DR JONATHAN M PAYNE (Orcid ID : 0000-0001-9565-3845)

Article type : Original Article

[Original article: 2 tables; 3 figures; 1 online appendix]

Attention to faces in social context in children with neurofibromatosis type 1

AMELIA K LEWIS[1]
MELANIE A PORTER[1]
TRACEY A WILLIAMS[2,3]
SAMANTHA BZISHVILI[1]
KATHRYN N NORTH[4,5]
JONATHAN M PAYNE[4,5]

1 Department of Psychology, Macquarie University, Sydney; **2** ARC Centre of Excellence in Cognition and its Disorders, Macquarie University, Sydney; **3** Kids Rehab, The Children's Hospital at Westmead, Sydney; **4** Murdoch Children's Research Institute, The Royal Children's Hospital, Parkville; **5** Department of Paediatrics, Faculty of Medicine, Dentistry and Health Sciences, University of Melbourne, Melbourne, Australia.

Correspondence to Jonathan M Payne, Murdoch Children's Research Institute
The Royal Children's Hospital, Flemington Road Parkville, Victoria 3052, Australia. E-mail: jonathan.payne@mcri.edu.au

PUBLICATION DATA

This is the author manuscript accepted for publication and has undergone full peer review but has not been through the copyediting, typesetting, pagination and proofreading process, which may lead to differences between this version and the Version of Record. Please cite this article as doi: 10.1111/dmcn.13928

Accepted for publication 00th Month 2018.
Published online 00th Month 2018.

ABBREVIATIONS

ASD	Autism spectrum disorder
FSIQ	Full-scale IQ
NF1	Neurofibromatosis type 1
SRS	Social Responsiveness Scale

AIM To examine visual attention to faces within social scenes in children with neurofibromatosis type 1 (NF1) and typically developing peers.

METHOD Using eye-tracking technology we investigated the time taken to fixate on a face and the percentage of time spent attending to faces relative to the rest of the screen within social scenes in 24 children with NF1 (17 females, 7 males; mean age 10y 4mo [SD 1y 9m]). Results were compared with those of 24 age-matched typically developing controls (11 females, 13 males; mean age 10y 3mo [SD 2y]).

RESULTS There was no significant between-group differences in time taken to initially fixate on a face (p=0.617); however, children with NF1 spent less time attending to faces within scenes than controls (p=0.048). Decreased attention to faces was associated with elevated autism traits in children with NF1.

INTERPRETATION Children with NF1 spend less time attending to faces than typically developing children when presented in social scenes. Our findings contribute to a growing body of literature suggesting that abnormal face processing is a key aspect of the social-cognitive phenotype of NF1 and appears to be related to autism spectrum disorder traits. Clinicians should consider the impact of reduced attention to faces when designing and implementing treatment programmes for social dysfunction in this population.

[First page footer]

DOI: 10.1111/dmcn.xxxxx

[Left page footer]

Developmental Medicine & Child Neurology 2018, 60: 000–000

What this paper adds

- Children with neurofibromatosis type 1 (NF1) demonstrated atypical gaze behaviour when attending to faces.
- NF1 gaze behaviour was characterized by normal initial fixation on faces but shorter face dwell time.
- Decreased attention to faces was associated with elevated autism traits in the NF1 sample.

[Main text]

Neurofibromatosis type 1 (NF1) is an autosomal dominant genetic disorder associated with reduced social skills and cognitive deficits.[1–3] There has been recent interest in better understanding how individuals with NF1 process social information, including faces. To this end, studies have identified deficits in social cognition more broadly,[4,5] and face perception more specifically,[5–7] in both adults and children with the condition. These findings suggest that aberrant face processing may be an important feature of the NF1 social phenotype. Eye-tracking technology has provided researchers with a useful means of exploring the manner in which attention, as measured by eye gaze, is allocated to different classes of visual stimuli. While eye gaze is not completely analogous to attention, evidence suggests that scan path analysis allows researchers to map the spatiotemporal locations of visual attention to develop a better understanding of how individuals analyse and perceive visual material.[8–10] To date, only one study has used eye-tracking technology to investigate face processing in children with NF1 and found that the amount of time spent viewing key individual facial features, such as the eyes, did not differ from a typically developing comparison group.[7] Nevertheless, children with NF1 spent significantly less time viewing internal facial features overall than controls, suggesting the degree of visual attention they allocate to faces, at least when presented in isolation, is reduced.

While studying attention to faces presented in isolation may yield valuable information, important aspects of genuine social interactions are absent from these stimuli.[11] Complex social scenes containing both social and non-social information represent a particularly useful class of stimuli for eye-tracking studies as they allow investigation of attention to faces in a more ecologically valid manner. While no studies to date have examined social scene processing in individuals with NF1, reduced attention to faces or people in social scenes has been documented in other neurodevelopmental disorders characterized by impaired social processing, including autism spectrum disorder (ASD), Williams syndrome, and Fragile X syndrome.[9–12] It is important to investigate whether children with NF1 display aberrant scene-processing patterns, particularly in light of the growing body of evidence suggesting impaired social information processing in this population.[4,6]

The aim of this study was to investigate attention (as measured by eye gaze) to faces in children with NF1 within a semi-naturalistic setting, in the context of static social scenes. We investigated whether children with NF1 take longer to first fixate on a face and/or spend less time overall fixated on faces within a scene than typically developing peers. A secondary aim was to investigate relationships between eye gaze parameters and participant characteristics, including age, intelligence, quantitative ASD traits, and face perception. We hypothesized that children with NF1 would take longer to first fixate on face stimuli and spend less time viewing faces within social scenes than typically developing controls. We predicted that a longer time to first face fixation and less time viewing faces would be associated with elevated ASD traits in NF1 participants. Based on theoretical links between face perception skills and attention to faces in the environment,[10,13] we also expected poorer face recognition skills in children with NF1 to be associated with a longer time to first face fixation and a smaller percentage of time spent attending to faces within a scene.

