

Northumbria Research Link

Citation: Lovell, Brian and Wetherell, Mark (2016) The psychophysiological impact of childhood autism spectrum disorder on siblings. *Research in Developmental Disabilities*, 49-50. pp. 226-234. ISSN 0891-4222

Published by: Elsevier

URL: <http://dx.doi.org/10.1016/j.ridd.2015.11.023>
<<http://dx.doi.org/10.1016/j.ridd.2015.11.023>>

This version was downloaded from Northumbria Research Link:
<http://nrl.northumbria.ac.uk/id/eprint/25037/>

Northumbria University has developed Northumbria Research Link (NRL) to enable users to access the University's research output. Copyright © and moral rights for items on NRL are retained by the individual author(s) and/or other copyright owners. Single copies of full items can be reproduced, displayed or performed, and given to third parties in any format or medium for personal research or study, educational, or not-for-profit purposes without prior permission or charge, provided the authors, title and full bibliographic details are given, as well as a hyperlink and/or URL to the original metadata page. The content must not be changed in any way. Full items must not be sold commercially in any format or medium without formal permission of the copyright holder. The full policy is available online: <http://nrl.northumbria.ac.uk/policies.html>

This document may differ from the final, published version of the research and has been made available online in accordance with publisher policies. To read and/or cite from the published version of the research, please visit the publisher's website (a subscription may be required.)

The psychophysiological impact of childhood autism spectrum disorder on siblings

Abstract

Objective: The negative impact of caring for a child with autism spectrum disorder (ASD) on parents' psychophysiological functioning has been widely evidenced. However, siblings, who also face emotional, social and physical challenges associated with having a brother/sister with ASD, have been less widely studied. This study examined the psychophysiological impact of childhood ASD on siblings. *Methods:* A sample of 25 siblings of children with ASD (and their mothers) and a control group of 20 siblings of neuro-typical children (and their mothers) completed questionnaires assessing: (a) demographic and lifestyle information, (b) family characteristics, (c) child behaviour problems, (d) social support and (e) depressive symptomology. Saliva samples were collected at several time points on two consecutive days, and estimates of the cortisol awakening response (CAR), diurnal cortisol slope and mean diurnal cortisol output were derived. *Results:* Total depressive symptoms were higher in siblings of children with ASD compared with controls. Group differences with respect to depressive symptomology were driven more by emotional than functional problems. With respect to physiological functioning, groups were comparable on all cortisol indices. In siblings of children with ASD, social support, especially from parents and close friends, predicted total depressive symptoms, as did the behaviour problems of their brother/sister with ASD. *Conclusion:* Siblings of children with ASD experience greater emotional problems and overall depressive symptoms compared with a control group. Interventions that enhance social support, as well as helping siblings better understand the behaviour problems of their brother/sister with ASD, might be effective for alleviating depressive symptoms.

Keywords: cortisol; behaviour problems; depression; siblings; social support

The psychophysiological impact of childhood autism spectrum disorder on siblings

Introduction

Caring for a child with a developmental disability (DD) such as ASD has been widely used as one model for examining the effect of chronic stress on psychophysiological functioning (Lovell & Wetherell, 2011). Indeed, the challenges of caring for a child with a DD, which include financial hardship (Kogan et al., 2008), social isolation (Yantzi, Rosenberg, & McKeever, 2006), negotiating a fragmented service system (Griffith & Hastings, 2013), stigma and social judgement (Cantwell, Muldoon, & Gallagher, in press), far outstrip those of parenting a neuro-typical child. That caregivers of children with a DD experience increased psychological distress has been widely evidenced, with studies reporting on higher levels of psychological symptoms such as perceived stress (Cantwell, Muldoon, & Gallagher, 2014), hopelessness (Bandeira et al., 2010), anxiety (Ruiz-Robledillo & Moya-Albiol, 2013) and depression (Cantwell et al., in press; Smith & Grzywacz, 2014). In addition, compared with parents of neuro-typical children, caregivers' levels of anxiety and depression were found to be more likely to satisfy criterion for clinical mood disorder (Gallagher, Phillips, Oliver, & Carroll, 2008). That caregiving stress is associated with poorer psychological functioning was also demonstrated in two recent meta analyses (Easter, Sharpe, & Hunt, 2015; Hayes & Watson, 2013) and review studies (Cousino & Hazen, 2013; Fairthorne, de Klerk, & Leonard, 2015). The stress of caring for a child with a DD has also been linked with alterations in a variety of health relevant, physiological parameters. For example, perturbations in cardiovascular (Gallagher & Whiteley, 2012), neuroendocrine (Bella, Garcia, & Spadari-Bratfisch, 2011; Ruiz-Robledillo & Moya-Albiol, 2013; Seltzer et al., 2009) and immune (Gallagher, Phillips, Drayson, & Carroll, 2009; Lovell, Moss, & Wetherell, 2012) functioning have been widely implicated as plausible physiological pathways linking chronic caregiving stress with downstream disease.

Fewer studies have examined the psychophysiological impact of childhood disability on other family members living at home. This is surprising given that siblings, much like their parents,

face a number of challenges associated with having a brother/sister with a DD. For example, to alleviate some of the burden on their parents, siblings often take on additional household chores and responsibilities (Dyke Mulroy, & Leonard, 2009), thus limiting opportunities for social and peer interaction, and extracurricular activities (Barak-Levy, Goldstein, & Weinstock, 2010; Moyson & Roeyers, 2012). In addition, parents, who are overburdened by their caregiving role, have been found to be less physically and emotionally available for their other children (Macks & Reeve, 2007). Indeed, in two recent qualitative studies, sibling adjustment was, along with lack of support and stigma, one of the biggest concerns raised by caregivers of children with a DD (Ludlow, Skelly, & Rohleder, 2011; Oruche, Gerkenmeyer, Stephan, Wheeler, & Hanna, 2012). Perhaps most challenging, many siblings actively contribute to the caretaking role, participating in activities such as feeding, dressing and bathing their disabled brother/sister (Randall & Parker, 1999).

