

**Looking and Thinking: How individuals with Williams syndrome make judgements about  
mental states**

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Abstract

Individuals with the neuro-developmental disorder Williams syndrome (WS) are characterised by a combination of features which makes this group vulnerable socially, including mild-moderate cognitive difficulties, pro-social drive, and indiscriminate trust. The purpose of this study was to explore a key socio-communicative skill in individuals with WS, namely, mental state recognition abilities. We explored this skill in a detailed way by looking at how well individuals with WS recognise complex everyday mental states, and how they allocate their attention while making these judgements. Participants with WS were matched to two typically developing groups for comparison purposes, a verbal ability matched group and a chronological age matched group. While eye movements were recorded, participants were shown displays of eight different mental states in static and dynamic form, and they performed a forced-choice judgement on the mental state. Mental states were easier to recognise in dynamic form rather than static form. Mental state recognition ability for individuals with WS was poorer than expected by their chronological age, and at the level expected by their verbal ability. However, the pattern of mental state recognition for participants with WS varied according to mental state, and we found some interesting links between ease/difficulty recognising some mental states (worried/ don't trust) and the classic behavioural profile associated with WS (high anxiety/indiscriminate trust). Furthermore, eye-tracking data revealed that participants with WS allocated their attention atypically, with less time spent attending the information from the face regions. This challenges the widely held understanding of WS being associated with prolonged face and eye gaze, and indicates that there is more heterogeneity within this disorder in terms of socio-perception than previous reports would suggest.

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1. Introduction

Individuals with Williams syndrome (WS) are known for their friendly, sociable and outgoing nature (Jones et al., 2000; Mervis & Klein-Tasman, 2000); but this characterisation hides an array of atypicalities of social perception, social cognition and struggles with everyday functioning. WS is a genetic disorder caused by a hemizygous deletion of around 1.6 megabase, containing 26 – 28 genes on chromosomal 7q11.23 (Eisenberg, Jabbi, & Berman, 2010). The disorder is characterised by a unique pattern of relative strengths and difficulties of cognition; for instance, more severe difficulties with visuo-spatial processing compared to a relative proficiency of verbal skill, all against a backdrop of mild-moderate intellectual impairment and cognitive heterogeneity (Searcy et al., 2004; Porter & Coltheart, 2005). Although the cognitive phenotype has attracted attention of cognitive scientists since the 1990s, much of the recent focus on WS have been on the social characteristics of individuals with the disorder (e.g. for a review see Jarvinen-Pasley, Korenberg, & Bellugi, 2013)

It has been proposed that many individuals with WS are highly motivated towards social engagement and that they exhibit a ‘pro-social’ drive (Frigerio et al, 2006) that may be difficult to inhibit (e.g. Little et al., 2013) and which increases social approach behaviours to both familiar and unfamiliar people (e.g. Jones et al., 2000). It has also been proposed that once individuals with WS engage in an interaction with others they show an array of subtle social engagement atypicalities, such as prolonged attention to a person’s face at the expense of attending other information in their environment. This prolonged attention to faces has been observed in both toddlers with the disorder (Mervis et al., 2003) and has been shown experimentally with older individuals who have WS using eye tracking (Riby & Hancock, 2008). This prolonged attention to a person’s face does not, however, lead to an increase or

heightened proficiency at interpreting information from that face, such as cues as to how the person is feeling (Plesa-Skwerer, Faja, Schofield, Verbalis, & Tager-Flusberg, 2006a) or where they might be attending (Riby, Hancock, Jones & Hanley, 2013). This is particularly important, as increased drive for social interaction (Jones et al., 2000), a lack of understanding of the dangers associated with interactions with unfamiliar people (e.g. Riby et al., 2013) and reduced ability to interpret sophisticated social cognitive cues once in that interaction, in parallel with a general reduction in intellectual capacity can leave individuals with WS highly vulnerable (see Jawaaid, Riby, Owens, White, Tarar & Schulz, 2012 for discussion). We aim to provide a detailed investigation of how well individuals with WS make judgements about mental states (complex emotions) from unfamiliar faces because this type of judgement can be important during an interaction and the ability to make this type of judgement requires socio-cognitive capacity. As an important extension to current knowledge, we aim to explore the information that individuals with WS use when making this type of socio-cognitive judgement by using eye-tracking techniques (thus also exploring social perception and linking these skills).

### *1.1 Looking at Faces*

Looking at faces is critical for social development as they provide a wealth of social and communicative cues that are central to social interaction (Haxby, Hoffman, & Gobbini, 2000). The role that looking at faces plays in the WS social profile has attracted attention partly because the pattern of atypicality appears to be syndrome-specific. Due to the link between looking at faces and social behaviour in WS, comparisons are often made between WS and autism (Tager-Flusberg et al., 2006; Riby & Hancock, 2008, 2009 a,b). Individuals functioning on the autism spectrum display a very different, but equally atypical, social profile to those with WS, both in the way people with autism look at faces (Hanley, McPhillips, Mulhern & Riby, in press), and in the nature of their everyday social behaviours, often characterised by social withdrawal. It has been proposed that both over- and under-attending to faces, as illustrated by these two disorders of development, has a

negative consequence throughout development for learning to interpret social information and developing social expertise (Klin, Jones, Schultz & Volkmar, 2003; Riby et al., 2013).