METHOD

Participants

Participants were 24 individuals with NF1 and 24 typically developing controls matched individually for chronological age (within 6mo) and matched at the group level on handedness. All participants displayed normal/corrected-to-normal vision.

Participants with NF1 were recruited via the Neurogenetics Clinic at The Children's Hospital at Westmead, Sydney, Australia. This clinic caters for over 1300 individuals with NF1 and has a wide referral base, with all socio-economic groups represented. Children were

recruited if they met the following selection criteria: (1) confirmed diagnosis of NF1 based on clinical criteria;[14] (2) absence of diagnosed intracranial pathology (e.g. traumatic brain injury, symptomatic optic glioma); (3) a Full-scale IQ (FSIQ) greater than or equal to 70; (4) no current/previous diagnosis of anxiety, mood, or psychotic disorder; and (5) competency in the English language. Review of clinical records revealed that five NF1 participants had a diagnosis of attention-deficit–hyperactivity disorder (ADHD). They were included in our study in order to provide the most representative sample of children with NF1 in light of the high rates of comorbid ADHD reported in this population.[15] No participant with NF1 had been diagnosed with ASD. Approval for the study was granted by The Children's Hospital at Westmead and Macquarie University Human Research Ethics Committees. Written informed consent was obtained from all participants' carers.

Typically developing controls were recruited through Neuronauts, a children's research participation club at Macquarie University, Sydney, Australia. Exclusion criteria were a history of developmental delay, sensory impairment (that would impact on normal development and on their ability to complete the research tasks), intellectual or other cognitive impairment, neurological or psychiatric disorder, or English as a second language. No children were excluded based on these criteria.

Table I shows the demographic characteristics of each group. χ^2 analysis revealed no significant difference in sex distribution between the groups, although a non-significant trend was observed whereby there was a somewhat higher proportion of female participants in the NF1 group. χ^2 test confirmed no group difference in handedness. An independent samples *t*-test confirmed that the groups were well-matched on chronological age.

Materials

Stimuli included 18 images of social scenes taken from the International Affective Picture System.[16] The International Affective Picture System is a set of photographs depicting various stimuli, including animals, social scenes, and landscapes that is widely used in studies of emotion, arousal, and visual tracking.[17] Each image used in the current study contained at least one person in a natural scene, with at least one face visible (IDs: 2235, 2272, 2299, 2393, 2396, 2398, 2480, 2514, 2560, 2575, 2579, 2590, 2593, 2594, 2598, 2749, 5875, 7550).

FSIQ was assessed with the Wechsler Intelligence Scales for Children, Fourth Edition in the NF1 group,[18] and the Wechsler Abbreviated Scale of Intelligence in typically developing controls.[19] FSIQs generated from these two scales are substantially correlated (0.86) and have very high convergent validity.[20] The parent version of the Social

Responsiveness Scale (SRS) was used to identify quantitative ASD traits in participants with NF1.[21] The SRS consists of 65 items rated on a 4-point Likert scale that form five subscales of social awareness, social cognition, social communication, social motivation, and autistic mannerisms. Scores obtained across the subscales were summed to provide an SRS total score, which was reported in this study. SRS questionnaires were obtained from a parallel study of social competence in NF1 (*n*=18) and were only available for participants with NF1.[22] Face perception was examined using the Facial Recognition Test, which assessed identification and discrimination of unfamiliar human faces based on black-and-white photographs.[23] Our examination and scoring procedures have been previously described.[6]

Procedure

Participants viewed the images on a Dell 16-inch FP monitor in a darkened room. Scenes were presented in the centre of the computer screen at a standardized size of 25.14cm (950 pixels) × 18.84cm (712 pixels), width by height. The eye-tracking task was completed in a single testing session.

Eye-tracking procedure

Participants' eye movements were recorded using an Eyelink 1000 (SR Research, Ottawa, Canada) remote eye-tracking camera at a sampling rate of 500Hz. Calibration, validation, and scan path recordings were made for the right eye for most participants; however, calibration for the right eye was unsuccessful for two participants in the NF1 group. For these participants, the calibration procedure was repeated for the left eye and subsequent eye movement recordings were made for the left eye.

A 9-point calibration method was used before the social scene trials. Participants were instructed to fixate on a centrally placed black dot (10mm in diameter), which appeared in the centre of the screen. The dot then moved to eight different locations around the screen, and participants were instructed to follow its movements with their eyes. The dot moved to a new location after participants had fixated on the dot for at least 1000ms. The experimental procedure proceeded once adequate calibration was achieved to ensure accurate fixation recording was possible at all points on the computer screen.

A black dot was presented in the centre of the screen for 1000ms immediately before each social scene to control for the initial point of retinal attention. Participants were only able to progress to the next social scene trial once they had fixated continuously on this central dot for 200ms. Once adequate central fixation was obtained, manual experimenter

control initiated the next trial, and the central fixation dot disappeared and was replaced by a social scene, ensuring that all participants were attending to the centre of the screen as soon as the scene appeared. Central fixation reappeared in between each social scene trial. In order to record the most naturalistic scan path information, participants were instructed only to 'look at' each image. Scene stimuli were viewed passively for 10 000ms each.

