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Siblings of children with Williams syndrome: Correlates of psychosocial adjustment and sibling relationship quality

Katie Cebula[a,*], Amanda Gillooly[a], Laura K. Coulthard[b], Deborah M. Riby[b], Richard P. Hastings[c,d]

[a] Moray House School of Education and Sport, University of Edinburgh, Edinburgh, UK
[b] Department of Psychology, Durham University, Durham, UK
[c] Centre for Educational Development, Appraisal and Research, University of Warwick, Coventry, UK
[d] Centre for Developmental Psychiatry and Psychology, Department of Psychiatry, School of Clinical Sciences at Monash Health, Monash University, Melbourne, VIC, Australia

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ABSTRACT

Background: Previous research has examined adjustment in parents of children with Williams syndrome (WS), but little is known about sibling outcomes.
Aims: To explore sibling adjustment and relationship quality, and their demographic, psychological and behavioural phenotypic correlates from the perspective of caregivers and siblings in families of children with WS.
Methods and procedures: Forty-one caregivers of children with WS participated in this questionnaire study on the adjustment and relationship quality of the siblings. In 31 of these families, self-report data were also provided by the siblings themselves. Data were also gathered on potential correlates, including anxiety and social functioning in the child with WS, caregiver mental health, and sibling social support.
Outcomes and results: Sibling adjustment was similar to population norms, though significantly increased caregiver-reported emotional difficulties were found. Siblings reported greater behavioural, emotional and relationship difficulties than caregivers perceived them to have. Some significant associations were found between the behaviour of the child with WS, sibling behaviour problems and sibling relationship quality.
Conclusions and implications: A picture of relatively positive sibling adjustment and relationships emerged, but findings of individual differences and some emotional difficulties emphasise the need for an individualised approach to support in families of children with WS.

What this paper adds

Almost all family studies of Williams syndrome to date have focused on parents. Our study adds to the Williams syndrome family research literature by exploring adjustment and relationship quality in the neurotypical siblings, as well as the demographic, psychological and behavioural phenotypic correlates of sibling outcomes. It is also, to our knowledge the first study to include self-report data from siblings themselves. The findings add to knowledge of support needs in families with a child with WS: positive adjustment was found in siblings overall, emphasising the need to avoid taking a pathologising approach. However, there were some

* Corresponding author.
E-mail address: katie.cebula@ed.ac.uk (K. Cebula).

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discrepancies in sibling and caregiver views of sibling adjustment and relationships, emphasising the importance of practitioners listening to all family members. There was also variability in sibling adjustment, with poorer outcomes associated with caregiver symptoms of depression, and conflict in the sibling relationship, indicating the need for an individualised approach to support.

1. Introduction

A simplistic view of children with developmental disabilities as a universal, inevitable and unidirectional cause of family stress has been challenged (Hastings, 2016; Stoneman, 2005). In particular, any negative impact on neurotypical siblings (hereafter, 'siblings') is small overall, with prevalence of clinically-significant emotional difficulties generally comparable to that of the general population (Hayden, Hastings, Totsika, & Langley, 2019; Rossiter & Sharpe, 2001; Tudor, Rankin, & Lerner, 2018). Indeed, the experience of growing up with a child with developmental disabilities is associated in some studies with positive experiences and outcomes for many siblings (Cianfaglione, Hastings, Felce, Clarke, & Kerr, 2015; Macks & Reeve, 2007; Verté, Roeyers, & Buysse, 2003). Nevertheless, there is evidence that, at least some siblings may experience difficulties, at some points across the lifespan (Lobato et al., 2011; Neece, Blacher, & Baker, 2010; Shivers, Deisenroth, & Taylor, 2013). It is therefore important to identify siblings who might require higher levels of support. Alongside demographic factors and psychological variables, the diagnosis of the child with developmental disabilities has been recognised as being associated with family outcomes and support requirements (Cianfaglione et al., 2015; Hodapp, Wijma, & Masino, 1997; Kaminsky & Dewey, 2001; Love, Richters, Didden, Korzilius, & Machalicek, 2012; Usher, DaWalt, Greenberg, & Mailick, 2018). However, there has been little research to date in relation to siblings of children with the developmental disability Williams syndrome (WS), the focus of the present study.

Research into siblings of individuals with developmental disabilities has tended to rely on family systems theories (Cridland, Jones, Magee, & Caputi, 2014; Minuchin, 1985) and bioecological systems theories (Bronfenbrenner, 1979, 2005; Kovshoff, Cebula, Tsai, & Hastings, 2017; Saxena & Adamsons, 2013; Stoneman, 2005). These frameworks are helpful in understanding the complex mechanisms through which sibling outcomes may be influenced by the experience of growing up with a child with developmental disabilities. However, when exploring outcomes for siblings of children with genetic intellectual disabilities, some consideration of behavioural phenotype theory may also be helpful. According to this approach (Dykens, 1995; O'Brien, 1993) particular genetic syndromes are associated with an increased likelihood of an individual displaying specific patterns of behavioural or cognitive development. Hodapp (1997) refers to this as 'direct effects'. He also describes 'indirect effects': the idea that a particular developmental disability may evoke specific family experiences and outcomes. Seeking to understand how specific aspects of a behavioural phenotype are associated with sibling relationships and outcomes may therefore offer an additional approach to identifying family support requirements.

