



Examining reports of mental health in adults with Williams syndrome

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ABSTRACT

Prior research suggests that individuals with Williams syndrome (WS) have a disposition towards anxiety. Information regarding this is typically derived from parents and carers. The perspectives of the individuals with WS are rarely included in research of this nature. We examined the mental health of 19 adults with WS using explicit (psychiatric interview) and implicit (modified Stroop task) measures and compared informant (parents/carers) and respondent (adults with WS) reports of psychiatric symptoms. Informants and respondents both reported more symptoms of anxiety ($n = 7-9$) than depression ($n = 2$). Strong positive correlations were found between informant and respondent reports of symptoms of mental health problems. Compared to informants, respondents reported significantly more symptoms overall and somewhat more symptoms of anxiety. Results from the Stroop task indicated that the adults with WS were more vigilant to anxiety-related words than to depression-related words. The adults with WS provided reliable information regarding their mental health, thus providing further evidence that anxiety is part of the behavioural phenotype of the syndrome.

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1. Introduction

Williams syndrome (WS) is a genetic disorder caused by a deletion of genes on one copy of chromosome 7 (Ewart et al., 1993). It is associated with mild-to-moderate intellectual disability (ID) and a distinctive pattern of cognitive abilities that comprises weaknesses in aspects of spatial and number cognition and strengths in receptive vocabulary and face processing (Ansari et al., 2003; Farran, Jarrold, & Gathercole, 2003; Howlin, Davies, & Udwin, 1998; Paul, Stiles, Passarotti, Bavar, & Bellugi, 2002). Many individuals with WS display a behavioural phenotype that includes hypersociability, impulsivity and emotional difficulties (Davies, Udwin, & Howlin, 1998; Einfeld, Tonge, & Florio, 1997; Udwin & Yule, 1991). One of the most notable features of this behavioural phenotype is a disposition towards anxiety, with authors reporting significantly higher rates of anxiety among individuals with WS compared to those with Autism, Prader–Willi syndrome, ID of mixed/unknown aetiology and typically developing children (Dimitropoulos, Ho, Klaiman, Koenig, & Schultz, 2009; Dykens, 2003; Einfeld et al., 1997).

In recent years, research groups have sought to categorise the emotional and behavioural difficulties of individuals with WS in terms of diagnosable psychiatric disorders (e.g., Cherniske et al., 2004; Kennedy, Kaye, & Sadler, 2006; Leyfer, Woodruff-Borden, Klein-Tasman, Fricke, & Mervis, 2006; Stinton, Elison, & Howlin, 2010). These studies have supported prior assertions of high rates of anxiety, indicating that up to 54% of participants meet ICD/DSM diagnostic criteria for anxiety disorders (e.g., agoraphobia, generalised anxiety disorder and specific phobia). In addition to anxiety, a small number of

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studies has reported on affective disorders (e.g., bipolar disorder, depression and hypomania) and psychotic disorders (e.g., schizophrenia), indicating rates of 3–25% and 1–2%, respectively (e.g., Cherniske et al., 2004; Dodd & Porter, 2009; Kennedy et al., 2006; Stinton et al., 2010).