Areas of interest

Areas of interest were traced using the Eyelink Data Viewer freehand drawing function. For all scenes, a 'Faces' area of interest was defined as the sum of all faces (traced around the hairline) within the scene.

Visual scan path parameters

Visual scan path parameters selected for analysis included 'mean time to first fixation' (mean length of time in milliseconds for the first fixation to enter a defined area of interest); 'mean dwell time per cent' (mean percentage of time spent attending to an area of interest relative to the total time spent attending to the screen); and 'mean fixation per cent' (mean percentage of fixations made within a defined area of interest). Similar patterns of results were found for 'mean dwell time per cent' and 'mean fixation per cent', and so only the former is reported. An adjusted 'mean dwell time per cent to faces' was generated, defined as 'mean dwell time per cent to faces' divided by the 'mean dwell time per cent' to the whole scene image. The adjusted 'mean dwell time per cent' was used for analyses investigating whether children with NF1 spent less time overall viewing faces than controls (hypotheses 2 and 3), as it enabled us to ensure that the experimental results reflected 'true' attention to face stimuli in the context of the social scenes rather than 'off-task' behaviour (e.g. off-screen eye movements). This was considered particularly important given the rate of comorbid ADHD in the NF1 group.

Statistical analysis

Data were analysed using Predictive Analytics SoftWare Statistics Version 18 (SPSS Inc., Chicago, USA) for Windows. Group differences were explored using an independent samples *t*-test. Given the uneven male:female ratio between groups, Pearson product–moment correlations were conducted to explore relationships between sex and our eye-tracking variables of interest to address the possibility of bias due to sex effects. Pearson correlations were also used to investigate relationships between eye-tracking variables and participant

characteristics, including age, FSIQ, ASD traits, and face perception. A *p*-value of 0.05 was used for all analyses to indicate statistical significance.

RESULTS

Do children with NF1 take longer initially to look at faces within a scene?

Figure 1 displays the 'mean time to first fixation' to face stimuli in milliseconds, averaged across all social scenes for both groups. There was no significant difference between groups in the amount of time taken to initially fixate on faces ($t_{(46)}$=0.50, p=0.617, Cohen's d=0.15), suggesting that children with NF1 initially focused their visual attention on faces within a social scene as quickly as the comparison group. There were no significant relationships between sex and 'mean time to first fixation' in the NF1 or typically developing control groups (both $p \geq 0.82$).

Do children with NF1 spend less time viewing faces within a scene?

Figure 2 displays the 'mean dwell time per cent to faces' (averaged across all social scenes) for NF1 and typically developing control groups. An independent samples *t*-test revealed a significant difference between the groups, such that, on average, the NF1 group spent less time attending to faces in the scenes than typically developing controls ($t_{(46)}$=–2.03, p=0.04; Cohen's d=0.59). There were no significant relationships between sex and adjusted 'mean dwell time per cent to faces' in the NF1 or typically developing control groups (both $p \geq 0.343$). Box plots of 'mean time to first fixation' and 'mean dwell time per cent' to faces are included as supplementary data (Appendix S1, online supporting information).

Is attention to faces within social scenes related to participant characteristics?

Table II shows correlations between the eye-tracking variables ('mean time to first fixation', 'mean dwell time per cent to faces') and the participant characteristics of age, FSIQ, ASD traits, and face perception abilities. There were no significant relationships between gaze behaviours and age in either group (all $p \geq 0.30$). While higher FSIQ was associated with a quicker 'mean time to first fixation' (p=0.04) and a longer 'mean dwell time per cent to faces' (p=0.02) in the typically developing comparison group, there were no significant associations between these gaze behaviours and FSIQ within the NF1 sample (all $p > 0.44$). Associations between ASD symptoms and gaze behaviours revealed a significant positive correlation between SRS total scores and 'mean time to first fixation' in the NF1 group (p=0.03; Fig. 3). There was also a significant negative correlation between SRS total scores

and ‘mean dwell time per cent to faces’ (p=0.02) for NF1 participants. No correlations between gaze behaviours and face perception approached statistical significance for either the NF1 (all $p \geq 0.25$) or typically developing group (all $p \geq 0.21$).

Although numbers are small, we conducted an exploratory analysis investigating whether there were any demonstrable differences in eye-tracking parameters between participants with NF1 only and those with comorbid ADHD. There were no differences in either ‘time to first fixation’ ($t_{(22)}$=–1.69, p=0.16) or ‘dwell time per cent to faces’ in NF1 participants with versus without ADHD ($t_{(22)}$=–1.93, p=0.11).

DISCUSSION

The aims of this study were to investigate the manner in which children with NF1 attend to faces within a social scene and to investigate the relationship between attention to faces and the participant characteristics of age, FSIQ, ASD symptoms, and face perception skills. Contrary to our predictions, children with NF1 did not take longer than the comparison group to first fixate on a face within a social scene. This suggests that children with NF1 do not initially avoid looking at faces and are initially attracted to faces as readily as typically developing controls. However, as predicted, children with NF1 spent significantly less time overall looking at faces within the scenes compared with controls. This is consistent with the limited available research in the area,[6] which suggests that children with NF1 spend less time looking at faces than typically developing children when presented in isolation. This study extends previous findings by replicating this effect using more ecologically valid stimuli, which included both social and non-social information. Furthermore, by analysing the adjusted ‘mean dwell time per cent’, our results reflect attention allocated to faces within social scenes rather than time spent looking off-screen, thereby accounting for elevated levels of ‘off-task behaviour’ that might be expected owing to the attention deficits that are common in children with NF1.[24] These findings add to the growing body of research supporting face-processing abnormalities in individuals with NF1.[5–7]