WS is caused by the hemizygous deletion of around 28 genes on chromosome 7q11.23 (Tassabehji, 2003), and has a prevalence of approximately 1 in 20,000 (Korenberg, Bellugi, Salandanan, Mills, & Reiss, 2003). It is associated with mild to moderate intellectual disability, and a range of physical and medical features (Bellugi, Lichtenberger, Jones, Lai, & St. George, 2000; Morris & Mervis, 2000). It is also associated with a characteristic cognitive profile which includes relative strengths in verbal ability (Bellugi et al., 2000), although there is substantial individual variation (Jarrold, Baddeley, & Hewes, 1998).

Two socio-emotional aspects of the WS behavioural phenotype may be particularly important in terms of sibling relationships and adjustment. First, WS is associated with a characteristic social phenotype which includes a strong interest in people ('hypersociability'), high levels of affective expression, and a greater tendency to approach strangers when compared to neurotypical children or children with other developmental disabilities of a similar age (Doyle, Bellugi, Korenberg, & Graham, 2004; Hauser-Cram, Howell-Moneta, & Young, 2011; Järvinen-Pasley et al., 2008; Jones et al., 2000; Riby, Kirk, Hanley, & Riby, 2014; Weisman et al., 2015). Additionally, young children with WS show higher levels of empathic concern than developmentally-matched controls (Plesa Skwerer & Tager-Flusberg, 2016). Reported social difficulties include problems establishing and maintaining friendships, social isolation, and atypicalities in social cognition and interpretation of social cues (Davies, Udwin, & Howlin, 1998; Riby, Hancock, Jones, & Hanley, 2013; Weisman et al., 2017).

Second, individuals with WS are susceptible to heightened anxiety, both when compared to neurotypical individuals and those with some other developmental disabilities, with specific phobia the most commonly reported anxiety type (Dykens, 2003; Leyfer, Woodruff-Borden, Klein-Tasman, Fricke, & Mervis, 2006; Leyfer, Woodruff-Borden, & Mervis, 2009; Rodgers, Riby, Janes, Connolly, & McConachie, 2012; Royston, Howlin, Waite, & Oliver, 2017).

Despite this characteristic behavioural phenotype, to date, there has been relatively little research on its association with outcomes and relationships in siblings of children with WS. Almost all of the WS family studies to date have focused on parent-reports of either their own wellbeing or of family functioning (Ashworth, Palikara, & Van Herwegen, 2019; Brawn & Porter, 2014; Lanfranchi & Vianello, 2012; Leyfer et al., 2009; Papaeliou et al., 2012; Scallan, Senior, & Reilly, 2011; Van Lieshout, De Meyer, Curfs, & Fryns, 1998). Whilst a few studies have taken a behavioural phenotype 'indirect effects' approach, this has again been parent-focused (Fidler, Hodapp, & Dykens, 2000; Sarimski, 1997). Few have gathered data about siblings and none, to our knowledge, have included sibling self-report data.

Leyfer et al. (2009) did focus on siblings of children with WS, examining prevalence of anxiety disorders, using a parent-report structured diagnostic interview. They found that siblings had similar prevalence of anxiety disorders as children in the general population, with the exception that they had significantly higher prevalence of specific phobia, than has been found in some studies of neurotypical children. No significant associations were found between sibling anxiety and a number of factors, including sibling

demographics, maternal anxiety, and the anxiety, behaviour, and intellectual functioning in the children with WS. However, as Leyfer et al. focused on anxiety, broader sibling outcomes (e.g. behaviour problems) and sibling relationship quality were not explored. A later qualitative study on parent views of family experiences found the majority of parents reported some positive impact of children with WS on their siblings, such as helping siblings to be more caring (Scallan et al., 2011).

The present questionnaire study is, to our knowledge, the first to gather quantitative data on sibling behavioural and emotional adjustment, and on sibling relationships in WS families, and to include self-reports from siblings. Self-report data are important because there are often discrepancies between parent and sibling reports of sibling adjustment and relationship quality in developmental disability research (Rankin, Tomeny, & Barry, 2017; Tsai, Cebula, & Fletcher-Watson, 2016). The study drew on family systems and behavioural phenotype theories to examine the following questions: (1) What is the psychosocial adjustment of siblings of children with WS, according to parent- and self-report? (2) To what extent are sibling psychosocial adjustment and relationship quality associated with demographic variables, psychological variables, and the behavioural phenotype of the child with WS?

In terms of the specific variables associated with sibling adjustment and relationship outcomes, we were guided by the Siblings Embedded Systems Framework (Kovshoff et al., 2017) and examined: demographics; behaviour, social functioning and anxiety in the child with WS; caregiver mental health; sibling relationship quality; and social support perceived as available by the sibling. In considering how the WS behavioural phenotype might be associated with sibling adjustment and relationships, it may be that hypersociability in WS will be associated with a warm sibling relationship, as a result of positive affect towards the sibling (see Weisman et al., 2015). In relation to the heightened anxiety in WS, it is possible that this will be associated with higher sibling anxiety, as a result of transmission of anxiety via social contagion (see Serra Poirier, Brendgen, Vitaro, Dionne, & Boivin, 2017). However, given previous reports of relatively normative anxiety levels in siblings of children with WS (Leyfer et al., 2009), significantly raised sibling anxiety was not expected.