The first observational evidence of atypical attention to the faces of others came from Mervis and colleagues (2003) who suggested that toddlers with WS stared for longer than toddlers with other developmental disorders at the faces of their paediatrician. Experimental evidence from older children and adolescents was provided by Riby and Hancock in a series of studies that showed atypically prolonged attention to faces of people within social scenes (Riby & Hancock, 2008) and to actors within movies (Riby & Hancock, 2009a). On average, individuals with WS spent longer than typical looking at the eye region within the faces of actors in the stimuli (Riby & Hancock, 2009a) and this finding has since been replicated using faces illustrating basic expressions of emotion (Porter, Shaw, & March 2010). Therefore it has been proposed that the individuals with WS i) show atypically prolonged attention to faces and ii) within faces individuals with WS show atypically prolonged attention to the eye region. However, these previous eye tracking studies have used matched-group designs and reported the mean pattern for the WS group compared to their matches and have not explored within-group variability. This is important as we know that there is within-syndrome heterogeneity of both cognition (Porter & Coltheardt, 2005) and social behaviour (Little et al., 2013). The current study will explore the within-syndrome variability of attention allocation to faces and face regions.

### *1.2 Processing Emotions in WS: Basic and Complex Expressions*

Many individuals with WS seem highly sensitive to the emotions of others but perform relatively poorly on measures of emotion recognition in experimental settings. Several studies have provided evidence to show that individuals with WS perform at a level expected for their mental age (but not as accurately as expected by chronological age) when recognising basic expressions of emotion (see

various studies, Karmiloff-Smith et al., 1995; Gagliardi, Figerio, Burt, Cazzaniga, Perrett, & Borgatti, 2003; Plesa Skwerer et al., 2006a; Porter et al., 2010). For example, Plesa Skwerer et al. (2006a) compared the emotion recognition abilities of 47 individuals with WS to individuals with a learning disability (LD) and TD individuals. Participants were shown static images of basic emotions (happy, sad, angry, fearful) and both the WS and LD groups were significantly less accurate for recognition than the TD group. Group differences were particularly evident for the negative expressions, but not for 'happy'. The authors concluded that even when using basic expressions of emotion individuals with WS had some difficulties. A similar pattern was shown by Porter et al., (2010) for the recognition of static basic expressions, as participants with WS performed at the level of mental age, but worse than CA age matches.

Using animated facial expressions to better replicate the nature of changing expressions in everyday situations, Gagliardi et al. (2003) showed participants animated expressions portraying anger, disgust, fear, happiness and sadness (based on Ekman and Friesen). Participants with WS performed significantly less accurately at identifying the emotions in comparison to CA controls, but no differently to MA matches. Therefore, across both static and moving stimuli, when processing basic expressions of emotion individuals with WS perform as predicted by their mental age.

In everyday social interactions we need to process more complex expressions and mental states. These expressions are called mental states and examples include worried, jealous, thoughtful, and flirtatious among others. The ability to ascribe such 'mental states' to others is critical for understanding others' minds and therefore adapting our own behaviour. Tager-Flusberg, Boshart and Baron-Cohen (1998) and Plesa Skwerer et al. (2006b) have explored mental state understanding in WS using the 'Reading the Mind from the Eyes' task (Baron-Cohen, Wheelwright, Hill, Raste, & Plumb, 2001) and report variable findings (due to differences across studies in task design, see Riby & Back 2010 for discussion) but importantly both show that performance is below that expected by CA in typical

development. In research using more ecologically valid whole faces expressing different mental states, and linking directly to the task used in the current study, Riby and Back (2010) showed that when the whole face was available for interpretation individuals with WS were proficient at processing mental states but that they were largely relying on the eye region to do so, when the eye region became uninformative through computer manipulation performance significantly reduced for individuals with WS compared to those developing typically. However, in that study it would have been useful to track eye movements to explore the processing of different face regions. Indeed, in the only published study that has tracked eye movements during emotion recognition in WS, Porter et al. (2010) showed that participants with WS had an unusual attraction to the eyes when passively looking at faces expressing basic emotions in comparison to mental age matched TD participants.

Finally, the facial information that we process in everyday life is fluid and dynamic, and facial expressions can change on a moment-to-moment basis. Back, Jordan and Thomas (2009) demonstrated how providing dynamic facial information to TD individuals led to better mental state recognition than static facial information. The majority of WS research on emotion/mental state recognition however, has relied on static images of facial expressions.