Relative to normative controls, siblings of children with a DD such as ASD have been found to report higher levels of separation anxiety (Lobato et al., 2011), emotional problems (Petalas, Hastings, Nash, Lloyd, & Dowey, 2009) and internalising behaviours (Fisman, Wolf, Ellison, & Freeman, 2000). Findings from a review paper (Barlow & Ellard, 2006) and recent meta analysis (Vermaes, van Susante, & van Bakel, 2012) also suggested that siblings of children with a DD might be at greater risk for psychological adjustment difficulties, with internalising behaviours and depressive symptoms found to be higher when compared with a control group. However, not all findings have been consistent, such that several studies observed no effect (Benson & Karlof, 2008; Di Biasi et al., in press; Tomeny, Barry, & Bader, 2012), or an adaptive effect (Macks & Reeve, 2007), of childhood disability on siblings' psychological functioning. These inconsistent findings highlight the need for additional research in this area.

Siblings' psychological adjustment to childhood disability has been shown to be moderated by a range of variables. For example, family characteristics such as socioeconomic status (Macks & Reeve, 2007; Petalas et al., 2009), family size (Kaminsky & Dewey, 2002; Labay & Walco, 2004) and

parental stress (Giallo & Gavidia-Payne, 2006) have been shown to predict emotional and internalising problems in siblings of children with a DD. In addition, research has shown that siblings' psychological well being is, at least in part, affected by characteristics of their disabled brother/sister, especially their behavioural problems. For example, in two recent studies, child behaviour problems (CBP), and in particular, hyperactivity and conduct behaviours positively predicted depression symptoms in their siblings (Mayer, Ingersoll, & Hambrick, 2011; Neece, Blacher, & Baker, 2010). These findings resonate with studies involving parental caregivers of children with a DD, where CBP were found to account for much of the variance in caregivers' feelings of depression (Gallagher et al., 2008; Lovell, Moss, & Wetherell, 2015). Most recently, in a study involving siblings of children with ASD, Shivers, Deisenroth and Taylor (2012) found that CBP, along with maternal stress, uniquely predicted siblings' feelings of anxiety. Perceived availability of social support has also been shown to be influential for the psychological adjustment of siblings of children with a DD. for example, socially supported siblings, much like their parents, have been found to report lower levels of negative affective symptoms such as depression (Barrera, Fleming, & Khan, 2004), loneliness (Kaminsky et al., 2012) and anxiety (Nolbris et al., 2010).

The impact of childhood disability on siblings' physiological functioning has, we believe, yet to be determined. Cortisol, the final effector hormone of the hypothalamic-pituitary-adrenal (HPA) axis, displays a robust basal diurnal pattern; levels are high in the morning upon waking, reach an acrophase 30-45 minutes post waking (i.e., the cortisol awakening response: CAR), gradually decline across the day (i.e., diurnal cortisol slope) and reach a nadir around midnight (Saxbe, 2008). However, if overused by chronic (i.e., repeated) stress, the normal operating levels of physiological, stress responsive systems such as the HPA axis have been shown to shift. For example, atypical patterns of cortisol secretion characterised by flatter cortisol slopes (Seltzer et al., 2009), greater CAR magnitude (Ruiz-Robledillo & Moya-Albiol, 2013) and hypo-secretion of cortisol across the day (Bella et al., 2011; Seltzer et al., 2010) have been observed in parental caregivers of children with a

DD. Moreover, alterations in basal HPA activity have been implicated as one physiological indicator for downstream disease outcomes such as infectious disease (Edwards, Hucklebridge, Clow, & Evans, 2003), cardiovascular pathologies (Seldenrijk, Hamer, Lahiri, Penninx, & Steptoe, 2012) and sleep problems (Lasikiewicz, Hendrickx, Talbot, & Dye, 2008).

Like their parents, the sustained social, physical and emotional sequelae associated with having a brother/sister with a DD might, via repeated activation of physiological processes such as the HPA axis, confer potential health implications for siblings. Indeed, basal stress hormone activity, though yet to be investigated in siblings of children with a DD, has been shown to be altered in the context of other chronic childhood stressors. For example, reduced CAR magnitude (Gunnar, Morison, Chisholm, & Schuder, 2001) and flatter diurnal cortisol slopes (Wolf, Nicholls, & Chen, 2008) have been observed in the context of chronic childhood stressors such as maltreatment and family discord. Most recently, flattening of the diurnal cortisol slope, which is indicative of HPA dysregulation, was also observed in peer victimized children (Knack, Jensen-Campbell, & Baum, 2011), as was diminished cortisol reactivity to stress in the lab (Vaillancourt et al., 2011).

The current study had several aims: (a) to assess the impact of childhood ASD on siblings' depressive symptoms, (b) to move research in the area forward by assessing the impact of childhood ASD on siblings' basal stress hormone activity, and (c) to determine whether variations in siblings' psychological and physiological adjustment to childhood ASD might be explained by individual difference variables such as social support and CBP.

Method

Participants

A sample of (N=25) siblings of children with ASD (and their mothers) was recruited from caregiver support groups, charities, and special schools. All siblings were screened against the following criteria: (a) aged 7-17 years, (b) at least one brother/sister with ASD aged between 3-21

years living at home full time, (c) not experiencing long term stressors such as parent divorce, bereavement or recent change of school, (d) not experiencing serious medical or psychiatric problems, and (e) not taking steroid or statin based medication. A control group of (N=20) siblings (and their mothers) of neuro-typical children was recruited according to the same criteria, but in order to be eligible must not: (a) have a brother/sister, or any other person living in the family home (e.g., parent, grandparent, friend etc) living with chronic illness. This study and all its procedures were approved by the Institutional Ethics Review Board; all siblings (and their mothers) provided written consent to take part. Of (N= 45) participants recruited, (N=5) failed to return any questionnaires or saliva samples, and (N=2) reported a delay between waking and collection of the waking cortisol sample on one saliva collection day. As non compliance with the saliva collection protocol can invalidate the reliability of resultant cortisol data (Clow, Hucklebridge, Stalder, Evans, & Thorn, 2010), only protocol adherent data, i.e., from the remaining collection day, was used for statistical analysis. Statistical analysis was conducted on a final sample of (N=22) siblings of children with ASD and (N=18) siblings of neuro-typical children. Sample characteristics by group are presented in Table 1.