In relation to our secondary aims, our hypothesis that attention to faces would be correlated with ASD traits in children with NF1 was supported. Specifically, results indicated that elevated ASD traits were associated with a longer time taken to first fixate on a face and shorter times fixating on faces overall. These findings suggest that participants with NF1 with elevated ASD symptoms have a tendency towards decreased face gaze and an atypical allocation of attention. Thus, for participants with NF1, faces did not capture attention above and beyond other aspects of the environment to the same degree as typically developing

children, a trend greater in individuals with elevated ASD symptoms. From a developmental perspective, reduced attention to faces is expected to impact on the social functioning and maturation of social cognitive abilities of affected children. If children with NF1 spend less time than is typical viewing faces, it is likely that they will have decreased access to the important social cues contained within the face. This will result in less opportunities to perfect skills of social communication that are derived from facial expressions and will likely affect the learning of appropriate social behaviours.

For the ages studied here, there were no associations between face-directed gaze behaviour and age, in either group. However, we cannot assume that the patterns observed here will be evident in patients outside of this age range, particularly infants, toddlers, and young children with NF1. Indeed, evidence from a large pooled sample suggests that severity of SRS-derived ASD traits vary considerably from preschool-aged children (less elevated) to school-aged children (more elevated).[25] Given the developmental importance of viewing faces in infancy and early childhood, it will be important to examine face gaze characteristics in a younger cohort of children with NF1, to determine whether the patterns evident in the current study will be replicated, or whether, like parent-reported ASD symptoms,[25] face-directed gaze behaviour is also attenuated in younger children.

The results of the current study are somewhat consistent with similar studies reported in the idiopathic ASD literature. While we did not demonstrate the reduced time to first fixation that is typical in idiopathic ASD studies, our finding of shorter overall times focused on faces is consistent with the idiopathic ASD literature.[26,27] The significant relationships we report between length of face gaze and level of severity of ASD symptoms – with more severe ASD symptoms related to decreased face gaze – have also been replicated in previous idiopathic ASD studies.[27] This finding adds important behavioural data to the growing evidence base linking the NF1 phenotype to ASD-typical behaviours.[25,28] However, there are some differences between behavioural features typically seen in ASD and those reported in NF1. For example, our previous eye-tracking study reported normal face-scan paths in children with NF1, including attention to the eye region.[7] While too few studies exist to form firm conclusions, emerging findings also suggest that ASD features observed in NF1 are characterized by better eye contact than idiopathic ASD.[29] So while the current findings indicate atypical attention to faces in NF1, the broader NF1 literature suggests relatively well-preserved eye contact and scanning of face features.

Our final hypothesis, that attention to faces would be associated with face perception skills in children with NF1 was not supported. Although significant face perception deficits

were evident in our NF1 sample, correlational analyses failed to detect a relationship between these skills and attention to faces. Of note, these skills were also not significantly related in the comparison group. These findings are consistent, in part, with those of Wilson et al.,[10] who failed to detect a significant relationship between time spent viewing people and face perception skills in their sample of children with ASD, but identified a significant relationship between poorer face-matching skills and a preference for initially fixating on objects over faces in this group. We did not investigate relative attention to faces versus objects in the present study; however, this will be an important task for future research.

Further study will be necessary to explore the mechanisms underlying the reduced attention to faces identified in children with NF1. In particular, it would be interesting to examine the influence of different task instructions on scene scanning behaviour in this cohort. While the present study investigated more naturalistic scanning of social scenes with minimal task instructions (i.e. to 'look at' each scene), the addition of an explicit social-cognitive task (e.g. 'describe what is happening in this scene') may alter the allocation of visual attention to faces in children with NF1. It would also be interesting to repeat our study using dynamic social scenes that more closely approximate real-world social interactions.[11]

Investigating the relationship between attention to faces and day-to-day social functioning in children with NF1 will also be an important focus for future research. There is increasing evidence to suggest that NF1 is associated with both face-processing abnormalities and significantly poorer social outcomes compared with the general population.[1,5–7] While the findings of the current study indicate preliminary support for a relationship between atypical face-directed attention and ASD-related behavioural difficulties, the associations between these variables in children with NF1 remain unclear and the psychological constructs driving the observed poor attention to faces are not known. Future research should examine relationships between attention to faces in children with NF1 and social cognitive outcomes, observer-reported measures of day-to-day social functioning, and psychological well-being. Developing a better understanding of how reduced attention to faces might impact on daily social functioning in children with NF1 would inform the development of social intervention and remediation programmes at home and school.

Our present findings suggest that clinicians and education professionals should consider the potential impact of reduced attention to faces in children with NF1 when designing and implementing treatment programmes for social dysfunction in this population. Faces are a unique class of visual stimuli, containing large amounts of socially relevant information, including another person's age, sex, thoughts and feelings, and familiarity.[30] As

such, the ability to attend to and decode information from faces is critical for successful reciprocal social interactions. The present results suggest that children with NF1 may benefit from explicit instruction and practice aimed at teaching them how to attend to information from faces in their day-to-day social interactions. Designing and trialling intervention programmes and examining their efficacy in children with NF1 will be important tasks for future studies in the field of NF1.

