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Stranger Danger Awareness in Williams syndrome

Deborah M Riby¹, Hannah Kirk¹, Mary Hanley², & Leigh M Riby³

1. School of Psychology, Newcastle University, UK
2. School of Psychology, Queen's University Belfast, UK
3. Department of Psychology, Northumbria University, UK

Correspondence should be addressed to: Dr Deborah Riby, School of Psychology, Newcastle University, Ridley Building 1, Framlington Place, Newcastle upon Tyne, NE1 7RU, UK. Tel: +44 (0) 191 222 6557. Email: Deborah.riby@ncl.ac.uk

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Hannah Kirk has since moved to the Department of Psychology & Psychiatry, Monash University, Clayton, Vic 3800, Australia.

Conflict of Interests Statement

The authors report no conflict of interests

Abstract

The developmental disorder Williams syndrome (WS) is characterised by a distinctive cognitive profile and an intriguing social phenotype. Individuals with the disorder are often highly social engaging in interactions with familiar and unfamiliar people and once in those interactions they often show subtle abnormalities of social behaviour. Atypically increased approach to unfamiliar people is widely reported in the existing literature for both children and adults. Parents frequently report interactions with unfamiliar people as a major concern. In this study we aimed to evaluate ‘Stranger Danger’ awareness in individuals with WS. When linked to other components of the WS phenotype (e.g. reduced intellectual ability, increased social approach) an awareness of stranger danger is particularly important. Using a video vignette task we show that young people with WS have difficulties making judgements about whether or not to trust and engage in conversation with unfamiliar people. Qualitative data showed that individuals with WS often suggested that they would engage in an interaction with an unfamiliar person. The findings have substantial implications for the safety of young people with the disorder and emphasise the timely need for further intervention regarding this behaviour.

Keywords: Williams syndrome, social approach, approachability, trust, stranger danger

Stranger Danger Awareness in Williams syndrome

In our everyday lives we must interact with many different people in different ways. Some of those people will be more familiar to us than others and it is critical that from a relatively early age we begin to distinguish between people that we know and those we do not know; modifying our behaviour and interactions accordingly. In typical development, infants begin to show an instinctive awareness of strangers (e.g. stranger anxiety) with behaviours associated with wariness and fear from about 6-12 months of age (e.g. Rheingold & Eckerman, 1973). At a slightly older age, young children are often made explicitly aware (for example, by parents or teachers) that they should not interact with people they do not know (e.g. see Moran, Warden, Macleod, Mayes, & Gillies, 1997). With increasing age we learn to make evaluations of when it might, or might not, be appropriate to interact with someone we have not met before. Not only must we become competent at differentiating familiar and unfamiliar faces through development, we must also learn to decipher the sophisticated and subtle social cues that can be derived from those faces, for example a person's feeling or intentions. Entwined in both our awareness of unfamiliar people and our evaluation of face cues is the assessment of trust. Making an evaluation of trust will effect our decision to approach or avoid someone and is involved in recognising the possible consequences of making good or bad social judgements (for example, trusting and approaching an inappropriate individual). A wrong decision at this point could have serious negative consequences.

Difficulties evaluating social cues from faces can lead to interaction atypicalities and this may be the case for individuals with the sporadically occurring developmental disorder Williams syndrome (WS). WS has an estimated prevalence of 1:20,000 (Morris & Mervis,

1999, but see Strømme, et al., 2002) and is caused by the deletion of approximately 25-28 genes on chromosome 7 (Ewart et al., 1993). The disorder is characterised by atypical social behaviours (e.g. Jones et al., 2000; Doyle, Bellugi, Korenberg, & Graham, 2004) and mild-moderate intellectual difficulty (e.g. Searcy et al., 2004). Cognitively, individuals with WS are often more proficient in the use of verbal than spatial skills, though deficits occur even within language (e.g. better receptive vocabulary alongside deficits of grammar; for a comprehensive review see Martens, Wilson, & Reutens, 2008). Socially, individuals with WS are reported to show atypical social interaction styles that are often characterised by a propulsion towards social engagement (Frigerio et al., 2006) irrespective of the familiarity of the person they are interacting with (e.g. Jones et al., 2000). Specifically, many individuals with WS are described as ‘people-oriented’, ‘affectionate’, ‘sensitive’, ‘empathetic’ and ‘friendly’ (e.g. Tomc, Williamson, & Pauli, 1990; Tager-Flusberg & Sullivan 2000; Klein-Tasman & Mervis 2003). However, despite these seemingly positive attributes, many adults with WS struggle with peer relations in adulthood (e.g. Davies et al., 1998) and suffer social isolation (e.g. Mervis & Klein-Tasman, 2000). One critical feature is that some individuals with WS may be vulnerable to engaging with strangers, as shown by their increased approach choices to unfamiliar people. When coupled with evidence of mild-moderate intellectual difficulties (e.g. Searcy et al., 2004) and atypical social engagement styles once in an interaction (e.g. prolonged face gaze, Mervis et al., 2003; Riby & Hancock, 2008) this combination of behaviours becomes of paramount importance (for discussion of vulnerability issues in WS see Jawaid et al., 2012).

