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Atypical Sensory behaviours in children with Tourette's Syndrome and in children with Autism Spectrum Disorders

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Abstract

Certain visual disturbances make it difficult to read text and have been attributed to visual stress, also called "pattern-related visual stress". 12 Children with ASD, 12 children with Tourette's syndrome and without ASD and 12 controls, all matched on age and non verbal ability, participated in an experiment exploring sensory behaviours and visual stress. Reading rate and accuracy were assessed with the Wilkins Rate of Reading test with and without the Intuitive Overlays. Both the children with Tourette's and the children with ASD showed a higher prevalence of atypical sensory behaviours and symptoms of visual stress than the typically developing control children. Six out of twelve children with Tourette's syndrome (50%) read more accurately and over 15% more quickly with a coloured overlay. Four of the 12 children with ASD and none of the control children read over 15% more quickly with an overlay. The findings are discussed in relation to problems in sensory modulation.

Key Words: ASD, Tourette's Syndrome, Sensory Processing, Visual Stress, Cortical Hyperexcitability

What this paper adds:

Tourettes Syndrome and Autism Spectrum Disorders have been shown to overlap clinically and prevalence rates for the comorbidity of Tourette's syndrome and ASD are high. Yet despite the many research papers addressing how children with ASD attend to sensory stimuli, there is a paucity of research in other developmental disabilities. To our knowledge this is the first paper to directly assess sensory experiences both in children with ASD and in children with Tourette Syndrome who do not have ASD.

Many of the perceptual atypicalities reported in ASD such as hyper- sensitivity to lights and colours and experiences of visual distortion are often classified in the literature as sensory abnormalities (Dunn, 1999). However, some of these symptoms are reminiscent of those referred to in other literature as visual stress and treated using coloured filters (Wilkins 1995, 2003). Visual stress here refers to perceptual distortions and discomfort, most notably when reading printed text. If visual stress and sensory difficulties are related problems, then the prevalence of visual stress is likely to be higher in children with sensory disorders. Our findings show the beneficial effect of colour overlays in two different clinical populations associated with sensory atypicalities. Both groups of children showed similar levels of atypical sensory behaviours, and higher levels of sensory avoidance behaviours were correlated with symptoms of visual stress in both groups. It is suggested that colour overlays may be particularly beneficial to children showing a higher prevalence of sensory behaviours.

Introduction

Abnormalities in visual processing are one of the most commonly reported sensory symptoms in children with Autism Spectrum Disorders (Lord, Storoschuck, Rutter& Pickles, 1993; Mottron et al., 2007). Many of these children demonstrate poor eye contact, looking through or beyond objects, extreme aversion to light and/or unusual reaction to visual stimuli (Kanner, 1943; Ritvo& Laxer, 1983; Chawarska, Klin, & Volkmar, 2003). Whilst some of these symptoms can be caused by undiagnosed vision problems such as binocular vision anomalies (eye co-ordination problems) (Wilson, Paterson, & Hutchinson, 2015), there is also evidence to suggest that the visual difficulties are attributable to the processing of sensory information in the environment. The focus of this paper is on visual sensory abnormalities and their potential remediation.

Visual sensory symptoms in ASD are generally characterized by a hypersensitivity to environmental stimulation. For example, individuals with ASD may show an enhanced ability to select and process visual stimuli (Happé & Frith, 2006; Mottron, Dawson, Soulieres, Hubert & Burack, 2006) including searching for objects and distracters and/or noticing subtle changes in the scenery (Simmons et al., 2009). Bright lights may cause visual pain and can be distracting to a child (Kleinhans et al., 2010; Doherty-Sneddon et al., 2002). In contrast, other children with ASD are also reported to be hyposensitive and show a diminished response to visual stimuli (O'Neil & Jones, 1997). They may require additional sensory input to register and to be aware of what other children would usually perceive normally. Common autistic symptoms such as stereotyped behaviour, when a child repeats movements like rocking or waving their hands have been attributed to "underactive" senses (Mottron et al., 2006).

Whilst these perceptual behaviours are commonly referred to in anecdotal and autobiographical accounts from individuals with ASD (Grandin, 1992; Williams, 1999), empirical studies addressing atypical visual behaviours are scarce (O'Neil & Jones,

1997). To date, visual behaviours have mainly been categorised as reactions to sensory events and measured by use of parental questionnaires (Dunn, 1999). Items in these questionnaires focus on visual processing abnormalities commonly reported in ASD including hyper-sensitivity to lights and colours (Myles, Cook, Miller, Rinner & Robbins, 2000; Olney 2000; Attwood, 2006) and experiences of visual distortion, which may, for example, alter the perceived dimensions of rooms (White & White, 1987).

Many of the perceptual atypicalities reported in ASD are reminiscent of those referred to in other literature as visual stress and treated using coloured filters (Wilkins 1995, 2003). Visual stress refers to perceptual distortions and discomfort, most notably when reading printed text. Associated symptoms such as discomfort with bright lights are similar to those classified previously in the ASD literature as sensory atypicalities (Dunn, 1999). This apparent overlap between sensory symptoms and visual stress merits further exploration.

Children experience symptoms of ‘visual stress’ most commonly when reading. The symptoms include discomfort and a variety of perceptual distortions of text (e.g illusory colours, and instability). Placing a coloured overlay on top of a page whilst reading has been shown to be beneficial in reducing these symptoms and increasing reading speed. For example, 5% of 7-8 year old school children show an increase of 25% or more in reading speed when one is used (Wilkins, Lewis, Smith, Rowland, & Tweedie, 2001). The benefits of coloured filters in reducing symptoms have also been found to be greater in neurological disorders in which it is reasonable to hypothesise a cortical hyperexcitability: photosensitive epilepsy (Wilkins et al., 1999), migraine (Harle, Shepherd & Evans, 2006), head injury (Jackowski, Sturr, Taub & Turk, 1996) and stroke (Beasley & Davies, 2013). The cortical hyperexcitability theory proposes that different colours cause a shift in the major locus of activation away from hyperexcitable areas of the visual cortex to areas that are less hyperexcitable (Wilkins, 2003).