The main limitation of the present study was our relatively small sample size. Although an NF1 sample of 24 children is reasonable in relation to other studies in this population,[4,6] a larger sample size would be beneficial in future studies so that relationships between face processing, cognitive abilities, behavioural features, and external factors (e.g. family environment, socio-economic status) can be explored in more depth. The two groups were also not matched on socio-economic status. As NF1 is associated with significantly higher rates of ADHD, ASD, and social anxiety disorder than the general population,[15,25,31] future research should include subtypes of these comorbidities. For example, to better understand how syndromic ASD in individuals with NF1 affects face processing and attention to the eye region, NF1 cohorts should be split into those with and without ASD and preferably compared with a matched idiopathic ASD group. As face-processing abnormalities have been identified in individuals with idiopathic ADHD, ASD, and social anxiety disorder, it may also be valuable to consider the potential mediating effects of these comorbid psychopathologies on face processing in NF1.[10,12,32,33]

In summary, our findings suggest that children with NF1 spend less time attending to faces than typically developing children when presented in the context of static social scenes. Exploring the mechanisms underlying this effect, as well as the relationships between reduced attention to faces and real-world social functioning in children with NF1 will be essential areas for future research.

ACKNOWLEDGEMENTS

The authors gratefully acknowledge Naomi Sweller for her statistical expertise. This work was partially supported by a Murdoch Children's Research Institute fellowship awarded to JMP. The authors have stated that they had no interests that might be perceived as posing a conflict or bias.

Supporting information

The following additional material may be found online:

Appendix S1: Box plots demonstrating participant performances on the two primary eye-tracking outcomes.

REFERENCES

1. Barton B, North K. Social skills of children with neurofibromatosis type 1. *Dev Med Child Neurol* 2004; **46**: 553–63.

2. Hyman SL, Shores EA, North KN. The nature and frequency of cognitive deficits in children with neurofibromatosis type 1. *Neurology* 2005; **65**: 1037–44.

3. Pride NA, Crawford H, Payne JM, North KN. Social functioning in adults with neurofibromatosis type 1. *Res Dev Disabil* 2013; **34**: 3393–9.

4. Payne JM, Porter MA, Pride NA, North KN. Theory of mind in children with neurofibromatosis type 1. *Neuropsychology* 2016; **30**: 439–48.

5. Pride NA, Korgaonkar MS, Barton B, Payne JM, North KN. The genetic and neuroanatomical basis of social dysfunction - lessons from neurofibromatosis. *Hum Brain Mapp* 2014; **35**: 2372–82.

6. Huijbregts S, Jahja R, de Sonneville L, de Breij S, Swaab-Barneveld H. Social information processing in children and adolescents with neurofibromatosis type 1. *Dev Med Child Neurol* 2010; **52**: 620–5.

7. Lewis AK, Porter MA, Williams TA, Bzishvili S, North KN, Payne JM. Facial emotion recognition, face scan paths, and face perception in children with neurofibromatosis type 1. *Neuropsychology* 2017; **31**: 361–70.

8. Noton D, Stark L. Eye movements and visual perception. *Sci Am* 1971; **224**: 34–43.

9. Williams TA, Porter MA, Langdon R. Viewing social scenes: a visual scan-path study comparing fragile X syndrome and Williams syndrome. *J Autism Dev Disord* 2013; **43**: 1880–94.

10. Wilson CE, Brock J, Palermo R. Attention to social stimuli and facial identity recognition skills in autism spectrum disorder. *J Intellect Disabil Res* 2010; **54**: 1104–15.

11. Risko EF, Laidlaw K, Freeth M, Foulsham T, Kingstone A. Social attention with real versus reel stimuli: toward an empirical approach to concerns about ecological validity. *Front Hum Neurosci* 2012; **6**: 143.

12. Riby DM, Hancock PJ. Viewing it differently: social scene perception in Williams syndrome and autism. *Neuropsychologia* 2008; **46**: 2855–60.

13. Nelson CA. The development and neural bases of face recognition. *Infant Child Dev* 2001; **10**: 3–18.

14. Neurofibromatosis. Conference statement. National Institutes of Health Consensus Development Conference. *Arch Neurol* 1988; **45**: 575–8.

15. Payne JM, Hyman SL, Shores EA, North KN. Assessment of executive function and attention in children with neurofibromatosis type 1: relationships between cognitive measures and real-world behavior. *Child Neuropsychol* 2011; **17**: 313–29.

16. Lang PJ, Bradley MM, Cuthbert BN. International Affective Picture System (IAPS): technical manual and affective ratings. Gainesville, FL: University of Florida, Centre for Research in Psychophysiology, 1999.

17. Mikels JA, Fredrickson BL, Larkin GR, Lindberg CM, Maglio SJ, Reuter-Lorenz PA. Emotional category data on images from the International Affective Picture System. *Behav Res Methods* 2005; **37**: 626–30.

18. Wechsler D. Wechsler Intelligence Scales for Children – Fourth Edition. San Antonio, TX: The Psychological Corporation, 2003.

19. Wechsler D. Wechsler Abbreviated Scale of Intelligence: Manual. San Antonio, TX: Pearson, 1999.

20. Flanagan DP, Kaufman AS. Essentials of WISC-IV assessment. 2nd edn. Chichester: John Wiley and Sons, 2009.

21. Constantino JN. The Social Responsiveness Scale. Los Angeles: Western Psychological Services, 2002.

22. Lewis AK, Porter MA, Williams TA, North KN, Payne JM. Social competence in children with neurofibromatosis type 1: relationships with psychopathology and cognitive ability. *J Child Dev Disord* 2016; **2**: 2.

