perception of facial features

The current study has highlighted that developmental perceptual impairments may affect some regions of the face more than others. It is interesting to contrast AL's pattern of feature salience with that of normal children. We demonstrated that AL's perception and discrimination of facial features (within a face context) was characterized by relatively good performance with the "mouth" region and very poor performance (at chance) with the "nose" and "eye" regions, relative to that of the control subject. This pattern of feature salience contrasts with that of normal children.

Very few studies have investigated feature processing in children, and conclusions are difficult because of differences in task demands, stimuli type, and age groups studied. Nevertheless, a number of studies suggest a similar hierarchy of feature salience in children to that seen in adults for both familiar-face recognition and unfamiliar-face tasks (Chung, 1991; Chung & Thomson, 1995; Flin & Dziurawiec, 1989)—that is, the relative importance of upper regions over lower regions, and the eyes and mouth being most salient and the nose least important for face processing (Chung, 1991; Flin, Markham, & Davies, 1989; Goldstein & Mackenberg, 1966; Hay & Cox, 2000; Langdell, 1978; Pedelty, Levine, & Shevell, 1985). However, there is some evidence that younger children may in general (for both familiar- and unfamiliar-face processing) rely more on external features than on internal features (Campbell, Walker, & Baron-Cohen, 1995; Newcombe & Lie, 1995) when external features are available to them.

So, it is clear that AL differs most markedly from young, normally developing children (as reported in the literature) in his relative difficulty with processing information from the eye and nose regions. The eyes are particularly important in normal facial-feature salience studies, so we would suspect that AL's poor processing of the eye region might have the largest impact on his face processing.

Structural encoding impairments and development of facial-feature representations

A clear dissociation between face recognition and face imagery has been documented in acquired prosopagnosia (e.g., Behrmann, Moscovitch, & Winocur, 1994; Young, Humphreys, Riddoch, Hellawell, & De Haan, 1994). However, it has been argued that early processing deficits can prevent the acquisition of adequate face recognition units (or internal stored face representations; Barton, Cherkasova, & O'Connor, 2001; De Haan & Campbell, 1991). Barton et al.

[12] Although KD was not strictly a developmental case, her early insult (meningitis at 14 months followed by numerous shunt infections and surgical shunt revisions and also anaesthetic complications when aged 3) warrants her to be mentioned here.

(2001), in their discussions about covert processing, state that "developmental prosopagnosia . . . likely precludes the establishment of a store of accurate face memories" (p. 1161). They conclude that "any perceptual, amnestic, or disconnection deficit would undoubtedly prevent the acquisition of a store of facial memories in [developmental] subjects" (p. 1167). Some cases support this prediction—case KD (Young & Ellis, 1989), YT (Bentin et al., 1999), and EP (Nunn et al., 2001)—and present with documented structural encoding impairments and impaired face imagery, though assessment of face imagery in these cases was not thorough.

In contrast, AL provides preliminary evidence that despite structural encoding or perceptual impairments developmental prosopagnosics can form memories for faces. In other words, in our terminology, they can form internal representations of familiar facial features (though these may not be fully specified) and can access these via activation of person identity nodes and face recognition units. Again, we stress that our use of the term "facial features" incorporates a range of face characteristics including facial features, face shape, skin tone, and spatial relations between features. In addition, we conceive that the retrieval of face memories in imagery tasks occurs following activation of and interaction between face recognition units and feature units. Thus we argue that despite longstanding deficits in face perception AL performed surprisingly well on face imagery tasks, suggesting that he had at least partially formed internal representations of facial features of highly familiar faces and could access these via face recognition units. It is also interesting to note that AL also had a surprisingly intact store of knowledge regarding the visual features of objects, again despite a longstanding visual agnosia characterized by perceptual difficulties (Joy & Brunsdon, 2002).[13] Additional evidence for a dissociation between impaired face recognition and relatively intact (but not normal) face imagery skills can be found in Michelon and Biederman's (2003) investigation of MJH (an adult who acquired his prosopagnosia at age 5).

How might this occur? Although AL had never been able to perceive faces normally, in terms of the theoretical framework (Figure 1) he most probably was able to gradually form a selection of feature units and feature groups, albeit with distorted or incomplete contents. This in turn enabled the formation of face recognition units of known faces (most likely partial or incomplete). We propose that for familiar faces the pattern of activation between feature units and face recognition units have been constantly reinforced through repeated exposure to the same face (this would be particularly the case for his immediate family whose faces he had been exposed to most often). This proposal seems consistent with the very gradual improvement in his face recognition accuracy during early childhood (e.g., at preschool age AL was totally unable to recognize himself or immediate family members from photographs). We cannot argue that AL's internal representation of familiar faces or facial features was entirely normal; however, assessment results suggest that AL had surprisingly good face imagery skills relative to his very impaired face perception abilities.

FINAL COMMENTS

The current study represents the first known successful treatment of familiar-face recognition in developmental prosopagnosia. Treatment aims were achieved rapidly in the context of long-standing deficits in early visual perception and severe face recognition difficulties. AL's deficit in early face perception (or structural encoding) in conjunction with his positive response to treatment and his at least partially developed internal representations of familiar-face features seems at

[13] Relatively intact visual object imagery has been reported in other cases of developmental visual agnosia and prosopagnosia characterized by structural encoding deficits (KD, Young & Ellis, 1989; EP, Nunn et al., 2001; LG, Ariel & Sadeh, 1996).

variance with previous research findings and opinions regarding developmental prosopagnosia.

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APPENDIX

Treatment instructions for homework sessions

"We are going to learn about looking at faces and working out who they are. We are going to practice looking at some faces of people that you know. Remember, do not only use one thing to remember who the person is like a beard, hairstyle or glasses. We need to use all the clues that we practice."

For each photo:

1. Present the photo to AL and say "who is this?"
2. Give feedback to AL regarding whether his response is correct or incorrect and provide the correct answer (i.e., "yes, this is. . . ." or "no, this is. . . .").
3. Say for each photo "Now, how can we tell who this is, how can we remember who this is?"—then work through all the face cues with AL (i.e., talk about each cue, point to facial characteristics on the photo).