
Stephanie A. Hayes · Shelley L. Watson

Published online: 13 July 2012
© Springer Science+Business Media, LLC 2012

Abstract Researchers commonly report that families of children with autism spectrum disorder (ASD) experience more parenting stress than families of typically developing (TD) children or those diagnosed with other disabilities [e.g., Down syndrome (DS), cerebral palsy, intellectual disability]. The authors reexamined the research using comparison groups to investigate parenting stress and conducted a meta-analysis to pool results across studies. The experience of stress in families of children with ASD versus families of TD children resulted in a large effect size. Comparisons between families of children of ASD and families with other disabilities also generated a large effect size however, this result should be interpreted with caution as it may be associated with the specific experience of parenting a child with DS.

Keywords Autism spectrum disorder · Parenting stress · Family · Meta-analysis

Introduction

The experience of parenting any child can be stressful (Cameron et al. 1991) and yet researchers have suggested that families generally respond well and adjust in order to maintain stability and manage life’s challenges (Minnes 1988). Conversely, researchers have posited that families of a child diagnosed with a disability are negatively impacted and therefore experience more instability and dysfunction than “typical” families (see Watson et al. 2011, for a review). Of the various paradigms in family research aimed at capturing the experience of families of children with disabilities, the most widely investigated topic is that of parenting stress (Davis and Carter 2008; Pisula 2003). In particular, it has become common place for researchers to introduce their articles by stating that families with a child with an autism spectrum disorder (ASD) experience more stress than other families (e.g., Estes et al. 2009; Griffith et al. 2010; Hamlyn-Wright et al. 2007; Kasari and Sigman 1997; Wolf et al. 1989). Investigations of parenting stress are important as they provide a framework within which to identify key variables that may contribute to the experience of stress. Understanding what contributes to stress will then lead to more targeted interventions to support families and facilitate family functioning. Therefore it is valuable to pause and take stock of the current research on parenting stress focused on families of children with ASD to explore whether or not they are experiencing the most stress in comparison to other families.

What is Parenting Stress?

In its simplest definition, parenting stress is the experience of distress or discomfort that results from demands associated with the role of parenting (Deater-Deckard 1998). Frequently articles investigating the impact of a child with a disability on the family have used one or two factors associated with distress (e.g., depression, anxiety, or marital discord; Webster-Stratton 1990) as a primary indicator of stress (e.g., Benson and Karlof 2009; Davis and Carter 2008; Ekas and Whitman 2010) despite the challenges of establishing a relationship between stress and distress.
(Wolf et al. 1989). It is important to consider that distress may arise for many reasons that are not directly linked to having a child with a disability (e.g., pre-existing pathology or other environmental stressors). It is therefore necessary to be aware of our limited conceptualization of stress and that it is an oversimplification to measure stress based on one or two indicators of distress (Webster-Stratton 1990).

According to Folkman and Lazarus’s (1985) general model of stress, stress results from the interaction of an individual (or family) with their environment. When an individual deems that environmental stressors have overwhelmed their resources they engage coping mechanisms to restore functioning. However, if the individual’s coping mechanisms are either maladaptive or cannot meet the new demands, the outcome is stress. Although beyond the scope of this paper, the effects of stress may include physical and/or psychological symptoms such as depression, fatigue, restlessness, elevated neural and hormonal pathways or an increased risk for ulcers or heart diseases (Carpenter and Steffen 2004). What Folkman and Lazarus (1985) emphasized was that stress is individual and therefore subjective. In parallel, parenting stress is when the family is unable to restore functioning following the introduction of a stressor (related to parenting, i.e., a child’s difficult behavior) by engaging in their regular family-coping strategies.

The Impact of ASD on Parenting Stress

While it is valuable to apply a theoretical framework to investigations of parenting stress, it is also important to understand the broader familial context. Researchers have suggested that the experience of parenting stress may vary based on the specific diagnosis of disability of the child due to the associated behavioral phenotype, which are the expressions of behaviors related to a diagnostic label (e.g., intelligence, social skills, agreeableness; Dykens and Hodapp 2001; Hodapp et al. 1998; Seltzer et al. 2004). Therefore, examining the impact of ASD versus other diagnostic groups on the experience of families is helpful in furthering our understanding of both the unique and common experiences (Seltzer et al. 2004) associated with parenting stress.