### *1.3 Current Study*

The current study pulls together the issues addressed above to provide insight into the way that individuals with WS attend to and process complex mental states from unfamiliar faces. To make the stimuli ecologically valid we use moving faces as well as static faces. We use eight mental states that have been utilised in studies of WS individuals (Riby & Back, 2010), individuals with autism and both typical children and adults (Back, Ropar & Mitchell, 2007; Back et al., 2009). We also include eye tracking methods to explore attention allocation within the faces (e.g. eyes, mouth). Finally we

incorporate parental reports of everyday social behaviour to link task performance and eye tracking patterns to everyday abilities. We hypothesise that for individuals with WS mental state understanding will be at level predicted by mental age, not chronological age, and that attention allocation will be atypical.

## 2. Method

### 2.1 Participants

Nineteen individuals with Williams Syndrome aged between 7 years 2 months and 38 years 10 months (mean 21 years 6 months; 8 males, 11 females) were recruited via the Williams Syndrome Association of Ireland and the Williams Syndrome Foundation UK. Data from all WS participants were used in the analysis of the mental state accuracy assessment. However, it was not possible to obtain eye tracking data for four individuals with WS, leaving eye-tracking data from fifteen WS participants to be used in eye tracking analysis (see Table 1 for participant characteristics). Reasons for failure to obtain eye tracking data included inattention and strabismus, which are known to be common issues associated with WS (Atkinson et al., 2001; Leyfer et al., 2006).

All WS participants had been previously been phenotypically diagnosed by experienced clinicians and 15 of the 19 had previously had their diagnosis confirmed with positive fluorescence in situ hybridization (FISH) testing. The FISH test is a more recent diagnostic tool and those who had not had the FISH test were older members of the WS group (all were over the age of 28 years)<sup>1</sup>.

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<sup>1</sup> The 4 individuals who had not previously been FISH tested were all older adults (>28years) who were diagnosed prior to routine genetic testing for this disorder. Except for one result (see Footnote 2), the pattern of results remains consistent if these individuals are removed from the sample. As this does not change the main



Participants with WS were matched to two typically developing groups on the basis of i) chronological age (CA), and ii) verbal ability (VA) based on raw scores on British Picture Vocabulary Scale 2<sup>nd</sup> Edition (BPVS II; Dunn, Dunn, Whetton & Burley, 1997). There were 17 participants in the CA matched group, including six males and eight females, although eye-tracking data from two participants were not obtained (age range 6 years 10 months to 46 years 2 months; mean 22 years 0 months). There were 14 participants in the VA matched group, including four males and ten females (age range 6 years 6 months to 20 years 5 months; mean 10 years 11 months). All TD participants were also screened for history of neurological or psychiatric disorder, developmental disorder or learning difficulty, and no such issues were reported. All participants had normal or corrected-to-normal vision.

*[insert Table 1]*

Table 1 provides a summary of the participant characteristics for all groups. Individuals with WS did not differ from the CA group on the basis of chronological age, [WS  $n = 19$ , CA  $n = 18$ :  $t(35) = -.129$ ,  $p = .898$ ; WS  $n = 15$ , CA  $n = 15$ , :  $t(28) = .268$ ,  $p = .732$ ], and they also did not differ from the VA group on the basis of BPVS raw score, [WS  $n = 19$ , VA  $n = 14$ ,  $t(31) = -.589$ ,  $p = .560$ ; WS  $n = 15$ , VA  $n = 14$ ,  $t(27) = -1.044$ ,  $p = .306$ ]. Ethical approval for the research was granted by the ethics committee in the School of Psychology at Queen's University Belfast. Informed consent was obtained from all of the parents of participants with WS and participants in the VA and CA groups under the age of 18 years. Participants over 18 years in the CA group gave informed consent themselves.

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story of this manuscript, and to allow us to analyse all available data, these individuals have been left in the analysis.

## 2.2 Materials

### 2.2.1 *Experimental Stimuli*

Comprehensive details on the stimuli development are available elsewhere (Back et al., 2007; 2009). The stimuli used in the current study consisted of a series of eight mental states (deciding, don't trust, disapproving, not interested, not sure, relieved, surprised, worried) portrayed by a female actor; her face and top of her shoulders were visible onscreen in front of a blank white background. All the stimuli were in colour.

Each of the eight mental states had a static and a dynamic version. The dynamic versions began and ended with the actress showing a neutral face, and, in between, her emotional expression was shown for approximately 5 seconds. The static versions showed a still image of the actress portraying an emotional expression captured at the apex (most exaggerated part of the expression formation) for the same duration as the respective dynamic version. Following the presentation of a mental state stimulus, a slide appeared with four choices, where the correct mental state term was accompanied by three mental state 'foils' which shared a similar valence. The stimuli and foils used in this experiment have been validated and were used previously by Back et al. (2007). The order of appearance of the correct answer was counterbalanced.

The eight mental states were shown four times to each participant: twice as dynamic, and twice as static. The same mental state did not occur twice in succession. The order of the stimuli was otherwise random. Four predetermined counterbalanced stimuli sets were constructed (A, B, C, D), and participants were randomly assigned to a stimuli set. The first sixteen stimuli in each set (which consisted of the first presentation of each dynamic and static stimulus) were used for the eye tracking analysis, to capture the first attentional response to seeing each stimuli (as memory effects from repetition of stimuli can have an influence on eye movements; see Foulsham & Underwood, 2008).