23. Benton AL, Sivan AB, Hamsher Kd, Varney NR, Spreen O. Contributions for neuropsychological assessment. 2nd edn. Orlando, FL: Psychological Assessment Resources, 1994.

24. Payne JM, Arnold SS, Pride NA, North KN. Does attention-deficit–hyperactivity disorder exacerbate executive dysfunction in children with neurofibromatosis type 1? *Dev Med Child Neurol* 2012; **54**: 898–904.

25. Morris SM, Acosta MT, Garg S, et al. Disease burden and symptom structure of autism in neurofibromatosis type 1: a study of the international NF1-ASD consortium team (INFACT). *JAMA Psychiatry* 2016; **73**: 1276–84.

26. Guillon Q, Hadjikhani N, Baduel S, Rogé B. Visual social attention in autism spectrum disorder: Insights from eye tracking studies. *Neurosci Biobehav Rev* 2014; **42**: 279–97.

27. Riby DM, Hancock PJ. Do faces capture the attention of individuals with Williams syndrome or autism? Evidence from tracking eye movements. *J Autism Dev Disord* 2009; **39**: 421–31.

28. Plasschaert E, Descheemaeker MJ, Van Eylen L, Noens I, Steyaert J, Legius E. Prevalence of autism spectrum disorder symptoms in children with neurofibromatosis type 1. *Am J Med Genet Part B* 2015; **168B**: 72–80.

29. Garg S, Plasschaert E, Descheemaeker MJ, et al. Autism spectrum disorder profile in neurofibromatosis type I. *J Autism Dev Disord* 2015; **45**: 1649–57.

30. Kanwisher N, Moscovitch M. The cognitive neuroscience of face processing: an introduction. *Cogn Neuropsychol* 2000; **17**: 1–11.

31. Pasini A, Lo-Castro A, Di Carlo L, et al. Detecting anxiety symptoms in children and youths with neurofibromatosis type I. *Am J Med Genet Part B* 2012; **159B**: 869–73.

32. Chen YP, Ehlers A, Clark DM, Mansell W. Patients with generalized social phobia direct their attention away from faces. *Behav Res Ther* 2002; **40**: 677–87.

33. Marsh P. Impaired emotion perception in ADHD and first-episode schizophrenia: How are they different? Saarbrucken: VDM Verlag, 2008.

Table I: Demographic characteristics of study and comparison groups

	NF1 group	TD group	T score
Sex: male/female (*n*)	7/17	13/11	3.09[a]
Right-handed/left-handed (*n*)	22/2	22/2	0.00[a]
Age (y:mo) Range (y:mo)	10:4 (1:9) 6:8–12:1	10:3 (2:0) 6:8–13:5	0.05
FSIQ[b]	90.54 (9.83)[c]	106.71 (12.19)[d]	–5.06[g]
SRS total[e]	60 (15.61)	–	–
Face perception[f]	–0.56 (1.03)	0.50 (1.05)	–3.56[g]

Values are mean (SD) unless otherwise noted. [a]χ^2 statistic. [b]Standard score, mean 100 (SD 15). [c]Wechsler Intelligence Scale for Children, Fourth Edition. [d]Wechsler Abbreviated Scale of Intelligence. [e]*n*=18, T score mean 100 (SD 15). [f]Z-score, mean 0 (SD 1). [g]$p<0.01$. NF1, neurofibromatosis type 1; TD, typically developing; FSIQ, Full-scale IQ; SRS, Social Responsiveness Scale.

Table II: Pearson's product–moment correlation coefficients between eye-tracking variables and face perception and Full-scale IQ (FSIQ)

Eye-tracking variable	Age	FSIQ	SRS total	Face perception[a]
NF1 group				
Mean time to first fixation to face	0.09	–0.17	0.51[b]	0.25
Mean dwell time per cent	–	0.06	–0.51[b]	–0.22
TD group				
Mean time to first	–0.22	–0.41[b]	–	–0.01

