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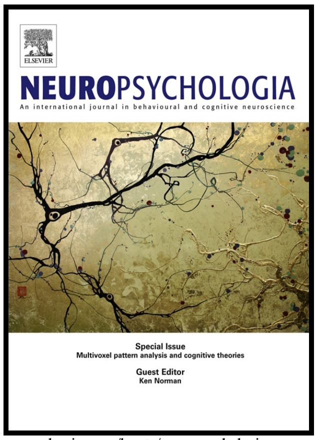

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Developmental visual perception deficits with no indications of prosopagnosia in a child with abnormal eye movements

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Abstract

Visual categories are associated with eccentricity biases in high-order visual cortex: Faces and reading with foveally-biased regions, while common objects and space with mid- and peripherally-biased regions. As face perception and reading are among the most challenging human visual skills, and are often regarded as the peak achievements of a distributed neural network supporting common objects perception, it is unclear why objects, which also rely on foveal vision to be processed, are associated with mid-peripheral rather than with a foveal bias.

Here, we studied BN, a 9 y.o. boy who has normal basic-level vision, abnormal (limited) oculomotor pursuit and saccades, and shows developmental object and contour integration deficits but with no indication of prosopagnosia. Although we cannot infer causation from the data presented here, we suggest that normal pursuit and saccades could be critical for the development of contour integration and object perception. While faces and perhaps reading, when fixated upon, take up a small portion of central visual field and require only small eye movements to be properly processed, common objects typically prevail in mid-peripheral visual field and rely on longer-distance voluntary eye movements as saccades to be brought to fixation. While retinal information feeds into early visual cortex in an eccentricity orderly manner, we hypothesize that propagation of non-foveal information to mid and high-order visual cortex critically relies on circuitry involving eye movements. Limited or atypical eye movements, as in the case of BN, may hinder normal information flow to mid-eccentricity biased high-order visual cortex, adversely affecting its development and consequently inducing visual perceptual deficits predominantly for categories associated with these regions.

Keywords: object recognition, contour integration, geometry, eye movements, pursuit, saccades, developmental visual agnosia, face perception

1. Introduction

Many regions in high order visual cortex show category-specific sensitivity such as to faces, places, words, common objects, and even to body-parts (Malach et al. 1995, Kanwisher et al. 1997, McCarthy et al. 1997, Epstein and Kanwisher 1998, Gauthier et al. 2000, Kourtzi and Kanwisher 2000, Downing et al. 2001, Grill-Spector et al. 2001, Levy et al. 2001, Cohen et al. 2002, Dehaene et al. 2002, Hasson et al. 2002, Malach et al. 2002, Downing et al. 2006, Pitcher et al. 2009, Konen et al. 2011). Furthermore, lesion studies, electrophysiological recordings, microsimulation and TMS studies indicate that the integrity and activation in some of these regions are critical for the perception of these categories. For example, regions in the right fusiform are critical for face perception (Barton et al. 2002, Parvizi et al. 2012, Behrmann and Plaut 2013, Gilaie-Dotan et al. 2013c, Behrmann and Plaut 2014, Rangarajan et al. 2014), the left fusiform for word perception (Sekuler and Behrmann 1996, Behrmann et al. 1998, Behrmann and Plaut 2014, Habekost et al. 2014), the lateral occipital complex/cortex (LOC), especially in the right hemisphere, for object perception (Milner and Goodale 1993, James et al. 2003, Konen et al. 2011, Gilaie-Dotan et al. 2013c, Gilaie-Dotan et al. 2015) and parahippocampal cortex for scene perception (Epstein et al. 2000). Until recently the organizational principles guiding the sensitivities in high-order visual cortex remained elusive; it is now clear that eccentricity guides not only retinotopic cortex, but also high-order visual cortex organization. For example, faces and words are associated with foveally biased regions (Levy et al. 2001, Hasson et al. 2002, Malach et al. 2002, Weiner et al. 2014), places with peripherally biased regions (Levy et al. 2001, Levy et al. 2004, Weiner et al. 2014), and common objects with mid-peripheral bias (Hasson et al. 2002, Hasson et al. 2003, Sayres and Grill-Spector 2008). The association of faces and words with a foveal bias seems intuitive given that foveal vision is required for their perception. Also intuitive is the association of places and houses with peripheral bias given the spatial integration required for place perception (Levy et al. 2004). However, the association of objects with mid-peripheral bias in high-order visual cortex remains unclear.

Neuropsychological and visual neuroscience have debated whether faces, common objects and words are supported by a joint (as a distributed) processing network (Ishai et al. 1999a, Haxby et al. 2001, Avidan et al. 2002, Behrmann and Plaut 2013, Behrmann and Plaut 2014) or by more modular category-specific regions or networks (Kanwisher et al. 1997, Epstein and Kanwisher 1998, Downing and Kanwisher 1999, Kanwisher 2000, Downing et al. 2001, Spiridon and Kanwisher 2002, Schwarzlose et al. 2005, Baker et al. 2007, Germine et al. 2011, Pitcher et al. 2012, Susilo et al. 2013, Susilo et al. 2015). Intermediate proposals of a joint network for faces and objects or for words and objects are also supported by lesions adversely affecting multiple categories ((Behrmann et al. 1994, Moscovitch et al. 1997, Gilaie-Dotan et al. 2013c) and see (Farah 2004) for review). While the evidence is complex and this debate is far from being resolved, the idea that objects, that are associated with mid to peripheral bias in high order visual cortex, would be jointly processed within the same network as faces or as words, that are each associated with a foveal bias, is somewhat puzzling.

Eye movements have long been implicated in visual perception (Yarbus 1967, Noton and Stark 1971a, Noton and Stark 1971b, Martinez-Conde et al. 2004, Schutz et al. 2011), and much more so in visual attention (e.g. (Corbetta et al. 1998, Torralba et al. 2006, Rolfs et al. 2011). For example, Yarbus showed that different eye movement patterns are associated with different visual perceptual categories (e.g. faces, outdoor woods (Yarbus 1967)), and it has also been shown that eye movements are attracted to salient features of the visual scene, such as animals, contours, or visual motion (Rolf et al. 1998, Krieger et al. 2000, Itti 2005, Guyonneau et al. 2006, Kirchner and Thorpe 2006, Bacon-Mace et al. 2007). Yet, how critical eye movements are for the development of normal visual perception has not been widely addressed.

Here we report on our investigation of BN, a 9 y.o. boy, whose face (and person) perception skills provide no indication of prosopagnosia. However, BN has significant perceptual deficits that are most evident but not limited to animals, geometry, and contour integration. Interestingly, he also shows abnormal eye movements as limited pursuit and imprecise saccades. Following these findings we discuss the possibility that pursuit and saccade eye movements play a significant role in the development of normal object and geometry perception.

2. Case history

BN is a bright and intelligent 9 year old boy, studying at school at his age level (in the Israeli general education system). He receives special education support in school to improve his gross and fine motor skills, and to assist in overcoming his perceptual difficulties. For a certain period he also attended occupational therapy to overcome his graphomotor problems. His parents reported that despite his normal basic level vision, he experienced many visual perceptual difficulties (some of which persist), and also difficulties in placing himself and manipulating objects in the environment. They recalled that he was unable to assemble simple puzzles of four pieces and did not play with toy blocks. In school BN experiences severe difficulties in geometry, a subject which he fails in, in contrast to his normal or above normal performance in math. They also recalled he would hold a cup obliquely or would not usually stand in the “appropriate” place in a queue or elevator. Until the age of five he was unable to distinguish between common fruits (e.g. apple and plum), whereupon his mother explicitly taught him these distinctions using children’s picture booklets and verbal explanations. BN has animal recognition impairments; he can confuse a dog and a cat, he thought a nearby ostrich was a cow, and that a big dog he saw in the park was a tiger. His visual perception may at times be “fragmented”; when shaking hands with someone, he reported perceiving the other person’s arm separated from their body. These observations and incidents, recalled by his parents, fit findings by a number of developmental specialists that examined BN (see details below). We present here a synopsis of their findings.

At the age of five, a psychologist diagnosed BN as having visual perception deficits, difficulties distinguishing between figure and ground, and not scanning a picture as expected.

At the age of 5.8 (years.months), clinical educational psychologists administered the Hebrew version of the Wechsler Preschool and Primary Scale of Intelligence (*WPPSI* (Lieblich 1971)) to BN. His verbal score was 106 (categorized as Average) and his performance score was 71 (categorized as Borderline), leading to a total score of 88 (Low Average). The verbal score was based on his scores in the following subtests: general knowledge (scored 8), vocabulary (scored 13), math (scored 11), common-side (measuring verbal abstraction, scored 13), and comprehension (measuring practical intelligence knowledge, scored 10). The performance score was based on his scores in zoo locations (scored 7), picture completion (scored 7), mazes (fine motor and spatial planning, scored 7), geometric shapes (scored 2), and block design (scored 6) subtests.

At the age of 6 a psychiatrist diagnosed BN with “fragmentation” after BN drew a person with scattered body parts.
During that period (age 5 to 6), a psychologist and a psychiatrist diagnosed BN with difficulties in graphomotor and visuomotor movements, sensory modulation, and in assembling puzzles, yet with normal language comprehension, general knowledge and mathematics.

2.1. Earlier basic visual functions assessments

At the age of 7.11 (years.months), a visual screening examination by an optometrist at his school showed that BN’s distance visual acuity (VA) was normal (5/5) in both eyes.
At the age of 8.4, an *expert orthoptist* examined BN’s basic level vision and found orthophoria present (i.e. normal balance between the eye muscles permitting lines of sight to meet at the point of fixation), and no strabismus. Distance VA in each eye was 6/6. Normal convergence and accommodation were found. Binocular vision measures were normal: normal depth perception, positive fusion range, and normal binocular vision. The orthoptist observed difficulties in pursuit eye movements during the ocular motility test.