Demographic and family characteristics

Participating siblings completed a short questionnaire to assess their age, gender, weight, and exercise frequency. Mothers of participating siblings completed a short questionnaire to assess their age, marital status and number of children living at home. As a proxy measure of socioeconomic status, mothers also disclosed their annual household income and highest level of education.

Depressive symptomology

Participating siblings completed the Children's Depression Inventory-2 (CDI-2), a 27 item questionnaire that yields a total score (which can range between 0-56 and is derived by summing across all items), two scale scores (emotional and functional problems) and four subscale scores (negative self esteem, negative mood, interpersonal problems and ineffectiveness). Scale responses range from 0, *symptom free* to 2, *showing definite symptoms*, with higher scores reflecting greater depressive symptoms. The CDI-2 has been shown to have good internal consistency in previous studies involving siblings of children with ASD (Macks & Reeve, 2007), and this was also the case here ($\alpha = 0.90$).

Social support

Participating siblings completed the Social Support Scale for Children (SSSC), a 24 item questionnaire that measures support from: (a) parents, (b) classmates, (c) teachers, and (d) close friends (Harter, 1985). Scale responses range between 1, *lowest level of support* and 4, *highest level of support*. A total score for each subscale can range between 1 and 24, with higher scores reflecting greater perceived support. The SSSC has been shown to have excellent psychometrics in previous studies involving siblings of children with ASD (Kaminsky & Dewey, 2002), and internal consistency in the current sample was also good ($\alpha = 0.91$).

Child behaviour problems (CBP)

The 25 item *Strengths and Difficulties Questionnaire* (SDQ) was used to measure the behaviour problems of the child with ASD. Mothers of participating siblings were asked to rate whether behaviours were, 0 (*not true*), 1 (*somewhat true*), or 2 (*certainly true*) for their child with ASD. The SDQ measures child problematic behaviours across four subscales: *emotional symptoms* (e.g., nervous or clingy in new situations), *conduct problems* (e.g., often fights with other children),

hyperactivity (e.g., restless, overactive, cannot sit still for long), and *peer relationships* (e.g., generally liked by other children). A total SDQ score, which can range between 0-40, was derived by summing across all four subscales, with higher scores reflecting more CBP. The SDQ has achieved good psychometrics in previous studies of a similar nature (Meyer, Ingersoll, & Hambrick, 2011), as was the case here ($\alpha = 0.75$).

Physiological measures

Basal HPA axis functioning was assessed by measuring cortisol in saliva at waking, 30 minutes post waking, 1200h and 2200h on two consecutive weekdays. Collected samples were centrifuged for 10 minutes, 400 x g at 20°C and tested in-house using an enzyme-linked immunosorbant assay (ELISA), Salimetrics Ltd, Suffolk, England. Mean inter and intra assay coefficients were 7.1% and 10.7%, respectively. Raw cortisol data was \log^{10} transformed to correct for positive skew. Log data for each sampling day was treated in two ways to provide different markers of HPA axis activity. Findings from a recent meta analysis indicated that cortisol reactivity from waking is, compared with measures of overall cortisol volume (i.e., area under the curve), a more appropriate measure for estimating HPA activity during the post waking period (Chida & Steptoe, 2009). Therefore, in accord with other recent studies (Kudielka, Gierens, Hellhammer, Wust, & Schlotz, 2012), the cortisol awakening response (CAR) was calculated as the difference between cortisol values at waking and 30 minutes post waking. Second, to capture the diurnal cortisol slope, a linear regression line was estimated for each participant that predicted cortisol decline from time since waking (Smyth et al., 1997). Steeper cortisol slopes, which indicate a greater rate of diurnal decline, are represented by smaller β values (i.e., larger negative values). Higher β values (as they approach, or cross zero) on the other hand reflect flatter diurnal slopes and are indicative of dysregulated cortisol secretion. In accord with other work, and to avoid any influence of the CAR, cortisol values at 30 minutes post waking were removed from estimates of the diurnal

slope (Brant, Wetherell, Lightman, Crown, & Vedhara, 2009). In keeping with recent work, cortisol values were averaged across sampling days to provide more reliable estimates of basal HPA axis functioning (Holland, Thompson, Zsuang, & Gallagher-Thompson, 2010),

Procedure

A participation pack containing self report measures of demographic and lifestyle factors, depressive symptomology (CDI-2) and social support (SSSC) was sent to participants by post. Mothers were asked to oversee and to ensure siblings' accurate completion of these questionnaires. The pack also included questionnaires to measure family characteristics (e.g., mother's age and marital status, and number of other children living in the home) and child behaviour problems (SDQ). These questionnaires were to be completed by the mother only. Materials for the ambulatory collection of salivary cortisol were also included in the pack, as were detailed written instructions emphasising the time sensitive nature of the hormone. Poor adherence with the saliva collection protocol can invalidate the reliability of resultant cortisol data (Kudielka et al., 2012). That is, inaccurately timed morning samples, both in relation to waking (Okun et al. 2010) and to each other (Kudielka, Hawkley, Adam, & Cacioppo, 2007), have been linked with erroneous interpretations of cortisol indices such as the CAR. Therefore, to encourage protocol adherence, siblings were asked to record waking and saliva collection times on each sampling day using a paper diary (Lovell, Moss, & Wetherell, 2015). A delay >10 minutes between waking and collecting the waking sample was used as criterion for the exclusion of inaccurate cortisol data (Lovell et al., 2012). Physiologic data were also excluded if collection of the waking and subsequent post waking saliva sample deviated by >10 minutes from the requested 30 minute interval (Bhattacharyya, Molloy, & Steptoe, 2008). In addition, for 30 minutes prior to the collection of any sample, siblings were instructed to abstain from behaviours known to affect the measurement of cortisol in saliva (Fries, Dettenborn, & Kirschbaum, 2009). These behaviours included: consuming food and/or caffeinated drinks, exercising, brushing teeth and/or using mouthwash. Saliva was collected using the salivette device

(Sarstedt, UK), whereby participants are asked to chew a sterile cotton swab for 1-2 minutes before depositing the saturated swab into a plastic collection tube. For the present study, mothers were instructed to oversee and ensure participants' accurate collection of saliva. Completed questionnaires and collected saliva were returned to the research team using prepaid addressed envelopes. As recompense for their time, all participants were entered into a prize draw to win an Apple iPad.