All 32 stimuli from each set were used to score the accuracy of each participant in naming the mental states.

### *2.2.2 Parental Report of Social Behaviour*

The Social Responsiveness Scale (SRS; Constantino & Gruber, 2005) was administered to parents of the participants with WS who completed the eye-tracking part of the study. The SRS is a 65 item scale that provides an indication of social reciprocity, but which also contains sub-domains for social awareness, social cognition, social communication, social motivation and autistic mannerisms. The total score and the subscale scores can be converted to T scores to determine if behaviour is within the ‘normal’ range (T score below 55), the ‘mild – moderate impairment’ (55 to 75 range) or shows ‘severe impairments’ (76 to 90 range) that impact on every day functioning and ability. This measure has previously been used with individuals who have WS (van der Fluit, Gaffrey & Klein-Tasman, 2012; Klein-Tasmin, Li, Barber & Margaree, 2011). The adult version of the SRS was used for the adults in the sample (Constantino, & Todd, 2005).

Thirteen out of 15 SRS questionnaires were completed by parents (two parents did not return the questionnaires). The mean total score for the group was 64.8. Two participants total scores were in the normal range (15%), 2 were in the mild difficulties ranges (15%), and the remaining 9 (69%) scored in the severe range for difficulties with social reciprocity. The profile of functioning for these individuals with WS on the SRS is very similar to that reported elsewhere using the same measure (see van der Fluit, Gaffrey & Klein-Tasman 2012).

### *2.2.3 Eye-tracking Data Collection*

An SMI Remote Eye tracking Device (RED) 250 (SMI Germany) was used to record participants' eye movements. It sampled at 250Hz, with a gaze position accuracy of less than 0.5° of the visual angle. The stimuli were presented on a 22 inch monitor, and the infrared camera was mounted below the monitor. Participants sat in front of the RED, and although required to keep relatively still, they were not constrained in any way. BeGaze 2.0 (SMI, Germany) was used for analyses of the eye tracking data. Each mental state stimulus was divided into eleven Areas Of Interest (AOIs): eyes, nose, mouth, face (excluding the eyes, nose, and mouth regions), hair, body, background.

### 2.3 Procedure

We ensured that participants had the appropriate level of understanding for the mental states terms and foils, by reading aloud a series of sentences which each included one of the mental states (or one of the foils) in context (replicating Riby & Back, 2010). Participants were encouraged to ask questions about any of the words and were asked to demonstrate their understanding by producing their own sentence including the same mental state, or by describing a different situation in which someone may feel the same emotion, or by offering a synonym of the named mental state. In cases where participants struggled to demonstrate an understanding, the experimenter provided an explanation and a further example of the term, and the participant was required to show their understanding. All participants showed appropriate level of understanding.

The participant sat 0.5 metres from the monitor and eye-tracker and underwent a 13-point calibration. A 4-point validation was used to confirm the accuracy of the calibration (values of < .5 degrees were accepted).

Participants were given the following instruction: “There will be lots of faces appearing on the screen in front of you, please look at each face carefully. After each face four words will appear on the screen; please choose the word that best describes what the person was thinking or feeling and say it out loud”. The experiment began with two practice trials (one static stimulus and one dynamic stimulus) that were different to the experimental stimuli. After each stimulus, participants were presented with a forced choice; the correct mental state and three specific mental state foils were shown on screen and read aloud by the experimenter. The order of the multiple choice answers was randomised. At this point participants were asked to name the correct mental state. The next stimulus was not shown until they answered, thus making the task self-paced. No feedback was given and each stimulus was shown only once per trial.

The procedure, including the completion of the BPVS, took approximately 50 minutes. All WS participants were tested in their homes; typically developing participants were tested either in their homes or in the eye tracking lab at Queens University Belfast. On completion of the experiment, the participants (and, where appropriate, their parents) were given the opportunity to ask any questions.

### 3. Results

#### 3.1 Mental State Recognition Accuracy

Accuracy for mental state recognition was explored using a three-way ANOVA with factors Group (WS, CA, VA), Display, (Static, Dynamic) and Mental State (deciding, don't trust, disapproving, not interested, not sure, relieved, surprised, worried). Accuracy differed significantly between groups,  $F(2, 47) = 8.598, p = .001, \eta^2 = .267$ , as the WS group was less accurate than the CA matched group ( $p < .001$ ), but comparable to the VA group ( $p = .184$ ). There was no difference between the CA and VA

groups ( $p = .103$ ). In relation to chance performance, the CA group performed above chance on all mental states, and the VA group performed above chance on all but disapproving,  $t(13) = 1.099$ ,  $p = .292$ , and not interested,  $t(13) = 1.979$ ,  $p = .06$ . The WS group performed above chance on all but disapproving,  $t(18) = -1.837$ ,  $p = .08$ , don't trust,  $t(18) = .490$ ,  $p = .630$  and relieved,  $t(18) = 1.723$ ,  $p = .102$ .