In a hearing test (audiology examination) at the age of 6, BN showed normal hearing in both ears. He also showed normal hearing at an auditory screening test at school.

3. Current study

We met BN and his family to examine his visual perception and function, and consequently to consider possible means for alleviating some of his visual deficits. Here we report the findings from the examinations that started when BN was 9.0 and continued until he was 9.4. The study was approved by the Sheba Medical Center Helsinki committee. BN’s parents agreed to his participation in the study and signed a consent form. A concise summary of the findings, described in detail below, is given in Table 1.

3.1. Oculomotor assessments

3.1.1. Standard clinical tests for evaluating pursuit and saccades

We started our examination by focusing on the orthoptist’s predominant finding, BN’s difficulties in pursuit and saccades.

3.1.1.1. General procedures

The Northeastern State University College of Optometry (NSUCO) Oculomotor test (Maples and Ficklin 1988, Maples et al. 1992) and additional assessments were used to evaluate BN’s monocular and binocular pursuit, and binocular saccade functions. These observational tests are often used by clinicians (as optometrists, ophthalmologists, and neurologists) to diagnose oculomotor dysfunctions (Tassinari 2007, Harris 2012, Scheiman and Wick 2013). The experimenter who tested BN (R.D.) was a certified optometrist. Since we noticed in the first testing session that the tasks were difficult for BN, we repeated these oculomotor behavior assessments in two testing sessions on different days (see below). In the first session we tested pursuit (see below), and in the second session we tested pursuit again and then saccadic movements.

3.1.1.2. General observations
Before providing specific details we emphasize that BN became very stressed when asked to perform eye movement tasks. His body became restless and somewhat tense, eye and facial tics were observed. After completing the testing sessions, each lasting only a few minutes, BN seemed exhausted as if after strenuous physical effort and confirmed that the tasks were very difficult for him. This type of stressful or effortful behavior was not observed in the other tasks we administered.
During all the examinations no visible nystagmus was presented.

3.1.1.3. Pursuit

3.1.1.3.1. Procedure
BN was asked to follow a small painted target held by the examiner, while no instructions about head movements were initially given. The target was positioned at first directly in front of him (along the midline) at an approximate viewing distance of 40 cm, and then, as the pursuit task began, the target traced an imaginary letter H, an ellipse, or the horizontal and vertical meridians in the plane in front of him, all of which BN had to follow with his eyes. We measured mostly binocular pursuit.
3.1.1.3.2. Results
Instead of following the targets with his eyes, BN swung his body and head from side to side during the horizontal movements. Following that and throughout these testing sessions, he was instructed and reminded to keep his head still and use only eye movements during the task. After a few attempts he succeeded in keeping his head and body still, which was clearly a great effort for him. Abnormal pursuit was observed. During the test BN made many jerky eye movements, especially in vertical gaze. Sometimes, during the vertical gaze his right eye moved to adduction position (nasal) and during horizontal gaze his eyes almost closed. Since it seemed that he had greater difficulty with the vertical movements (up-down), we were able to obtain more information about horizontal movements (right-left) in the first testing session. As the greater difficulties in vertical movements may have been due to fatigue, in the second testing session on a different day, we deliberately started the session by testing vertical pursuit movements. However, we still observed great difficulty with vertical eye movements, including his right eye moving to adduction position, as in the first testing session. We note that BN’s monocular pursuit measurements (in each eye) were similar to the binocular ones.

3.1.1.2. Saccades

3.1.1.2.1. Procedure
BN was instructed to move his eyes quickly between two targets (colored fixation sticks) whose locations differed along the horizontal axis. He was asked to move his eyes as quickly as possible to the target indicated by the examiner (e.g. “blue, red, blue, etc...”). The targets were approximately 10 - 25 cm to the left or to the right of his midline (at a distance of about 40 cm). We repeated this procedure five times. We also tested him on the saccade test at longer distances of 1-1.5 meters and at 3 meters which may not be as informative as the closer test since the examiner was situated too far to precisely observe possible dysfunction.

3.1.1.2.2. Results

In the near distance tests (~40 cm from him) BN was unable to maintain fixation (he did it only for a short while), his saccadic movements were performed with moderate over- and under- shoots, and he did not complete two consecutive saccades without displaying a deficit or making an error. In the more distant tests (at longer distances of 1-1.5 meters and at 3 meters) we were also under the impression that BN had difficulties, and that he did not precisely fixate the targets as instructed.

3.1.2. Norm-based quantitative evaluation of eye movements

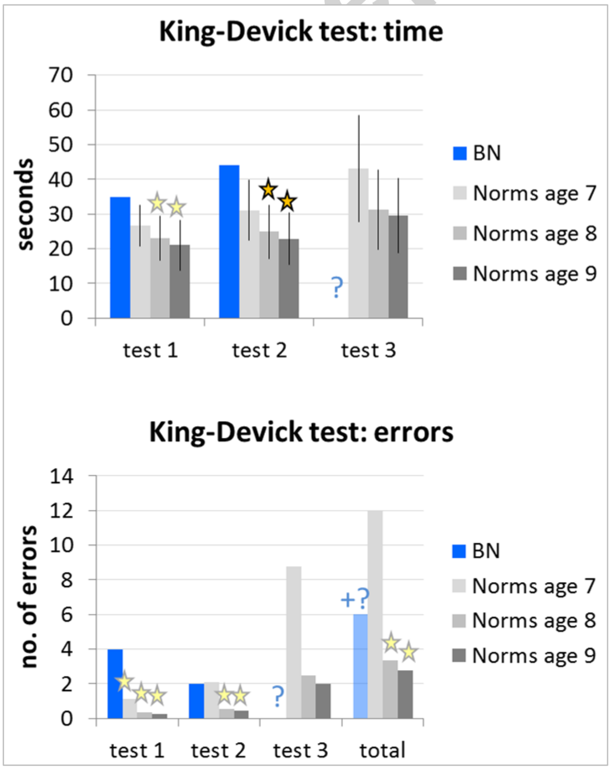
Because of BN's qualitative deficits in extraocular movements, in the third session we tested BN's performance on the age-normed King-Devick (K-D) test for eye tracking skills (King and Devick 1976, Lieberman et al. 1983, Leong et al. 2014). The K-D test relies on saccadic eye movements, and is associated with attention, language, reading ability, and many other perceptual and cognitive functions (Galetta et al. 2013, Leong et al. 2015). We assumed that since BN's eye movements were qualitatively impaired, his performance on the K-D test would also be significantly deficient relative to age-based normative scores (time to complete the test, and number of errors).

3.1.2.1. Procedure

The K-D test requires quickly and precisely reading aloud single digit numbers from left to right, line by line. It starts with a practice card which is the easiest, followed by three test cards with growing difficulty.

3.1.2.2. Results

After completing the practice card successfully, BN completed the first two (of three) K-D subtests but was disinclined to complete the third. As can be seen in Figure 1, he showed marginal impairment on the time to complete the first subtest (34.88 seconds, 1.925 SD longer than would be expected at his age) with 4 errors (vs. 0.28 expected errors at his age). BN's time to complete the second test was significantly longer than expected at his age (44.18 seconds, 2.83 SD longer than his age mean), making 2 errors (vs. 0.45 expected errors for his age). While we cannot assess his total time performance (as he did not complete the third subtest), the number of errors he made on the first two subtests are more than the total errors expected for his age (BN made 6 errors on the two tests vs. 2.75 errors expected at the age of 9 on all three tests).



3.1.3. Oculomotor assessment summary

Overall, we found that BN had significant difficulties in performing saccades and pursuit, which were evident both in standard clinical observational tests and in the well-established norm-based King-Devick test.

Figure 1. ***BN's performance on the King-Devick eye movement test relative to age level norms.*** Although BN completed the practice and only two of the three subtests, his performance was similar to that of 7 year olds, significantly slower (top), and less accurate (bottom) in the first two tests than his age-level norms (age 9).

3.2. Visual perceptual assessments

3.2.1. Controls

Eighteen children (eight boys) aged 9.45 ± 2.04 (SD) years (range 6.6 - 12.6 years) served as controls for BN on a subset of the following tasks. The parents of all these children agreed to their children's participation in this study.
All comparisons between BN's performance and that of the control group were according to established single case vs. group modified t-test comparisons (Crawford and Howell 1998, Crawford and Garthwaite 2002, Crawford et al. 2009, Gilaie-Dotan et al. 2009, Gilaie-Dotan et al. 2011, Brooks et al. 2012, Gilaie-Dotan et al. 2013a, Gilaie-Dotan et al. 2013c, Gilaie-Dotan et al. 2014, Gilaie-Dotan et al. 2015, Freud et al. 2016), using the SINGLIMS software (http://homepages.abdn.ac.uk/j.crawford/pages/dept/SingleCaseMethodsComputerPrograms.HTM).

3.2.2. Mid-level vision and figure-ground segregation

We report the following examinations together as we consider each of them to have a dominant component requiring distinguishing figure-from-ground for successful performance. These are not the only tasks engaging figure-ground assignment processes, and additional tasks are reported below (see 'object perception' and 'geometry and space' sections).

3.2.2.1. Stereopsis

3.2.2.1.1. Procedure

We used a standard stereo-vision test (Randot, Stereo Optical Co., Inc) (Simons 1981, Birch et al. 2008) to assess BN's stereopsis. The stimuli consist of random dot stereograms, some of which are presented with luminance based elements, and these are viewed via polarizing glasses. The task is to point to the protruding elements and in some cases report their shape. There are no time constraints for providing the response.

3.2.2.1.2. Results

BN performed normally, reaching stereo acuity of 60", which lies in the normal range according to the optometry clinical standards for this age. This was consistent with the orthoptist's findings.