Numerous studies have relied on parental reports of the behaviour of individuals with WS to make claims of atypically increased approach to unfamiliar people. Such studies have consistently found that individuals with WS, even as young as 13 months, are rated as more

sociable towards strangers than typically-developing children and children with other developmental disorders (Doyle et al., 2004; Dykens & Rosner, 1999; Jones et al., 2000; Sarimski, 1997). Other studies have assessed social approach behaviour by asking individuals with WS to rate the approachability of pictures of unfamiliar faces. For example, Bellugi, Adolphs, Cassady and Chiles (1999) found that individuals with WS rated unfamiliar faces as more approachable than typically developing children. Several more recent studies have indicated higher than typical approach ratings for faces depicting either positive or negative expressions (e.g. Jones et al., 2000; Frigerio et al., 2006; Martens et al., 2009, see also Porter, Coltheart & Langdon, 2007). The only other study to have included a more direct evaluation of approach to a stranger has been conducted by Dodd, Porter, Peters and Rappe (2010) who used an observational paradigm to study the approach behaviour of pre-school children with WS. The young children with WS were significantly more willing than typically developing children to engage with a stranger in a real interaction. Other than this work with particularly young children, there has been little insight into other methods of evaluating how well an individual with WS understands the implications of approaching unfamiliar people or indeed the possible consequences. Qualitative data may be particularly useful here (providing insights into thoughts and feelings of individuals with WS) to explore the phenomenology of atypical social interaction tendencies in individuals with the disorder.

The aim of the current study was to probe an awareness of 'stranger danger' in individuals with Williams syndrome. To meet this aim we used a video vignette task to probe understanding of strangers, especially interactions with an unfamiliar adult, by young people with the disorder. Using this method would allow the collection of qualitative data to explore the thoughts of individuals with and without WS when they watched unfamiliar people interacting in different situations. Questions were used to encourage the participant to talk

about the appropriateness of interactions with unfamiliar adults and the study aimed to add a new dimension to this type of research, which was previously dominated by the use of social approach rating scale tasks involving quantitative data collection. It was hypothesised that individuals with WS would show a lack of awareness of appropriate interactions styles for engaging with unfamiliar people in the answers they gave when probed about issues of stranger danger. This prediction was made based on reports in the literature of increased approach towards unfamiliar others (e.g. Jones et al., 2000). The scenarios were purposefully set up so that interacting with the stranger would be associated with a possible negative outcome (e.g. increase the vulnerability of the character in the video) . Any lack of awareness could have important practical implications and would warrant further work on the development of social skills training programs covering components of stranger danger specific to the needs of individuals with this disorder. We also analysed any emerging themes from the information provided in responses about strangers to explore common issues and understanding for the individuals with WS as well as for the typically developing participants. Finally, as an exploration of the relationship between stranger danger awareness and everyday social skills in WS (a question not thoroughly probed in the existing literature), we correlated a parent report measure of social skills (using the Strengths & Difficulties Questionnaire; Goodman, 2001) with a quantitative measure of stranger danger awareness shown on the video vignette task. We hypothesised that those individuals with more problems in terms of their social behaviour in everyday situations would be the same individuals that showed problems with adequately evaluating the appropriateness of interactions with unfamiliar people.

Method

Participants

Sixteen individuals with WS (mean 12 years 1 month; ranging 8 to 17 years; 8 male) were recruited via the Williams syndrome Foundation. All participants had previously been diagnosed phenotypically and their diagnosis had been confirmed with positive *fluorescent in situ hybridisation* testing indicating the absence of one copy of the elastin gene on chromosome 7. Inclusion in the WS group was reliant upon a previous genetic and phenotypic diagnosis. Individuals were also excluded if they had an existing co-morbid diagnosis (e.g. ADHD, Autism etc). Each individual with WS was matched to a typically developing individual on the basis of verbal ability as measured by the verbal IQ (VIQ) component of the Wechsler Intelligence Scale for Children – Third Edition Short Form (WISC-III; Wechsler, 1991). The ‘Similarities’ and ‘Vocabulary’ components of the WISC were combined to provide an indicator of VIQ (See Minshew, Turner & Goldstein, 2005 for a discussion of the use of these specific subtests as part of an extensive short form IQ measure). VIQ scores ranged from 49 – 91 (mean 61.2; standard deviation 12.7) and gave a range of verbal mental ages between 5 and 13 years. For information, the full scale IQ scores for the WS group ranged between 47 – 86 (mean 58.4; standard deviation 12.2), showing within-syndrome variability but a mean within the mild intellectual difficulty range, which is highly indicative of WS (e.g. as reported by Searcy et al., 2004).