There was a significant main effect of Display,  $F(1, 47) = 5.311$ ,  $p = .026$ ,  $\eta^2 = .101$ , as dynamic displays were recognised more accurately than static displays [mean dynamic = 61%, static = 56%]. Finally, there was a significant main effect of Mental State,  $F(7, 329) = 11.642$ ,  $p < .001$ ,  $\eta^2 = .187$ . Pairwise comparison post hoc tests (with Bonferroni adjustment) were used to unpack the effect of mental state on accuracy. Disapproving was the most difficult mental state to identify (36.5% accuracy), and was significantly harder to identify than deciding ( $p < .001$ ), not sure ( $p < .001$ ), relieved ( $p = .004$ ), surprised ( $p < .001$ ) and worried ( $p < .001$ ). Worried was the easiest to identify (76% accuracy), and was significantly easier to identify than don't trust ( $p = .001$ ), not interested ( $p < .001$ ) and relieved ( $p = .005$ ). Finally, not interested was also difficult to identify (46.5% accuracy), more difficult than deciding ( $p = .008$ ), not sure ( $p = .016$ ), and surprised ( $p = .044$ ). Table 2 shows the mean accuracy scores for each mental state in order of difficulty (for all participants and separately for each display type).

*[Insert Table 2]*

There were significant interactions between display and mental state,  $F(7, 329) = 4.646$ ,  $p < .001$ ,  $\eta^2 = .088$ , and mental state and group,  $F(14, 329) = 1.739$ ,  $p = .047$ ,  $\eta^2 = .055$ , but no significant interactions between display and group,  $F(2, 47) = .002$ ,  $p = .998$ ,  $\eta^2 = .000$ , or no three-way interaction between display, group and mental state,  $F(14, 329) = .537$ ,  $p = .911$ ,  $\eta^2 = .02$ .

The interaction between display and mental state indicated that although the mean accuracy for the dynamic displays was highest, this pattern differed across the eight mental states. Paired samples *t*-tests revealed that for three mental states, accuracy rates differed significantly across display types, in the direction of better accuracy for dynamic displays. The mental states don't trust (static: 40.2%; dynamic: 58.6%;  $t(49) = -3.447, p = .001$ ), not interested (static: 36.9%; dynamic: 53.2%;  $t(49) = -2.687, p < .05$ ) and surprised (static: 50%; dynamic: 75%;  $t(49) = -4.096, p < .001$ ) were all significantly easier to identify in the dynamic display form. However, deciding was easier to identify in the static condition (static: 75%; dynamic: 60%;  $t(49) = 2.605, p = .012$ ) [see Table 2].

A series of one-way ANOVAs were carried out on the data to unpack the interaction between group and mental state. The groups differed for accuracy on three mental states: disapproving,  $F(2, 47) = 10.668, p < .001, \eta^2 = .312$ , don't trust,  $F(2, 47) = 9.236, p < .001, \eta^2 = .282$ , and relieved,  $F(2, 47) = 4.371, p = .018, \eta^2 = .156$ . No other effects were significant (all  $F$ s  $< 2.732$ , all  $p$ s  $> .075$ ). Tukey post hoc tests revealed that the WS and the VA groups were less accurate at identifying disapproving than the CA group (WS-CA:  $p < .001$ ; VA-CA:  $p = .033$ ), but that there was no difference between the WS and VA group ( $p = .216$ ). The WS group were significantly less accurate at identifying don't trust by comparison to the CA group ( $p = .001$ ) and the VA group ( $p = .008$ ), but the CA and VA groups did not differ ( $p = .726$ ). Finally, the WS group were significantly less accurate than the CA group at identifying relieved ( $p = .013$ ), but there was no difference between the WS and VA groups ( $p = .353$ ) or the CA and VA groups ( $p = .362$ ).

Therefore accuracy was dependent upon mental state and the type of display of those mental states. Overall, participants with WS performed at the level expected by their verbal ability, with some important exceptions. Participants with WS had particular difficulty recognising don't trust,

performing worse at recognising this mental state than VA participants and actually performing no different to chance for that mental state when it was shown in the dynamic form. In contrast, they performed better (and at the level expected by their CA) at recognising the mental states deciding, not sure and worried [WS-CA: deciding,  $t(34) = -.629, p = .553$ ; not sure,  $t(34) = -1.101, p = .279$ ; worried,  $t(34) = -.629, p = .553$ ].

### 3.2 Attention Patterns during Mental State Recognition

#### 3.2.1 Attention to face regions during mental state judgements

Using a three-way ANOVA we explored how participants in each group (WS, CA, VA) allocated attention to the face regions (eyes, nose, mouth, face non-feature areas) while making mental state judgements across both display types (static, dynamic). For this analysis, only ET data from the first viewing of each mental state were used<sup>2</sup>. The ANOVA revealed that the groups spent different percentages of time fixating the whole face,  $F(2, 41) = 4.365, p = .019, \eta^2 = .175$ , that attention was allocated differently within the face AOIs,  $F(3, 123) = 50.612, p < .001, \eta^2 = .529$ , but that the face AOIs were viewed similarly across the two display types,  $F(1, 41) = 2.333, p = .134, \eta^2 = .05$ .