3.2.2.2. Illusory contours

3.2.2.2.1. Procedure

Three Kanizsa illusory figures (Kanizsa 1976, Kanizsa 1979) were presented in a consecutive manner (triangle, rectangle, and then inverted triangle) on a computer screen, each for an unlimited time until the expected response was provided or until the experimenter decided that such response cannot be provided for that figure. BN was asked to verbally describe the display.

3.2.2.2.2. Results

Initially, with the first triangular display, BN reported seeing the pacmans, and only after we inquired whether he saw something else, he reported on a small triangle (as in the mouth of the pacman), and then saw the big triangle. In the following displays, he was able to report the "big" illusory shape straight away. For this simple shape observation task, *this type of "local vision" behaviour is more typical of younger ages up to 6 years of age*, while at the age of 9 the global pattern should already emerge (Abravanel 1982, Nayar et al. 2015).

3.2.2.3. Pop outs

3.2.2.3.1. Procedure

In the first session, we presented BN with 8 different array displays, where one odd element in the array was visually different and usually "pops out". The distinctive dimension of the odd element included color (red square among green squares), orientation (among Gabor patches, or among blue bars), size (a thicker bar among other equally sized and oriented bars), shape (O among Xs, circle among squares), or shading (bottom light source among top light sources). In the second session (on a different day), we presented BN with 35 arrays, 14 of which were based on color cues (red or yellow circle among blue circles), and 21 which were based on Gabor orientations. The targets (across the displays) were located in most areas of the visual array (centre, surround, right, left, up, down). The stimuli were presented for an unlimited time until the expected response was provided or until the experimenter decided that such response cannot be provided for that display.

3.2.2.3.2. Results

In the first session BN was able to detect all the odd elements instantaneously, *apart from the oriented Gabor patch that he could not detect*, even though there were no time constraints. At the end we showed him that same display but rotated, and then he was succeeded in detecting it without trouble.

In the second session BN recognized the odd elements in all the displays, but took more time with the orientation task and said that the colored displays were easier for him.

3.2.2.4. Contour integration

3.2.2.4.1. Procedure

We assessed BN's contour integration ability using the contour integration card set (Kovacs et al. 1999, Kovacs et al. 2000, Doron et al. 2015) and compared it to previously published age-typical performance as well as to the group of age-matched controls participating in our study. Each card in the set consists of a round contour made of collinear Gabor patches ("signal") that is embedded in randomly oriented Gabor patches ("noise"). The task is to detect the contour and scan it with the finger once detected without touching the card. The cards are ordered by growing difficulty as the signal to noise ratio reduces. A practice card, easier than all the cards, is used to explain the task.

3.2.2.4.2. Results

As can be seen in Figure 2, all the control participants performed at or above the level of performance expected for their age (Doron et al. 2015). In contrast, *BN performed at the level of 3-4 year olds, significantly below his age level* (Kovacs et al. 1999, Doron et al. 2015). He successfully detected the contours up to and including card 5 (equivalent to a threshold of 0.975); at age 9, it is expected that the contours in cards 8 or 9 are detected. It is important to emphasize that the task has no time limit, giving participants as much time as they need to find the contour. We retested BN again on this same task three months later, and found that his performance had not improved and was still impaired (again he successfully detected only the contours up to and including card 5). To statistically compare his performance to age-level controls, and since contour integration skills continue to mature until adolescence (Kovacs et al. 1999), we compared BN's performance to 8 of the controls whose ages were within a year of BN's (aged 8.5-10.1 years, mean 9.46 ± 0.53 SD). BN's performance on the Kovacs cards was significantly worse than that of the closely age-matched controls' ($t(7) = -5.790$, p(2-tailed) = 0.001).

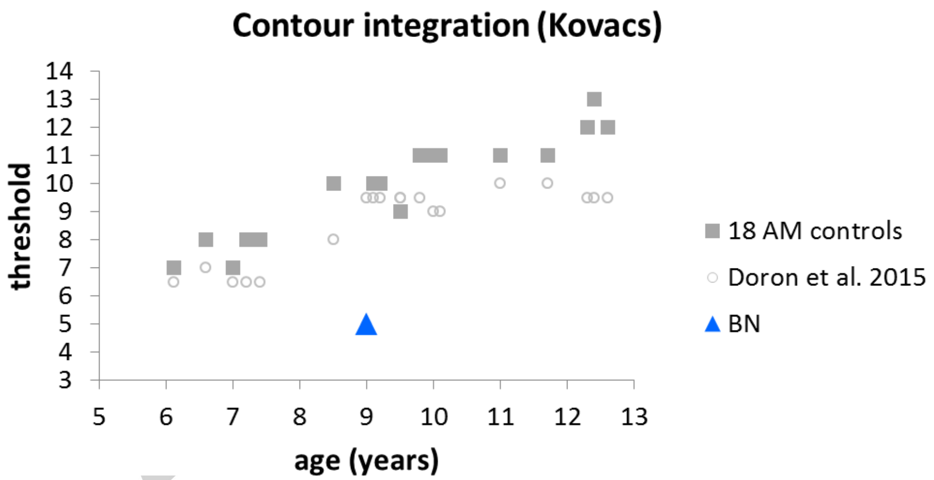


Figure 2. ***Collinear contour integration abilities in BN and age matched controls.*** While the age-matched controls (gray filled squares) performed at or above their age-level (data from (Doron et al. 2015) are indicated by gray empty circles), BN's performance (blue triangle) was significantly below that expected for his age, and matched that of 3-4 year olds (Doron et al. 2015). Note that there were no time restrictions while performing the task.

3.2.2.5. Figure-ground assignment

3.2.2.5.1. Procedure

Each stimulus comprised two overlaid surfaces, one white, the other black, and the task in each trial was to report which surface seemed in front (Brooks and Driver 2010, Brooks et al. 2012). The first two displays relied on local (non-contextual) cues and were presented together (one on the right and one on the left), each subtending approximately 4° x 3° of visual angle (width x height). The left display comprised a plain white rectangle that seemed to be continuing behind a slightly taller plain black rectangle (hence the black rectangle seemed in front); the right display comprised a plain white rectangle that seemed overlaid on a slightly taller plain black rectangle (hence the white rectangle seemed in front). The next four motion-based displays also relied on local non-contextual cues (i.e. were locally biased, see (Brooks and Driver 2010, Brooks et al. 2012)), and were presented consecutively. Each of these displays subtended approximately 2° x 2° of visual angle; one comprised a white rectangle with a black dotted pattern on it, the other a black rectangle with a white dotted pattern superimposed on it. These dots on the rectangles provided non-ambiguous local shape information, such that as they were both moving one seemed to be moving on top of the other. The last three motion-based displays required relying on non-local (contextual) cues as the decision was to be based on locally ambiguous information (Brooks et al. 2012). Each stimulus subtended a visual angle of approximately 2° x 5° and comprised two elongated moving rectangles, one white and one black, such that their upper part was as in the previous motion displays (dotted), their lower part contained no dots, and their upper and lower parts were separated by a red central horizontally-oriented occluding bar. The task was to attend the lower part of the display (without dots, hence ambiguous information) and report (based on the locally ambiguous information) which rectangle was in front. Despite attending the locally ambiguous area of the display, information from the top half of the display provided sufficient contextual information that allowed succeeding in the task.

3.2.2.5.2. Results

BN responded correctly to the two static displays based on local (non-contextual) cues, and to the four motion-based displays with local (non-contextual) cues. However, when required to rely on contextual cues, he answered incorrectly in one out of the three displays. Note, however, that this assessment was only observational; and we do not have sufficient data or any age-matched controls with which to compare his performance. Thus we cannot conclude if there was any deficit or if such behaviour is associated with normal development.

3.2.3. Object perception

One of BN's main perceptual deficits relates to object perception, and more particularly, as reported by his parents, to animal perception. We therefore tested BN with multiple tests, to define his deficits more precisely.

3.2.3.1. Object naming from colored or black and white photos

3.2.3.1.1. Procedure

BN was shown 18 colored or black and white photos and was asked to name them. These included a rooster, a penguin, a cow, a guitar, a plate, a shoe, a bottle, a key, a cup, glasses, scissors, a calculator, a paper clip, a baseball, a clock, a flashlight, and a bear. The control group was only shown three of these images (the rooster, penguin, and bear).

3.2.3.1.2 Results

BN correctly recognized almost all the images (including precisely naming the baseball), except for the bear which he incorrectly named as "dog". The eighteen age-matched control children correctly identified the images they were shown. No statistical comparisons were done for this task due to the small number of items the controls were shown.

3.2.3.2. Animal naming from line drawings, with or without occluding bars

3.2.3.2.1. Procedure

We assessed BN's animal naming performance using a small subset of the line drawn animals (penguin, bird, rooster, zebra and cat) which could appear behind occluding bars and that were used in previous studies ((Lerner et al. 2002, Gilaie-Dotan et al. 2009); see Figure 3C). We then compared his performance to that of age matched controls. There were no time restrictions. In the first testing session BN was shown the penguin, bird, and rooster without occluding bars, and the zebra and cat behind occluding bars. On a different day he was shown all the five line drawn animals behind occluding bars first, then (in a different order) the animals behind thinner occluding bars, and eventually without occluding bars. The controls were shown the images in the same order as BN in the second session.

3.2.3.2.2. Results

In the first testing session BN correctly identified the penguin, bird, and rooster (all shown without occluding bars), while he mistakenly named a zebra behind bars as "horse", and a cat behind bars as "dog". The second time this examination was repeated (on a different day) he, again, named the occluded zebra a "horse", and had difficulty with the cat behind the occluding bars (it took him a while to name it correctly). Apart from these he correctly identified all the other images.