Statistical analysis

Chi square (χ^2) and univariate ANOVA were used to assess group differences with respect to sibling demographic and lifestyle factors, and family characteristics. One way ANOVA was also used to compare groups on total CDI-2 and SSSC scores, and on aspects of the diurnal cortisol pattern, CAR magnitude and diurnal cortisol slope. Mixed ANOVA was used to test for group differences with respect to cortisol values per individual sampling point and mean cortisol output across the day. Variations in degrees of freedom reflect missing values and, where appropriate, Huynh Feldt was applied to correct violations of sphericity.

Subsequent analysis used multiple regression analysis to determine whether psychophysiological outcomes in siblings of children with ASD might be differentially affected by social support and CBP.

Results

Demographic and family characteristics

Groups were comparable with respect to gender, age, weight and exercise (all $ps > 0.11$). Groups were also indistinguishable on all family characteristics: mother's age ($F(1, 32) = .93, p = .34, \eta_p^2 = .03$), mother's marital status ($\chi^2(1) = 1.13, p = .29$), mother's highest level of education ($\chi^2(3) = 4.53, p = .21$), annual household income ($F(1, 30) = 1.72, p = .20, \eta_p^2 = .05$), and total number of

children living at home ($F(1, 34) = 2.20, p = .15, \eta_p^2 = .06$). As such, none of these variables were included in subsequent analyses.

Table 1.

Summary Characteristics by Group (N=38)

	Siblings of children with autism (N=20)	Siblings of neuro-typical children (N=18)	$p =$
Gender (%)			0.64
Female	45.5	53.0	
Male	55.5	47.0	
Mean age (SD), yrs	12.6 (3.1)	11.0 (3.0)	0.12
Mean weight (SD), lbs	103.2 (36.3)	85.4 (48.0)	0.23
Mean exercise^a (SD)	2.2 (2.0)	2.9 (1.7)	0.29
Mean NOC (SD)	2.8 (1.4)	2.3 (0.5)	0.15
Mean AHI (SD)	41,937.5 (19,964.9)	52,562.5 (25,554.7)	0.20
Mean age of mother (SD), yrs	42.9 (5.7)	40.9 (6.2)	0.34
Mother's LOE (%)			0.21
School	11.1	5.6	
College	16.7	0.0	
Undergraduate	16.7	11.1	
Postgraduate	55.5	83.3	
Mother's marital status (%)			0.29
Partnered	83.3	94.4	
Not partnered	16.7	5.6	

Note. ^aMeasured as number of occasions per week. NOC = number of children (total number of children living in the family home). AHI = annual household income in pounds sterling. LOE = level of education.

Self report measures

Groups could not be differentiated with respect to scores on the negative self esteem ($F(1, 38) = 1.00, p = .32, \eta_p^2 = .03$), ineffectiveness ($F(1, 38) = 2.97, p = .09, \eta_p^2 = .07$) and interpersonal problems ($F(1, 38) = 1.57, p = .22, \eta_p^2 = .04$) subscales of the CDI-2. Levels of negative mood were, however, higher in ASD siblings compared with controls ($F(1, 38) = 6.98, p = .01, \eta_p^2 = .16$). Scores on the emotional problems scale of the CDI-2 were also higher in siblings of children with ASD ($F(1, 38) = 4.38, p = .04, \eta_p^2 = .10$), as were total (CDI-2 scores) depressive symptoms ($F(1, 38) = 4.22, p = .05, \eta_p^2 = .10$). There was also a trend for greater functional problems in siblings of children with ASD ($F(1, 38) = 2.86, p = .09, \eta_p^2 = .07$). These results reflect the fact that depressive symptoms were higher in siblings of children with ASD, and this was largely due to greater emotional, rather than functional, problems.

Data indicated that groups could not be differentiated on SSSC subscales: parents ($F(1, 37) = .24, p = .63, \eta_p^2 < .01$), close friends ($F(1, 37) = .05, p = .83, \eta_p^2 < .01$), peers ($F(1, 37) = .43, p = .52, \eta_p^2 = .01$), and teachers ($F(1, 37) = .06, p = .81, \eta_p^2 < .01$).

Physiological measures

A two (**group**: siblings of children with ASD vs. siblings of neuro-typical children) x four (**time**: waking, 30 minutes post waking, afternoon, before bed) mixed ANOVA yielded a significant main effect of time ($F(2.4, 84.6) = 99.84, p < .01, \eta_p^2 = .74$), reflecting normal variation in diurnal cortisol secretion. However, groups were comparable with respect to mean diurnal cortisol output ($F(1, 35) = .28, p = .60, \eta_p^2 < .01$) and pattern of change across the day ($F(2.4, 84.6) = .43, p = .55, \eta_p^2 = .01$). One way ANOVA also revealed that groups could not be differentiated on HPA markers, CAR magnitude ($F(1, 35) = .94, p = .34, \eta_p^2 = .03$) or diurnal cortisol slope ($F(1, 35) = .10, p = .75, \eta_p^2 < .01$). Means and standard deviations for psychophysiological outcomes by group are presented in Table 2.