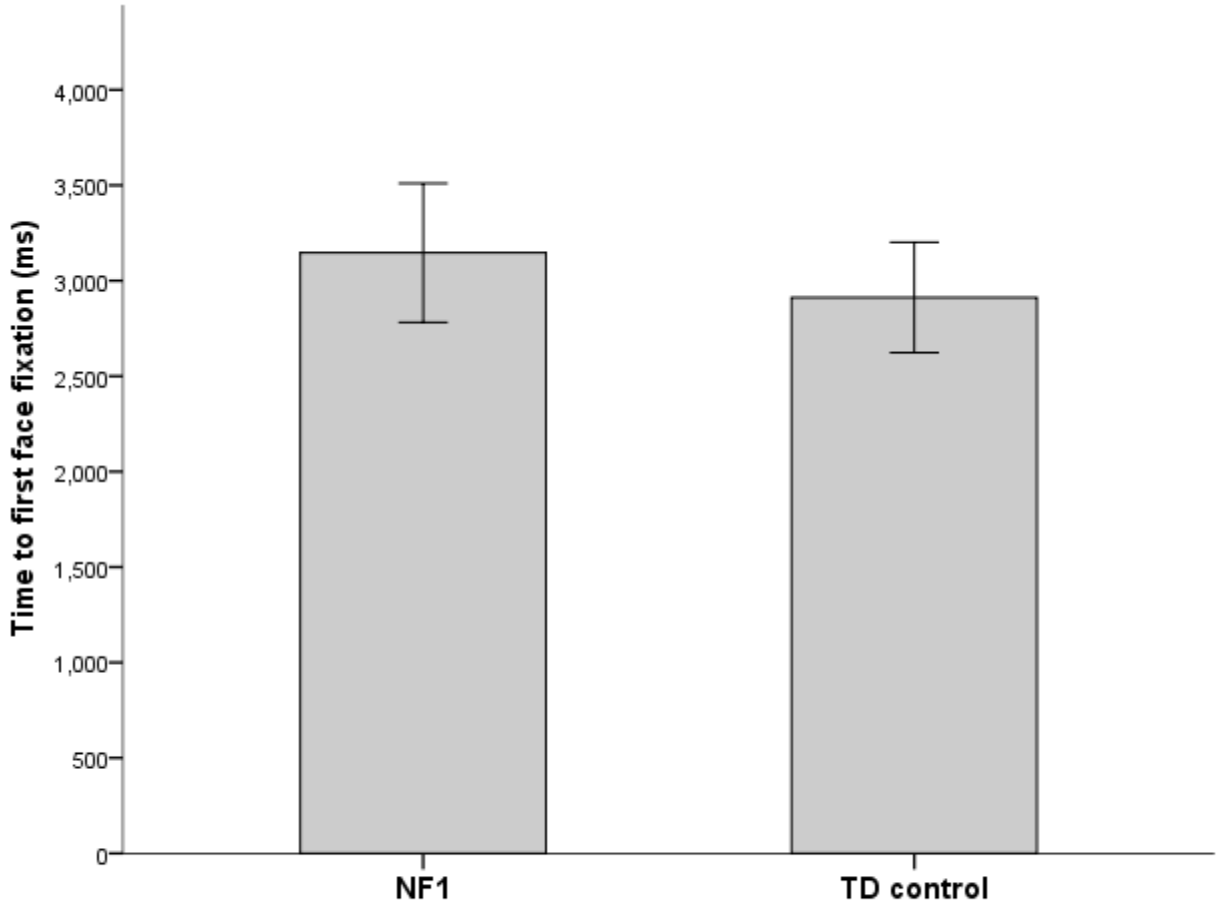
fixation to face				
Mean dwell time per cent	–0.16	0.49[b]	–	0.26

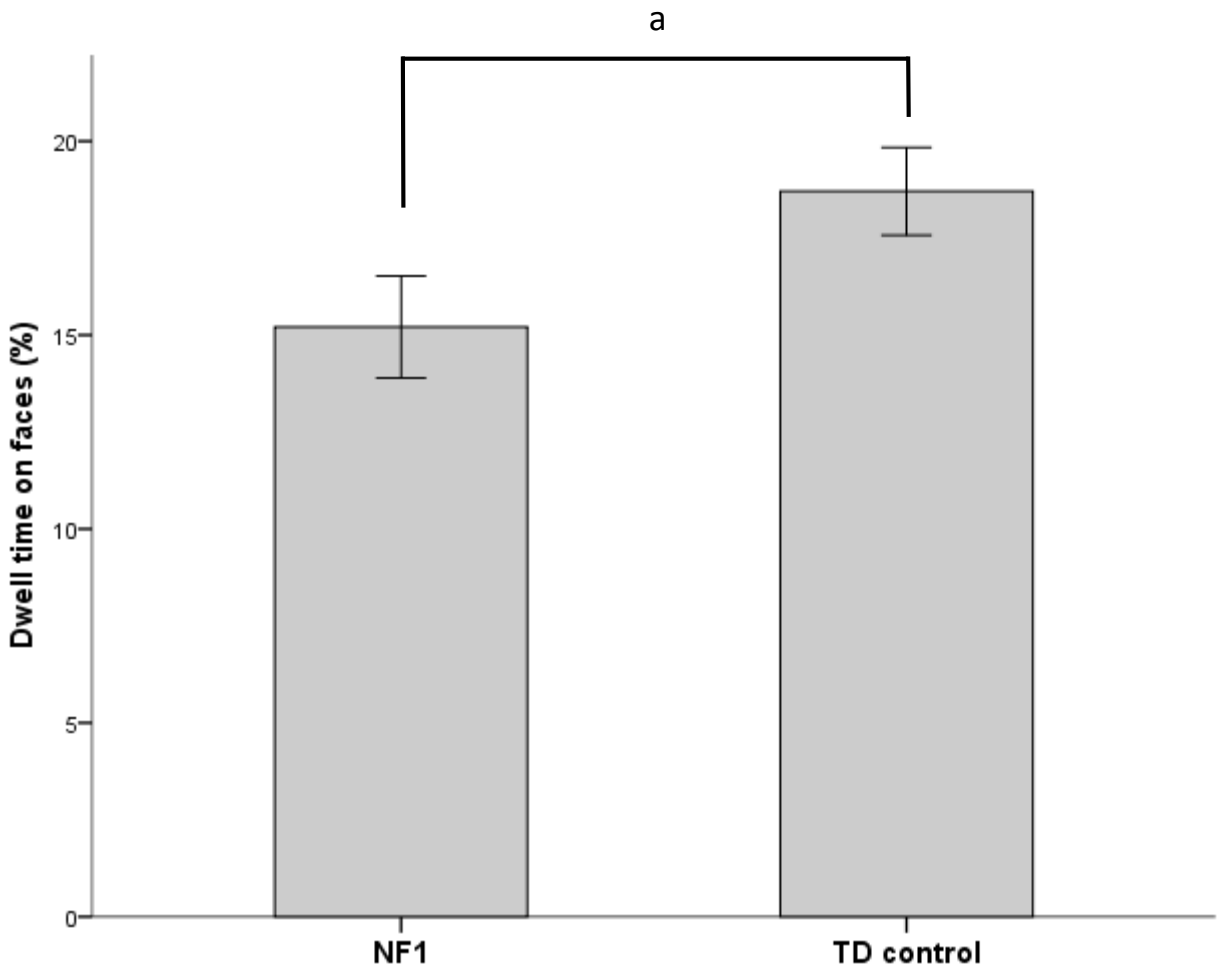
[a]Facial recognition test. [b]$p<0.05$. SRS, Social Responsiveness Scale; NF1, neurofibromatosis type 1; TD, typically developing.