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<sup>2</sup> We also conducted a further analysis of attention during correct mental state judgements only, in order to explore in detail what information participants were looking at when they made correct judgements. We conducted an ANOVA with factors group (WS, CA, VA) and AOI (eyes, nose, mouth, face non-feature areas) on looking time for the first correctly answered trial for both display types of each mental state. The pattern of results was the same as above, in that there was a main effect of group,  $F(2, 41) = 4.549, p = .016, \eta^2 = .221$ , driven by the WS participants spending less time looking at the face regions than the CA group ( $p = .018$ ); a main effect of AOI,  $F(3, 123) = 55.336, p < .001, \eta^2 = .55$ , driven by the eyes being viewed for longer than all other face regions [eyes-nose:  $p < .001$ ; eyes-mouth:  $p < .001$ ; eyes-face non-feature areas:  $p < .001$ ]; but no interaction effect between group and AOI,  $F(6, 123) = 1.900, p = .086, \eta^2 = .03$ .



In terms of the main effect of group, Tukey post-hoc tests showed that the WS group spent a smaller proportion of time viewing the whole face than the CA group ( $p = .024$ ). There was a trend towards significance for the WS/VA comparison ( $p = .068$ ), but the two typically developing groups did not differ significantly (CA-VA:  $p = .912$ ). Follow-up analyses to the main effect of AOI (pairwise comparisons with Bonferroni adjustment) revealed that the eyes were viewed for a longer proportion of time than all other face AOIs [eyes-nose:  $p < .001$ ; eyes-mouth:  $p < .001$ ; eyes-face:  $p < .001$ ], but there were no differences between the other face AOIs (all  $p$ 's above .875).

*[Insert Figure 1]*

In terms of interactions, the ANOVA revealed no significant interaction between group and display for viewing of face AOIs,  $F(2, 41) = .551, p = .581, \eta^2 = .024$ , and no three-way interaction between group, display and AOI,  $F(6, 123) = .427, p = .860, \eta^2 = .018$ . The interaction between group and AOI approached significance,  $F(6, 123) = 2.005, p = .070, \eta^2 = .04$ . The pattern of the trend was explored further, given that looking to the eyes in particular has been highlighted as a syndrome specific atypicality in WS. A one-way ANOVA revealed the effect of group (WS, VA, CA) on looking to the eyes approached significance,  $F(2, 41) = 3.019, p = .06, \eta^2 = .128$ , and was driven by the WS group spending less time looking at the eyes than the CA group, although this was only a trend towards significance ( $p = .09$ )(WS-VA:  $p = .104$ ). There were no main effects of group on looking to any other face AOIs (all  $F$ 's  $< 1.082$ , and all  $p$ 's  $> .348$ ).

There was an interaction between display and AOI,  $F(3, 123) = 3.700, p = .014, \eta^2 = .018$ , and follow-up paired t-tests showed that this was because the nose and mouth regions were viewed for a

larger proportion of time in the dynamic as opposed to static condition [Nose:  $t(43) = 2.596, p = .013$ ; Mouth:  $t(43) = 2.228, p = .031$ ].

*[Insert Figure 2]*

### 3.2.2 *Priority of attention to the face regions*

We explored the priority of attention to the face regions by looking at which areas of the face participants fixated first. 45% of the WS group's first fixations were made to the non-feature areas of the face; 31.1% were directed towards the eyes; 13.5% were directed towards the nose; and 9.8% were directed towards the mouth region. For the CA group, 48.6% of first fixations were directed towards the non-feature areas of the face; 34.2% towards the eyes; 13.5% towards the nose, and 3.6% towards the mouth. For the VA group, 42.7% of their first fixations to the face were directed to the non-feature areas; 39.7% to the eyes; 10.6% to the mouth; and 7.0% to the nose.

We explored the pattern of first fixations using a mixed ANOVA with factors group (WS, VA, CA) and AOI (eyes, nose, mouth, face non-feature areas) on number of first fixations. There was a main effect of AOI on the number of first fixations,  $F(3, 120) = 23.781, p < .001, \eta^2 = .36$ , but no effect of group,  $F(2, 40) = 1.557, p = .223, \eta^2 = .07$  and no interaction between group and AOI,  $F(6, 120) = .469, p = .830, \eta^2 = .014$ . Planned comparisons with Bonferroni adjustment revealed that the main effect of AOI was driven by eye region being fixated more often than the nose ( $p < .001$ ) and the mouth ( $p < .001$ ), and the face non-feature area being fixated first more often than the nose ( $p < .001$ ) and the mouth ( $p < .001$ ), but not the eyes ( $p = .230$ ).