2. Methods

2.1. Participants

Forty-one families with a child with WS and a neurotypical sibling, from four English-speaking countries (UK: N = 37; USA: N = 2; Australia: N = 1; Canada: N = 1) participated in this postal questionnaire study. The 41 caregivers (39 mothers, 1 father and 1 grandmother) each completed measures about the child with WS in their family and about the child's neurotypical sibling. In 31 of these families, the siblings also completed self-report measures. As can be seen in Table 1, the mean (SD) age of the 41 siblings for whom there was caregiver data was 10.58 (3.76) years (range: 4.32–17.93 years), and the majority were older than the child with WS. Forty families reported on a full-sibling relationship, and one on a half-sibling relationship (living together full-time). In terms of ethnicity, all family members were white. For the 31 siblings for whom there was self-report data, the mean (SD) age was 11.69 (3.16) years (range 5.22–17.93 years), with 7 male and 24 female siblings.

In the full sample, the mean age of the children with WS was 9.78 years (range: 4.02 years–17.70 years), with the majority being boys and having mild/moderate intellectual disability. Parents reported that most of the children with WS were diagnosed within the first two years of life, with their diagnosis confirmed with fluorescent in situ hybridisation testing (N = 31) or chromosomal microarray (N = 9), (unknown method: N = 1). Health issues were common amongst the child with WS as expected, with hypersensitivity (e.g. to noise) (N = 38), cardiac problems (N = 26), sleep difficulties (N = 24), mobility difficulties (N = 20), visual impairment (N = 19), slow growth/feeding problems (N = 17) and dental anomalies (N = 21) the most common.

2.2. Procedures

Ethical approval was obtained from the research ethics committee at the first author's institution prior to commencement of the study. Families were included if they had a child with WS and another neurotypical sibling both between 4 and 18 years old, who were both living at home together with the caregiver respondent for the majority of the time. For consistency, in families with more than one neurotypical sibling, the one whose age was closest to that of the child with WS was asked to participate.

Families were recruited via social media, with the assistance of the Williams Syndrome Foundation charity, and via emails to family support charities and schools about the research. Families were supplied with caregiver and sibling project information and consent sheets, and informed consent was obtained from the caregiver and the participating sibling. Caregivers were also asked, where possible (e.g. according to the child's age and receptive language ability), to confirm that the child with WS was happy for the caregiver and sibling to complete questionnaires about them. Families were then sent questionnaire packs for the caregiver and, where required, the participating sibling. Caregivers were encouraged to look through the siblings' blank questionnaire pack, to ensure that the sibling understood the instructions, but were then asked to allow the sibling to complete their pack as independently as possible. Siblings were encouraged to contact the research team via their caregiver if they required assistance in completing the questionnaire (none did so). They were provided with an envelope within which to seal their questionnaires, and they then returned this via their caregiver, to ensure the confidentiality of their responses. For the four youngest siblings who completed self-report data, the questionnaires were completed by the researcher who asked the questions orally during a home visit (these siblings lived in the UK) for another study. The remaining 27 siblings received the questionnaires in the post, and completed them themselves.

Table 1
Demographic characteristics of sample (N = 41).

Characteristic	Mean (SD) or N (%)
Sibling:	
Age (years)	10.58 (3.76)
Age compared to child with WS:	
Older/younger/same	25 (61%)/13(32%) /3 (7%)
Gender (M/F)	9/32
Gender match with child with WS	16 (39%)
Lives with caregiver respondent:	
Full-time	38 (93%)
Part-time	1 (2%)[a]
Missing data	2 (5%)
Child with Williams syndrome:	
Age (years)	9.78 (3.98)
Gender (M/F)	26/15
Lives with caregiver respondent:	
Full-time	39 (95%)
Part-time	2 (5%)[a]
Missing data	1 (2%)
Level of intellectual disability:	
Mild/moderate	31 (76%)
Severe/profound	10 (24%)
School type:	
Nursery/preschool	5 (12%)
Mainstream	15 (37%)
Special unit in mainstream	3 (7%)
Special school	18 (44%)
Caregiver:	
Education to university degree level or higher	24 (59%)
Employed	26 (63%)
Family:	
Two-parent/blended two-parent household	37 (90%)
Annual Household Income[b]	5.31 (1.80)
Managing financially[c]	1.98 (0.85)
Total no. children in family[d]	2.78 (1.28) (range 2 – 8)

[a] lives with caregiver respondent for the majority of each week.
[b] Less than £10k = 1; £10,001 - £15k = 2; £15,001 - £30k = 3; £30,001 - £45k = 4; £45,001 - £60k = 5; £60,001 - £75k = 6; £75,001 - £90k = 7; £90,001 - £100k = 8; over £100k = 9).
[c] 1 = Living comfortably; 2 = Doing alright; 3 = Just about getting by; 4 = Finding it quite difficult; 5 = Finding it very difficult.
[d] Includes all sibling relationships (full, step, and half).