In children with ASD it has been shown that when using overlays having a colour chosen individually to increase the “clarity” of text, the increase in reading speed is substantially greater than in controls matched for age and verbal intelligence (Ludlow, Wilkins & Heaton, 2006). Several small-scale studies have suggested that the increase in reading speed is not readily attributable to placebo effects and demand characteristics (Ludlow, Heaton & Wilkins, 2008; Jeanes et al., 1997). In one study involving children with ASD and matched controls, a rate of reading test was carried out in five different conditions: (1) with an overlay chosen for clarity of text, (2) with an overlay of their favourite colour, (3 and 4) with two other coloured overlays from the same set, and (5) without an overlay. Children’s performance on the rate of reading was superior only with the overlay chosen for clarity (Ludlow, Wilkins & Heaton, 2008).

Perceptual improvements have also been found to extend to visual search and matching to sample tasks (Ludlow, Wilkins & Heaton, 2008). Beneficial effects of the overlays in ASD have been also shown on the “reading the eye in mind task” (Baron-Cohen, Joliffe, Mortimore & Robertson, 1997). Children were presented with photographs of the periocular region of various faces and were asked to judge which emotion was being expressed in the eyes. In children with ASD, the perception of the emotion was significantly improved when using a coloured overlay, providing further evidence that perceptual impairment may contribute to wide range of impairments in ASD, such as their social difficulties (Ludlow, Taylor-Whiffen & Wilkins, 2012). These results were given further support by a recent study by Whitaker, Jones, Wilkins, & Roberson, (2015), who showed that the judgement of emotional intensity in faces by individuals with ASD improved significantly with the addition of a colored tint. Evidently there may be potential benefits of colour filters in alleviating a range of perceptual distortions in ASD.

If visual stress and sensory difficulties are related problems, then the prevalence of visual stress is likely to be higher in children with sensory disorders. For example, sensory behaviours can also occur in disorders other than ASD, such as Tourette’s syndrome. Tourette’s syndrome (TS) is a condition characterized by the occurrence of chronic multiple motor and vocal tics, with onset usually around school age (4–6 years) and with a fluctuating course (APA, 2013). It is a complex disorder with an unknown aetiology, which has shown a strong genetic component, yet to be defined. Importantly, ASD and Tourette’s syndrome have been consistently shown to overlap clinically, and prevalence rates for the comorbidity of Tourette’s syndrome in ASD range between 6.5 and 22 percent (Baron-Cohen, Scahill, Izaguirre, Hornsey, Robertson, 1999; Canitano & Vivanti, 2007).

Sensory experiences of children with Tourette’s syndrome are often noted as occurring alongside repetitive behaviours in the form of tics (Kwak, Dat Vuong & Hewitt, 2003). For example, in children with Tourette’s syndrome it has been reported that tics are preceded by aversive sensations, which develop through a process in which sensory events, such as tactile perceptions, take on a special significance. A large range of auditory or visual cues can also prompt tics, but the nature of these cues is usually highly selective for individual patients, such as a cough, a particular word, or an alignment of angles of specific shapes (Leckman, 2002). Tics are thus produced to relieve these sensations, although they allow only a fleeting reprieve. Eighty per cent of individuals with Tourette’s syndrome reported that tics were produced as intentional responses to aversive sensory phenomena (Cohen & Leckman, 1992). Some researchers have even speculated that Tourette’s syndrome is characterised by a hypersensitivity to somatosensory stimulation (Orth, Amann, Robertson & Rothwell, 2005).

It is not clear whether visual stress occurs in syndromes such as Tourette’s, but given the overlap between sensory behaviours and visual stress symptoms this seems likely. In addition there has also been an abundance of research suggesting cortical hyperexcitability in Tourette’s syndrome. This has included the demonstration that clinical measurements of tic severity in Tourette’s are associated with a reduction in intra-cortical inhibition and a hyper-excitability in the motor cortex (Jackman, Mueller, Hambleton & Hollis, 2007; Mueller, Jackson, Dhalla, Datsopoulos, & Hollis, 2006; Leckman, Bloch, Scahill & King, 2006); and the findings that people with Tourette’s syndrome appear to have an increased risk of epilepsy, particularly those children with additional co-morbid conditions such as an ASD (Williams, Stern, Grabecki, Simmons & Robertson, 2013).

There were two key aims of the current study. First, the study was designed to compare sensory behaviours in two separate and different clinical populations: children with ASD and Tourette’s syndrome without ASD. It was predicted that there would be a similar occurrence of sensory behaviours in both groups of children, although they may differ in regard to types of sensory problems experienced. Second, the study was designed to assess whether children with Tourette’s syndrome would show visual

stress, and/or benefit from the use of a coloured filter. Both the children with ASD and children with Tourette's were expected to show higher levels of visual stress and consequently show more benefits from the use of colour when reading.

Method

Participants

Twelve children with a formal diagnosis of Tourette's syndrome (aged 6 years 5 months - 12 years 2 months) were recruited via the charity Tourette's Action. The age range was typical and encompassed the time period when the severity of tics is thought to be maximal (Jackson et al., 2012). Of the twelve children, 9 were boys and 3 were girls. These children were selectively recruited on the basis they exhibited physical tics. Physical tics were chosen as a criterion because these behaviours often mirror the visual exploratory behaviours exhibited in children with ASD, including biting, gestures with hands, finger movements and touching objects (Hollander et al., 2005). In order to interpret the results in the context of a diagnosis of Tourette's syndrome and to rule out any additional comorbid diagnosis of ASD, *no child taking part had a formal diagnosis of ASD or ADHD*, although two displayed autistic traits, and one, ADHD (undiagnosed). The selection criteria meant that the groups were small. All children's diagnoses conformed to the DSM-5: (1) onset before the age of 18, (2) multiple motor tics, (3) one of more vocal tics, (4) waxing and waning of symptoms, (5) presence of symptoms for more than 1 year. Children's mean onset of the disorder was 7 years 1 month of age, which agrees with previous studies (Comings & Comings 1985).