### 3.3 Relationships between Accuracy, Attention & Participant Characteristics

### 3.3.1 *Typically developing participants*

A series of two-tailed correlations were conducted to explore the relationships between participant characteristics (BPVS, CA) and performance measures from the task (accuracy, attention).

In relation to performance accuracy on the mental state judgement task (across all mental states and for both display types), no significant relationships were found between either BPVS raw score or chronological age and task accuracy for the typically developing individuals ( $p > .1$ ).

In relation to measures of attention (time spent looking at the eyes/mouth), for all typical participants there was no significant relationship between looking at the eyes and chronological age, ( $r = -.118, p = .541$ ), nor for looking at the mouth and chronological age, ( $r = -.330, p = .08$ ).

For the VA group, there was a significant relationship between percentage of time spent looking at the eyes and BPVS raw score ( $r = .619, p = .018$ ), and for looking to the mouth and BPVS raw score ( $r = -.577, p = .031$ ), indicating that more time spent looking at the eyes was related to higher receptive vocabulary scores, and less time spent looking at the mouth was related to higher receptive vocabulary scores.

### 3.3.2 *Williams syndrome participants*

A series of two-tailed correlations were conducted to explore the relationships between participant characteristics (BPVS, CA) and performance measures from the task (accuracy, attention).

In relation to performance accuracy on the mental state judgement task , there was no significant relationship between either BPVS raw score or chronological age and accuracy for the WS group ( $p > .1$ ).

In relation to measures of attention (time spent looking at the eyes/mouth), the relationship between percentage of time spent looking at the eyes and BPVS raw score was not significant, ( $r = .430, p = .110$ ) nor was the relationship between BPVS raw score and looking at the mouth, ( $r = .204, p = .466$ ). No relationships were found between CA in the WS group and looking at the eyes, ( $r = .365, p = .181$ ), or the mouth, ( $r = -.077, p = .785$ ).

### 3.3.2.1 Attention to the Eyes & Mouth and SRS

A series of two-tailed correlations were conducted between scores on the SRS and attention to the eyes and mouth (for all stimuli, i.e., for all mental states and for both display types) for the WS group only.

The percentage of time spent looking at the eyes was found to correlate negatively with SRS total score ( $r = -.730, p = .005$ ), indicating that less time spent looking at the eyes was related to higher scores on the SRS (higher levels of social functioning difficulties). In terms of the SRS sub-domains, less time spent looking at the eyes was also correlated with poorer social cognition ( $r = -.781, p = .002$ ), poorer social communication ( $r = -.599, p = .031$ ), and more autistic mannerisms ( $r = -.637, p = .019$ ), but not with social awareness ( $r = -.435, p = .138$ ) or social motivation ( $r = -.430, p = .143$ ).

A relationship between percentage of time spent looking at the mouth and SRS total score was not found ( $r = .483, p = .094$ ). In terms of SRS sub-domains, more time spent looking at the mouth correlated with poorer social motivation ( $r = .564, p = .045$ ), but not with any of the other sub-domains (all  $r$ 's  $< .468$ , all  $p$ 's  $> .107$ ).

#### 4. Discussion

The aim of the study was to provide a detailed investigation of mental state understanding in WS, by exploring how well individuals with WS made judgements about complex mental states, and where they looked when making those judgements. In line with previous research, participants with WS generally performed at a level expected from their verbal ability, and poorer than expected from their chronological age (Gagliardi et al., 2003; Plesa Skwerer et al., 2006). Importantly however, attention patterns revealed that participants with WS allocated their attention atypically, in ways that may impact upon their ability to make socio-cognitive judgements, and which were related to their social functioning. So even though individuals with WS and verbal ability matched TD individuals performed to a similar level in terms of accuracy, attention distribution patterns indicated that the groups were fixating on and therefore using, different information to make their judgements.

##### 4.1 *Mental State Judgements*

Using stimuli that better replicate realistic social information is important when trying to understand how this information is used in real life. We used a range of complex mental states in two presentation forms and this was significant for recognition as dynamic stimuli were more easily identified by all participants than static stimuli. This is in line with Back et al. (2009) who showed how dynamic expressions facilitate recognition. They suggest that the additional information contained in dynamic facial expressions, such as temporal information, gives rise to greater recognition. In real life, the

facial expressions we encounter are dynamic and fluid, and our results show how participants find it easier to recognise complex mental states in this form.

Using a range of complex everyday mental states we found that some were easier to recognise than others (worried was the easiest, disapproving was the hardest). Participants with WS generally recognised mental states at the level expected by their verbal ability, which was poorer than expected from their CA (Plesa Skwerer et al., 2006a). However, they had some specific difficulties. 'Relieved' and 'don't trust' were particularly difficult for the WS group to identify, and they had more difficulty with these mental states even in comparison to VA participants (VA participants recognised both mental states above chance levels). Specific difficulties with the mental state 'don't trust' is especially interesting in context of the issues that people with WS have with stranger danger. Indiscriminate trust (Riby et al., 2013) and increased social approach (e.g. Jones et al., 2000) have become hallmarks of the WS social phenotype. Here we can see how understanding the socio-communicative cues surrounding trust is a particular difficulty for individuals with WS. Further research may explore in more detail the link between trust judgements and approach decisions in this group, as inappropriate judgments at either stage may lead to increased social vulnerability.