Commonly, studies have compared an overall measure of stress between families of children with ASD to those of families of children with typical development (TD; Brobst et al. 2009; Hoffman et al. 2009; Lee et al. 2009; Rao and Beidel 2009) or those diagnosed with Down syndrome (DS), intellectual disability (with no known etiology; ID), cerebral palsy, fragile X syndrome, cystic fibrosis, fetal alcohol spectrum disorder (FASD), or externalizing behaviors (e.g., Abbeduto et al. 2004; Blacher and McIntyre 2006; Bouma and Schweitzer 1990; Dabrowska and Pisula 2010; Donenberg and Baker 1993; Eisenhower et al. 2005; Estes et al. 2009; Griffith et al. 2010; Hamlyn-Wright et al. 2007; Konstantareas et al. 1992; Pisula 2007; Watson et al. 2012; Wolf et al. 1989) and have identified higher rates of stress in families of children with ASD. In addition, researchers have identified higher incidence of depression and anxiety (e.g., Dumas et al. 1991; Eisenhower et al. 2005; Hamlyn-Wright et al. 2007; Koegel et al. 1992), less overall well-being (Blacher and McIntyre 2006) and more general life stress and daily hassles in parents of children with ASD when compared to control groups (Quintero and McIntyre 2010). In fact, it may be the challenging behaviors associated with the ASD phenotype that contribute to the overall experience of stress (Kasari and Sigman 1997; Wolf et al. 1989). For example, parents have reported that two of the key diagnostic traits of ASD, impairments in social communication (Bebko et al. 1987; Davis and Carter 2008) and restricted or repetitive behaviors (Gabriels et al. 2005), are particularly stressful. Researchers continue to identify child characteristics and ASD symptom severity as strongly associated with the experience of parenting stress (e.g., Brobst et al. 2009; Ekas and Whitman 2010; Estes et al. 2009; Hastings et al. 2006; Lecavalier et al. 2006; Tomanik et al. 2004). However, it is important to note that a recent study by Totsika et al. (2011) found that the impact of intellectual disability was distinct from the impact of ASD and that higher intellectual functioning of the diagnosed child did not appear to moderate a family’s experience of stress (Rao and Beidel 2009; Totsika et al. 2011) and therefore any family with a child on the ASD spectrum may be at an increased risk for parenting stress.

To the authors’ knowledge, no systematic review has yet been completed that summarizes the parenting stress literature comparing families of children with autism to children with TD or those with other disabilities. In addition, a review highlights not only what has been accomplished already in the literature, but provides researchers with an opportunity to identify where future studies may be needed. A meta-analysis was therefore conducted to summarize the variability among the outcomes of stress as reported by families of children with ASD in comparison to families of children with TD, and those diagnosed with other disabilities. Conducting a meta-analysis permits researchers to combine various studies addressing a common construct by calculating a standardized statistic known as an effect size and answers questions about the magnitude, variability and generalizability of findings (Field and Gillett 2010).

Methods

Search and Selection Criteria

Literature searches were conducted using various databases including PsychInfo, Scholars Portal, Dissertations
Measures

The purpose of this review was to examine the construct of parental stress in comparative studies of parents of children with ASD versus parents of TD children or those diagnosed with other disabilities. Consequently, it was important to ensure that the outcome variable of “stress” was not identified as “depression”. As a result, this meta-analysis only included studies where the outcome measures had been designed to capture the larger construct of parental stress. Tables 2 and 3 include a column identifying the measure used in each study included herein.

The majority of studies used established measures of parenting stress; however, one study included in the analysis used a researcher-developed measure with a reliability index of α = 0.76 (Hamlyn-Wright et al. 2007), designed to include factors outside of the parent–child relationship known to impact stress (e.g., support from educational and health professionals). Parents answered 12 questions such as “I find my child very hard work” or “I think others sometimes see me as a bad parent” using a 6-point Likert scale (where 1 was “strongly agree” and 6 was “strongly disagree”) and higher scores denoted more stress; Hamlyn-Wright et al. 2007).

Two other studies (Blacher and McIntyre 2006; Eisenhower et al. 2005) used the Family Impact Questionnaire developed by Donenberg and Baker (1993). Both of these studies used a “negative impact composite”, which included the Negative Impact on Relationships and the Negative Feelings Toward Parenting subscales (Donenberg and Baker 1993). The authors suggested that this composite score captured the perception of parents of the overall impact of their child on their family. The composite score contained 20 items endorsed using a 4-point Likert-like scale (where 0 is “Not at all”, and 3 is “Very much”; Donenberg and Baker 1993). Questions included “Compared to children and parents with children the same age as my child… I feel like I should have better control over his/ her behavior” (Donenberg and Baker 1993).