All the controls correctly named all the images, except for the youngest control (aged 6.6) who named the zebra behind bars "giraffe", and another young control who named the occluded cat a unicorn. A statistical comparison between BN's and the controls' (even when assuming he made only two mistakes) indicates BN's naming performance was significantly worse than that of the controls ($t(17) = -3.467$, p(2-tailed) = 0.003).

3.2.3.3. SHEMESH object naming test

3.2.3.3.1. Procedure

We also administered to BN an object naming test comprising 100 drawn color images of objects from different visual categories that has age-based norms (the SHEMESH naming test, (Biran and Friedmann 2005)). Although this test was designed to detect language related deficits, its age-based norms allowed us to assess BN's naming performance according to his age-level. Furthermore, since the test includes items from multiple perceptual categories (food, body, electronics, tools, clothing, musical instruments, jewellery, animals, nature, outdoors, etc..), it allowed us to examine whether BN's deficits are indeed specific to animals or are more extensive. The age-matched controls were also asked to name a subset of the SHEMESH stimuli comprising 7 stimuli which BN correctly identified (bird, mushroom, elephant, key, camel, kettle, and clay vase), and 7 which he incorrectly identified (giraffe, leaf, ring, shell, jar, owl, and bear). We then compared his performance to theirs.

3.2.3.3.2. Results

As can be seen in Figure 3A, BN's visual naming performance was *significantly impaired* relative to his age level ((Biran and Friedmann 2005) and see http://www.tau.ac.il/~naamafr/shemesh.pdf), matching *the lower performance range expected at 7-8 years of age* (approximately two years younger than his chronological age). Specifically, he made 14 mistakes, 6 from the 25 rarest words in the test, but none from the most frequent 25 words. His mistakes included "stick" instead of axe (he said he did not know what that was), "red light" instead of traffic light, "crow" instead of owl, "box" instead of jar, "a very big dog" instead of a bear, "mouth" instead of tongue, "ship" instead of boat, "plant" instead of leaf, "watch" instead of ring, "black shirt" instead of a necklace (although the background behind the necklace could have resembled a shirt), and "zebra" instead of giraffe. Most of these mistakes appear semantic (Biran and Friedmann 2005), and yet they are not limited to the animate domain.

Only one young control (aged 7.2) made two naming mistakes (pot instead of kettle, glass instead of jar), while all the others correctly identified all 14 stimuli. We statistically compared BN's naming relative to that of the controls' according to very lenient statistical criteria. We chose 9 images that included the 7 images BN correctly named and only two of the mistakes BN made, and also accounted for the two mistakes the control participant made. Even with this liberal comparison, BN's naming was significantly worse than that of the controls ($t(17) = -3.899$, p(2-tailed) = 0.001).

3.2.3.4. Object size comparisons

3.2.3.4.1. Procedure

We asked BN to judge which of 4 pairs of colored photos was bigger in real life. The images appeared side by side and occupied the same visual angle.

3.2.3.4.2. Results

BN correctly named and identified the bigger item in reality (car vs shoe, dog vs apple, plate vs flower, table vs glasses).

3.2.4. Geometry

We designed the following tests especially for BN to more precisely define the substantial difficulties he had been having in geometry at school, as described by his parents.

3.2.4.1. Size comparisons

3.2.4.1.1. Procedure

BN was asked to determine which line was longer (with the same or different orientations), which rectangle was larger, which was the largest in an ensemble of six circles or six triangles, which was tallest of six differently shaped triangles whose bases were not always horizontally aligned.

3.2.4.1.2. Results

BN performed correctly on all of these tasks without any apparent difficulty.

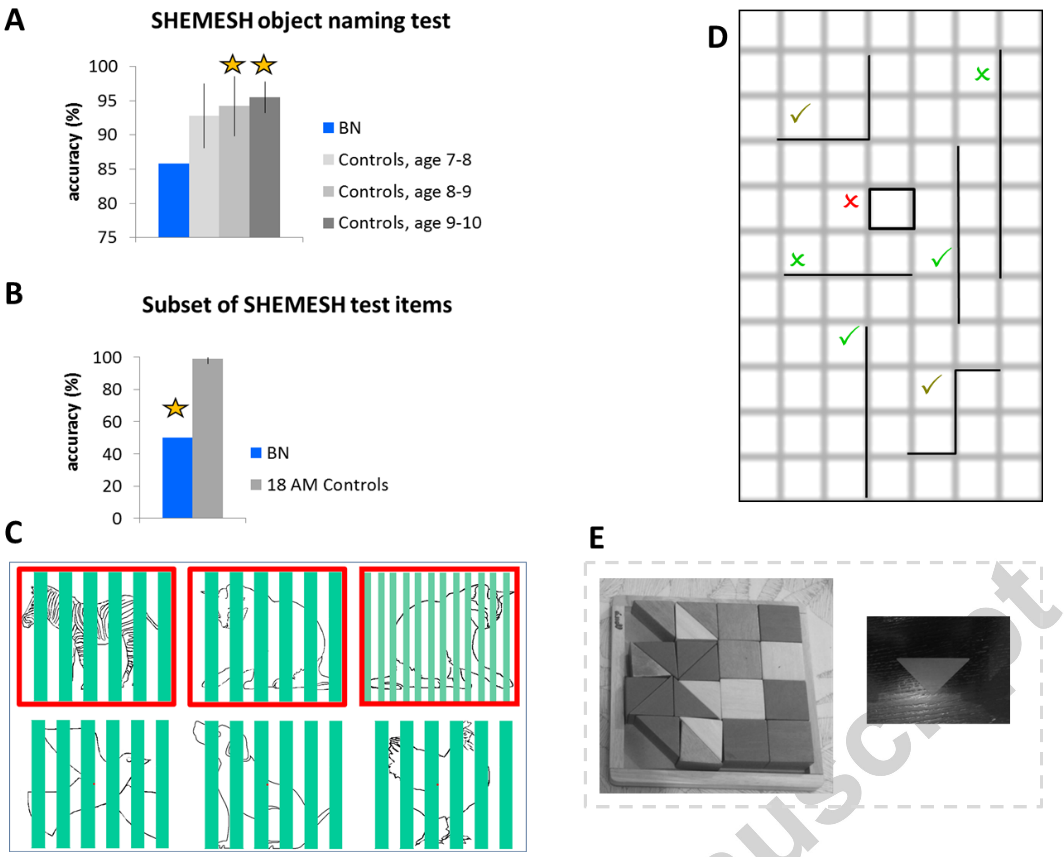


Figure 3. ***BN's object perception and naming (A,B and C), and geometrical difficulties (D and E)***. (A) BN was significantly impaired in naming 100 objects from multiple perceptual categories (the SHEMESH naming test (Biran and Friedmann 2005)) relative to age-matched controls (control data from http://www.tau.ac.il/~naamafr/shemesh.pdf), making 14 mistakes and performing at the lower end of children two years younger, or similarly to children three to four years younger. (B) The age matched controls in our study performed at ceiling when tested with seven of the items incorrectly named by BN and seven items that BN correctly named. (C) Outlined in red are occluded line drawn animals that BN misidentified ("horse" instead of zebra, "dog" instead of cat), and animals he correctly identified (the test set included only five line drawn animals). He repeated these mistakes in a different session. (D) BN's difficulties in estimating contour length, were evident when the contour became longer, was not straight or was closed. BN was not able to perceive the closed contour (the circumference of the rectangle) as a contour and treated it as a surface (answering "1"). (E) When asked to identify the shape on the right in the array on the left, BN had major difficulties and made multiple erroneous attempts (see Results).

3.2.4.2. Overlapping and occluding 2D geometric shapes

3.2.4.2.1. Procedure

We presented BN with 20 trials, each consisting of isolated or overlapping simple 2D geometric shapes (circles, rectangles, circles), either line drawn or filled with colors (red, light orange, or blue). BN was asked to describe what was in the image (name the shapes), describe their color, determine which was in front (closer to him), etc.

3.2.4.2.2. Results

His descriptions were very accurate, including when line drawn shapes were arranged between colored shapes or when three line drawn shapes overlapped one another.

3.2.4.3. Perception of 3D geometric shapes: matching images to real shapes

3.2.4.3.1. Procedure

We first placed three colored wooden toy blocks on a table (green squared, and red and green triangular blocks) and showed BN “3D” photos of each block, asking him to match the images to the real life blocks. The “3D” photos exposed two surfaces of each block and included perspective. This task was repeated with “2D” images (with only one characteristic surface of the block showing, without perspective, as in Figure 3E but in color), or with line drawings of the blocks.

3.2.4.3.2. Results

BN succeeded in the first task easily without touching the blocks; he also succeeded in the additional tasks without difficulty.

3.2.4.4. Perception of 3D geometric shapes based on 2D images

3.2.4.4.1. Discrimination

3.2.4.4.1.1. Procedure

BN was asked to point to the “odd one out” of three “3D” or three “2D” images (see above). The task was based on shape discrimination (regardless of color), or based on color discrimination (regardless of shape). One shape discrimination display comprised three “3D” images: two of square green blocks from very different viewing angles inducing very different appearances, and one of a triangular green block. Another shape discrimination display comprised three “2D” images: of triangular red, triangular green, and square green blocks. The color discrimination display was identical to the second shape discrimination display.

3.2.4.4.1.1. Results

BN performed on these tasks correctly.

3.2.4.4.2. Matching with color cues

3.2.4.4.2.1. Procedure

The task in each trial was to find the isolated block appearing on the right (as in Figure 3E on the right, but in color) in an array of blocks appearing on the left (as in Figure 3E on the left, but in color).

3.2.4.4.2.2. Results

BN performed this task flawlessly, but as he could have relied on color cues, in following sessions we removed the color cues.