Table 2**Means and Standard Deviations for Psychophysiological Outcomes by Group**

	Siblings of Children with ASD (N=22)	Siblings of Neuro- Typical Children (N=18)	<i>p</i> =
Child behaviour problems (SDQ)	19.5 (8.8)	6.8 (4.2)	< .01
Social support (SSSC)			
Parents	21.5 (2.6)	21.9 (2.7)	.63
Classmates	19.4 (3.8)	20.1 (2.5)	.52
Teachers	20.5 (3.0)	20.8 (3.7)	.78
Close friends	21.0 (4.4)	20.8 (3.9)	.83
Depression (CDI-2)			
Negative mood	3.8 (2.9)	1.8 (1.7)	.01
Negative self esteem	1.8 (2.3)	1.2 (1.2)	.32
Ineffectiveness	3.7 (2.4)	2.4 (2.5)	.09
Interpersonal problems	1.0 (1.6)	.05 (1.0)	.22
Functional problems	4.8 (3.7)	2.9 (3.2)	.09
Emotional problems	5.6 (4.9)	2.9 (2.3)	.04
Total CDI-2 score	10.4 (8.2)	5.8 (4.9)	.05
HPA markers			
CAR (nmol/L)	-1.48 (5.4)	0.96 (6.8)	.34
Diurnal slope (β)	-0.87 (0.16)	-0.89 (0.15)	.72
Mean diurnal output (nmol/L)	5.5 (2.1)	6.0 (2.9)	.60

Note: CAR – cortisol awakening response

Within group analysis

No association between any of the demographic and family characteristics, and siblings' total CDI-2 scores were observed (all $ps > .06$). Similarly, siblings' total depressive symptoms were not related to age of the child with ASD ($r = .35, p = .14$). As such, none of these variables were included in the regression model.

In competing analysis where all SSSC subscale scores and total SDQ scores were entered simultaneously, support from parents ($\beta = -.32, t = -2.40, p = .03$) and close friends ($\beta = -.89, t = -3.99, p < .001$), and CBP ($\beta = .30, t = 2.27, p = .04$) predicted siblings' total scores on the CDI-2. The model accounted for 82.0% of the variance in siblings' depressive symptoms ($F = 13.61, p < .001$). Subsequent analysis found that social support and CBP did not predict siblings' basal levels of cortisol (all $ps > 0.20$).

Discussion

This study examined the psychophysiological impact of childhood ASD on siblings. Data indicated that total depression symptoms were higher in siblings of children with ASD compared with a normative control group. Data suggested that group disparities with respect to depressive symptoms were driven more by emotional than functional problems. That is, ASD siblings scored higher on the emotional problems scale of the CDI-2; scores on the functional problems scale, while higher in siblings of children with ASD, approached, but failed to reach statistical significance. These findings are commensurate with several recent studies, in which depression and other psychological markers such as emotional problems and internalising behaviours were found to be higher in the context of having a brother/sister with ASD (Lobato et al., 2011; Petalas et al., 2009). These findings also resonate with those from a recent meta analysis, in which siblings of children with DD, including ASD, reported higher levels of depressive symptoms compared to a control group (Vermaes et al., 2012). That some studies have observed no effect (Benson & Karlof, 2008, Di Biasi et al., in press;

Tomeny et al., 2012) or an adaptive effect (Macks & Reeve, 2007) of childhood disability on siblings' psychological functioning might be explained by methodological variations. That is, many previous studies used parent reports to measure siblings' psychological adjustment to having a brother/sister with a DD. However, as used in the current study, sibling self report data has been found to be more reliable (Macks & Reeve, 2007). In fact, it has been suggested incumbent on researchers to collect psychological data directly from the participating sibling (Guite, Lobato, Kao, & Plante, 2004; Stoneman, 2005).

Groups were comparable with respect to basal HPA axis activity, such that both groups displayed the typical descending pattern of cortisol secretion across the day and were comparable on HPA markers, CAR magnitude and diurnal slope. This finding might be deemed unexpected given that atypical patterns of cortisol secretion have been observed in the context of other chronic childhood stressors such as family discord (Wolf et al., 2008), maltreatment (Gunnar et al., 2001) and peer victimization (Knack et al., 2011; Vaillancourt et al., 2011). Age related changes in basal stress hormone activity might provide one explanation. For example, in the current study, siblings of children with ASD (mean = 12.6 years) were, on average, almost two years older than controls (mean = 11.0 years). Basal (Gunnar, Wewerka, Frenn, Long, & Griggs, 2009) and reactive cortisol levels (Stroud et al., 2009) undergo marked changes during the transition from childhood to adolescence. As such, in the context of the current study, any effect of childhood ASD on siblings' basal HPA axis functioning might have been masked by age related variations. In addition, failure to observe cortisol disparities between the groups might reflect having recruited a relatively high functioning group of children with ASD. Indeed, recent research has highlighted the predictive value of ASD symptom severity for the physiological functioning of parental caregivers (Ruiz-Robeldillo et al., 2014).

Data reported here indicated that siblings' depressive symptoms were, at least in part, influenced by their availability of social support. Indeed, along with the behaviour problems of their brother/sister with ASD, social support accounted for a significant portion of the variance in siblings'

total scores on the CDI-2. In particular, support from parents and close friends inversely predicted siblings' depressive symptoms. These results reflect the fact that greater support, especially from family and close friends, might buffer against the psychological sequelae associated with having a brother/sister with ASD. These findings resonate with several other studies, in which socially supported siblings of children with a DD reported lower levels of depression (Barrera et al., 2004) and other psychological symptoms such as loneliness (Kaminsky & Dewey, 2002). The adaptive effect of social support for siblings' psychological adjustment to childhood disability was also highlighted in recent qualitative research. In a study by Moyson and Roeyers (2012), siblings of children with intellectual disabilities, including ASD, commented that support, particularly from parents and close friends, helped them forget about the stress associated with being a sibling. Moreover, social support groups, which allow siblings of children with a DD to come together, make friends and share experiences, have been linked with positive psychological changes such as fewer depressive symptoms (Houtzager, Grootenhuis, & Last, 2001). Based on findings reported here, interventions that enhance siblings' perceived availability of support, especially support from parents and close friends, might be adaptive for alleviating depressive symptoms.