The remaining studies used different versions or interpretations of the two most commonly used parenting stress measures; the Questionnaire on Resources and Stress (QRS; Holroyd 1987) or the Parenting Stress Index (PSI; Abidin 1983). These two parenting stress measures were analyzed for concurrent validity and a strong bivariate correlation of their overall stress score was reported (r = 0.63, p < .001; Sexton et al. 1992). In addition, multivariate correlations were calculated using factor scores of the PSI and a short form of the QRS (QRS-F; Friedrich et al. 1983), indicating three concurrent functions; a general factor related to the adjustment of the family to the needs of the child, a factor related to the dependence and abilities of the child, and a factor related to the emotional reactions of the mother to their child (Sexton et al. 1992). The largest difference between the two measures was related to the physical abilities of the child, whereby the QRS includes a Physical Incapacitation subscale but the PSI subscales (Restrictions Imposed by Parental Role, Child Mood, Child Adaptability/Plasticity, Child Demandingness/Degree of Bother, and Child Distractibility/Hyperactivity) do not overlap (Sexton et al. 1992).
<table>
<thead>
<tr>
<th>Study</th>
<th>Parents by diagnosis</th>
<th>Age in years</th>
<th>Child’s age in years</th>
<th>Marital status (% married)</th>
<th>Additional population characteristics</th>
<th>ASD group characteristics</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Mothers of DS (59)</td>
<td>Latino: 49.5 (8.10)</td>
<td>Latino: 20.2 (2.80)</td>
<td>Latino 61.4</td>
<td>–</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Mothers of CP (87)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Mothers of ID (113)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>2. Bouma and Schweitzer (1990)</td>
<td>Mothers of ASD (24)</td>
<td>–</td>
<td>7.7 (2.50)</td>
<td>95.8</td>
<td>–</td>
<td>Autism with moderate to severe intellectual disabilities</td>
</tr>
<tr>
<td></td>
<td>Mothers of CF (24)</td>
<td>–</td>
<td>8.4 (2.23)</td>
<td>83.3</td>
<td>–</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Mothers of TD (24)</td>
<td>–</td>
<td>8.3 (2.39)</td>
<td>66.7</td>
<td>–</td>
<td></td>
</tr>
<tr>
<td>3. Brobst et al. (2009)</td>
<td>Mothers of ASD (25)</td>
<td>38.48 (6.54)</td>
<td>6.6 (2.66)</td>
<td>100.0</td>
<td>–</td>
<td>Autism, Asperger’s syndrome, or PDD-NOS</td>
</tr>
<tr>
<td></td>
<td>Fathers of ASD (25)</td>
<td>40.13 (7.36)</td>
<td>6.8 (3.19)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Mothers of TD (20)</td>
<td>35.10 (5.52)</td>
<td>6.8 (3.19)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>4. Eisenhower et al. (2005)</td>
<td>Parents of ASD (14)</td>
<td>Mothers 35.6 (5.60)</td>
<td>3.0 (0.30)</td>
<td>85.7</td>
<td>No n reported for Mothers versus Fathers</td>
<td>No Asperger’s diagnoses, all with intellectual disabilities</td>
</tr>
<tr>
<td></td>
<td>Mothers of TD (136)</td>
<td>Fathers 35.4 (4.90)</td>
<td>2.9 (0.26)</td>
<td>88.2</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Parents of ID (43)</td>
<td>Mothers 31.8 (6.70)</td>
<td>3.0 (0.21)</td>
<td>69.8</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Parents of DS (12)</td>
<td>Mothers 33.6 (5.60)</td>
<td>2.9 (0.30)</td>
<td>91.7</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Fathers of TD (20)</td>
<td>36.20 (5.94)</td>
<td>6.8 (3.19)</td>
<td>70.0</td>
<td></td>
<td></td>
</tr>
<tr>
<td>5. Estes et al. (2009)</td>
<td>Mothers of ASD (50)</td>
<td>35.99 (5.29)</td>
<td>3.6 (0.36)</td>
<td>92.0</td>
<td>–</td>
<td>Autism 71 %, PDD-NOS 29 %</td>
</tr>
<tr>
<td></td>
<td>Mothers of ID (22)</td>
<td>36.23 (4.55)</td>
<td>3.6 (0.37)</td>
<td>100.0</td>
<td>–</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Mothers of DS (19)</td>
<td>38.63 (5.71)</td>
<td>10.16 (3.86)</td>
<td>73.7</td>
<td>–</td>
<td>Low average IQ 3 %, moderate ID 37 %, and severe/profound ID 60 %</td>
</tr>
<tr>
<td></td>
<td>Mothers of ID (19)</td>
<td>43.89 (13.12)</td>
<td>9.98 (4.04)</td>
<td>84.2</td>
<td></td>
<td></td>
</tr>
<tr>
<td>6. Griffith et al. (2010)</td>
<td>Parents of ASD (18)</td>
<td>34.3</td>
<td>4.90</td>
<td>88.0</td>
<td>Fathers n = 8</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Parents of ID (18)</td>
<td>33.9</td>
<td>5.50</td>
<td>67.0</td>
<td>Fathers n = 2</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Parents of TD (18)</td>
<td>33.7</td>
<td>4.30</td>
<td>72.0</td>
<td>Fathers n = 2</td>
<td></td>
</tr>
<tr>
<td>7. Guess (1996) unpublished dissertation</td>
<td>Parents of ASD (265)</td>
<td>43.98 (7.73)</td>
<td>13.51 (7.65)</td>
<td>82.6</td>
<td>Fathers n = 20</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Parents of DS (223)</td>
<td>43.35 (7.00)</td>
<td>12.35 (7.43)</td>
<td>78.0</td>
<td>Fathers n = 15</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Parents of TD (131)</td>
<td>43.44 (7.66)</td>
<td>12.35 (4.47)</td>
<td>90.8</td>
<td>Fathers n = 7</td>
<td></td>
</tr>
<tr>
<td>8. Hamlyn-Wright et al. (2007)</td>
<td>Mothers of ASD (104)</td>
<td>37.52 (7.63)</td>
<td>8.61 (2.77)</td>
<td>69.2</td>
<td>–</td>
<td>Autism with ID 38 %, or with other co-morbid condition 15 %</td>
</tr>
<tr>
<td></td>
<td>Mothers of TD (342)</td>
<td>34.85 (8.15)</td>
<td>8.03 (3.61)</td>
<td>70.5</td>
<td></td>
<td></td>
</tr>
<tr>
<td>9. Hoffman et al. (2009)</td>
<td>Parents of ASD (89)</td>
<td>42.2 (6.20)</td>
<td>9.5 (2.00)</td>
<td>83.3</td>
<td>Fathers n = 27</td>
<td>Asperger’s, PDD-NOS, High Functioning Autism (IQ ≥ 70), and Autism</td>
</tr>
<tr>
<td></td>
<td>Parents of TD (46)</td>
<td>38.5 (5.40)</td>
<td>9.7 (2.10)</td>
<td>82.6</td>
<td>Fathers n = 14</td>
<td></td>
</tr>
<tr>
<td>10. Lee et al. (2009)</td>
<td>Parents of ASD (15)</td>
<td>42.5</td>
<td>Range 8–14</td>
<td>–</td>
<td>Fathers n = 3</td>
<td>High Functioning Autism (IQ ≥ 85)</td>
</tr>
<tr>
<td></td>
<td>Parents of TD (14)</td>
<td>41.6</td>
<td></td>
<td></td>
<td>Fathers n = 2</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Parents of ADHD (47)</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td>Parents of TD (22)</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td></td>
<td></td>
</tr>
<tr>
<td>12. Rao and Beidel (2009)</td>
<td>Parents of ASD (15)</td>
<td>42.5</td>
<td>Range 8–14</td>
<td>–</td>
<td>Fathers n = 3</td>
<td>High Functioning Autism (IQ ≥ 85)</td>
</tr>
<tr>
<td></td>
<td>Parents of TD (14)</td>
<td>41.6</td>
<td></td>
<td></td>
<td>Fathers n = 2</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Mothers of FXS (51)</td>
<td>–</td>
<td>5.9 (4.70)</td>
<td>–</td>
<td></td>
<td></td>
</tr>
<tr>
<td>14. Watson et al. (2012)</td>
<td>Parents of ASD (17)</td>
<td>41.29 (8.65)</td>
<td>9.54 (5.13)</td>
<td>100.0</td>
<td>Fathers n = 8</td>
<td>Asperger’s, PDD-NOS, and Autism</td>
</tr>
<tr>
<td></td>
<td>Parents of FASD (19)</td>
<td>53.37 (9.16)</td>
<td>18.40 (10.10)</td>
<td>84.0</td>
<td>Fathers n = 7</td>
<td></td>
</tr>
</tbody>
</table>
Parenting Stress Index