3.2.4.4.3. Matching without color cues

3.2.4.4.3.1. Procedure and results

In the following session we showed him grayscale images of the blocks, in some images leaving only the edges. With the edges he seemed to perceive the shapes but did not excel in the task. In the display that appears in Figure 3E, he was asked to indicate where the shape on the right appeared in the array on the left. **He said he could only find two such**

blocks (there were 14). We later realized that he might have based his answer on the shades (i.e. not on the shape of the block alone), and therefore we showed him that display again and instructed him to disregard colors and answer again based on shape alone. He reported that he **could still see no more than 3 appearances, maybe 5**. We presented this display to the 18 age-matched controls, and only 65% of them answered correctly (with 58.3% accuracy for the controls aged 10 or younger), 3 of the youngest controls chose not to answer as it was too difficult for them. BN's performance was not significantly different from the controls' on this task ($t(17) = -1.083$, p(2-tailed) = 0.294). In another similar display BN was asked to detect the squared block in the same array (with 8 such items in the display, see Figure 3E). BN pointed to them and said he could **see "all six"**. As this answer was incorrect, we asked him to count again and he said **"all 12"**, again being incorrect. He might have perceived all the items but might have had difficulty in estimating how many there were.

3.2.4.5. Geometry related tasks

3.2.4.5.1. Procedure

We created multiple geometry related tasks for BN to practice on using a computer, which were related to the material he was studying at school. These included matching line orientations (finding all the lines in a clutter of differently oriented lines that were oriented the same as a target line), matching shapes under 2D rotations, identifying a piece from a variety of pieces as the missing part of an incomplete shape, determining the number of edges that formed some of the shapes, and identifying lines of a specific length.

3.2.4.5.2. Results

BN correctly matched line orientations. He also matched shapes under 2D rotations (again most of these correctly), and successfully identified a piece from a variety of pieces as the missing part of an incomplete shape.

BN experienced the greatest difficulty in tasks related to comprehension of line contour. This was evident in his difficulty in determining the number of edges that formed some of the shapes; for example, he could not determine for specific triangle or trapezoid if they had 3 or 4 edges. Most of the controls did not have trouble with determining which shapes had four edges (three of the younger controls, 7.4 years or younger, made one error each). BN made 3 mistakes in this task. Comparing his accuracy to the controls' showed a significant impairment on this task ($t(17) = -7.191$, $p<0.001$).

When BN was asked to identify lines of a specific length (e.g. 4 units) on a grid/squared paper background, he was reasonably successful for short lengths, but *had difficulties when the contours were not straight lines, or were longer* (see Figure 3D). The *most conspicuous difficulty in estimating contour length was when the contour was of a closed shape*. In this case he was unable to view it as a contour line, instead regarding only its surface area (in the example of Figure 3D he said that contour of the small square had a length of 1).

Five of the six youngest controls (aged 8.5 or younger) found the advanced contour estimation trials (e.g. Fig. 3D) difficult and were reluctant to do them. Overall for a subset of 5 contour estimation trials, the controls performed at 73.3% ± 11.9% (SD) accuracy (controls under 10 with 68.3% ± 10.3% (SD)), while BN performed at 60% accuracy ($t(17) = -1.09$, p(2-tailed) = 0.291), not significantly worse than the controls.

We emphasize that we did not compare his performance on all of these tasks to that of the controls, and we did not record response times, which also could have been informative. As

BN had significant difficulties with geometry at school (e.g. being unable to classify angle types, difficulty in calculating circumference of a shape), further investigations are required to better characterize his perceptual difficulties with geometry.

3.2.5. *Famous places recognition*

As place recognition may be related to geometry or space perception (with which BN has difficulties), we also assessed this ability.

3.2.5.1. Paradigm

Recognition of 21 famous places around the world (as Statue of Liberty, Arc de Triumph, Sydney Opera House, the Grand Canyon, Big Ben, Leaning Tower of Pisa, Eiffel Tower, Taj Mahal, Petra, Christ the Redeemer statue in Rio, Ayers Rock in Australia, Golden Gate Bridge, Colosseum in Rome, and others) was tested using color photos presented on a computer display, with no time restrictions. Accuracy levels were assessed.

3.2.5.2. Results

Figure 4 shows that BN recognized 9 out of 21 places (43% accuracy), which is slightly above but not significantly different from the mean of the controls (37.6% ± 18.03% (SD), $t(17) = 0.286$, p(2-tailed) = 0.778).

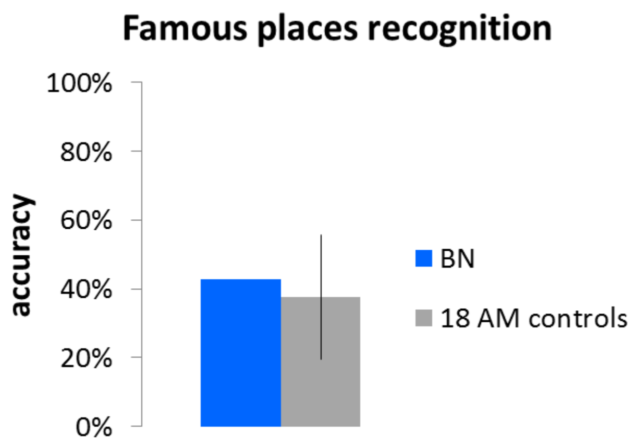


Figure 4. *BN's performance on a short famous places recognition test.* BN's recognition of famous places was similar to that of the controls.

3.2.6. *Motion vision*

We briefly assessed BN's visual motion perception using the following tasks.

3.2.6.1. 3D structure-from-motion (SFM)

3.2.6.1.1. Procedure

We presented BN with four consecutive displays of rotating 3D shapes composed of dots on a black background, three spheres, and one cube. Their directions of rotation varied, and the task was to describe the display.

3.2.6.1.2. Results

BN's description was very precise, including details about the direction of rotation ("like earth", "towards me", etc.).

3.2.6.2. Biological motion

3.2.6.2.1. Procedure

First, three point-light Johansson animations (Johansson 1973) of a leftward walking walker, a pitcher, and a person kicking sideways were presented without any noise dots (Gilaie-Dotan et al. 2011, Gilaie-Dotan et al. 2013b, Gilaie-Dotan et al. 2015). The task was to verbally describe the display. Second, a short blurred clip depicting a scene of children playing football in a playground was presented, and the task was to describe the display. Lastly, six filmed actions of actors (hand waving, skipping, running, or hopping) were presented (see Figure 5 in (Gorelick et al. 2007)) and the task was to describe the display in the most precise manner.

3.2.6.2.2. Results

BN correctly identified the human movements presented in point-light Johansson displays (Johansson 1973) and described them correctly: "like a baseball serve or tennis throw", "person stretching", and "martial arts". He correctly described that there were children playing football in a playground area in the blurred scene, and he correctly identified the actions of the actors in the six short videos.

3.2.6.3. Motion-induced-blindness (MIB)

3.2.6.3.1. Procedure. A MIB display (Bonneh et al. 2001) was presented on a computer screen for an unlimited time. The display subtended 6° x 6° of visual angle and consisted a green central fixation dot and three static yellow dots (at 1-1.5° from fixation) that were presented in front of a rotating array of blue crosses (on a black background). The task was to fixate on the green dot and report if any of the yellow dots disappeared.

3.2.6.3.2. Results

Upon viewing a MIB display, it took BN a while to report the disappearance of the yellow dots, but eventually he noticed that some of them disappeared at times, as is typically expected.

3.2.7. Face perception

BN's face perception has never been considered a problem as in his daily life he recognizes people and faces, even from images, and his family and educational staff have never encountered incidents of misidentification or difficulty in person recognition. To the contrary, his parents recall that once, when they were strolling down the street, to their surprise, BN spotted his grandmother behind a shop window and drew their attention to her (his parents note that they were not expecting to meet the grandmother on that occasion). In another incident, BN recognized his uncle in a charcoal drawing on the internet. We also felt that BN recognized people from images. For example, in one of the tasks where he had to discriminate between faces (same/different person) and was *not* supposed to name or recognize them, he pointed to one of them and said he was the president of the United States, and indeed it was an image of President Barack Obama. Importantly, this was not the only Afro-American looking man in the image set, and Obama was presented alongside his lookalike who also had his suit collar showing, so BN's recognition could not merely have been due to skin color or dress style.

As BN's face recognition and perception were not the main cause of our investigation and since we wished to understand the breadth of BN's visual deficits, we used a short face perception test that is part of a comprehensive visual screening battery for children we are developing. As this test is still under development, it does not yet have age-based norms, and has not been validated or compared with other face recognition tests (as the Cambridge Face Memory test for unfamiliar faces (Duchaine and Nakayama 2006) or the Benton Face Memory test (Benton et al. 1994)). We were not able to test BN on a normed face perception test (see (Dalrymple and Palermo 2016)) as he was no longer available for testing. Therefore we cannot determine the statistical reliability of the results presented here. We present BN's performance on this test in comparison to the 18 age-matched controls in this study.

3.2.7.1. Short face perception test

3.2.7.1. 1. Paradigm

Briefly, the short face perception test started with a face discrimination task (based on 23 trials of mostly unfamiliar people, see details below), followed by a face memory task for the unfamiliar people who had just been seen (22 trials) and ending with a short recognition test for familiar people (12 trials).

The first *face discrimination* phase required discriminating identities of unfamiliar people (i.e. 'same person' or 'different person' judgements) that were presented together. The test comprised 23 trials, 17 were based on images of children, 6 on images of adult. 19 of these trials included two photos presented side by side (10 'same person', 9 'different person'), and four included triplets of images ('indicate the odd person out'). Age, gender, hair color, hair type, and skin color were always matched in a trial. Ten of these trials included females, 13 included males. 17 trials were based on Caucasians, and six on Asians or Afro-Americans. All images were colored real life photos, within each trial all images were different, and across trials only 4 images reappeared. Participants were not informed that they would need to remember the people seen in these trials.