Data also indicated that siblings' depressive symptoms were partially contingent on the behaviour problems of their brother/sister with ASD. That is, CBP were found to positively predict siblings' total CDI-2 scores. That CBP might affect psychological outcomes such as depression in siblings of children with a DD has also been observed in other recent studies (Mayer et al., 2011; Shivers et al., 2012). These findings might have implications for the design and delivery of psychotherapeutic interventions. For example, researchers might look to corroborate and extend the findings of Lobato and Kao (2005), in which positive psychological adjustments such as higher levels of self competence were reported by siblings who participated in a psychoeducational skills training intervention to learn more about the behavioural manifestations of ASD.

Findings of this study should be evaluated in the context of its limitations. First, the cross sectional nature of the work precludes drawing inferences about the relationship between study variables. As such, future studies might use longitudinal designs to better delineate the direction of the relationships between social support, CBP and depressive symptoms in siblings of children with ASD. Second, the small size of the sample is a notable limitation of the study, with post hoc power analysis indicating that (N=43) participants would be needed to provide adequate power (80%; $\alpha = .05$) to detect a moderate effect size ($f^2 = .15$). However, despite being underpowered, significant relationships between social support and CBP, and siblings' depressive symptoms were observed. Moreover, owing to the demands of the caregiving experience, parents of children with ASD (who were asked to oversee siblings' participation in the study) have little free time (Yantzi et al., 2006), thus making it difficult to engage with research opportunities. As such, the modest size of the sample, which matches the size of other studies of a similar nature (Quintero & McIntyre, 2010), might be expected. Third, characteristics of the child with ASD such as symptom severity (Rao & Beidel, 2009) and birth order (Tomeny, Barry & Bader, 2014) have been shown to be associated with the psychological adjustment of neuro-typical siblings. These important measures were omitted from the current study and, as such, caution should be exercised when interpreting the findings.

In conclusion, relative to a normative control group, siblings of children with ASD reported greater depressive symptoms. Data indicated that support from parents and close friends, and the behaviour problems of their brother/sister with ASD predicted siblings' total scores on the CDI-2. These findings might inform the design and delivery of interventions with a view to better supporting siblings of children with ASD, and their families.

Acknowledgements

We would like to thank the Baily Thomas Charitable Trust for funding this research.

Conflict of interest

None declared.

References

- Bandeira, D. R., Pawlowski, J., Gonçaves, T. R., Hilgert, J. B., Bozzetti, M. C., & Hugo, F. N. (2007). Psychological distress in Brazilian caregivers of relatives with dementia. *Aging & Mental Health, 11*(1), 14-19.
- Barak-Levy, Y., Goldstein, E., & Weinstock, M. (2010). Adjustment characteristics of healthy siblings of children with autism. *Journal of Family Studies, 16*, 155-164.
- Barlow, J. H., & Ellard, D. R. (2006). The psychosocial well-being of children with chronic disease, their parents and siblings: An overview of the research evidence base. *Child: Care, Health & Development, 32*(1), 19-31.
- Barrera, M., Fleming, C. F., & Khan, F. S. (2004). The role of emotional social support in the psychological adjustment of siblings of children with cancer. *Child: Care, Health & Development, 30*(2), 103-111.
- Bella, G. P., Garcia, M. C., Spadari-Bratfisch, R. C. (2011). Salivary cortisol, stress, and health in primary caregivers (mothers) of children with cerebral palsy. *Psychoneuroendocrinology, 36*, 834-842.
- Benson, P. R., & Karlof, K. L. (2008). Child, parent, and family predictors of latter adjustment in siblings of children with autism. *Research in Autism Spectrum Disorders, 2*, 583-600.
- Bhattacharyya, M. R., Molloy, G. R., & Steptoe, A. (2008). Depression is associated with flatter cortisol rhythms in patients with coronary artery disease. *Journal of Psychosomatic Research, 65*, 107-113.

- Brant, H., Wetherell, M. A., Lightman, S. L., Crown, A. L., & Vedhara, K. (2010). An exploration into physiologic and self-report measures of stress in pre-registration doctors at the beginning and end of a clinical rotation. *Stress, 13*(2), 155-162.
- Cantwell, J., Muldoon, O. T., & Gallagher, S. (2014). Social support and mastery influence the association between stress and poor physical health in parents caring for children with developmental disabilities. *Research in Developmental Disabilities, 35*, 2215-2223.
- Cantwell, J., Muldoon, O. T., & Gallagher, S. (in press). The influence of self-esteem and social support on the relationship between stigma and depressive symptomology in parents caring for children with intellectual disabilities. *Journal of Intellectual Disability Research*.
- Chida, Y., & Steptoe, A. (2009). Cortisol awakening response and psychosocial factors: A systematic review and meta-analysis. *Biological Psychology, 80*, 265-278.
- Clow, A., Hucklebridge, F., Stalder, T., Evans, P., & Thorn, L. (2010). The cortisol awakening response: More than a measure of HPA axis function. *Neuroscience and Biobehavioural Reviews 35*, 97-103.
- Cousino, M. K. & Hazen, R. A. (2013). Parenting Stress among caregivers of children with chronic illness: A systematic review. *Journal of Paediatric Psychology, 1-20*.
- Di Biasi, S., Trimarco, B., D'Ardia, C., Melogno, S., Meledandri, G., & Levi, G. (in press). Psychological adjustment, social responsiveness and parental distress in an Italian sample of siblings of children with high-functioning autism spectrum disorder. *Journal of Child & Family Studies*.
- Dyke, P., Mulroy, S., & Leonard, H. (2009). Siblings of children with disabilities: challenges and opportunities. *Acta Paediatrica, 98*, 23-24.
- Easter, G., Sharpe, L., & Hunt, C. J. (2015). Systematic review and meta-analysis of anxious and depressive symptoms in caregivers of children with asthma. *Journal of Paediatric Psychology, 40*(7), 623-32.