The PSI was originally developed by Abidin (1983) and has undergone several revisions with the most recent published in 1995 (PSI-3; Abidin 1995). It is a parent self-report questionnaire and is available in a long form (101-items) or a short form (36-items). According to the developer, the PSI was intended as a screening instrument used to identify parent–child systems at risk for dysfunction (Loyd and Abidin 1985). Both the short and long forms ask parents to read a statement and answer based on a 5-point Likert-scale (where 1 is “strongly disagree” and 5 is “strongly agree”) and examples include; “My child rarely does things for me that make me feel good” or “My child gets upset easily over the smallest thing”.

The long form yields a total stress score, and two general domain scores (Child Domain and Parent Domain) and an optional Life Stress Scale. Scores above the 75th or 90th percentile (depending on how conservative the researcher is) are indicative of significant stress and may require referral for professional interventions or consultation (Abidin 1995; Loyd and Abidin 1985). The Child Domain contains six subscales addressing a variety of child characteristics which may be impacting the parent–child system including; Adaptability, Reinforces Parent, Distractibility/Hyperactivity, Demandingness, Mood and Acceptability (Abidin 1995). The Parent Domain contains seven subscales addressing various sources of stress which may impact parental functioning including; Competence, Isolation, Attachment, Health, Role Restriction, Depression, and Spouse (Abidin 1995).