The VIQ matched group of typically developing individuals was aged between 5 and 12 years (mean 7 years 9 months; 8 male; groups were also matched on gender). The WS and VIQ groups did not statistically differ on the basis of their verbal ability as measured by the WISC VIQ components $p=.72$ but chronologically the WS group was significantly older than the typically developing group $t(30)=6.38, p<.001$. Inclusion / exclusion for the typical matched group was reliant upon ‘normal’ functioning on the Strengths & Difficulties Questionnaire (Goodman, 2001) completed by parents indicating a lack of problems in the domains of

emotion and behaviour ('total difficulties' score for all participants in this group was within the 'normal' range). Participants would also not meet inclusion criteria if they have known diagnosed developmental disorders.

Stimuli and Procedure

Two video extracts were used that included strangers (an unfamiliar adult) interacting with young children. The videos were taken from material specifically designed to raise the awareness of young children about stranger danger. Therefore, the choice of stimuli was appropriate to the age range of our participants as the individuals would be able to closely relate to the situations and characters involved in the clips. The clips were relevant to children and therefore the participants would either have previously had experience of the types of settings seen in the video extracts (e.g. a park) or they would be something familiar to their current experiences. Each participant was exposed to both videos, which were played as part of a Microsoft PowerPoint presentation; however the order in which participants viewed the clips was randomised. Each participant was tested individually in a quiet environment. The experimenter sat by their side, with the computer screen in front of the participant, and no other people were in the room at the time of testing.

A brief introduction was given, whereby the experimenter gave a verbal outline of what would happen in each clip prior to the participant being exposed to the visual events. This was important as no sound accompanied the video extracts (this was partly due to the accent of actors within the clips as we did not want this to influence the participants and it was also partly because we wanted to be able to set out own background story for these extracts which would not have otherwise been possible). Participants were told to watch carefully as a

discussion with the experimenter would follow. After the viewing of each video the experimenter asked the participant if they had any questions before they started their discussion of the video clips (thus ensuring understanding).

The experimenter explained that the first clip would involve a young boy called Jamie playing in a playground with lots of other children. Jamie's mum is said to be sat on a bench nearby relaxing (she can be seen in the video sitting on a bench). While Jamie is playing a man is shown to walk up to him, Jamie does not know the man. The man states that he has lost his dog and asks for Jamie to help him find the dog. The video lasted 19 seconds.

As soon as the clip finished the individual was then asked two questions, specifically: 'What do you think Jamie should do?', 'Should he help the man find his dog?' and are asked to give reasons for their answers. They are then asked what they would do in the same situation. See Appendix A.

Clip 2 showed three boys playing on their bikes, as they cycled down the road a man in a car pulls up and calls them over. The participant is told that the boys do not know the man that is driving the car. The man tells one of the boys that his mum has been in an accident and that he should go with him to get help. The video lasted 12 seconds. Again the participant was asked two questions about the clip they had just watched: 'What do you think the boys should do?' and 'Do you think they should go with the man or find a different way to check if their mother is ok?'. The marking matches that used for Clip 1 in Appendix A.

Once the participant had viewed both clips they were also asked two more general questions about 'stranger danger' that was not directly linked to the clips. These questions were 'what is a stranger?' and 'tell me something you know about stranger danger.'

At the end of the session the participant was thanked and the experimenter answered any additional questions that arose.

As well as the participant completing the video extract task, their parent was also asked to complete the Strengths & Difficulties Questionnaire (SDQ; Goodman, 2001). The SDQ is a parent rated 25-item questionnaire which provides measures of ‘emotional symptoms’, ‘conduct problems’, ‘hyperactivity’, ‘peer problems’, and ‘pro-social behaviour’. A ‘total difficulties’ score can be calculated from the addition of each subscale (with the exception of pro-social behaviour). We were particularly interested in the peer problem and prosocial behaviour scores. On the Peer Problems subscale the scoring can range from 0-10: scores ranging 0-2 are considered ‘normal’, 3 is considered ‘borderline’ and scores 4-10 are considered ‘abnormal’. The scale is inverted for the Prosocial Behaviour score with the ‘normal’ range being 6-10, ‘borderline’ being 5 and ‘abnormal’ range being 0-4. Five items contribute to each of these categories (each scores 0-2). In all cases the SDQ was completed by the mother.