The children with Tourette's syndrome were compared to 12 typically developing control children (9 boys, 3 girls) with no clinical diagnosis and 12 boys with a diagnosis of an Autism Spectrum Disorder. The Autism Diagnostic Observation Schedule Generic (ADOS-G; Lord et al., 2000) was carried out on the ASD group to confirm their diagnosis of autism and to gain additional information about their social and language patterns of behaviour. The ADOS-G was carried out by researchers trained to use it for research purposes. The protocol consists of a series of structured and semi-structured tasks that involve social interaction between the examiner and the participant. All ASD participants had an unambiguous clinical diagnosis of autistic disorder or Asperger's syndrome according to DSM-IV criteria, and scored above threshold for ASD on the ADOS-G diagnostic algorithm. None had identifiable medical conditions underlying their ASD. In order to control for co-occurring reading disorders, no child included in the study had a diagnosis of dyslexia.

Across groups, all children were matched on age and non-verbal IQ (see Table 1). Non-verbal IQ scores were obtained using Raven's Standard Progressive Matrices test (Raven, Court & Raven, 1990). It is a test commonly used in clinical neuropsychology to assess general intellectual abilities, and is readily used to match clinical samples with a non-clinical sample of children (e.g. Swettenham, Remington, Murphy, Feuerstein, Grim, & Lavie, 2014). There were no differences between groups in age, $F(2,35) = .88, p = .43$ or non-verbal IQ, $F(2,35) = 1.72, p = .19$. The children were recruited from various schools and social groups in the East of England, London and the Midlands. Full Ethical approval was obtained from the university. All children who took part in the study gave verbal consent to take part in the study, and written consent was also obtained from all children's parents/guardians.

Table 1. Mean Age (years and months) and Non-verbal IQ of each group of participants

	Age range		Age		Non-Verbal IQ*	
	Min - Max	Mean	SD	Mean	SD	
Tourette's Syndrome (N=12)	6.5-12.2	9.8	1.8	102.8	12.5	
ASD (N=12)	6.9-13.1	9.8	2.3	105.8	16.2	
Controls (N=12)	6.5-13.5	10.7	2.2	93.8	19.6	

Note: * = Standard score (M=100; SD=15)

Materials and Procedure

All children included in the study were tested on the following tasks at a room in the university and the children completed the tests in the following order. A standardised procedure was used for the selection of an overlay and for the administration of the rate of reading test (Wilkins, 2003). All of the testing was carried out by a developmental psychologist trained to administer both overlay selection and rate of reading.

2.2.1. *City colour test* (Third edition, Fletcher, 1998) was first used to screen for any colour anomaly and none was found.

2.2.2. *The Intuitive Overlays* (Wilkins, 1994) are supplied by iOO Sales Ltd. The pack contains 2 A5 size overlays of each of the following colours; Rose, Pink, Purple, Orange, Yellow, Lime Green, Mint Green, Blue and Aqua. The overlays can be combined in pairs of neighbouring chromaticity so as to provide 30 shades of colour, sampling chromaticity systematically (Wilkins, 1994).

2.2.3. *Overlay selection*: The selection of overlays followed the procedure recommended in the instructions (iOO sales, London UK). Two identical passages of text were printed side by side, and overlays were placed over one passage. Any overlay that made the text clearer and more comfortable to read was left in place and the next overlay placed beside it for comparison, until the best had been chosen by a process of elimination. The final choice of overlay was compared with the uncovered page. Stronger colours formed by placing one overlay on top of another of the same or neighbouring chromaticity were then compared with the final choice of single overlay.

2.2.4. The '*Rate of Reading Test*' (Wilkins et al., 1996) was administered according to the published instructions (iOO sales, London, UK). This test looks like a passage of prose, but consists of randomly ordered common words. Therefore reading is independent of syntactic and semantic constraints but requires some of the usual visual and visuo-perceptual processing. At the outset all children were required to read the list of the words included in the rate of reading test to ensure familiarity with the

vocabulary. All children were then asked to read the text out loud for one minute with and without an overlay and the order of these conditions was randomised. The total number of words accurately read in the correct order in one minute was calculated.

2.2.5. *Visual Stress*: The children were next asked about symptoms of ‘visual stress’ related to reading: “Do the letters stay still or *do they move?*”; “Are they clear or *are they blurred (fuzzy, difficult to see)?*”; “Are the words *too close together* or far enough apart?”; “Is the *page too bright*, not bright enough, or just about right?”; “Does the *page hurt your eyes to look at* or is it ok?”. All of the participants in the study were verbal, and gave ready responses to the questions asked. No child asked for clarification. A score of 1 was given for each visual stress symptom expressed (these are italicised above), others responses scored 0.

2.2.6. ‘*The Sensory Profile*’ (Dunn, 1999) is a questionnaire completed by parents. It contains 125 items across 14 subsections. Sensory behaviours are grouped into three main types 1) sensory processing, 2) modulation, and 3) behavioural outcomes of sensory processing. Items on the questionnaire form nine meaningful factors: Sensory Seeking, Emotionally Reactive, Low Endurance/Tone, Oral Sensory Sensitivity, Inattention/Distractibility, Poor Registration, Sensory Sensitivity, Sedentary, and Fine Motor/Perceptual. Score across the 125 items can also be collapsed into four sensory quadrants, Low Registration, Sensory Seeking, Sensory Sensitivity and Sensory Avoidance. Parents indicated the frequency with which their child engaged in the behaviour as described in each item on a five-point Likert scale (i.e., 1 = always, 100% of the time; 2 = frequently, 75% of the time; 3 = occasionally, 50% of the time; 4 = seldom, 25% of the time; and 5 = never, 0% of the time).

Results

Sensory Behaviours

Children with Tourette’s syndrome showed levels of sensory abnormalities similar to those of the ASD group. On over 85% of the measures, the children from both clinical groups showed *definite* (2SD below the mean) to *probable* differences (1SD below the mean) according to the population norms in the Sensory Profile (Dunn, 1999). As expected, the typically developing controls showed normal performances across the majority of the measures (Table 2).

Table 2. Mean Sensory Behaviour Scores: Sensory Processing, Modulation, Behavioural Outcomes.