2.3. Measures

2.3.1. Measures completed by caregivers and siblings

Strengths and Difficulties Questionnaire (*SDQ: Goodman, 1997*)

The SDQ is a 25-item measure assessing five constructs of behaviour: emotional symptoms, conduct problems, hyperactivity, peer problems and prosocial behaviour in children aged 4–17 years. This questionnaire is available in a parent and self-report format. Respondents rate the extent to which the item is applicable to the child, on a 3-point scale (0 to 2). A score is generated for each subscale, from 0 to 10. The participants' scores in all subscales except prosocial behaviour are combined to generate a total difficulties score. For both parent and self-report a total difficulties score of 20 or higher is classified as 'very high'. Caregivers were asked to complete SDQ forms about both the child with WS and the sibling. Although the self-report form is designed for children aged 11–17 years, it has previously been used with children as young as 8 years (Muris, Meesters, Eijkelenboom, & Vincken, 2004). In the present study, the total difficulties scale showed acceptable internal consistency (.72, .77, and .78, for the caregiver-report on the child with WS, their report on the sibling, and the sibling self-report respectively). The prosocial behaviour scale showed somewhat lower internal consistency (.73, .55, and .58, for the caregiver-report on the child with WS, their report on the sibling, and the sibling self-report respectively).

Spence Children's Anxiety Scale (*SCAS: Spence, 1998*)

The SCAS is a 38-item measure of a child's symptoms of anxiety. It is available in a parent (SCAS-P) and self-report (SCAS-C) format (aged 8 years and above). The SCAS measures six subscales: panic attack and agoraphobia, separation anxiety, physical injury fears, social phobia, obsessive compulsive, and generalised anxiety disorder. The respondent rates how often an item applies to them/their child on a 4-point scale (0–3). Possible total scores range from 0 to 114, with higher scores indicating a higher frequency of anxiety symptoms. Although there is no formal clinical cut-off for the SCAS-P, a score of 24 or above has been suggested as indicative of 'clinical caseness' (as reported in Rodgers et al., 2012). Caregivers were asked to complete SCAS-P forms about both the child with WS and the sibling. In the present study, good internal consistency was found for the SCAS-P (α = .87 and .93, for the child with WS and the sibling respectively), and for the SCAS-C (α = .85).

The Sibling Relationship Questionnaire (*SRQ: Buhrmester & Furman, 1990*)

The SRQ is available in a parent report and self-report format (aged 8 years and above). Ten items from the scale were administered in the present study, measuring two sibling relationship constructs: warmth/closeness (six items) and conflict (four items). Respondents rate the extent to which the item applies to the child and their sibling, on a 5-point scale (1–5). Possible scores were 6–30 for warmth and 4–20 for conflict. Higher scores equate to greater warmth and greater conflict in the sibling relationship. The SRQ demonstrated acceptable to good internal consistency in the present study (warmth/closeness $\alpha = .87$ and .71; conflict $\alpha = .81$ and .77, for the caregiver and self-reports respectively).

2.3.2. Measures completed by caregiver only

Demographic questionnaire

A demographic questionnaire was used to gather information about the caregivers (e.g. marital status, ethnicity, occupation and family income), the child with WS (e.g. level of intellectual disability; method and age of diagnosis; health issues) and the other children living in the household (e.g. number, age, special educational needs).

The Social Responsiveness Scale (*SRS: Constantino & Gruber, 2005*)

The SRS is a 65-item scale measuring a child's social functioning. The school-aged version of this questionnaire was utilised, which assesses social functioning in children aged 4–18 years. Respondents indicate the extent to which each item is applicable to their child on a 4-point scale (1–4). Caregivers were asked to complete SRS forms about the child with WS. The total scale demonstrated strong internal consistency in the present study ($\alpha = .89$).

Hospital Anxiety and Depression Scale (*HADS: Zigmond & Snaith, 1983*)

The HADS is a 14-item self-report questionnaire which measures symptoms of anxiety and depression (7 items each). The participant reads each statement and rates the extent to which he or she has experienced that feeling within the past week. All items are scored on a 4-point scale (0–3). The possible scores for each subscale range from 0 to 21, with higher scores indicating a higher level of perceived symptoms. In the present study, the HADS demonstrated good internal consistency (anxiety: $\alpha = .84$, depression: $\alpha = .80$).

2.3.3. Measures completed by sibling only

Survey of Children's Social Support: short version (*SCSS-SV: Dubow, Edwards, & Ippolito, 1997*)

The SCSS-SV is a 9-item questionnaire measuring a child's perceptions of social support, from their family, teachers and peers. It is an abbreviated version of the original 41-item scale (Dubow & Ullman, 1989). Respondents indicate the extent to which they believe that they receive different types of support on a 5-point scale, (1–5). The possible range of scores for each subscale was 3 to 15, with higher scores indicating a perception of greater social support. The subscales of this questionnaire showed good internal consistency (family: $\alpha = .80$, teachers: $\alpha = .92$, peers: $\alpha = .85$) in the present study.

2.4. Data analysis

There was a small amount of missing data. The rules for missing data for individual measures were followed, with mean replacement used where a minority of items were missing from a subscale. Where whole measures were missing ($N = 4$ caregiver measures; $N = 2$ sibling measures), participants were excluded from analyses involving the missing measure.

Analysis of caregiver data was conducted using the data from all 41 caregiver participants. Analysis of sibling data was conducted using data from the 31 siblings for whom there was self-report data. Analysis which directly compared caregiver and sibling data used only the data from the 31 families which had caregiver and sibling self-report data available.