Scoring for Quantitative Analysis

The data obtained at the end of the video vignette task were qualitative and we therefore devised a scoring guide which allowed us to rate each individual's answer and to provide them with a score, thus transferring the data to a quantitative state. For Clip 1, a maximum score of 6 was possible if they stated appropriately that the boy should speak to his mum and shouldn't go with the man as well as giving a good reason why. See Appendix A for clear indication of marking of this question.

The scoring for Clip 2 was based on replicating the procedure used for clip 1. Again the highest score was achieved by appropriate answers stating that the boys should not go with the man in his van, and that they should contact an adult. As for Clip 1, high marks were only achieved if valid reasons for their responses were given.

For the two general questions on stranger danger at the end of the session, points were awarded if a stranger was described as someone they don't know, in addition to giving an explanation as to why they would not interact with them. A maximum of 2 points was available for each question. Importantly, across all questions, the higher the score the more appropriate and detailed the responses.

As this was a bespoke, newly devised task and scoring system information concerning validity is not available. However, the reliability of the coding of participant responses was explored. Using transcripts of the sessions, 10% of the data were independently coded by a second experimenter who was blind to the participant group and to the aims of the study. The scoring of this portion of the transcripts showed a significant positive correlation between scorers $r(16)=.81, p<.01$ therefore indicating that the devised scale was applied reliably.

Results

Quantitative Analysis:

Using the percentage of appropriate responses given by each participant we explored the mean 'overall score' to investigate group differences in awareness of stranger danger and

appropriate social interactions. An independent samples t test explored the effect of Group (WS, TD) on awareness of stranger danger. The results indicated that individuals with WS gave significantly less appropriate answers than the verbal ability matched typically developing sample, even though they have had significantly more years of interacting within a social environment $t(30)=4.70, p<.001$. Specifically, 27% of the answers given by individuals with WS were deemed to show appropriate knowledge and awareness of issues relating to interactions with unfamiliar adults and aspects of stranger danger (although there was a great deal of individual variability as shown by a standard deviation of 21% accuracy and scores ranging between 0-64 for the appropriateness of answers). The typically developing matches showed a mean appropriate response rate of 60% (standard deviation 17%; range 21-86) indicating the provision of, on average, significantly more awareness and expression of appropriate issues.

Qualitative insights and emerging themes:

As well as the statistical analysis provided above, for this study the insights provided by participants in the transcripts are particularly informative and reveal the level of understanding of individuals with WS and those developing typically. For example, when asked in clip one what the boy should do and whether he should leave to find the dog with the unfamiliar man, an 8-year old female with WS responded “*he can, he should find his dog*” (talking about the boy going and finding the dog) and asked why the same child responded “*I think because he’s lost him*” (linking this to the strange man and helping the man) indicating a lack of awareness of the possible danger related to going with this stranger. Similarly a 15-year old male with WS responded that “*I think the boy should try and help the man find his dog.*” (the individual was unable to provide an adequate explanation for this). These

examples show a lack of understanding of the possible dangers, especially when compared to the awareness shown by typically developing children, such as an 8-year old boy who responded “*no because he might be tricking him*” when asked if the boy should leave in the same question / clip and a 6-year old girl who said “*no he shouldn't help because he's a stranger*”. Further qualitative insights into participant responses are provided in the Appendix B.

To attempt to encapsulate the qualitative nature of the responses we applied a thematic analysis across participant groups. We followed the methodology proposed by Braun and Clarke (2008) to use a flexible approach to identifying emerging patterns of responses given by the participants in relation to the vignettes (see Patton, 1990). This method allowed us the opportunity to try to capture the key issues evident for all individuals when making interactional decisions of the nature used here. We used the transcripts for all questions and all individuals, irrespective of diagnosis, to explore emerging themes as summarised in Figure 1. This Figure shows the final thematic map of the main themes extracted from the transcripts. Theme I related to the awareness that familiarity might impact upon the nature of the interaction – therefore whether or not the characters were known to each other or whether they were unfamiliar to each other. Participants who showed a level of awareness regarding familiarity often qualified their responses with statements concerning familiarity, evaluation of familiar / unfamiliar people and a knowledge that the level of familiarity will modulate the nature of the interaction (for example allowing for decisions regarding safety to be made). More individuals developing typically showed this awareness of familiarity evaluation in guiding their decisions than those with Williams syndrome.