	Group					
	Tourettes		ASD		Controls	
	<i>Mean</i>	<i>SD</i>	<i>Mean</i>	<i>SD</i>	<i>Mean</i>	<i>SD</i>
<i>Sensory Processing</i>						
Auditory	23.0**	6.5 ^a	24.9**	6.5 ^{ab}	31.8	3.8 ^b
Visual	31.3*	7.6 ^a	31.5*	6.2 ^{ab}	36.7	6.1 ^a
Vestibular	42.3**	4.6 ^a	43.9**	5.8 ^{ab}	47.7*	4.8 ^a
Touch	62.2**	13.4 ^a	61.2**	13.7 ^{ab}	80.9	7.7 ^b
Multisensory	25.9*	4.2 ^a	26.8*	5.1 ^{ab}	29.5	6.0 ^a
Oral	44.9*	11.2 ^a	44.7*	11.6 ^{ab}	50.1	12.8 ^a
<i>Modulation</i>						
Sensory processing related to endurance/tone	40.2	8.1 ^a	40.7	6.5 ^a	41.5	7.3 ^a
Modulation to body position and movement	33.8**	6.4 ^a	33.8**	6.8 ^a	43.0	4.7 ^b
Modulation of movement affecting activity	20.7*	6.1 ^a	22.5*	6.7 ^a	24.2	5.1 ^a
Modulation of sensory input affecting emotion	12.8**	4.6 ^a	13.9**	4.7 ^a	17.2	3.4 ^a
Modulation of visual input affecting emotion	11.9**	3.1 ^a	13.8*	3.6 ^a	16.2	4.6 ^a
<i>Behavioural Outcomes</i>						
Emotional Social Responses	49.9**	9.9 ^a	54.0**	14.4 ^a	68.8	8.2 ^b
Behavioural outcomes of sensory processing	16.7**	4.2 ^a	15.5**	2.7 ^a	24.5	7.1 ^b
Items indicating thresholds for response	11.0*	2.1 ^a	10.4*	6.3 ^a	13.5	1.2 ^a

Note: * = Probable Difference, ** = Definite Difference. Standard deviations are in parenthesis. For each sensory factor, means in the same row with different subscripts are significantly different with Bonferroni adjusted significance level $p < .008$ Sensory processing, $p < .01$ Modulation, $p < .02$ Behavioural Outcomes.

The investigation sought to determine whether the three main groups of sensory behaviours (sensory processing, modulation and behavioural and emotional responses) as identified by the Sensory Profile (Dunn, 1999) differed among the three groups. Section scores of the three groups across these three groups of sensory behaviours using a multivariate analysis of variance (Wilks's Lambda), were statistically significant, $F(6, 62) = 4.03, p < .01, \eta_p^2 = .28$. Univariate tests showed that there were significant differences across the groups in sensory processing, $F(2, 33) = 4.97, p < .05, \eta_p^2 = .23$, modulation, $F(2, 33) = 3.42, p < .05, \eta_p^2 = .17$ and behaviour and emotional responses $F(2, 33) = 11.94, p < .001, \eta_p^2 = .42$. Follow-up comparisons (Bonferroni-corrected, $p = .016$), found that the children with Tourette’s syndrome displayed significantly more sensory behaviours than controls as regards overall sensory processing, $t(22) = 3.34, p < .01$; modulation, $t(22) = 2.87, p < .01$; and behavioural and emotional responses $t(22) = 4.61, p < .001$. The ASD group showed significantly more behavioural and emotional responses than controls $t(22) = 6.27, p < .001$. There were no significant differences between the children with ASD and Tourette’s syndrome.

A multivariate analysis of variance (MANOVA) was then used to determine differences between the three groups of children, Tourette's syndrome, ASD and the control group with respect to the nine sensory factors identified by the Sensory Profile. With group serving as the independent variable, the dependent variables were the nine sensory factors including sensory seeking, Inattention/Distractibility and Sensory Sensitivity. Section scores of the three groups using a MANOVA (Wilks's Lambda), were statistically significant, $F(18,50) = 4.39, p < .001, \eta_p^2 = .61$. Means and standard deviations, and ranges for the sensory variables are shown in Table 3.

Table 3. Mean Sensory Behaviour Scores: Sensory Processing, Modulation, Behavioural Outcomes.

	Group					
	Tourettes		ASD		Controls	
	Mean	SD	Mean	SD	Mean	SD
Sensory Seeking	43.3**	10.6 ^a	50.1**	10.1 ^{ab}	59.1*	10.3 ^b
Emotionally Reactive	44.6**	15.3 ^a	50.6*	11.3 ^{ab}	60.4	9.1 ^b
Low Endurance/Tone	35.1**	10.3 ^a	40.0	6.2 ^a	42.3	7.1 ^a
Oral sensory sensitivity	32.2*	9.7 ^a	34.9	11.5 ^a	37.5	6.0 ^a
Inattention/Distractibility	21.1**	7.9 ^a	21.6**	4.3 ^a	30.5	4.2 ^b
Poor registration	31.5*	6.7 ^{ab}	27.0**	6.3 ^a	37.3	2.1 ^b
Sensory Sensitivity	19.3	4.4 ^{ab}	14.2*	4.1 ^a	19.0	1.4 ^b
Sedentary	14.8	4.5 ^a	15.4	4.4 ^a	15.5	2.8 ^a
Fine Motor/Perceptual	10.3	2.2 ^a	12.1	2.5 ^a	13.5	1.7 ^a

Note: * = Probable Difference, ** = Definite Difference. Standard deviations are in parenthesis. For each sensory factor, means in the same row with different subscripts are significantly different with Bonferroni adjusted significance level $p < .006$