Figure 1: Mean time to first fixation to face stimuli for neurofibromatosis 1 (NF1) and typically developing (TD) control groups. Error bars represent ±1 standard error.

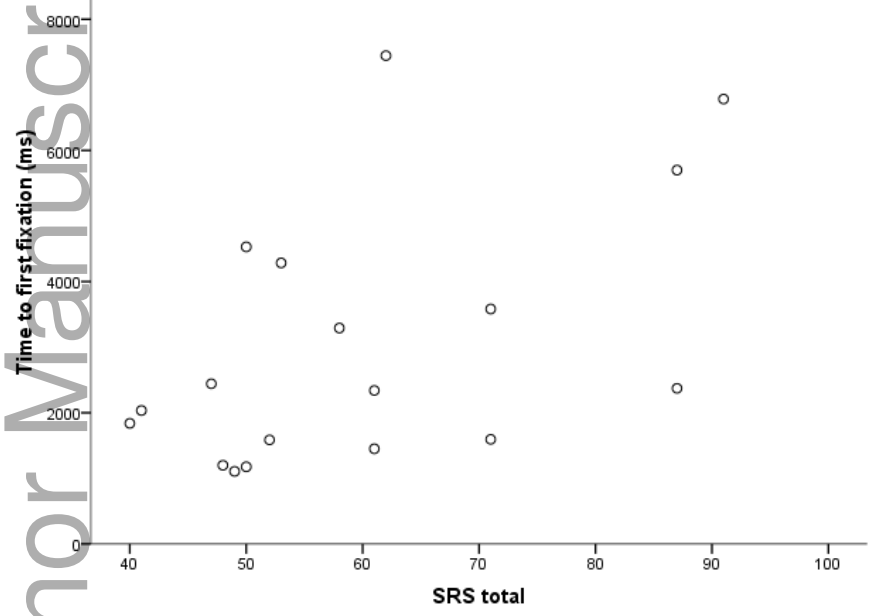
Figure 2: Adjusted mean dwell time per cent to faces for neurofibromatosis 1 (NF1) and typically developing (TD) control groups. Error bars represent ±1 standard error. [a]$p<0.05$.

Figure 3: Scatter plots representing relationships between T scores as measured by the Social Responsiveness Scale (SRS) total score and the eye-tracking parameters of (a) time to first fixation and (b) mean face dwell time.

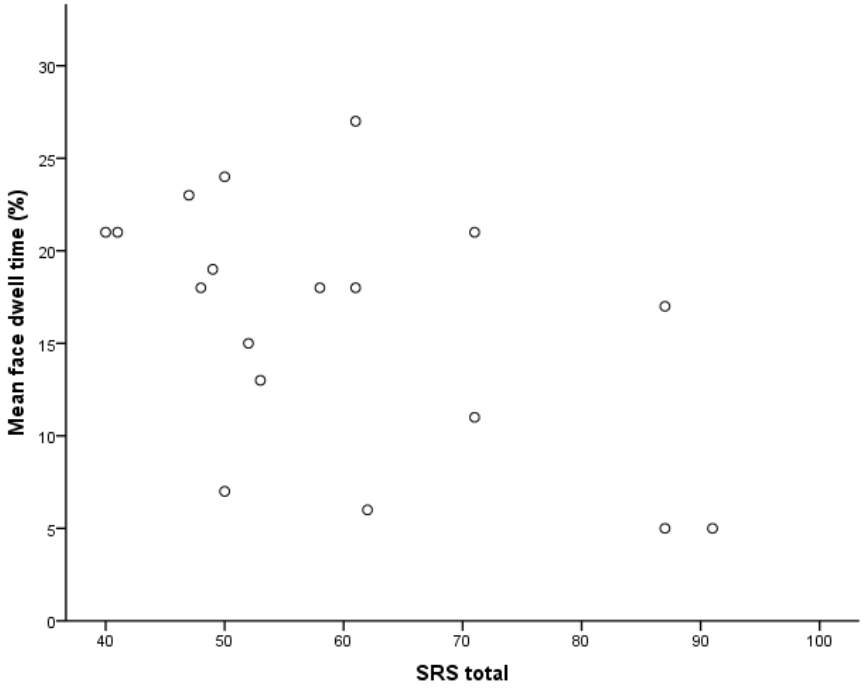




a



b



Author/s:
Lewis, AK; Porter, MA; Williams, TA; Bzishvili, S; North, KN; Payne, JM

Title:
Attention to faces in social context in children with neurofibromatosis type 1

Date:
2019-02-01

Citation:
Lewis, A. K., Porter, M. A., Williams, T. A., Bzishvili, S., North, K. N. & Payne, J. M. (2019). Attention to faces in social context in children with neurofibromatosis type 1. DEVELOPMENTAL MEDICINE AND CHILD NEUROLOGY, 61 (2), pp.174-+. https://doi.org/10.1111/dmcn.13928.