Mean scores for sibling outcomes (caregiver and self-report scores on the SDQ and SCAS) were compared to normative data using one-sample t-tests. Binomial tests were used to compare the proportion of siblings who scored in the 'very high' range of SDQ scores to the proportions in the normative data. Caregiver and sibling SDQ, SCAS, and SRQ data were then compared using paired t-tests. To determine the extent to which sibling psychosocial adjustment and relationship quality were associated with demographic factors, psychological factors and aspects of the behavioural phenotype of the child with WS, a series of univariate analyses (correlations and t-tests) were run on the caregiver and sibling self-report data. To allow exploration of how multiple family variables were associated with sibling outcomes, exploratory multiple regression models were then run on the caregiver data for each main sibling outcome (total difficulties and pro-social behaviour SDQ scores, anxiety (SCAS-P), and the two SRQ sibling relationship scales). As the number of potential predictor variables was large, and the sample size relatively small, variables were included in a regression model only if they were initially associated with the outcome variable at $p < .01$.

3. Results

3.1. Psychosocial adjustment of siblings of children with Williams syndrome

Caregiver (full sample for whom data were available) and sibling self-report SDQ mean scores and proportions in the 'very high' range are reported in Table 2, alongside normative data. According to caregiver report, siblings had fewer adjustment difficulties overall than the general population. One sample t-tests showed significantly lower levels of caregiver-reported difficulties for hyperactivity, peer problems and total difficulties, with small effect sizes for peer problems and total difficulties, but an effect size close to large for hyperactivity. A significantly smaller proportion scored in the very high range in the conduct and hyperactivity subscales compared to the normative population. In contrast, caregiver reports indicated a significantly greater proportion of siblings in the

Table 2
Sibling psychosocial adjustment (SDQ data).

	Normative sample		Caregiver data (N = 39[b])		Mean score t-test effect size (*d*)	Normative sample		Self-report scores (N = 31)		Mean score t-test effect size (*d*)
	Parent-report (N = 10, 298)[a]					Self-report (N = 4,228) [a,c]				
	Mean (SD)	% 'very high'[d]	Mean (SD)	% 'very high'[d]		Mean (SD)	% 'very high'[d]	Mean (SD)	% 'very high'[d]	
Emotional	1.9 (2.0)	3.4	2.96 (2.67)*	15.4**	0.45	2.8 (2.1)	5.2	3.68 (2.64)	9.7	0.37
Conduct	1.6 (1.7)	3.5	1.15 (1.50)	0***	0.28	2.2 (1.7)	4.6	1.97 (1.64)	3.2	0.14
Hyperactivity	3.5 (2.6)	5.2	1.69 (1.92)***	0***	0.79	3.8 (2.2)	5.4	3.58 (2.35)	3.2	0.10
Peer	1.5 (1.7)	6.2	0.90 (1.53)*	5.1	0.37	1.5 (1.4)	3.7	1.52 (1.31)	0***	0.01
Total Difficulties	8.4 (5.8)	4.8	6.71 (4.86)*	2.6	0.32	10.3 (5.2)	5.1	10.74 (5.66)	6.5	0.08
Prosocial	8.6 (1.6)	5.1	8.87 (1.38)	2.6	0.18	8.0 (1.7)	1.8	8.32 (1.40)	0***	0.21

$^{*}p < .05$, $^{**}p < .01$, $^{***}p < .001$.

[a] Meltzer, Gatward, Goodman, and Ford (2000).

[b] SDQ caregiver data were missing for 2 siblings.

[c] the normative data relates to children aged 11–15 years, whereas the self-report data in the present study relates to children 6.88–17.93 years.

[d] for the prosocial behaviour subscale this is 'very low' i.e. a low score in prosocial behaviour.

Table 3
Sibling anxiety symptoms (SCAS-P and SCAS-C data).

	Normative sample Parent-report (N = 261)[a] Mean (SD)	Caregiver data (N = 41) Mean (SD)	t-test effect size (*d*)	Normative sample Self-report (N = 4916)[b] Mean (SD)	Self-report scores (N = 31) Mean (SD)	t-test effect size (*d*)
Separation anxiety	2.68 (2.65)	3.15 (3.18)	0.16	3.73 (3.21)	3.77 (3.19)	0.01
Social phobia	4.20 (2.75)	6.05 (3.92)**	0.55	6.06 (3.61)	6.13 (3.78)	0.02
Generalised anxiety	2.73 (2.05)	3.07 (2.09)	0.16	5.86 (3.36)	5.77 (2.97)	0.03
Panic/agoraphobia	1.0 (1.63)	1.20 (1.78)	0.12	3.62 (4.11)	3.61 (3.33)	0.002
Physical injury fears	2.60 (2.23)	2.90 (2.54)	0.13	3.15 (2.68)	3.52 (2.57)	0.14
Obsessive compulsive disorder	1.05 (1.75)	0.88 (1.03)	0.12	4.97 (3.63)	3.84 (3.59)	0.31
Total	14.08 (9.5)	17.24 (10.38)	0.32	27.38 (16.50)	26.65 (13.23)	0.05

*$p < .05$, ** $p < .01$.
[a] Nauta et al. (2004).
[b] Self-report norm data was derived from an Australian community sample (N = 4916) of school children aged 8–15 years (https://www.scaswebsite.com), whereas the self-report data in the present study relates to children 6.88–17.93 years.

very high range for the emotional problems subscale compared to the normative population, with significantly higher scores on this subscale and an effect size in the small-medium range. In relation to the siblings' self-report data, mean scores did not differ significantly from the general population in relation to total difficulties or any of the subscales. However, the binomial test showed a significantly lower proportion in the 'very high' range of self-report difficulties for peer problems and prosocial behaviour (i.e. proportion with very low prosocial behaviour scores) in comparison to the general population.