Subsequent univariate tests of each variable revealed that there were group differences on six of these measures: Sensory seeking, $F(2, 33) = 7.04, p < .005, \eta_p^2 = .29$; Emotionally reactive $F(2, 33) = 5.17, p < .05, \eta_p^2 = .24$; Inattention and distractibility, $F(2, 33) = 7.29, p < .01, \eta_p^2 = .31$; Poor emotion registration, $F(2, 33) = 10.85, p < .001, \eta_p^2 = .39$; Sensory sensitivity, $F(2, 33) = 13, p < .01, \eta_p^2 = .31$ and finally Fine motor control, $F(2, 33) = 3.78, p < .05, \eta_p^2 = .18$. The groups were not significantly different across the three factors of Low endurance, Oral sensitivity and Sedentary. Follow up comparisons (Bonferroni-corrected, $p = .006$) found that the children with Tourette's syndrome displayed significantly more sensory behaviours than controls, as regards Sensory seeking, $t(22) = 3.71, p < .01$ Emotional reactivity, $t(22) = 3.08, p < .001$ and Inattention and distractibility $t(22) = 3.07, p < .006$. The children with ASD showed more sensory behaviours compared to controls as regards Inattention and Distractibility; $t(22) = 4.06, p < .001$, Poor registration, $t(22) = 5.38, p < .001$ and Sensory sensitivity $t(22) = 3.81, p < .001$. There were no significant differences across any of the measures between the children with ASD and the children with Tourette's syndrome.

Choice of Colour Overlays

All children selected a colour overlay as improving the clarity of text. The children selected a wide range of colours and no single colour was chosen by the majority. Similar to previous studies, approximately half of each group chose a single overlay (6 children with Tourette's syndrome, 5 children with ASD and 6 control children) and the remaining half chose a double overlay (Ludlow, Wilkins & Heaton, 2006). Four out of the 12 children with Tourettes opted for a pink overlay, making this the most popular selection.

Rate of Reading

The 95% confidence interval for the number of words read per minute with an overlay was 88.2 words to 107.9 words, ($M = 98.1, sd = 28.4$) and without an overlay was 81.9 words to 101.7 words ($M = 91.8, sd = 29.3$). In order to examine the difference in rate of reading (number of words accurately read per minute), a 3*2 mixed analysis of variance was carried out adding a within participants factor (Condition: with vs. without overlays) to the analysis. This revealed a significant effect of Condition, $F(1, 33) = 13.4, p < .001, \eta_p^2 = .29$, more words being read with an overlay ($M = 98.1; sd = 28.4$) than without ($M = 91.7; sd = 29.3$). There was no significant effect of group, $F(2, 33) = .39, n.s., \eta_p^2 = .02$. However, there was a significant group x condition interaction, $F(2, 33) = 8.3, p < .001, \eta_p^2 = .35$. Further analysis revealed the children with Tourette's syndrome to show significantly better performance with an overlay ($M = 96.5, sd = 21.4$) than without ($M = 83.6, sd = 21.1$), $t(11) = 5.2, p < .001$. Children with ASD also showed significantly better performance with an overlay ($M = 99.7, sd = 38.8$), than without ($M = 90.3, sd = 35.5$), $t(11) = 3.3, p < .05$. In contrast, the typically developing controls showed no significant difference in performance with an overlay ($M = 98.0, sd = 24.5$) and without ($M = 101.4, sd = 29.3$), $t(11) = .9, ns$.

In previous studies, the criterion for clinically significant improvements in reading speed when using overlays has been set at 5%. Using the 5% improvement threshold, it was found that a high proportion of children with Tourette's syndrome 11/12 (92%) and children with ASD 8/12 (67%) read more than 5% faster with an overlay, compared to 3/12 (25%) of the typically developing children.

It has recently been suggested that an increase of 15% exceeds that expected from chance variation and offers a suitable clinical threshold (Wilkins, Allen, Monger, & Evans (in press). When applying the same threshold to the current sample, 6/12 (50%) children with Tourette's syndrome read >15% faster with an overlay, with levels of improvement of up to 54%. Four out of the 12 children with ASD (32%) read > 15% faster with an overlay, with levels of improvement of up to 39%. None of the typically developing children read more than 15% faster with an overlay, with levels of improvement less than 13%.

Visual Stress symptoms when reading

The number of visual stress symptoms with and without an overlay were calculated across each group (maximum score 5). Number of visual stress symptoms reported by each group of children were as follows: Children with Tourette's syndrome without an overlay ($M=1.2$, $sd=1.8$) with an overlay ($M=.6$, $sd=.9$); Children with ASD without an overlay ($M=.5$, $sd=.7$) with an overlay ($M=.08$, $sd=.3$); The typically developing controls without an overlay ($M=.5$, $sd=1.2$) and with ($M=.6$, $sd=.7$). A 3*2 mixed analysis of variance was carried out between groups and visual stress symptoms with and without an overlay. This revealed a significant effect of condition; $F(1, 33) = 23.4$, $p < .001$, $\eta_p^2 = .41$. Children reported significantly more symptoms of visual stress without an overlay than with one, $t(35) = 4.3$, $p < .001$. There was no significant effect of group, $F(2, 33) = 1.9$, $p = .16$, $\eta_p^2 = .11$. However, there was a significant group x condition interaction, $F(2, 33) = 5.5$, $p < .01$, $\eta_p^2 = .25$. Children from both clinical groups reported significantly more symptoms of visual stress without an overlay: Tourettes, $t(11) = 3.7$, $p < .05$; ASD, $t(11) = 2.6$, $p < .05$, however there was no significant difference in symptoms reported with and without overlays for the Controls, $t(11) = 2.2$, $p = .07$.

Visual Stress and Sensory Behaviours

Multiple regressions were used in order to explore the relationship between the scores across the sensory behaviours and the symptoms of visual stress without an overlay across all three groups. The results of the regression indicated the entire 14 sections of the sensory profile when used together as predictors explained 78% of the variance in visual stress symptoms when reading ($R^2 = .78$, $F(14, 35) = 5.13$, $p < .001$).