The PSI short form (PSI-SF) is similar to the long form as it provides a total stress score; however, it re-interprets the subscales and domains and provides three subscale scores; Parental Distress, Parent–Child Dysfunctional Interaction, and Difficult Child (Abidin 1995). The PSI-SF was a factor-analysis of the long form and the correlation for the total stress score between the two versions was \( r = 0.94, p < .001 \) (Abidin 1995). According to the author, both PSI versions capture stress of parents of children between the ages of 1 month and 12 years (Abidin 1995); however, they have frequently been used with parents of children with disabilities or chronic illness of any age (e.g., Fedele et al. 2010; Hastings et al. 2006). A recent analysis of the PSI-SF using item response theory, suggests that both the parent–child dysfunctional interaction and the difficult child subscales should be used with caution with children with ASD (and likely children with other disabilities; Zaidman-Zait et al. 2010). Results of the study by Zaidman-Zait et al. (2010) suggest that some items did not adequately discriminate the total severity of stress experienced by parents because they were either too easy or too difficult to endorse. In addition, one study included in this meta-analysis (Rao and Beidel 2009) used the Stress Index for Parents of Adolescents (Sheras et al. 1998), which is an adaptation of the PSI designed for parents of adolescents.

Questionnaire on Resources and Stress

The QRS can be used with any age-range and was originally developed by Holroyd (1974) to assess the negative impact having a child with a disability has on the family (Konstantareas et al. 1992). The QRS contains 285-items and asks parents to answer “true” or “false” to items measuring 15 dimensions related to family stress; Poor Health/Mood, Excessive Time Demands, Negative Attitude Toward Index Case, Overprotection/Dependency, Lack of Social Support, Overcommitment/Martyrdom, Pessimism, Lack of Family Integration, Limits on Family Opportunity, Financial Problems, Physical Incapacitation, Lack of

<table>
<thead>
<tr>
<th>Table 1</th>
<th>Study</th>
<th>Parents by diagnosis (n)</th>
<th>Age in years M (SD)</th>
<th>Child’s age in years M (SD)</th>
<th>Marital status (% married)</th>
<th>Additional population characteristics</th>
</tr>
</thead>
<tbody>
<tr>
<td>15. Wolf et al. (1989)</td>
<td>Mothers of ASD (30)</td>
<td>33.97 (11.67)</td>
<td>9.34 (4.16)</td>
<td>–</td>
<td>–</td>
<td>ID in 61 %</td>
</tr>
<tr>
<td></td>
<td>Fathers of ASD (27)</td>
<td>38.31 (8.05)</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Mothers of DS (30)</td>
<td>37.13 (9.14)</td>
<td>9.11 (4.21)</td>
<td>–</td>
<td>–</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Fathers of DS (29)</td>
<td>39.69 (7.59)</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Mothers of TD age-matched (31)</td>
<td>33.46 (8.45)</td>
<td>7.62 (4.43)</td>
<td>–</td>
<td>–</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Mothers of TD IQ-matched (31)</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Fathers of TD age-matched (30)</td>
<td>38.76 (7.21)</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td></td>
</tr>
<tr>
<td></td>
<td>Fathers of TD IQ-matched (31)</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td>–</td>
<td></td>
</tr>
</tbody>
</table>
Activities, Occupational Limitations, Social Obtrusiveness, and Difficult Personality Characteristics (Holroyd 1988). The QRS seeks to identify multiple variables that may interact and impact the child, parent, and family system (Holroyd 1988). Limitations of this measure (e.g., the length, the binary forced choice format, and negative bias) have led to numerous revisions and short forms (Konstantareas et al. 1992).

This meta-analysis contains studies that used four different short forms of the QRS. The original author proposed a 66-item version referred to as the QRS-short form (QRS-SF; Holroyd 1987), while other authors have proposed a 78-item version called the Clarke modification (QRS-C; Konstantareas et al. 1992) or a 52-item version referred to as the QRS-F (Friedrich et al. 1983). The QRS-F is the most commonly used short form in published research and its psychometric integrity has been established (Scott et al. 1989). The QRS-F provides a total stress score as well as four factor scores related to; Parent and Family Problems, Pessimism, Child Characteristics, and Physical Incapacitation (Friedrich et al. 1983). The QRS-F includes items such as “Our family agrees on important matters” or “I worry about what will happen to ____ when I can no longer take care of him/her” (Friedrich et al. 1983).