Theme II was more frequently shown by participants with Williams syndrome than those developing typically. This theme relates to the inappropriateness of decisions made regarding the stranger and the interaction between characters in the vignettes. Individuals who showed this theme may couple this with a lack of emotional interpretation of the situation or how the characters might feel. The interpretation was often very 'matter of fact' regarding whether the character should help or interact with the unfamiliar adult, lacking emotional awareness of their interpretation. Often this lack of emotional reaction to the situation would indicate that they were unsuspecting that someone might trick the character into behaving in a certain manner. This would be interesting to follow up in relation to theory of mind ability in the future (we will discuss this further in the Discussion Section). Those individuals who showed a lack of awareness or suspicion and an inappropriate response to the nature of the interaction were those who gave responses that would place the characters in a situation that vulnerable to manipulation and danger.

Theme III was more frequently evident for the participants developing typically than those with Williams syndrome. These individuals showed that they understood the nature of the interaction and the precautions that needed to be taken. There was a clear sense of not only an emotional evaluation of the situation (especially in terms of trust) but also a situational evaluation of what was occurring in the vignette. Individuals who showed this theme in their responses would also give information about the setting and why that might effect decision making (e.g. the park setting). In the majority of examples of this theme the participants showed that the characters should use appropriate adults of support when making their decisions and seeking help; for example saying that the character should ask their mother or a friend what to do.

Relationship between Stranger Awareness and everyday Social Skills

To explore in more detail the relationship between an expression of awareness regarding stranger danger and everyday social capabilities we correlated the overall score obtained from converting the transcripts into accuracy information (quantitative data above) and specific socially relevant subscales of the Strengths & Difficulties Questionnaire (SDQ; Goodman, 2001) as completed by parents. We were particularly interested in the 'Peer Problems Scale' and the 'Prosocial Behaviour Scale'. A lower Total Score on the SDQ is more desirable and parents scored all individuals with WS above the cut off for 'normal' social competence (a total difficulties score between 0-11 indicates 'normal' social skills) implying that all individuals in the sample had some problems with their social behaviour. The mean score for the group was 21.1 (ranging 13-25¹). For peer problems a higher score indicated more abnormal behaviour and thus we predicted that this might be correlated with problems making appropriate social evaluations of trust and approach (predicting a negative correlation between score on the peer problems scale and the awareness of stranger danger). The mean score for peer problems was 6.1 (range 3-9; 0 rated as 'normal' 2 individuals scored as 'borderline', 14 were rated as 'abnormal') and a significant negative correlation was evident as individuals with greater abnormality of peer relationships (a higher peer problems total) scored lower for their awareness of stranger danger $r(16)=-.75, p<.05$. In terms of prosocial behaviour a higher score is more favourable and indicates 'normal' prosocial behaviour. Therefore, we predicted that there would be a significant positive correlation between appropriate prosocial behaviour and awareness of stranger danger. The mean score for the prosocial behaviour subscale was 6.5 (range 3-10; 6 rated as 'normal', 6 rated as 'borderline')

¹ On the SDQ a total difficulties score between 12-15 is considered 'borderline' and a score above 16 (max 40) indicates 'abnormal' social behaviours. In the current sample 2 individuals with WS were rated as borderline and 14 were rated as showing abnormal social behaviours. There was a trend for a significant negative correlation between Total Difficulties score on the SDQ and awareness of stranger danger issues on the video vignette task $r(16)=-.45, p=.07$.

4 'abnormal') and the relationship between prosocial behaviour and awareness of stranger danger showed a trend towards significance $r(16)=.41$, $p=.07$. Some caution is required for these exploratory correlations due to relatively small sample size ($n=16$) and further work with a larger sample size is required to explore this relationship further in WS. There was no significant relationship between chronological age and stranger danger awareness in the WS group ($p=.58$) and there was a trend towards a significant relationship between overall IQ and stranger danger awareness ($p=.082$ with higher levels of IQ associated with high levels of awareness and the ability to express this awareness).

Discussion

There is a need to understand how individuals with WS adapt their social behaviour to match the situation they are in. Parents report concern regarding the interactions of their children with unfamiliar others. Therefore the aim of the current study was to use a video vignette task to probe stranger danger awareness in young people with the disorder. Previous research exploring increased approach has relied on rating scale measures (predominantly with adults who have the disorder) and the only other skill that has been *directly* related to approach rating is basic emotion perception (e.g. Porter et al., 2007). In the current study we aimed to explore the value of qualitative insights into the awareness that individuals with WS could show in their evaluation of interactions with unfamiliar people. We therefore present evidence using a novel method to explore social interaction desires and awareness in young people with the disorder.