A multiple linear regression analysis using the four sensory quadrants (low avoidance, sensory seeking, sensory sensitivity, sensory avoidance, see Table 4) as predictors of symptoms of visual stress without an overlay revealed a significant overall model only for the children with Tourettes, $R^2 = .74$, $F(4, 11) = 5.1$, $MSE = 5.62$, $p < 0.05$. This also revealed that children with Tourettes who reported more symptoms of visual stress showed significantly lower levels of sensory seeking behaviours $\beta = -.89$, $t = -3.14$, $p < 0.05$, and to display higher levels of sensory avoiding behaviours, $\beta = 1.45$, $t = 3.42$, $p < 0.05$. A significant relationship was also found between the children with ASD reporting higher levels of visual stress and displaying higher levels of sensory avoiding behaviours, $\beta = 1.45$, $t = 3.42$, $p < .05$.

Table 4. Means Scores across the Four Sensory Quadrant

	Group					
	Tourettes		ASD		Controls	
	Mean	SD	Mean	SD	Mean	SD
Low Registration	58.7**	11.3	62.8*	6.1	69.8	10.3
Sensory Seeking	83.1**	19.3	81.3**	16.3	103.1	23.1
Sensory Sensitivity	74.2*	10.1	75.0*	8.6	87.7	17.9
Sensory Avoiding	108.0*	16.9	103.3*	13.8	122.5	13.8

Note: * = Probable Difference, ** = Definite Difference. Standard deviations are in parenthesis. For each of the sensory quadrants, no significant group differences occurred following Bonferroni adjusted significance level $p < .01$.

Discussion

This is the first study to show the beneficial effect of colour overlays in two different clinical populations associated with atypical sensory processing. A higher than normal proportion of the children with ASD and children with Tourette's syndrome were found to read more quickly with the use of a coloured overlay, with levels of improvement reaching 54%. Importantly, both groups of children showed similar levels of atypical sensory behaviors, and those who presented more sensory avoidance behaviors displayed higher levels of visual stress. It is suggested that colour overlays may be particularly beneficial to children showing a higher prevalence of sensory symptoms.

At the outset, one of the first aims of the study was to assess sensory behaviors for the three groups using the sensory profile. Children with ASD and Tourette's syndrome showed atypical responses across the three sections of the sensory profile (sensory processing, modulation and behavioural and emotional responses). Importantly, both groups of children were also identifiable by their atypical responses across several of these factors compared to controls: Sensory Seeking, Emotional reactivity, Inattention/Distractibility, Poor registration and Sensory sensitivity. The findings from the study clearly show that both the children with ASD and those with Tourette's syndrome exhibit problems in Sensory modulation, the ability to facilitate or inhibit responses to sensory stimuli. ASD and Tourette's syndrome have been consistently shown to overlap clinically (Baron-Cohen et al., 1999; Canitano & Vivanti, 2007) and this study is important in highlighting that sensory problems may underlie some of their shared characteristics.

The second aim of the present study was to determine whether children with Tourette's syndrome would show a higher level of symptoms of visual stress when reading, as alleviated with the use of a coloured overlay (Wilkins, 2003). Whilst the children with Tourette syndrome and ASD reported significantly fewer visual stress symptoms with an overlay than without, there was no significant difference between the groups in the number symptoms of visual stress reported. Symptoms of visual stress have been found to be a measure with only modest reliability in predicting reading speed increment with a colour overlay among children in mainstream education (Wilkins et al., 1994).

Finally, the study was able to establish a relationship between atypical sensory responses in the children with ASD and Tourette's syndrome and their levels of visual stress. Children with ASD have been shown to display both hypo- and hyper- sensory behaviours (Rogers & Ozonoff, 2005), and therefore it is important for research to consider visual stress in samples displaying both types of behaviour. The current findings suggest that it is children who are hypersensitive to sensory stimuli that may show higher levels of visual stress, as demonstrated by the relationship found between reported symptoms of visual stress and high levels of

sensory avoidance behaviours such as ‘avoid bright lights because they are painful’ (Kleinhans et al., 2010; Doherty-Sneddon et al., 2002).

Given that coloured filters have been found to benefit performance on tasks other than reading; including matching to sample, visual search and recognition of emotion in faces (Ludlow et al, 2008; 2012), the overlays might offer an important tool to children with sensory disorders in educational settings.

Limitations

This is a small-scale study. However, the modest sample sizes reflect our desire to select children with only one diagnosis, although these diagnoses often co-occur. Another possible concern is that the children who generally benefit most from overlays are usually those with lower levels of reading ability, who therefore had more of a margin to show improvements (Christ, Coolong-Chaffin, 2007). Whilst this is unlikely to be case in the current study, as no significant differences across groups were found in the number of words read without an overlay, reading ability needs to be controlled for in future studies.

It may appear that an uncharacteristically large number of clinical conditions have now been reported in the literature as showing a reduction in adverse symptoms using coloured overlays currently include ASD, Tourettes, migraine (Harle, Shepherd & Evans, 2006), head injury (Jackowski, Sturr, Taub & Turk, 1996) and stroke (Beasley & Davies, 2013). It is important to note that these are all neurological disorders in which it is reasonable to postulate a hyperexcitability of the visual cortex (Wilkins, 2003).

Conclusion

The results provide further evidence consistent with an association between sensory difficulties and cortical hyperexcitability that merits further exploration. It is suggested that children who are hypersensitive to sensory stimuli may experience the greatest benefits from the use of a colour filter.

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Declaration of interest

‘The Intuitive Overlays were designed by AJW when he was employed by the British Medical Research Council. The MRC own the rights. AJW formerly received an “Award to Inventors” from the MRC, based on royalties on sales of Intuitive Overlays. This award has now lapsed.