The second phase - *memory for unfamiliar faces* – comprised 22 trials of unfamiliar people, and required determining whether the person in the trial had been seen in the previous phase or not (11 were seen, 11 were not). Most of the images in the 'seen' trials were different from those used in the face discrimination trials (except for 3). 16 of these trials were photos of children, 6 of adults.

In the third phase - *familiar face recognition* - we included images of the experimenters, that all the children tested were familiar with, a popular figure from a children's show in Israel (but filtered to black and while and smeared, see Figure 5B), a figure of an unfamiliar child wearing a Santa hat, one of the experimenter's images with a Santa hat (see Figure 5C), and a display of five elliptic faces of Caucasian women with black hair. The child was asked to indicate if they recognized anyone, and if so, then who (two of these were the experimenters, the other three were unfamiliar women, see Figure 5D).

3.2.7.1. 2. Results

As can be seen in Figure 5A, BN accurately discriminated 69.6% of the unfamiliar people, which did not differ from the controls' performance (71.7% ± 8.5% (SD), BN vs. controls: $t(17) = -0.24$, $p(2\text{-tailed}) = 0.813$). BN's performance on the unfamiliar face memory (50%) was significantly worse than that of the controls' (84.3% ± 6.3% (SD), BN vs. controls: $t(17) = -5.299$, $p(2\text{-tailed}) < 0.00$)). BN did not perform significantly below the controls' mean on

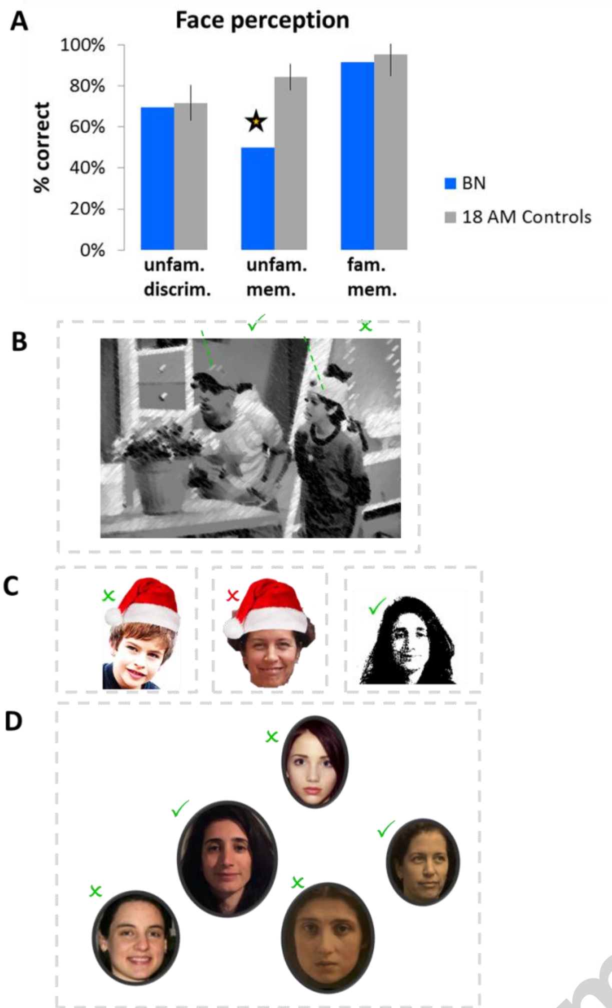


Figure 5. ***BN's performance on a short face perception test*****.** (A) While BN seemed to perform significantly below the control group on the memory for unfamiliar faces, he seemed to perform normally on unfamiliar face discrimination or with familiar faces, in line with his parents' reports that he has identified familiar people in unexpected places (see Results). We also observed that his recognition of familiar faces was adequate (see anecdotes in the text). (B) A familiar TV figure was correctly identified by BN (in green). (C, D) examples of identities that BN correctly identified (in green) or incorrectly (in red) or indicated he did not know. Note that in D pictures of the experimenters were embedded in additional pictures; BN correctly identified only the two experimenters and indicated he did not know the other women. None of these tasks was timed, but he was very quick to answer on all trials.

the familiar face recognition (BN: 91.7%, controls: 95.4% ± 10.5% (SD), BN vs. controls: $t(17) = -0.343$, p(2-tailed)=0.736).

3.2.8. Reading skills

We did not formally assess any of BN's reading skills in this study. His parents report that he can read Hebrew (his mother tongue), English, and possibly even another language. He reads books appropriate to his age level (as Dairy of a Wimpy Kid), frequently surfs the internet and for a few years has been very active on many English-based websites. There are also additional stories about him reading messages off school bulletin boards already in first grade. However, we cannot provide a veridical quantitative or qualitative estimate of these observations. During our testing sessions, some of the instructions appeared on the screen in English (not his mother tongue), and even though we did not expect him to read these, he once noted that he was not sitting at a proper distance indicated by the instructions (in English) on the screen (a sitting distance of 1.5 meters). He also read the words black and white that appeared during our experiments, without being asked to do so. We assume that if BN does experience some reading related difficulties, these may be minor in comparison to those he experiences in the perception of objects, geometry and space.

4. Discussion

In this study we have investigated a wide range of visual functions in BN, a 9 year old boy, concentrating on perceptual and oculomotor aspects. In line with his parents' reports, we found significant perceptual impairments that were most evident but not limited to the animate, contour and geometry domains. We also found abnormal oculomotor behavior of pursuit and saccades marked by prominent avoidance. As our investigations aimed to characterize BN's perceptual deficits, there are many domains which we only tapped into, and others that we did not cover at all. Despite these limitations, which we discuss below, we raise the possibility that his perceptual deficits are linked to his limited pursuit and saccadic ocular movements.

We have found that BN was impaired in contour integration (performed at an age level of 3-4 years) and in figure-ground separation (in some of the tests). These deficits are similar to those of LG, a young adult with developmental visual object agnosia (Ariel and Sadeh 1996, Gilaie-Dotan et al. 2009, Gilaie-Dotan 2016). Like BN, LG's contour integration was impaired when tested with the paradigm used here (Kovacs et al. 2000, Gilaie-Dotan et al. 2009, Lev et al. 2015). LG's performance at age 18 matched that of children aged 5-6. Similar to BN, LG's figure-ground segmentation was impaired for contextual cues while normal for local cues (Brooks et al. 2012). However, here we tested BN with a small subset of stimuli from LG's study, and found hints of impairments similar to LG's but these results are not substantiated due to insufficient data and lack of statistical power. Contour integration is also impaired in amblyopia (Polat et al. 1997, Chandna et al. 2001, Polat et al. 2004), a visual disorder assumed to reflect abnormal brain mechanisms, although the precise origins are still unclear (Levi 2013) . As opposed to basic visual functions, such as visual acuity, that develop early (Dobson and Teller 1978, Birch et al. 1983, Dobson 1993, Daw 1998), contour integration develops throughout childhood and matures at the age of 13-14 (Kovacs 2000, Doron et al. 2015). Both contour integration and figure-ground segmentation are assumed critical for facilitating many high level perceptual visual functions, such as edge assignment, grouping of elements in a scene, segmenting the scene, perceptual organization, and object and scene perception (Koffka 1935, Driver and Baylis 1996, Kovacs 2000, Polat and Bonneh 2000, Altmann et al. 2003, Sterkin et al. 2008, Brooks and Driver 2010, Silvanto et al. 2010, Brooks et al. 2012).

It is therefore not surprising that BN's object perception is impaired, given his impairments in contour integration and in separating figure from ground. His perceptual impairments are unlikely to arise from semantic origins for a number of reasons. First, the developmental specialists that have examined BN found normal or above normal language skills; his verbal and vocabulary scores in the Wechsler Preschool and Primary Scale of Intelligence (*WPPSI* (Lieblich 1971)) were also Average and above (see Case History). Second, in the contour integration test there was no reliance on semantic knowledge, and this is true for the illusory contours, the pop outs, the figure-ground assignment, and the geometry tasks. Finally, it would be hard to claim that BN misidentified a cat as a dog due to semantic deficits, when there is abundance of dogs and cats in the neighborhood he lives in.

Although we do not claim that BN has developmental visual agnosia, some of his perceptual deficits resemble those of LG, such as difficulties in recognizing animals behind bars (although caution should be exercised as LG underwent a more comprehensive examination), object naming, and reports about incidents that can be classified as "fragmented vision". LG also recalls having difficulties with geometry during his middle and high-school years, but not at elementary school (Gilaie-Dotan, personal communication). Similarly, BN's and LG's place recognition seem normal (Gilaie-Dotan et al. 2009). However there are also many dissimilarities. Neither LG nor his parents complained about specific difficulties in animal recognition, when these were distinct concerns raised by BN's parents. LG, unlike BN who is supported by a special educator, was never in need of any special education support, and does not have any difficulties in manipulating himself within his environment (Freud et al. 2016). The most obvious demonstrations of this being that LG used to work as a waiter, and one cannot tell that LG suffers from visual agnosia unless he himself speaks openly about it. However there are two dominant visual functions that LG and BN seem to differ in, face perception and eye movements, and these we discuss below.