SCAS-P (full caregiver sample) and self-report SCAS-C mean scores for siblings are reported in Table 3, alongside the significance and effect sizes of one-sample t-tests comparing the study data with normative data. According to caregiver report, siblings had levels of anxiety symptoms no greater than the general population, with the exception that mean social phobia levels were significantly higher, with a medium effect size. According to caregiver data, 26.8% of siblings were higher than the suggested 'clinical caseness' total score of 24. In relation to the siblings' self-report data, mean scores did not differ from the general population in relation to total anxiety or any of the subscales.

Mean (SD) self-report scores on the SCSS-SV for peer support, family support, and teacher support were 12.73 (2.29), 13.07 (2.46), and 10.43 (3.53) respectively, with a total score of 36.23 (6.0), indicating high levels of perceived support, particularly from peers and family.

3.2. Comparison of caregiver and self-report sibling data

SDQ, SCAS, and SRQ caregiver-report data were compared with sibling self-report data, using only the data from the families for whom both caregiver and self-report data were available (Table 4). On the SDQ there were no significant differences on mean scores

Table 4
Comparison of caregiver and sibling SDQ data (N = 29), SCAS (N = 31), and SRQ (N = 30).

Measure	Caregiver report Mean (SD)	Self-report Mean (SD)	t-test comparison
SDQ			*t(28), effect size (d)*
Emotional	3.45 (2.81)	3.66 (2.68)	−0.35, 0.04
Conduct	1.21 (1.37)	1.86 (1.64)*	−2.29, 0.43
Hyperactivity	1.55 (1.97)	3.69 (2.39)***	−4.36, 0.98
Peer	0.93 (1.28)	1.41 (1.30)	−1.55, 0.30
Total Difficulties	7.14 (4.84)	10.62 (5.82)**	−2.99, 0.65
Prosocial	8.76 (1.53)	8.41 (1.38)	1.26, 0.24
SCAS			*t(30), effect size (d)*
Separation anxiety	2.97 (3.19)	3.77 (3.19)	−1.27, 0.25
Social phobia	6.45 (4.00)	6.13 (3.78)	0.44, 0.08
Generalised anxiety	3.32 (2.14)	5.77 (2.97)**	−3.73, 0.95
Panic/agoraphobia	1.45 (1.96)	3.61 (3.33)**	−3.35, 0.79
Physical injury fears	2.77 (2.39)	3.52 (2.57)	−2.03, 0.30
Obsessive compulsive disorder	0.90 (1.04)	3.84 (3.59)***	−4.79, 1.11
Total	17.87 (10.44)	26.65 (13.23)**	−3.20, 0.74
SRQ			*t(29), effect size (d)*
Warmth/closeness	19.52 (4.33)	18.57 (3.22)	1.69, 0.25
Conflict	10.35 (4.24)	11.69 (4.17)*	−2.37, 0.31

*$p < .05$, **$p < .01$, ***$p < .001$.
[a]Data included only for families with both caregiver and self-report data available.

Table 5
Regression analyses of caregiver-reported sibling adjustment and relationship quality.

Predictor variable	Sibling adjustment		Sibling relationship	
	SDQ Total Difficulties β	SCAS-P Total β	Warmth/ closeness β	Conflict β
Caregiver HADS depression	–	.28*	–	–
Caregiver family satisfaction	–	–	–	–
Child with WS age	–	–	−.49**	–
Child with WS prosocial behaviour (SDQ)	–	–	.31*	–
Sibling total difficulties (SDQ)[a]	–	.47**	–	.45*
Sibling anxiety (SCAS-P)[a]	.50***	–	–	.13
Conflict in sibling relationship (SRQ)[a]	.32*	.12	–	–

*$p < .05$, **$p < .01$, ***$p < .001$.
Note. Dashes (-) signify that the variable was not entered into the regression analysis.
[a]Caregiver-report.
SDQ Total difficulties: $F(2, 36) = 17.46$, $p < .001$, $R^2 = .49$.
SCAS-P Total: $F(3, 35) = 10.81$, $p < .001$, $R^2 = .48$.
SRQ Warmth/closeness: $F(2, 38) = 12.24$, $p < .001$, $R^2 = .36$.
SRQ Conflict: $F(2, 36) = 7.45$, $p = .002$, $R^2 = .29$.

between caregiver and self-report for the subscales of emotional problems, peer problems, or prosocial behaviour, but siblings reported significantly greater difficulties in conduct problems (small effect size), hyperactivity (large effect size), and total difficulties (medium effect size) than did parents. On the SCAS there were no significant differences between caregiver and self-report for separation anxiety, social phobia, or physical injury fears. However, siblings reported significantly greater difficulties than caregivers in relation to the sibling's levels of generalised anxiety and obsessive-compulsive disorder (both large effect sizes) as well as panic/ agoraphobia and total anxiety symptoms (both medium effect sizes). For the SRQ, there were no significant differences between caregiver and self-report on warmth in the sibling relationship. However, siblings reported significantly greater conflict in the relationship than caregivers (small effect size).