References

- American Psychiatric Association (2013). *Diagnostic and statistical manual of mental disorders* (5th edition). Arlington, VA: American Psychiatric Publishing.
- Attwood, T. (2006). *The complete guide to Asperger's syndrome*. Jessica Kingsley Publishers.
- Baron-Cohen, S., Jolliffe, T., Mortimore, C., & Robertson, M. (1997). Another advanced test of theory of mind: Evidence from very high functioning adults with autism or Asperger syndrome. *Journal of Child Psychology and Psychiatry*, 38(7), 813-822. doi:10.1111/j.1469-7610.1997.tb01599.x
- Baron-Cohen, S., Scahill, V. L., Izaguirre, J., Hornsey, H., & Robertson, M. M. (1999). The prevalence of Gilles de la Tourette syndrome in children and adolescents with autism: a large scale study. *Psychological Medicine*, 29(5), 1151-1159.
- Beasley, I. G., & Davies, L. N. (2013). The effect of spectral filters on reading speed and accuracy following stroke. *Journal of Optometry*, 6(3), 134-40. doi: 10.1016/j.optom.2013.03.002
- Canitano, R., & Vivanti, G. (2007). Tics and Tourette syndrome in autism spectrum disorders. *Autism*, 11(1), 19-28. doi: 10.1177/1362361307070992
- Chawarska, K., Klin, A., & Volkmar, F. (2003). Automatic Attention Cueing Through Eye Movement in 2-Year-Old Children With Autism. *Child development*, 74(4), 1108-1122. doi:10.1111/j.1469-7610.2006.01685.x
- Christ, T. J., & Coolong-Chaffin, M. (2007). Interpretation of curriculum-based measurement outcomes: standard error and confidence intervals. *School of Psychology Forum: Research in Practice*, 1, 75-86.
- Cohen, A.J., & Leckman, J.F. (1992). Sensory phenomena associated with Gilles de la Tourette's syndrome. *Journal of Clinical Psychiatry*, 53, 319-323.
- Comings, D. E., & Comings, B. G. (1985). Tourette syndrome: clinical and psychological aspects of 250 cases. *American Journal of Human Genetics*, 37(3), 435.
- Doherty-Sneddon, G., Bruce, V., Bonner, L., Longbotham, S., & Doyle, C. (2002). Development of gaze aversion as disengagement from visual information. *Developmental psychology*, 38(3), 438-445. doi: 10.1037/0012-1649.38.3.438
- Dunn, W. (1999). *Sensory profile: User's manual*. San Antonio, TX: Psychological Corporation.
- Fletcher, R. (1998). *City colour vision test. Third Edition*. Windsor: Keeler Ltd.
- Grandin, T. (1992). An inside view of autism. High-functioning individuals with autism, 105-126.

- Happé, F., & Frith, U. (2006). The weak coherence account: Detail-focused cognitive style in autism spectrum disorders. *Journal of autism and developmental disorders*, 36(1), 5-25. doi:10.1007/s10803-005-0039-0
- Harle, D. E., Shepherd, A. J., & Evans, B. J. (2006). Visual stimuli are common triggers of migraine and are associated with pattern glare. *Headache: The Journal of Head and Face Pain*, 46(9), 1431-1440. doi:10.1111/j.1526-4610.2006.00585.x
- Hollander, E., Anagnostou, E., Chaplin, W., Esposito, K., Haznedar, M. M., Licalzi, E., Wasserman, S., Soorya, L., & Buchsbaum, M. (2005). Striatal volume on magnetic resonance imaging and repetitive behaviors in autism. *Biological psychiatry*, 58(3), 226-232.
- Jackowski, M. M., Sturr, J. F., Taub, H. A., & Turk, M. A. (1996). Photophobia in patients with traumatic brain injury: uses of light-filtering lenses to enhance contrast sensitivity and reading rate. *Neurorehabilitation*, 6, 193-201.
- Jackson, G. M., Mueller, S. C., Hambleton, K., & Hollis, C. P. (2007). Enhanced cognitive control in Tourette Syndrome during task uncertainty. *Experimental Brain Research*, 182(3), 357-364. doi.org/10.1007/s00221-007-0999-8
- Jeanes, R., Busby, A., Martin, J., Lewis, E., Stevenson, N., Pointon, D., & Wilkins, A. (1997). Prolonged use of coloured overlays for classroom reading. *British Journal of Psychology*, 88(4), 541-548
- Kanner, L. (1943). Autistic disturbance of affective contact. *Nervous Child*, 2, 217-250. Reprinted in L. Kanner. (1973). *Childhood Psychosis: Initial Studies and New Insights*. New York: John Wiley and Sons.
- Kleinmans, N. M., Richards, T., Weaver, K., Johnson, L. C., Greenson, J., Dawson, G., & Aylward, E. (2010). Association between amygdala response to emotional faces and social anxiety in autism spectrum disorders. *Neuropsychologia*, 48(12), 3665-3670. doi.org/10.1016/j.neuropsychologia.2010.07.022
- Kwak, C., Dat Vuong, & Jankovic, J. (2003). Premonitory sensory phenomenon in Tourette's syndrome. *Movement Disorders*, 18(2), 1530-1533. Leckman, J. F. (2002). Tourette's syndrome. *The Lancet*, 360(9345), 1577-1586. doi.org/10.1016/S0140-6736(02)11526-1
- Leckman, J. F. (2002). Tourette's syndrome. *The Lancet*, 360(9345), 1577-1586. doi:10.1016/S0140-6736(02)11526-1
- Leckman, J. F., Bloch, M. H., Scahill, L., & King, R. A. (2006). Tourette syndrome: the self under siege. *Journal of Child Neurology*, 21(8), 642-649. doi: 10.1177/08830738060210081001
- Lord, C., Storoschuk, S., Rutter, M., & Pickles, A. (1993). Using the ADI-R to diagnose autism in preschool children. *Infant Mental Health Journal*, 14(3), 234-252. doi:10.1002/1097-0355
- Lord, C., Risi, S., Lambrecht, L., Cook Jr, E. H., Leventhal, B. L., DiLavore, P. C., ... & Rutter, M. (2000). The Autism Diagnostic Observation Schedule—Generic: A standard measure of social and communication deficits associated with the spectrum of autism. *Journal of autism and developmental disorders*, 30(3), 205-223.
- Ludlow, A. K., & Wilkins, A. J. (2009). Case report: color as a therapeutic intervention. *Journal of autism and developmental disorders*, 39(5), 815-818. doi:10.1007/s10803-008-0672-5
- Ludlow, A. K., Wilkins, A. J., & Heaton, P. (2006). The effect of coloured overlays on reading ability in children with autism. *Journal of Autism and Developmental Disorders*, 36(4), 507-516. Doi. 10.1007/s10803-006-0090-5
- Ludlow, A. K., Wilkins, A. J., & Heaton, P. (2008). Colored overlays enhance visual perceptual performance in children with autism spectrum disorders. *Research in Autism Spectrum Disorders*, 2(3), 498-515. doi.org/10.1016/j.rasd.2007.10.001
- Ludlow, A. K., Taylor-Whiffen, E., & Wilkins, A. J. (2012). Coloured Filters Enhance the Visual Perception of Social Cues in Children with Autism Spectrum Disorders. *ISRN neurology*. doi:10.5402/2012/298098
- Mottron, L., Dawson, M., Soulières, I., Hubert, B., & Burack, J. (2006). Enhanced perceptual functioning in autism: An update, and eight principles of autistic perception. *Journal of autism and developmental disorders*, 36(1), 27-43. doi:10.1007/s10803-005-0040-7
- Mottron, L., Mineau, S., Martel, G., Bernier, C. S., Berthiaume, C., Dawson, M., Lemay, M., Palardy, S., Charman, T., & Faubert, J. (2007). Lateral glances toward moving stimuli among young children with autism: Early regulation of locally oriented perception?. *Development and psychopathology*, 24(5), 555-574. doi.org/10.1017/S0954579407070022
- Mueller, S. C., Jackson, G. M., Dhalla, R., Datsopoulos, S., & Hollis, C. P. (2006). Enhanced cognitive control in young people with Tourette's syndrome. *Current Biology*, 16(6), 570-573. doi.org/10.1016/j.cub.2006.01.064
- Myles, B. S., Cook, K. T., Miller, N. E., Rinner, L., & Robbins, L. (2000). *Asperger Syndrome and sensory issues: Practical solutions for making sense of the world*. Shawnee Mission, KS: Autism Asperger Publishing Company
- Neil, O. M., & Jones, RSP (1997). Sensory-perceptual abnormalities in autism: A case for more research. *Journal of Autism and Developmental Disorders*, 27(3), 283-293. doi. 10.1023/A:1025850431170
- Olney, M. F. (2000). Working with autism and other social-communication disorders. *Journal of Rehabilitation*, 66(4), 51-56.
- Orth, M., Amann, B., Robertson, M. M., & Rothwell, J. C. (2005). Excitability of motor cortex inhibitory circuits in Tourette syndrome before and after single dose nicotine. *Brain*, 128(6), 1292-1300. doi: 10.1093/brain/awh473
- Raven, J. C., Raven, J., & Court, J. H. (1990). *Standard progressive matrices*. Oxford, United Kingdom: Oxford Psychologists Press.