This study is the first to use a video vignette task to explore awareness of stranger danger in individuals with WS and those who are developing typically. Given evidence of heightened sociability and experimental reports of increased approach to unfamiliar people (e.g. Jones et al., 2000; Frigerio et al., 2006) this timely investigation tackles a question of extreme importance to a socially vulnerable group (e.g. see Jawaid et al., 2012, for issues of vulnerability in WS relating to social profiles). The results (both quantitative and qualitative in nature) suggest that many individuals with WS, here between the ages of 8 and 17 years, show a lack of awareness of the possible dangers of interacting with strangers. The thematic analysis specifically showed that when individuals gave responses that indicated interaction would be acceptable with the stranger they showed vulnerability and a lack of appropriate reaction to the situation (e.g. a lack of awareness that the child in the scenario should be wary or uncertain). The video vignettes enabled the study to probe these issues using clips of actors engaged in interactions in situations with which the participant may be familiar (or have had previous experience, e.g. a playground), as opposed to previous research that has required participants to rate unfamiliar faces using a Likert scale to obtain approach ratings (e.g. Jones et al., 2000).

Using insights from parents we were able to provide the preliminary suggestion of a link between difficulties making appropriate stranger danger awareness judgements (and expressing those judgements) and general everyday social behaviours (as measured by the SDQ, which has previously been widely used with typical and atypical populations as a measure of everyday social capabilities). Those individuals with WS who showed less awareness of stranger danger were also those who experienced more peer relation difficulties and who generally showed less adequate prosocial behaviour (as rated by the parents). Therefore, follow up research is required to confirm this preliminary suggestion that

problems evaluating stranger danger (and expressing the awareness of stranger danger) fall within a bigger picture of generalised problems functioning socially. Further research is required with a larger sample of individuals with WS to explore this relationship in more detail and to investigate how much of this effect is also related to overall level of intellectual functioning.

There was large variability in the responses given by individuals with WS as well as those developing typically (although slightly less so in the latter of these groups). Indeed previous research with typically developing children has suggested that the concept and awareness of ‘stranger danger’ continues to develop through childhood with evidence of individual variability and some children having problems up to 8 or 10 years of age (e.g. Moran et al., 1997). Therefore, while the aim of the current study was not to explore developmental changes in stranger danger awareness in typical and atypical development, further consideration of these age-related patterns might be the focus of future work. In the current study, while some of the variability in the WS group may relate to the previously mentioned intellectual capabilities of the cohort and how IQ varies within this syndrome (e.g. evidence of mild – moderate intellectual difficulties, Searcy et al., 2004), this variability requires further consideration. This is especially the case where previous research has provided a suggestion of within-syndrome variability of social behaviours in WS (Little et al., 2013). Exploring this within-syndrome variability may be particularly useful in identifying individuals who would benefit from targeted interventions for their social skills and as indicated here, for their awareness of stranger danger issues. While this issue would also be of interest when studying the typical matches given increased vulnerability associated with WS this is of heightened importance to that group. The current findings (both of a qualitative and quantitative nature) therefore have the ability to provide important preliminary awareness

concerning possible practical / applied issues that could inform social skills training programmes in the near future.

The participants with WS in the current study were children and adolescents who were old enough to have had significant experience interacting with familiar and unfamiliar people, but who would not yet have faced the full challenges of adulthood. Given that individuals of the age studied here show a lack of awareness (on average) of issues relating to stranger danger this may imply that this age range should be targeted before other challenges relating to adulthood and increased independence occur. The findings are particularly important given the suggestion that individuals with WS suffer peer relation problems and often interact with adults rather than peers. If they are not evaluating those adults in an appropriate manner this may make them particularly vulnerable. Furthermore, if individuals with WS who would benefit from social skills training related to interactions with unfamiliar people can be identified in childhood or adolescence this may mean that those individuals are subsequently protected from the high levels of adult social isolation and anxiety associated with WS. For example, it has been suggested that over 70% of adults with the disorder experience social isolation (Davies et al., 1998) and only about 30% are able to be independent enough to hold down employment in adulthood (Davies et al., 1997). Therefore, in summary, identifying and targeting these behaviours early could improve the longer term outcome for adults with the disorder. In the current study the scenarios were all set up in a way that meant that the 'correct' response would be to not interact with the unfamiliar adult. However, with increasing age we will interact with more new people in our everyday lives and therefore in future explorations of this nature it would be important to set up scenarios that vary in whether an approach / avoidance decision is more or less appropriate. This would allow us to capture the subtle nature of the social decision making process, especially in those older

individuals (older adolescents / adults) who might need to appropriately judge whether to approach or avoid an individual depending on other situational factors.