3.3. Variables associated with sibling psychosocial adjustment and relationship quality

To explore the variables associated with sibling outcome measures four regression models were run, one for each outcome measures (SDQ total difficulties, SCAS-P total anxiety, and SRQ warmth and conflict). This analysis drew on caregiver report only, using data from the full caregiver sample. Variables were selected from the univariate exploratory analysis for inclusion in a regression analysis if they were initially associated with the outcome variables at $p < .01$. The age of the child with WS and the sibling were both negatively associated with SRQ warmth, but they were highly correlated with each other, so only the age of child with WS (the variable more highly correlated with SRQ warmth) was entered into this regression. The age of the children with WS and the sibling were not significantly correlated with any of the other outcome variables, so were not entered into the other regression models. Regression was not run with SDQ pro-social behaviour as an outcome, due to the low number of significantly correlated variables. The results of the regression analyses are summarised in Table 5. There were few variables in the regression models which were found to be significant independent predictors of sibling outcomes. Siblings had higher levels of behavioural problems when they had higher levels of anxiety and higher levels of caregiver-reported conflict in the sibling relationship. They had higher levels of anxiety when there were higher levels of caregiver depression and higher levels of behavioural problems. Higher levels of caregiver-reported warmth/closeness in the sibling relationship was reported in families where the children with WS were younger and where they had higher levels of prosocial behaviour. Higher levels of caregiver-reported conflict in the sibling relationship was reported in families where the sibling had higher levels of behavioural problems.

Sibling self-report data were not included in the regression models, due to the smaller sample size. However, univariate analysis was conducted to explore which variables were associated with sibling self-reported outcome measures (SDQ, SCAS-C, and SRQ) (supplementary Table 1). This analysis used only the data from the 31 families for whom there was both caregiver and sibling self-report data. Self-report SDQ total difficulties were negatively correlated with perceived social support ($r = -0.48$, $p < .01$) and positively correlated with self-report total anxiety ($r = 0.80$, $p < .001$). Self-report SDQ prosocial behaviour was positively correlated with perceived social support ($r = 0.52$, $p < .01$) and with caregiver report SDQ prosocial behaviour ($r = .49$, $p < .01$). Self-report sibling relationship warmth and conflict were only positively correlated with the corresponding caregiver-report ($r = 0.62$, $p < .001$, and $r = 0.60$, $p < .001$, for warmth and conflict respectively). Given the large number of correlation analyses, only those with $p < .01$ are reported here.

4. Discussion

This study is, to our knowledge, the first to explore the behavioural and emotional adjustment, as well as the relationship quality, of siblings of children with WS, from both the perspective of caregivers and of the siblings themselves. It is also novel in exploring the factors which may be associated with adjustment and relationship quality.

The sibling relationships reported upon could be characterised as fairly warm, with reasonably low levels of conflict, and good adjustment in siblings overall. Caregiver-reported sibling behavioural outcomes were more positive than those of the general population in relation to some aspects of behaviour. From the perspective of the subsample of siblings for whom there was self-report data, behavioural and emotional adjustment was similar to that of the general population. Similarly, in terms of perceived symptoms of anxiety, the caregiver and sibling reports indicated very few differences from the general population. This accords with Leyfer et al. (2009) findings of low levels of parent-reported anxiety symptoms in siblings of children with WS. It also suggests that although anxiety symptoms may be raised in WS (Royston et al., 2017), there is no support for a social contagion model of anxiety (see Serra Poirier et al., 2017) in relation to siblings. Overall, our findings are positive, and provide further support for the argument that children with developmental disabilities are not an inevitable source of difficulty for siblings (Hastings & Petalas, 2014; Hastings, 2016; O'Neill & Murray, 2016; Rodgers et al., 2016).

However, there were some areas of sibling adjustment difficulty reported. Caregiver SDQ scores indicated significantly greater sibling emotional difficulties than in the general population. This suggests that, where siblings of children with WS do experience difficulties, this might manifest as worries, unhappiness or nervousness, rather than as difficulties around conduct or peer interaction. In terms of anxiety symptoms, whilst caregiver reports generally indicated normative levels of sibling anxiety symptoms, social phobia symptoms were greater than that reported in the general population. A possible explanation for our finding might lie in some of the specific social phobia items in the SCAS, e.g. '*My child feels afraid that s(he) will make a fool of him/herself in front of people*' and '*My child worries what other people think of him/her*' (Spence, 1998). Although the items do not refer specifically to the child with WS, raised scores on this subscale perhaps indicate that some caregivers are concerned about the response of the public when they are out with their child with WS, and the effect of this upon the sibling. If there are such public responses, this may relate to a lack of awareness of WS due to the rarity of the disorder (Fidler, Hodapp, & Dykens, 2002; Howlin & Udwin, 2006; Scallan et al., 2011). However, findings for the subsample of siblings for whom there was self-report data did not indicate significantly greater social phobia symptoms in comparison to population norms, suggesting that any issues of stigma (and social phobia more broadly) may be less of a concern for siblings than caregivers perceived them to be.