Importantly, the QRS has been criticized for its lack of validation with specific populations (Honey et al. 2005). To address this issue, the QRS-F was administered to parents of children with ASD and Honey et al. (2005) proposed a new interpretation based on 31-items (Parent and Family Problems, Pessimism) that omitted the Child Characteristics and Physical Incapacitation subscales because they over-emphasized abilities (or the lack thereof) inherently associated with a diagnosis of ASD. The studies by Griffith et al. (2010) and Watson et al. (2012) included in the meta-analysis used the 31-item interpretation based on these recommendations.

Calculation of Effect Sizes

In general, an effect size is a standardized indicator of the strength of the relationship between two outcome variables and allows for comparison of measures that employ different scales (Cohen 1992; Field and Gillett 2010). According to Cohen (1992), an effect size of 0.10 is considered small, 0.30 is medium, and anything above 0.50 is large. In this meta-analysis, effect sizes were calculated between the target group (parents of children with ASD) and the comparison group (TD children or those with other diagnosed disabilities). Tables 2 and 3 summarize the data from the studies included. The effect size $d$ was calculated and used in the analysis as recommended by Hedges and Olkin (1985) as an unbiased estimate. Also known as Hedges’ $g$, this procedure is preferred when there are uneven sample sizes between groups as it corrects any overestimate associated with small sample sizes while having a minor effect on larger studies (Hedges and Olkin 1985). To calculate the effect size, the following equation was used:

\[
\text{Effect size } = \frac{M_{\text{ASD}} - M_{\text{TD}}}{SD_{\text{pooled}}}
\]

where $M_{\text{ASD}}$ and $M_{\text{TD}}$ are the means of the target and comparison groups, respectively, and $SD_{\text{pooled}}$ is the pooled standard deviation.

<table>
<thead>
<tr>
<th>Study</th>
<th>Participant group</th>
<th>ASD group</th>
<th>TD group</th>
<th>Measure</th>
<th>Effect size</th>
<th>95% CI</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>n</td>
<td>M</td>
<td>SD</td>
<td>n</td>
<td>M</td>
<td>SD</td>
</tr>
<tr>
<td>Bouma and Schweitzer (1990)</td>
<td>Mothers</td>
<td>24</td>
<td>29.08</td>
<td>7.30</td>
<td>24</td>
<td>13.17</td>
</tr>
<tr>
<td>Brobst et al. (2009)</td>
<td>Mothers</td>
<td>25</td>
<td>101.71</td>
<td>23.48</td>
<td>20</td>
<td>66.00</td>
</tr>
<tr>
<td></td>
<td>Fathers</td>
<td>25</td>
<td>92.79</td>
<td>24.22</td>
<td>20</td>
<td>66.10</td>
</tr>
<tr>
<td>Eisenhower et al. (2005)</td>
<td>Parents</td>
<td>14</td>
<td>29.70</td>
<td>15.50</td>
<td>136</td>
<td>29.30</td>
</tr>
<tr>
<td>Guess (1996) unpublished dissertation</td>
<td>Mothers</td>
<td>17</td>
<td>264.71</td>
<td>56.71</td>
<td>18</td>
<td>247.50</td>
</tr>
<tr>
<td>Hamlyn-Wright et al. (2007)</td>
<td>Parents</td>
<td>265</td>
<td>45.66</td>
<td>8.61</td>
<td>342</td>
<td>94.79</td>
</tr>
<tr>
<td>Hoffman et al. (2009)</td>
<td>Mothers</td>
<td>104</td>
<td>147.90</td>
<td>25.70</td>
<td>131</td>
<td>29.30</td>
</tr>
<tr>
<td>Lee et al. (2009)</td>
<td>Parents</td>
<td>89</td>
<td>91.52</td>
<td>20.88</td>
<td>46</td>
<td>60.71</td>
</tr>
<tr>
<td>Markham (2000) unpublished dissertation</td>
<td>Parents</td>
<td>28</td>
<td>129.49</td>
<td>22.27</td>
<td>22</td>
<td>125.16</td>
</tr>
<tr>
<td>Rao and Beidel (2009)</td>
<td>Parents</td>
<td>15</td>
<td>266.67</td>
<td>43.91</td>
<td>14</td>
<td>198.71</td>
</tr>
<tr>
<td>Wolf et al. (1989)</td>
<td>Mothers</td>
<td>30</td>
<td>136.83</td>
<td>21.38</td>
<td>62</td>
<td>98.25</td>
</tr>
<tr>
<td></td>
<td>Fathers</td>
<td>27</td>
<td>130.96</td>
<td>21.29</td>
<td>61</td>
<td>97.34</td>
</tr>
</tbody>
</table>

TD = typically developing, CI = confidence interval, QRS-F = Questionnaire of Resources and Stress—Friedrich version, FIQ = Family Impact Questionnaire—Negative Impact composite, RD = Researcher developed, PSI-SF = Parenting Stress Index—Short Form, PSI = Parenting Stress Index, PSI+ = PSI administered to families with a child under 12, and the Stress Index for Parents of Adolescents for those over 12, PSIchild = only the PSI Child Domain.
The effect size \( (d_i) \) was determined for each individual study by subtracting the mean of the comparison group \( (X^c_i) \) from the mean of the target group \( (X^t_i) \) and dividing it by the weighted and pooled standard deviation \( (sd^*) \). In addition, 95% confidence intervals (CI) were calculated (see Figs. 1, 2 for forest-plots of the individual effect sizes and CI). It is important to note that if the CI contains zero, the assumption is that there is no difference between the levels of stress reported by parents of children with ASD in comparison to the other group.