It is important to note possible limitations of the current study, for example the relatively small sample size coupled with a high level of individual variability (e.g. see Little et al., 2013) means that further work is needed to capture information on those individuals who do struggle with these concepts and those who do not. Qualitative information / data as provided in the current study would be highly beneficial. It is not adequate to say that all individuals with WS will struggle with the notion of stranger danger and more work is required to capture the individual variability and to perhaps map that more directly onto general social functioning and intellectual capabilities. This is especially important where a relationship has been provided by the current study but caution is required due to the small sample size / lack of power. Although the use of parent insights through completion of the Strengths and Difficulties Questionnaire is a valuable secondary insight into social behaviours in everyday life, additional insights from a variety of sources would be beneficial. Indeed, for those individuals who are old enough self-report measures may be used to gain an insight into everyday capabilities versus struggles. This is also relevant to capturing the variance seen within the typically developing sample. The relationship between social approach behaviour, stranger danger awareness and emotional understanding clearly warrants further exploration. It would be critical to explore the relationship between the current findings and Theory of Mind in both typical development and in WS. Some of the individuals with WS may have struggled to place themselves within the situation and understand another person's perspective in the task that we used as it is known that some individuals with WS have Theory of Mind deficits (e.g. Tager-Flusberg & Sullivan, 2000). This issue definitely warrants future consideration. However, the current study does provide a valuable insight into

stranger danger awareness in WS using a new method that does not rely upon complex rating scales or solely on parental reports of social behaviour as previously used to explore social approach within the disorder.

To conclude, following up the study set out here with future investigations of social behaviours associated with WS will allow us to consider how best to feed these qualitative and quantitative insights into social skills training programmes. An evaluation of how existing interventions and social skills training programmes do and do not fit the needs of individuals with WS is particularly necessary. Identifying programmes that fit the very *specific* needs of individuals with WS is important as we can then consider how best to aid teaching / awareness of stranger danger in this group of individuals who may be particularly vulnerable in social settings due to their combined hypersocial personality and reduced intellectual capabilities.

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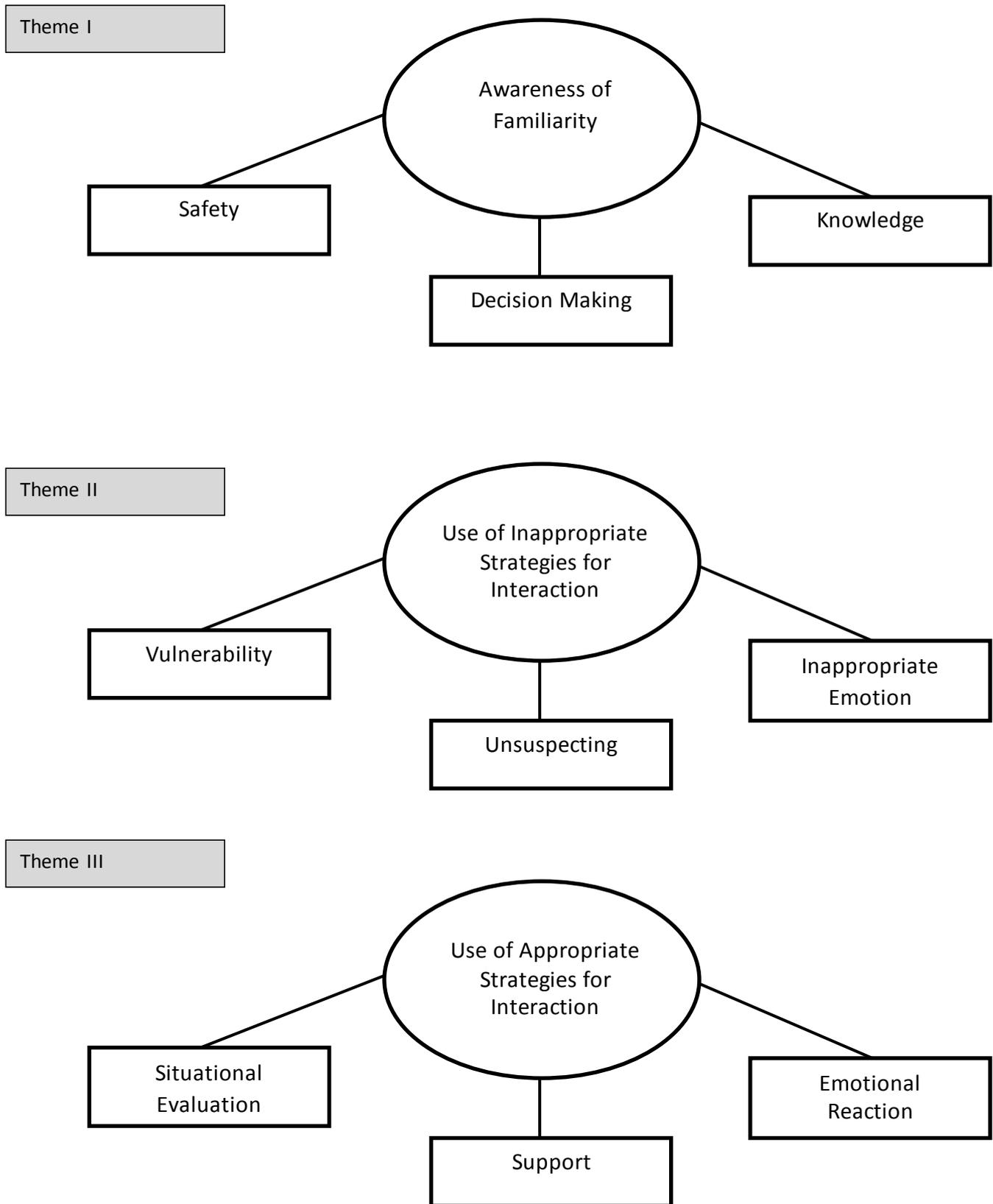
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Figure 1: Thematic Map