- Edwards, S., Hucklebridge, F., Clow, A., & Evans, P. (2003). Components of the diurnal cortisol cycle in relation to upper respiratory symptoms and perceived stress. *Psychosomatic Medicine*, *65*, 320-327.
- Fairthorne, J., de Klerk, N., & Leonard, H. (2015). Health of mothers of children with Intellectual disability or autism spectrum disorder: A review of the literature. *Medical Research Archives*, *3*, 1-21.
- Fisman, S., Wolf, L., Ellison, D., & Freeman, T. (2000). A longitudinal study of siblings of children with chronic disabilities. *Canadian Journal of Psychiatry*, *45*, 369-375.
- Fries, E., Dettenborn, L., & Kirschbaum, C. (2009). The cortisol awakening response (CAR): Facts and future directions. *International Journal of Psychophysiology*, *72*, 67-7.
- Gallagher, S., & Hannigan, A. (2014). Depression and chronic health conditions in parents of children with and without developmental disabilities: The growing up in Ireland cohort study. *Research in Developmental Disabilities*, *35*, 448-454.
- Gallagher, S., Phillips, A. C., Oliver, C., & Carroll, D. (2008). Predictors of psychological morbidity in parents of children with intellectual disabilities. *Journal of Paediatric Psychology*, *33*(10), 1129-1136.
- Gallagher, S., & Whiteley, J. (2012). Social support is associated with blood pressure responses in parents caring for children with developmental disabilities. *Research in Developmental Disabilities*, *33*, 2099-2105.
- Giallo, R., & Gavidia-Payne, S. (2006). Child, parent and family factors as predictors of adjustment for siblings of children with a disability. *Journal of Intellectual Disability Research*, *50*(12), 937-948.
- Griffith, G. M., & Hastings, R. P. (2013). He's hard work, but he's worth it. The experience of caregivers of individuals with intellectual disabilities and challenging behaviour: A meta-

- synthesis of qualitative research. *Journal of Applied Research in Intellectual Disabilities*, 27(5), 401-419.
- Guite, G., Lobato, D., Kao, B., & Plante, W. (2004). Discordance between sibling and parent reports of the impact of chronic illness and disability on siblings. *Children's Health Care*, 33, 77-92.
- Gunnar, M. R., Morison, S. J., Chisholm, K., & Schuder, M. (2001). Salivary cortisol levels in children adopted from Romanian orphanages. *Development and Psychopathology*, 13, 611-628.
- Gunnar, M. R., Wewerka, S., Frenn, K., Long, J. D., & Griggs, C. (2009). Developmental changes in hypothalamus–pituitary–adrenal activity over the transition to adolescence: Normative changes and associations with puberty. *Development and Psychopathology*, 21, 69-85.
- Harter, S. (1985). *The self-perception profile for children: Revision of the perceived competence scale for children*. Manual, University of Denver.
- Hayes, S. A., & Watson, S. L. (2013). The impact of parenting stress: A meta-analysis of studies comparing the experience of parenting stress in parents of children with and without autism spectrum disorder. *Journal of Autism and Developmental Disorders*, 43(3), 629-642.
- Holland J. M., Thompson, L. W., Tzuang, M., & Gallagher-Thompson, D. (2010). Psychosocial factors among Chinese American women dementia caregivers and their association with salivary cortisol: Results of an exploratory study. *Ageing International*, 35(2), 109-127.
- Houtzager, B. A., Grootenhuis, M. A., Last, B. F. (2001). Supportive groups for siblings of paediatric oncology patients: Impact on anxiety. *Psycho-Oncology*, 10, 315-324.
- Kaminsky, L., & Dewey, D. (2002). Psychosocial adjustment in siblings of children with autism. *Journal of Child Psychology and Psychiatry*, 43(2), 225-232.
- Knack, J. M., Jensen-Campbell, L. A., & Baum, A. (2011). Worse than sticks and stones? Bullying is associated with altered HPA axis functioning and poorer health. *Brain and Cognition*, 77, 183-190.

- Kovacs, M. (2011). *The Children's Depression Inventory 2: Manual*. North Tonawanda, NY: Multi-Health Systems, Inc.
- Kudielka, B. M., Gierens, A., Hellhammer, D. H., Wust, S., & Schlotz, W. (2012). Salivary cortisol in ambulatory assessment: Some do's, some don'ts, and some open questions. *Psychosomatic Medicine, 74*, 418-431.
- Kudielka, B. M., Hawkley, L. C., Adam, E. K., & Cacioppo, J. T. (2007). Compliance with ambulatory saliva sampling in the Chicago health, aging, and social relations study and associations with social support. *Annals of Behavioural Medicine, 34*(2), 209-216.
- Labay, L. E., & Walco, G. A. (2004) Brief report: Empathy and psychological adjustment in siblings of children with cancer. *Journal of Pediatric Psychology, 29*(4), 309-314.
- Lasikiewicz, N., Hendrickx, H., Talbot, D., & Dye, L. (2008). Exploration of basal diurnal salivary cortisol profiles in middle-aged adults: Associations with sleep quality and metabolic parameters. *Psychoneuroendocrinology, 33*, 143-151.
- Lobato, D. J., & Kao, B. T. (2005). Brief report: Family-based group intervention for young siblings of children with chronic illness and developmental disability. *Journal of Paediatric Psychology, 30*(8), 678-682.
- Lobato, D. J., & Kao, B. T., Plante, W., Seifer, R., Grullon, E., Cheas, L., ... et al. (2011). Psychological and school functioning of Latino siblings of children with intellectual disability. *Journal of Child Psychology and Psychiatry, 52*(6), 696-703.
- Lovell, B., Moss, M., & Wetherell, M. A. (2015). The psychophysiological and health corollaries of child problem behaviours in caregivers of children with autism and ADHD. *Journal of Intellectual Disability Research, 59*(2), 150-157.
- Lovell, B., Moss, M., & Wetherell, M. A. (2012). The psychosocial, endocrine and immune consequences of caring for a child with autism or ADHD. *Psychoneuroendocrinology, 37*, 534-542.