Method of Meta-analysis

A random-effects model was assumed and methodology outlined by Hedges and colleagues (Hedges and Olkin 1985; Hedges and Vevea 1998) and Field and Gillett (2010) was followed. A random-effects model is particularly salient for this analysis because of the differences between the comparison groups. Therefore if the studies are truly capturing what they are intending (i.e., that the experience differs between families based on the behavioral phenotype associated with a specific diagnosis of disability), the population samples included in the studies here are highly variable. Random-effect models result in larger effect size confidence intervals than fixed effect models; however, procedures were utilized in an attempt to minimize this difference (Borenstein et al. 2007). Instead of using sample size as a means to weight various studies, inverse variance was chosen to weight effect sizes by their standard error in order to account for both the between and within study sampling error (Borenstein et al. 2007; Sutton and Higgins 2008).
Fig. 1 Forest-plot of the effect sizes of included studies comparing parenting stress in families of children with ASD to those who are typically developing and results of the combined ASD versus TD meta-analysis.

Fig. 2 Forest-plot of the effect sizes of included studies comparing parenting stress in families of children with ASD to those with children with other disabilities and results of the combined ASD versus other meta-analysis.
Combining Effect Sizes

In some instances, researchers reported multiple effect sizes (e.g., studies with an ASD group and more than one comparison group, or studies that included subscale scores and did not report an overall stress score). Including multiple effect sizes from the same study may bias results; therefore, a simple average was computed as suggested by Rosenthal (1991) for any study that contained more than one outcome. For the study by Watson et al. (2012), which reported outcome measures for both the PSI and QRS-F, the 31-item QRS-F score, considered to be the most conservative stress indicator was included in the meta-analysis.

Heterogeneity

Q statistics were conducted for every analysis to determine if any violations of the assumption of homogeneity were detected for the distribution of effect sizes (Field and Gillett 2010). Q is a Chi-square test of homogeneity and if found to be significant ($z \geq 0.05$), the null hypothesis is that there is variation in the effect sizes associated with heterogeneity (Huedo-Medina et al. 2006). For all analyses conducted, the $Q$ test was significant, suggesting that the studies included here reflect heterogeneity and thus the use of a random-effects model was supported (Huedo-Medina et al. 2006). In addition, the $I^2$ statistic was calculated (Higgins and Thompson 2002) as an indicator of the impact of heterogeneity and as a compliment to the $Q$ statistic (Huedo-Medina et al. 2006). According to the classification system proposed by Higgins and Thompson (2002), the $I^2$ indexes calculated for the analyses comparing families of children with ASD to TD children or to another diagnosis of disability, indicated low heterogeneity (approximately 16–17% of the variability of effect sizes can be attributed to something other than sampling error).

Publication Bias

In order to address publication bias, which is the likelihood that more articles are accepted for publication if they contain significant results, the author included unpublished PhD dissertations and calculated two indices of publication bias. The first test of publication bias is Rosenthal’s “fail-safe $N$”, which estimates the number of studies required to negate significant findings (Field and Gillett 2009; Rosenthal 1995). The second test of publication bias applied Kendall’s tau to the standardized effect size and its associated variance, where a significant correlation suggests a publication bias (Field and Gillett 2009). Publication bias will be discussed further within the context of the results of each meta-analysis. All analyses reported in this article were conducted in PASW (Version 18.0) using a modified version of the syntax provided by Field and Gillett (2009, 2010).

Results

The Impact of ASD Versus Typically Developing Children on Parenting Stress

A meta-analysis was conducted to compare outcome measures of parenting stress between parents of children with ASD and parents of children who have TD. Studies included are described in Table 2. Based on a random-effects model (Hedges and Vevea 1998), the mean effect size was 1.58; the 95% CI was 1.16 (lower limit) to 2.0 (upper limit), with an associated $z = 7.36$, $p < .001$. According to Cohen’s (1992) guidelines, the overall effect size was large. As previously described, publication bias was assessed using two methods, results of Rosenthal’s fail-safe $N$ estimated that an additional 2,454 studies would be required to negate these findings, and Kendall’s tau was not significant, suggesting that there is a true difference between the experience of parents of children with ASD in comparison to those with children who have TD on comprehensive measures of parenting stress.