Some mental states posed less difficulty to participants with WS. Deciding, not sure and worried were recognised at the level expected by the WS participants' chronological age. Again, this is particularly interesting in the context of the behavioural and psychopathologic profiles associated with WS. Anxiety is the most common psychopathology seen in both children and adults (Porter, Dodd, & Cairns, 2009), and here we find that recognising the socio-communicative cues of anxiety on the faces of others is a strength in this group. This is an important finding and suggests that although individuals with WS are generally not proficient at discerning complex socio-communicative cues, they are very attuned to detecting anxiety. Although it is not possible to say from this study whether

there is a causal relationship between proficiency at recognising anxiety in others and experiencing high levels of personal anxiety, the link is interesting, and will be important to explore further.

#### 4.2 *Attention Allocation to Mental States*

We explored how accurate participants were at making mental state judgements, but also how they allocated their attention to the face while making those judgements, linking looking and thinking. The stimuli were viewed similarly regardless of whether they were static or dynamic, and the eyes were viewed more than any other face region. However, there were some group differences in attention allocation that indicate strategy differences in task completion.

Participants with WS spent less time looking at the face regions collectively (eyes, nose, mouth, non-face features) in comparison to both control groups. This was surprising, given the wealth of evidence that suggests participants with WS show prolonged face gaze (Mervis et al., 2003; Riby & Hancock, 2008). Prolonged attention to the eye region of emotionally expressive faces has been reported in WS (Porter et al., 2010), and so we explored attention allocation to the face regions between the groups. We did not find prolonged eye region fixations for our WS participants and instead report reduced attention to the eyes in comparison to typical participants. This is surprising, given that prolonged face and eye gaze is something that has been well-documented in WS (Riby & Hancock, 2008, 2009a, 2009b).

It is possible that the complexity of the mental state stimuli revealed different attention patterns in WS to that reported previously using basic emotions (Porter et al., 2010). Although under-researched in WS, the issue of the effect of stimuli factors on gaze behaviour has been considered in the literature

on social attention in autism in detail (Hanley et al., in press). It is generally accepted that for individuals with autism, atypicalities of social attention are most evident when experimental stimuli replicate realistic social information (stimuli with increased complexity, human actors and social interactions) (Hanley et al., in press). In the only study to address this issue in WS, Riby and Hancock (2009b) found that the ecological validity of experimental stimuli effected gaze behaviour in WS, and that the same participants with WS showed both prolonged face gaze *and* typical face gaze depending on the stimuli shown. Only with carefully designed studies using a range of stimuli similar to those that have been carried out with individuals with autism, can we understand that effect of stimuli choice on gaze behaviour in WS.

Finding reduced eye and face gaze in WS is intriguing not just because it contrasts with previous reports of these behaviours within the syndrome, but because of the cross-syndrome comparisons that are often made between WS and autism in the literature on social attention. Their apparently ‘opposite’ social profiles is the reason that studies often compare participants with WS to participants with autism to further understanding of social, cognitive and affective neuroscience (Tager-Flusberg et al., 2006). Whereas participants with autism have most often been found to show reduced social attention in a way that relates to their poor social functioning (Klin, Jones, Schultz, Volkmar, & Cohen, 2002), the links between attention and functioning in WS have not explored. Here, we found that looking patterns were associated with social functioning profiles in WS. We found that less time spent fixating the eyes was related to poorer social functioning in these participants, in particular poorer social cognition, communication and more autistic mannerisms. Although we do advise caution in interpreting the correlations here given the small sample size, the pattern we report for our WS participants is very similar to the pattern most often reported in studies involving participants with autism (Klin et al., 2002). Although heterogeneity has been highlighted in respect of the cognitive phenotype associated with WS, we are not aware of reports of such heterogeneity in respect of their social-cognitive profile. To the authors’ knowledge, this is the first study to report diminished eye region fixations in WS, which is associated with poor social functioning.



If these findings are taken into account in terms of the broader literature on WS, then they may not be considered as surprising. A small number of studies have highlighted phenotypic overlap between WS and autism using diagnostic assessments such as the Autism Diagnostic Observation Schedule (Klein-Tasman, Mervis, Lord, & Phillip, 2007; Lincoln, Searcy, Jones, & Lord, 2007; Lord, Rutter, DiLavore, & Risi, 1999). The converging evidence indicates that the characterisation of individuals with WS as being hypersociable and showing prolonged face gaze may not be true for all individuals and there is more heterogeneity than previously thought. We present the first evidence of this heterogeneity extending to the socio-perceptual domain, with evidence linking looking, thinking and functioning in WS. More work is needed as with most studies of WS, large sample sizes are difficult to obtain and achieving large samples will be key to understanding heterogeneity.

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