While the overall picture of sibling adjustment appears to be relatively positive, there was individual variation across siblings, and a key aim was to identify demographic, psychological and behavioural phenotypic variables which might be associated with variation in sibling adjustment and relationships. Whilst the regression analysis indicated relatively few significantly associated variables overall, the findings did emphasise the importance of adopting a family systems perspective: significant associations were found between the caregiver-reported data on the children with WS, the sibling, and the caregiver. Specifically: caregiver symptoms of depression were positively associated with sibling anxiety symptoms; conflict in the sibling relationship was positively associated with sibling total behavioural difficulties; and warmth in the sibling relationship was negatively associated with the age of the child with WS, and positively associated with the prosocial behaviour of the child with WS. Whilst warmer relationships in siblings with higher levels of prosocial behaviour is something which is likely to be typical across families both with and without developmental disabilities, the findings do indicate that one aspect of the WS behavioural phenotype, namely social functioning, may be associated with sibling relationship quality. In contrast, no significant associations were found between anxiety symptoms in the child with WS, and sibling adjustment or relationship quality, suggesting that this aspect of the WS behavioural phenotype may have little impact on siblings. This may be because, for example, siblings are not fully aware of the child's anxiety symptoms, or because caregivers mitigate any effects of the child's anxiety symptoms on the sibling.

In terms of the sibling self-report data, correlation analyses indicated relatively few significantly associated variables. However, perceived social support was consistently found to be associated with sibling adjustment. The value of social support from family, peers and teachers, has been highlighted in a number of developmental disability sibling studies (Cebula, 2012; Hastings, 2003; Tomeny, Rankin, Baker, Eldred, & Barry, 2018). Further exploration is warranted of pathways to effective social support for siblings of children with WS, the mechanisms by which support is associated with sibling behavioural and emotional outcomes, and the type of support which siblings consider most beneficial to address different issues at different ages.

Although previous research has examined discrepancies between multi-informant ratings of adjustment in siblings of children with autism (e.g. Rankin et al., 2017; Rodgers et al., 2016), this has not previously been examined in relation to siblings of children with WS. The current findings did indicate some discrepancy, in the direction of siblings generally reporting greater difficulties than the caregivers perceived them to have. Rankin et al. (2017) suggest that parents may slightly underestimate sibling difficulties because the behaviour of the child with developmental disabilities provides them with a skewed point of reference for normative behaviour. However, child and parent discrepancies are found even amongst families without a child with developmental disabilities (e.g. Achenbach, McConaughy, Howell, & Masters, 1987), and it would be premature to link the discrepancies in the current study to the presence of the child with WS. The findings do emphasise the importance of listening to any concerns raised by siblings of children with WS about adjustment or relationship difficulties, as these may be greater than those perceived by their caregivers.

It is important to consider the limitations of this study when interpreting the findings. As with much WS research, the overall sample size, was relatively small, and the age range relatively wide. Cautious interpretation of the findings, particularly for the regression modelling, is therefore important. As with all family developmental disability research, it is possible that families experiencing fewer difficulties were more likely to volunteer to participate in the study. The majority of caregiver respondents were mothers, so little is known about fathers' perspective on their children's sibling relationships. Two of the caregiver respondents in the present study were not mothers. Whilst this made the study more inclusive, it is possible that the responses of these other caregivers differed from those of mothers. Additionally, a small number of siblings who completed self-report data were younger than the age for which the measures were designed and the normative data to which they were compared. This may have influenced the findings, although steps were taken to ensure that all siblings understood the questions. The sample was diverse in terms of wealth, but limited

in ethnic diversity. Given the cultural differences in sibling experiences found in relation to other developmental disabilities (e.g. Tsai, Cebula, Liang, & Fletcher-Watson, 2018), exploring the experiences of siblings of children with WS in other cultural contexts is an important next step. Gathering data from the children with WS themselves in the future, would enable a more complete picture of family experiences. Doing so longitudinally would provide an insight into how family members influence each other behaviourally and emotionally over time.

In terms of practical implications, the positive nature of sibling outcomes in the present study, if replicated, suggest that interventions to ameliorate poor behavioural and emotional adjustment are not likely to be required for all neurotypical siblings of children with WS. However, there was variability in adjustment across our sample, indicating that support may be required in some families, with the regression analysis findings suggesting that support may be particularly required by families in which there are higher levels of caregiver depression symptoms, higher levels of sibling anxiety or behaviour problems or high levels of conflict between the siblings. Assessing whether the positive childhood outcomes found in this study are maintained as siblings transition into adulthood will also be important in ascertaining sibling support needs across the lifespan.

5. Conclusions

This study shows that the majority of siblings of children with WS are well adjusted, according to both caregiver and sibling report. Where difficulties do arise, these may more often be emotional difficulties than conduct or peer problems, and siblings may sometimes experience slightly greater difficulties than caregivers appreciate. Social support may be associated with better behavioural and emotional adjustment. The findings of this study do provide some support for both a family systems and a behavioural phenotype 'indirect effects' approach to understanding sibling relationships in WS. Overall, though, while siblings may have unique and important experiences growing up with a child with WS, the findings of this study emphasis the need to avoid taking a pathologising approach.

Declaration of Competing Interest

The authors declare that they have no conflicts of interest in presenting the results in this paper.

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Appendix A. Supplementary data

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