Appendix A

Information on scoring of the responses given by participants for Clip 1. This method was replicated for Clip 2.

Question	Scoring Protocol	Examples
<i>What do you think the boy should do?</i>	<p>2 point – appropriate response with justification</p> <p>1 point – appropriate response without any justification</p> <p>0 points – inappropriate response including ‘don’t know’</p>	<p>“he should seek his mums advice. He seems very young and she isnt far away” (2 points)</p> <p>“I think he should ask his mum” (1 point)</p> <p>“find the man’s dog quickly” (0 points)</p>
<i>Do you think the boy should go and help the man find his dog? Followed by why / why not?</i>	<p>2 point – appropriate response with justification</p> <p>1 point – appropriate response without any justification</p> <p>0 points – inappropriate response, including ‘don’t know’</p>	<p>“no he should go to a grown up, it’s not safe to go with the man” (2 points)</p> <p>“no he shouldnt” (1 point)</p> <p>“I dont know, he might like the dog” (0 point)</p>
<i>What would you do if you have been out playing or relaxing in the park and someone you didn’t know came and asked you to go with them to help them?</i>	<p>2 point – appropriate response with justification</p> <p>1 point – appropriate response without any justification / correct justification</p> <p>0 points – inappropriate response, including ‘don’t know’</p>	<p>“I would say no, just in case it is someone who is not a very nice person” (2 points)</p> <p>“I wouldn’t go with them, I might carry on playing” (1 point)</p> <p>“It might depend how I feel that day” (0 points)</p>

Appendix B

Example answers provided by participants in each group showing either awareness, or a lack of awareness, of stranger danger issues. These answers were provided in response to Clip 2.

The child's chronological age is provided in brackets after each quote.

<p>Williams syndrome</p> <p>Clear awareness of issues</p>	<p><i>"He should go and check if the mum is ok and then try and see what's wrong with her rather than trying to get help from the man"</i> (age 13)</p> <p><i>"You dont talk to him."</i> (age 12)</p> <p><i>"I think the boy should call the police and see if there is an accident, if the man's joking the police should go and arrest him"</i> (age 15)</p>
<p>Williams syndrome</p> <p>Lack of awareness</p>	<p><i>"I would say I would just quickly check if your right or not because you have got to trust a man ..."</i> (age 12)</p> <p><i>"Go with the man"</i> (age 9)</p> <p><i>"They should go with the man... because, in case he doesn't know the direction"</i> (age 9)</p>
<p>Typical development</p> <p>Clear awareness of issues</p>	<p><i>"Should ride their bikes back home without going in the car with the man and see if their mum is in trouble"</i> (age 6)</p> <p><i>"I don't think I should go with the man, cos he could be someone whose not very nice."</i> (age 7)</p> <p><i>"Find a different way to check their mum's ok it's a strange man in a white van"</i> (age 11)</p>
<p>Typical development</p> <p>Lack of awareness</p>	<p><i>"Go with him and try and help his mum"</i> (age 6)</p> <p><i>"Yeah go with him. Because the mum's had an accident..... because his mum got runover"</i> (age 7)</p> <p><i>"Go with the man So he can see their mum"</i> (age 6)</p>