Face perception skills are typically segregated into those relying on the form and appearance of the face (e.g. shape, features and their configuration, skin tone) known as "static" aspects, and those relying on the movements and kinematics of the face ("dynamic" aspects) (Haxby et al. 2000, Andrews and Ewbank 2004, Gilaie-Dotan 2014a, Gilaie-Dotan 2014b, Bernstein and Yovel 2015). As mentioned above, our investigations with BN were not focused on face perception, and thus, due to the limited time we had, we only briefly assessed some of BN's "static" face perception skills (without assessing his perception of facial expressions or other facial movements). To do this we used a short face perception test that is not yet fully developed and is not accompanied by normative data, and thus BN's performance was only compared to a group of age-matched controls. In this test we found that his discrimination of unfamiliar faces and his recognition of familiar faces were the same as those of the controls, while his short-term memory for the unfamiliar faces was significantly worse than that of the controls. BN's face perception was never considered a problem, and therefore it is difficult to reconcile this with the impaired short-term memory for unfamiliar faces. Therefore, it would have been beneficial to compare his face recognition skills to normed neuropsychological tests for children (e.g. those described in (Dalrymple and Palermo 2016)). However, BN was not available for further testing. Like other conditions (e.g. ADHD), there are still no absolute criteria for diagnosing prosopagnosia, and most diagnoses rely predominantly on self-reports that may be accompanied by neuropsychological face-related tests (Duchaine and Nakayama 2004, Duchaine and Nakayama 2006). Based on this "intuitive" definition of prosopagnosia, BN has never been suspected of prosopagnosia, and has surprised his parents more than once with recognizing individuals in unexpected occasions. There are also no recollections by his family or his educational staff of incidents where he confused or did not recognize familiar people, as is often the case with children suspected with prosopagnosia. If, as is now assumed, BN does not have prosopagnosia, his face recognition pattern would be different than that of many reported visual agnosic individuals who do suffer from prosopagnosia (e.g. LG (Gilaie-Dotan et al. 2009), SM, EC, CR (Gilaie-Dotan et al. 2013c, Freud et al. 2015, Gilaie-Dotan et al. 2015), but not CK (Behrmann et al. 1994, Moscovitch et al. 1997)). BN's performance on the three subsections of the short face test that we have administered was inconsistent, as he performed similar to controls in the unfamiliar face discrimination and familiar face memory sections, but significantly worse than controls in the unfamiliar face memory section. While this may seem unreasonable, it can indicate that even weak memory ability for unfamiliar faces may allow the formation of face memories when sufficient facial information is presented to the visual system, as in real world memory formations. As face recognition skills vary significantly among healthy sighted adults, from prosopagnosics (Behrmann and Avidan 2005, Kennerknecht et al. 2006, Dalrymple et al. 2012) to super-recognizers (Russell et al. 2009), without obtaining normative face perception scores (Benton et al. 1994, Duchaine and Nakayama 2006, Dalrymple and Palermo 2016) it is hard to determine where BN lies on this spectrum of face recognition abilities.

Oculomotor behavior of pursuit and saccades is another domain in which BN seems to differ from previously reported visual deficits like agnosia. We found that BN has significant impairments in both pursuit and saccades, differing from what is typically reported in visual agnosic individuals. For example LG, who was examined by an orthoptist at the age of 15, was not found to have any oculomotor dysfunction (Gilaie-Dotan, unpublished data). Other reported cases of visual agnosia we have studied (SM, CR, EC (Gilaie-Dotan et al. 2013c, Freud et al. 2015, Gilaie-Dotan et al. 2015) and JW (Marotta et al. 1997), but see (Rossit et al. 2010)), do not complain about and are typically not reported to suffer from oculomotor dysfunctions.

Thus, it is clear that oculomotor deficits are not the hallmark of visual perception deficits, but could they be sufficient to elicit visual perceptual deficits? Here we hypothesize that they could. Several oculomotor dysfunctions are associated with certain visual perceptual deficits, such as in subtypes of amblyopia (Levi and Klein 1985, Kiorpes et al. 1999, Kovacs et al. 2000, Ellemberg et al. 2002, Lerner et al. 2003, Polat et al. 2005, Lerner et al. 2006, Bonneh et al. 2007, Hayward et al. 2011) or in nystagmus (Merrill et al. 2011, Thomas et al. 2011, Barot et al. 2013, Barot et al. 2014). Some of these conditions involve strabismus or nystagmus and thus may be related to ongoing instability of the retinal image which may lead to noisy form or object representations in the brain (Rao and Ballard 1999, Friston 2009, Friston and Kiebel 2009). However, BN's oculomotor dysfunctions are very different: they do not involve nystagmus or strabismus, and therefore it is unlikely that his retinal image is instable. BN also seems to avoid making pursuits and his saccades are imprecise. But would such abnormally limited pursuit and imprecise saccades be involved in or cause his perceptual deficits? We have to acknowledge the possibility that his perceptual deficits are unrelated to his oculomotor deficits, and that they both result from an abnormally developed visual system. However, we hypothesize that a causal link does exist between his perceptual deficits and his oculomotor deficits. Thus, although we have no data to support such a causal link, we suggest that BN's oculomotor deficits adversely affected some of his perceptual abilities. BN was significantly impaired in object perception, perhaps more profoundly with animals, in contour integration and in some geometric tasks, with no clear indications of prosopagnosia. This pattern of results appears to differ from a number of reported cases of form perception deficits that co-occur with prosopagnosia (LG (Gilaie-Dotan et al. 2009), CR, SM, EC (Gilaie-Dotan et al. 2013c, Freud et al. 2015), but see CK (Behrmann et al. 1994, Moscovitch et al. 1997)), which have led to suggestions that faces and common objects are processed within a common joint network (for review see (Farah 2004)). Given our findings with BN, we propose that normal face perception and perhaps also reading, which both (i) rely on foveal vision, (ii) are associated with foveal bias in high-order visual cortex, and (iii) rely on limited eye movement range (approximately 5-10° visual degrees once the target is within the foveal vicinity), are not critically dependent on fluent pursuit and saccades. In contrast, we hypothesize that normal contour and common object perception may rely on fluent, non-limited and precise pursuit and saccades. We hypothesize that normal object and contour perception rely not only on information from across the visual field reaching the retina, but also on normal development of object selective cortex that shows a mid-peripheral bias (Hasson et al. 2002, Malach et al. 2002, Hasson et al. 2003). We hypothesize that pursuit and saccades might be involved in a circuit evaluating the saliency of all possible targets in peripheral vision (to determine to where the eyes will move next), and that this valuation component leads to the natural mid-peripheral bias in high-order object selective cortex. If this circuit does not function properly (as in BN), then object-selective high-order cortex may not develop properly (see Figure 6). Interestingly, recent studies show that normal observers make ultra-rapid saccades to the correct location when detecting objects such as animals in peripheral visual field (Guyonneau et al. 2006, Kirchner and Thorpe 2006, Bacon-Mace et al. 2007).

Since according to the hierarchical clustering of cortical responses, animals and animal bodies are clustered close to faces (Kriegeskorte et al. 2008, Grill-Spector and Weiner 2014), it is reasonable to hypothesize that they may rely on more central biased representations in high-order visual cortex. We assume that animals might be a special visual category for BN, given his limited pursuit and the animals' typical movements in the environment. These factors possibly lead to underdeveloped perceptual representations of animals in BN's high level visual cortex. The other categories BN has difficulties in (e.g. food, plants, and man-made objects) are more distant from human faces according to the hierarchical clustering scheme (Kriegeskorte et al. 2008, Grill-Spector and Weiner 2014), and therefore, even according to this scheme, it is reasonable to hypothesize that they rely on mid-peripheral eccentricity bias in high-order visual cortex.

We emphasize that these do not rule out the possibility that perception of objects and faces share and rely on common mechanisms. In fact, in Figure 6 we propose not only how early and high-order visual cortex are organized relative to eccentricity (Figure 6A), similarly to the proposal of Malach and colleagues (Levy et al. 2001, Hasson et al. 2002, Malach et al. 2002, Hasson et al. 2003), but also how specific visual categories are supported in high-order visual cortex according to eccentricity needs and biases (Figure 6B). Such eccentricity needs and biases may be partly based on normal pursuit and saccades. The heat map in Figure 6B illustrates the hypothesized weights that each area contributes to the processing of specific visual categories (faces, words, and common objects). Faces rely heavily on foveally biased regions predominantly in the right hemisphere (e.g. fusiform), but they also rely on similar foveal representations in the left hemisphere and on mid eccentricity cortical regions, but to a lesser extent (Figure 6B top right (Levy et al. 2001, Behrmann and Plaut 2014, Rangarajan et al. 2014)). The representation of words in high order visual cortex mirrors that of faces (right-left flip) so that the highest weights are found in the left ventral cortex (left fusiform VWFA (Cohen et al. 2002, Dehaene et al. 2002, Hasson et al. 2002, McCandliss et al. 2003, Weiner et al. 2014)). Objects, in contrast to faces and words, have the highest weight values in mid and peripheral biased right ventral cortex (Hasson et al. 2002, Malach et al. 2002, Sayres and Grill-Spector 2008), but also rely to a lesser extent on foveally biased high-order cortex (Avidan et al. 2002). Such a division of labor fits many previous reports, including neuroimaging activation studies that suggest a modular representation in high order visual cortex based on peaks of activation per category (Epstein and Kanwisher 1998, Downing et al. 2001, Spiridon and Kanwisher 2002, Downing et al. 2006, Kanwisher and Yovel 2006, Schwarzlose et al. 2008, Pitcher et al. 2009), and also with studies suggesting more distributed representations in high-order visual cortex (Ishai et al. 1999b, Haxby et al. 2000, Behrmann and Plaut 2013, Dundas et al. 2013, Behrmann and Plaut 2014). Furthermore, it also fits with neuropsychological cases, in which perceptual impairment stems from a lesion to the core (red on the heat map) or to a dominant portion of the network supporting a category. This model can explain how one can be impaired in face recognition but hardly in word or object recognition (Farah et al. 1998), or in words but to a much lesser extent in objects and faces (Sekuler and Behrmann 1996, Behrmann and Plaut 2014), or in faces and objects but not words (Behrmann and Kimchi 2003, Gilaie-Dotan et al. 2013c)), or in objects and words but not faces (Behrmann et al. 1994, Moscovitch et al. 1997), or with objects but much less so with faces and perhaps in words, like BN (also see (Rumiati et al. 1994, Germine et al. 2011)). We hypothesize that in BN, who is mostly impaired in objects and contour perception, mid to peripheral representations in high order visual cortex did not develop properly (see black dashed line in Fig. 6B, bottom). This may be attributed to BN's abnormally limited saccades and pursuit movements, which on the one hand limit his accessibility to mid and peripheral visual field, and on the other hand might also inhibit or hinder normal flow from mid and peripheral early vision (retina and early visual areas) to high-order cortex, and thus might adversely affect the development of object selective cortex.