- Lovell, B., & Wetherell, M. A. (2011). The cost of caregiving: Endocrine and immune implications in elderly and non elderly caregivers. *Neuroscience and Biobehavioural Reviews*, 35, 1342-1352.
- Ludlow, A., Skelly, C., & Rohleder, P. (2012). Challenges faced by parents of children diagnosed with autism spectrum disorder. *Journal of Health Psychology* 17(5), 702-711.
- Macks, R. J., & Reeve, R. E. (2007). The adjustment of non-disabled siblings of children with autism. *Autism and Developmental Disorders*, 37, 1060–1067.
- Meyer, K. A., Ingersoll, B., & Hambrick, D. Z. (2011). Factors influencing adjustment in siblings of children with autism spectrum disorders. *Research in Autism Spectrum Disorders*, 5, 1413-1420.
- Moyson, T., & Roeyers, H. (2012). ‘The overall quality of my life as a sibling is all right, but of course, it could always be better’. Quality of life of siblings of children with intellectual disability: The siblings’ perspectives. *Journal of Intellectual Disability Research*, 56, 87-101.
- Neece, C. L., Blacher, J., & Baker, B. L. (2010). Impact on siblings of children with intellectual disability: The role of child behaviour problems. *American Journal on Intellectual and Developmental Disabilities*, 115(4), 291-306.
- Okun, M. L., Krafty, R. T., Buysse, D. J., Monk, T. J., Reynolds, C. F., Begley, A., . . . Hall, M. (2010). What constitutes too long of a delay? Determining the cortisol awakening response (CAR) using self report and PSG-assessed wake time. *Psychoneuroendocrinology*, 35, 460-468.
- Oruche, U. K., Gerkensmeyer, J., Stephan, L., Wheeler, C. A., & Hanna, K. M. (2012). The described experience of primary caregivers of children with mental health needs. *Archives of Psychiatric Nursing*, 26(5), 382-391.
- Petalas, M. A., Hastings, R. P., Nash, S., Lloyd, T., & Dowey, A. (2009). Emotional and behavioural adjustment in siblings of children with intellectual disability with and without autism. *Autism*, 13(5), 471-.483.

- Quintero, N., & McIntyre, L. L. (2010). Sibling adjustment and maternal well-being: An examination of families with and without a child with an autism spectrum disorder. *Focus on Autism and other Developmental Disabilities, 25*(1), 37-46.
- Randall, P., & Parker, J. (1999). *Supporting the families of children with autism*. New York, NY: John Wiley & Sons.
- Rao, P. A., & Beidel, D. C. (2009). The impact of children with high functioning autism on parental stress, sibling adjustment, and family functioning. *Behavior Modification, 33*, 437-451.
- Saxbe, D. E. (2008). A field (researcher's) guide to cortisol: Tracking HPA axis functioning in everyday life. *Health Psychology Review, 2*(2), 163-190.
- Seldenrijk, A., Hamer, M., Lahiri, A., Penninx, B. W. J. H., & Steptoe, A. (2012). Psychological distress, cortisol stress response and subclinical coronary calcification. *Psychoneuroendocrinology, 37*(1), 48-55.
- Seltzer, M. M., Almeida, D. M., Greenberg, J. S., Salva, J., Stawski, R. S., Hong, J., . . . Lounds-Taylor, J. (2009). Psychosocial and biological markers of daily lives of midlife parents of children with disabilities. *Journal of Health and Social Behaviour, 5*, 1-15.
- Seltzer, M. M., Greenberg, J. S., Hong, J., Smith, L. E., Almeida, D. M., Coe, C., et al. (2010). Maternal cortisol levels and behaviour problems in adolescents and adults with ASD. *Journal of Autism and Developmental Disorders, 40*(4), 457-469.
- Sharpe, D., & Rossiter, L. (2002). Siblings of children with a chronic illness: A meta-analysis. *Journal of Paediatric Psychology, 27*(8), 699-710.
- Shivers, C. M., Deisenroth, L. K., & Taylor, J. L. (2012). Patterns and predictors of anxiety among siblings of children with autism spectrum disorders. *Journal of Autism and Developmental Disorders, 43*(6), 1336-1346.
- Smith, A. M., & Grzywacz, J. G. (2014). Health and well-being in midlife parents of children with special health needs. *Families, Systems, & Health, 32*(3), 303-312.

- Smyth, J., Ockenfels, M., Gorin, A., Catley, D., Porter, L., Kirschbaum, C., ... et al. (1997). Individual differences in the diurnal cycle of cortisol. *Psychoneuroendocrinology*, *22*, 89-105.
- Stoneman, Z. (2005). Siblings of children with disabilities: Research themes. *Mental Retardation*, *43*, 339-350.
- Stroud, L., Foster, E., Handwerker, K., Papandonatos, G. D., Granger, D., Kivlighan, K. T., et al. (2009). Stress response and the adolescent transition: Performance versus peer rejection stress. *Development and Psychopathology*, *21*, 47-68.
- Tomeny, T. S., Barry, T. D., & Bader, S. H. (2012). Are typically developing siblings of children with an autism spectrum disorder at risk for behavioral, emotional, and social maladjustment? *Research in Autism Spectrum Disorders*, *6*, 508-518.
- Tomeny, T. S., Barry, T. D., & Bader, S. H. (2014). Birth order rank as a moderator of the relation between behavior problems among children with an autism spectrum disorder and their siblings. *Autism*, *18*(2), 199-202.
- Vaillancourt, T., Duku, E., Becker, S., Schmidt, L. A., Nicol, J., Muir, C., ... et al. (2011). Peer victimization, depressive symptoms, and high salivary cortisol predict poorer memory in children. *Brain and Cognition*, *77*, 191-199.
- Vermaes, I. P. R., van Susante, A. M. J., van Bakel, H. J. A. (2012). Psychological functioning of siblings in families of children with chronic health conditions: A meta-analysis. *Journal of Paediatric Psychology*, *37*(2), 166-184.
- Wolf, J. M., Nicholls, E., & Chen, E. (2008). Chronic stress, salivary cortisol, and a-amylase in children with asthma and healthy children. *Biological Psychology*, *78*, 20-28.
- Yantzi, M. N., Rosenberg, M. W., & McKeever, P. (2006). Getting out of the house: The challenges mothers face when their children have long term care needs. *Health & Social Care in the Community*, *15*, 45-55.