- Ritvo, L., & Laxer, G. (1983). *Autisme. La verite' refusee*. SIMEP, Villeurbanne-Paris.
- Rogers, S. J., & Ozonoff, S. (2005). Annotation: What do we know about sensory dysfunction in autism? A critical review of the empirical evidence. *Journal of Child Psychology and Psychiatry*, *46*(12), 1255-1268. doi:10.1111/j.1469-7610.2005.01431.x
- Simmons, D. R., Robertson, A. E., McKay, L. S., Toal, E., McAleer, P., & Pollick, F. E. (2009). Vision in autism spectrum disorders. *Vision Research*, *49*(22), 2705-2739. doi.org/10.1016/j.visres.2009.08.005
- Swettenham, J., Remington, A., Murphy, P., Feuerstein, M., Grim, K., Lavie, N. (2014). Seeing the unseen: Autism involves reduced susceptibility to inattention blindness. *Neuropsychology*, *28*(4), 563-570. doi: 10.1037/neu0000042
- Wilkins, A. J. (1995). *Visual stress* (pp. 35-52). Oxford: Oxford University Press.
- Wilkins, A.J. (2003). *Reading through colour*. Chichester: John Wiley and Sons
- Wilkins, A. J., Evans, B. J. W., Brown, J. A., Busby, A. E., Wingfield, A. E., Jeanes, R. J., & Bald, J. (1994). Double-masked placebo-controlled trial of precision spectral filters in children who use coloured overlays. *Ophthalmic and Physiological Optics*, *14*(4), 365-370. doi:10.1111/j.1475-1313.1994.tb00126.x
- Wilkins, A. J., Jeanes, R. J., Pumfrey, P. D., & Laskier, M. (1996). Rate of Reading Test: its reliability, and its validity in the assessment of the effects of coloured overlays. *Ophthalmic and Physiological Optics*, *16*(6), 491-497. doi. 10.1046/j.1475-1313.1996.96000282.x
- Wilkins, A. J., Baker, A., Amin, D., Smith, S., Bradford, J., Zaiwalla, Z., Besag, M.C., Binnie, C.D., & Fish, D. (1999). Treatment of photosensitive epilepsy using coloured glasses. *Seizure*, *8*(8), 444-449. doi.org/10.1053/seiz.1999.0337
- Wilkins, A., Lewis, E., Smith, F., Rowland, E., & Tweedie, W. (2001). Coloured overlays and their benefit for reading. *Journal of research in reading*, *24*(1), 41-64. doi:10.1111/1467-9817.00132
- Williams, D. (1999). *Like Colour to the Blind*. London: Jessica Kingsley Publishers Ltd.
- Williams, D., Stern, J. S., Grabecki, K., Simmons, H., & Robertson, M. M. (2013). Epilepsy in Tourette Syndrome. *Journal of Neurology, Neurosurgery & Psychiatry*, *84*(9), e1-e1. doi:10.1136/jnnp-2013-306103.31
- Wilson, R. L., Paterson, K. B., Hutchinson, C.V. (2015). Increased Vulnerability to Pattern-Related Visual Stress in Myalgic Encephalomyelitis. *Perception*, *44*, 1422-1426.
- Whitaker, L., Jones, C. R., Wilkins, A. J., & Roberson, D. (2015). Judging the Intensity of Emotional Expression in Faces: the Effects of Colored Tints on Individuals With Autism Spectrum Disorder. *Autism Research*. doi:10.1002/aur.1506
- White, B. B., & White, M. S. (1987). Autism from the inside. *Medical Hypotheses*, *24*(3), 223-229.