Functional deficits of high-level vision can be complex. Attempts to correct them are still in their infancy and have not yet been successful (Polat et al. 2004, Behrmann et al. 2005, Lev et al. 2015). Given that we hypothesize that BN's visual perceptual deficits are linked to his impairments in pursuit and saccades, which was not the case of earlier treatment attempts, the odds of treating his perceptual deficits may be somewhat different. Non-invasive methods for correcting eye movement deficits, such as visual training (also termed visual therapy (VT)) paradigms, are easy to follow, usually accessible, and have high success rates (Fischer and Hartnegg 2000, Kerkhoff et al. 2013, Leong et al. 2014). We are hopeful that such training procedures and BN's young age will allow him to improve his pursuit and saccades, in turn allowing perceptual representations to develop more normally. BN now practices with an interactive whole body TV game, and at times with a dedicated computer practice we have created for him. Future examinations of his perceptual and oculomotor abilities may shed additional light on the hypotheses raised here on the relationship between oculomotor and perceptual functions.

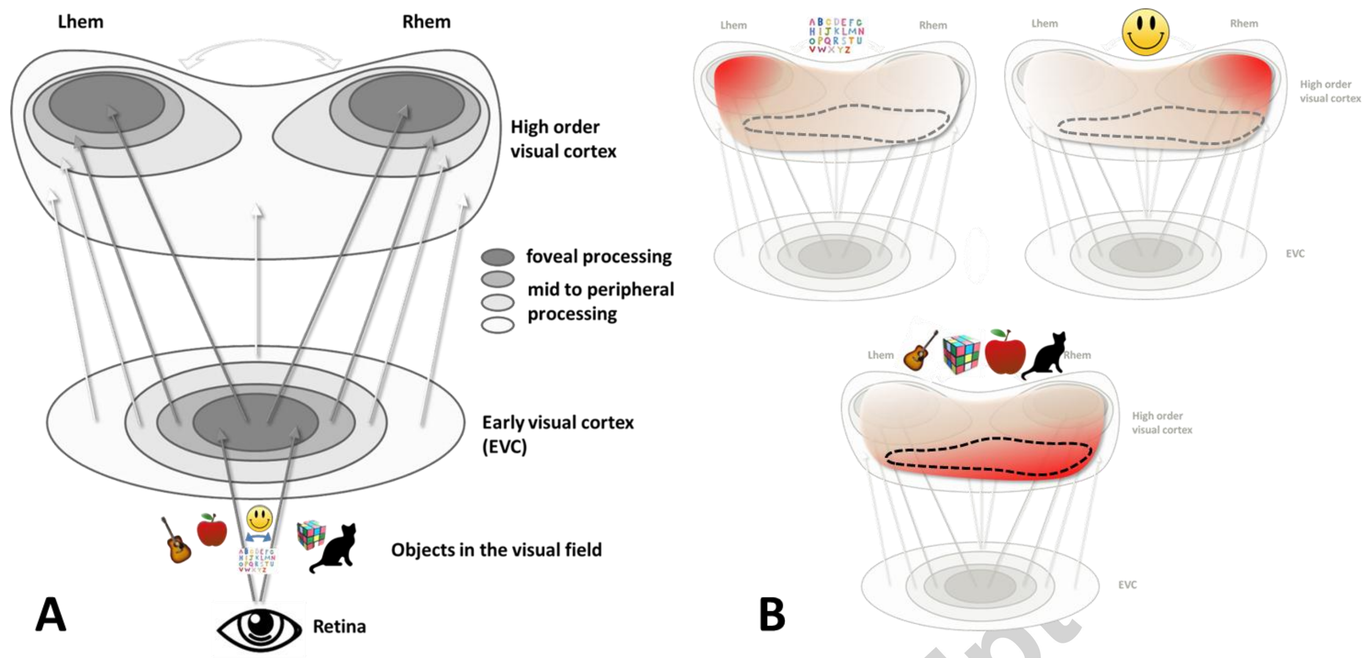


Figure 6. ***Model proposing why high-order visual object selective cortex shows a mid-peripheral bias.*** Based on BN's oculomotor and perceptual impairments, we propose that limited pursuit and saccades (as with BN) might be associated with inhibition of salient peripheral information. This can hinder information flow to peripheral high-order visual cortex and prevent the normal development of object selective cortex. Note that faces and words (A), once fixated upon, remain in the foveal vicinity and do not require extensive saccadic range to keep them in focus. In (B) the heat maps represent the weighted dependencies of each category representation (words - above left, faces – above right, objects – bottom) on hemisphere and eccentricity biased cortex, with red indicating the strongest reliance. Such weighted dependencies fit many neuroimaging studies and neuropsychological case reports. We hypothesize that given BN's limited pursuit and saccades, his object selective cortex - showing a mid-peripheral bias and relying on mid-peripheral inputs, did not develop normally (dashed dark contour), and that this significantly influenced his perception of common objects (bottom), while sparing face perception and perhaps reading (top right and left).

5. Conclusions

We report a child with significant visual perceptual deficits in contour integration and object perception, but with no indications of severe face recognition deficits such as prosopagnosia. His perceptual deficits are coupled with significant impairments in oculomotor pursuit and saccades. We propose that pursuit and saccadic eye movement may play a critical role in the development of normal object perception. Furthermore, we propose that BN's visual behavior can suggest why object representations in high-order visual cortex show a mid to peripheral eccentricity bias. We hope that future cases and studies will be able to test our hypotheses.

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Table 1. Summary of the functions BN was tested on.

	Test or task	Findings	Significant deficits	Determined by
Assessments prior to this study				
Basic visual functions				
	Distance visual acuity	Normal	No	Optometrist, orthoptist
	Convergence	Normal	No	Orthoptist
	Accommodation	Normal	No	Orthoptist
	Orthophoria	Present (Normal)	No	Orthoptist
	Binocular vision	Normal depth perception, positive fusion range, and normal binocular vision	No	Orthoptist
	Ocular motility	**Difficulties in pursuits**	NA	Orthoptist

	test			
	Other observations	No strabismus	NA	Orthoptist
Assessments in this study				
Eye movements				
	NSUCO Oculomotor optometric test	**Difficulties in pursuit and saccades**	Yes	Optometrist
	King-Devick test for eye tracking skills	**Impairment in time to complete**	Yes	Comparison to age-based norms
		Impairment in number of errors	Probably	Comparison to age-based norms
Basic to mid level vision				
	Randot stereopsis test	Normative stereo acuity	No	Optometrist
	Kanizsa (illusory) figures	**Local vision (typical up to 6 years of age)**	NA	
	Popouts	*Difficulties only with orientation-based items*	NA	
	Kovacs contour integration test	**Contour integration level of 3-4 year olds**	Yes	Comparison to age-matched controls and age-based reports
	Figure-ground assignment	Possible difficulty with contextual information	NA	
High level vision				
Object related				
	Object naming (color photos)	*Mistakenly naming bear a dog*	NA	
	Animal naming	**Impaired**	Yes	Relative to age-

	(line drawings, behind occlusion)			matched controls
	Object naming (SHEMESH test, color drawings)	**Impaired**	Yes	Relative to age-matched controls and to age-based reports
	Object size comparisons (photos)	No apparent difficulty	NA	
Geometry related				
	Size comparisons	No apparent difficulty	NA	
	Naming overlapping or occluded geometric shapes	No apparent difficulty	NA	
	Matching images to real shapes (of wooden blocks)	No apparent difficulty	NA	
	Counting shapes in an array (grayscale photos)	*Difficulty and errors*	No	Relative to age-matched controls
	Matching line orientations	No apparent difficulty	NA	
	Matching shapes under 2D rotation	No apparent difficulty	NA	
	Finding a missing piece	No apparent difficulty	NA	
	Determine which shapes have 4 edges	**Difficulty and errors**	Yes	Relative to age-matched controls

	Contour length estimation (on a grid/squared paper background)	*Difficulty at longer and complicated contours*	No	Relative to age-matched controls
Places				
	Famous places recognition	Normal	No	Relative to age-matched controls
Visual motion				
	Structure from motion (SFM)	No apparent difficulty	NA	
	Biological motion	No apparent difficulty	NA	
	Motion induced blindness (MIB)	No apparent difficulty	NA	
Face related				
	Unfamiliar face discrimination (short screening test)	No apparent difficulty	No	Relative to age-matched controls
	Unfamiliar face memory (short screening test)	**Impaired**	Yes	Relative to age-matched controls
	Familiar face memory (short screening test)	No apparent difficulty	No	Relative to age-matched controls

Significant deficits are indicated in bold, possible deficits in italics. NA – no available (or insufficient) information to determine significance of a deficit.

Highlights

- A boy with object and contour perception deficits, difficulty in geometry is reported
- No indication of prosopagnosia
- Normal basic level vision but limited pursuit and imprecise saccades
- Proposal: Normal pursuit and saccades could be critical for perceptual development
- Proposal: Object-selective cortex's mid-peripheral bias linked to eye movements