In many cases, information regarding the emotional difficulties and psychological states of individuals with WS has been derived from third party reports (i.e., “informants”), often with no attempt made to obtain information from the individuals with WS themselves (i.e., “respondents”). For example, Dodd and Porter (2009) state that ‘as is standard procedure when assessing an intellectually impaired population, only caregivers were interviewed’ (p. 95). Yet, there is a wealth of evidence indicating that individuals with ID (especially those with mild-to-moderate ID) are able to understand and provide meaningful information about their emotional states and experiences (e.g., Cuthill, Espie, & Cooper, 2003; Deb, Thomas, & Bright, 2001; Douma et al., 2006; Emerson, 2005; Lindsay et al., 1994; Matson, Kazdin, & Senatore, 1984). In addition, disregarding the perspective of the individuals themselves is likely to result in underdiagnosis of mental health problems due, for example, to diagnostic overshadowing and the necessity for information that can only be derived from respondents such as autonomic symptoms (Bramston & Fogarty, 2000; Moss, Prosser, Ibbotson, & Goldberg, 1996; Reiss, Levitan, & Szyszko, 1982). In the case of people with WS, while their range of abilities is broad, the vast majority of individuals have a mild ID and functional communication skills (Howlin, Elison, & Stinton, 2010; Martens, Wilson, & Reutens, 2008). Further, several studies have demonstrated that interviews and questionnaires about emotional difficulties can be successfully carried out with both adults and children with WS (e.g., Cherniske et al., 2004; Dykens, 2003; Freeman, Williams, Farran, & Brown, 2009; Kennedy et al., 2006; Stinton et al., 2010). These studies have highlighted the importance of including both informant and respondent reports. For example, using the revised Fear Survey Schedule for Children (Ollendick, King, & Frary, 1989), Dykens (2003) found that individuals with WS reported significantly more fears than their parents and Stinton et al. (2010) reported that anxiety disorders were more commonly derived from psychiatric interviews conducted with adults with WS than with their parents/carers, with the opposite true for depressive disorders. While limited agreement between informant and respondent reports of difficulties is common, the simple presence of this difference cannot be assumed to indicate that respondent reports are less reliable than informant reports. Differences are likely to reflect their different perspectives.

The finding that different individuals provide information that might result in very different interpretations of symptoms presents a considerable challenge in research and clinical practice. To better understand this it is necessary to examine mental health using alternative methodologies. One way in which mental health can be examined experimentally is through the use of emotional Stroop tasks. In the original format of this task, participants were presented with a series of items (words or meaningless letter strings such as XXXX) printed in various colours and were asked to name the colour that the stimuli were printed in while attempting to ignore the stimulus itself. Stroop (1935) found that participants were slower to name items that were incongruent (i.e. the word red written in blue) than those that were congruent (i.e. the word green written in green). This effect has been consistently observed in both typically developing individuals and individuals with ID (Das, 1970; Ellis, Woodley-Zanthos, Dulaney, & Palmer, 1989; Williams, Mathews, & MacLeod, 1996). More recent research has demonstrated that it is not only incongruent word/colour items that produce interference. Colour naming is significantly slower for clinically relevant emotional words (e.g., scared) than for neutral words (e.g., square) among clinical groups (i.e. those with anxiety or affective disorders) but not among control groups who do not have the given disorder (Mathews, Mogg, Kentish, & Eysenck, 1995; Mitterschiffthaler et al., 2008). As such, the Stroop task presents an opportunity to measure emotional problems implicitly and hence without the sorts of biases that are evident in reports obtained via interviews and questionnaires.

The present study examines the mental health of adults with WS via both explicit (psychiatric interview) and implicit (Stroop) measures. The Stroop task included anxiety-related (e.g., “alone”), depression-related (e.g., “crying”), and neutral words (e.g., “paper”). Convergence between explicit and implicit measures completed by the adults with WS would indicate that their reports are accurate, whereas a significant degree of divergence would suggest that their reports are inaccurate. These reports by the adults with WS were also compared to informant reports by their parents or carers. Convergence between respondent and informant reports would further support the adults with WS’s ability to provide meaningful information about their own emotional states and experiences.

2. Methods

2.1. Recruitment

Potential participants were identified via the Williams Syndrome Foundation’s database of research participants ($n = 110$). Inclusion criteria were: the individual with WS was aged 18 years or older, their diagnosis had been confirmed by genetic testing, they had a parent or other person who knew them well, agreement to participate from both parties. Exclusion criteria were: inability to read simple words and colour blindness.

2.2. Participants

The response rate to recruitment letters was 28.2% ($n = 31$). Of these families, 12 (38.7%) were excluded from the study for the following reasons: the adult with WS was not able to provide sufficient information to questions regarding their mental health ($n = 2$), the adult with WS was unable to complete the Stroop task ($n = 5$), the family withdrew before data collection

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