The Impact of ASD Versus Other Disabilities on Parenting Stress

A second meta-analysis comparing an outcome measure of parenting stress in parents of children with ASD to parents of children diagnosed with other disabilities was conducted. A summary of the 12 studies included as well as their individual effect sizes displayed by disability are provided in Table 3. Based on a random-effects model (Hedges and Vevea 1998), the mean effect size was 0.64; the 95% CI was 0.25 (lower limit) to 1.03 (upper limit), with an associated $z = 3.20$, $p < .001$. According to Cohen’s (1992) guidelines, the overall effect size is large (while the lower limit of the CI represents a small effect size). As previously described, publication bias was assessed using two methods, results of Rosenthal’s fail-safe $N$ estimated that an additional 361 studies would be required to negate these findings, and Kendall’s tau was not significant, suggesting that there is a true difference between the experience of stress between parents of children with ASD in comparison to those with children diagnosed with other disabilities.
Discussion

As suggested in the “Introduction” section of many journal articles, parenting a child with ASD is associated with greater parenting stress. This statement holds true when families of a child with ASD are compared to families of a child with TD or families of a child diagnosed with another disability. The overall effect size calculated was large for both analyses and thus suggests that parenting stress in families with a child diagnosed with ASD is a significant experience that warrants attention and intervention. Finding ways to moderate or mediate parenting stress may facilitate a family’s functioning. Furthermore, from a methodological standpoint, this analysis suggests that including families with a child who has TD as a comparison group for families of children with ASD when researching the construct of parenting stress will consistently result in significant findings.

Limitations and Suggestions for Future Research

Consideration needs to be made when designing future research on parenting stress, as it is not enough to compare families of children with ASD to others with children who have TD as “simply finding and reporting differences does not provide an explanation for those differences” (Seltzer et al. 2004, p. 46). Therefore efforts need to be made to identify and control for other variables such as child characteristics, family sociodemographics, biological and psychological vulnerability (Seltzer et al. 2004). Within family sociodemographics, many variables such as gender are important to examine further. As previously discussed, research is unclear about the differences between mothers and fathers. Table 1 provides information (when available) about the parent participants and highlights that the majority of research has been conducted on mothers only, or on parents without differentiating between mothers and fathers.

When considering some of the limitations of this systematic review, one challenge is the number of father participants included in research. Due to limited information, this meta-analysis could not make any conclusions about the overall effect of parenting stress on fathers in comparison to mothers. Beyond parent variables, it is also important to discuss child variables. It is of note to highlight that children with ASD as a group are highly heterogeneous and researchers commonly include the full range of ASD spectrum diagnoses as one homogeneous category. For example, Table 1 includes a column titled “ASD Characteristics” that reports group characteristics (when available) for the individual studies included in this analysis. As a source of confounding variables, the variability in the phenotype of ASD becomes a more salient issue when comparing families of children with ASD to those with children with other disabilities.

It is important to highlight the variability of results between the studies comparing children with ASD to other disabilities. Despite including five studies (Blacher and McIntyre 2006; Eisenhower et al. 2005; Guess 1996; Markham 2000; Watson et al. 2012) where the CI included zero (suggestive of no difference in stress between groups; see Fig. 2), the overall effect size still suggests that parents of children with ASD are impacted more by parenting stress. Furthermore, some studies that met inclusion criteria and may have strengthened the findings of this analysis were omitted due to lack of information (commonly due to not reporting the standard deviation of the mean). It is important to note that due to the limited number of studies per disability category, separate analyses by disability diagnosis were not possible at this time. However, it is also important to consider possible confounding variables related to shared child characteristics between the disability groups. For example, there are inherent difficulties related to high rates of comorbid disorders with ASD. Simonoff et al. (2008) found that seventy percent of participants in a population-based study diagnosed with ASD also qualified for at least one more comorbid disorder. The most commonly reported disorders include anxiety, attention-deficit/hyperactivity disorder (ADHD), depression, obsessive compulsive disorder (OCD) and oppositional defiance disorder (ODD; Kim et al. 2000; Leyfer et al. 2006; Mayes et al. 2011; Simonoff et al. 2008). Thus the high variability in the experience of ASD may confound results when comparing families of a child with ASD to families of children with other disabilities as many of these characteristics are present in other disabilities. For example, when trying to compare children with ASD to those with ADHD, it is important to consider that despite many children not having a formal diagnosis of ADHD, they may display many of the core characteristics of hyperactivity or impulsivity (Mayes et al. 2011). If characteristics of other disorders are present (which is more common than not), there are inherent challenges associated with comparative research in this domain. As suggested by Seltzer et al. (2004), future studies should either attempt to match participants by behavioral phenotypes or control for these effects statistically.

When discussing the differences between ASD and other disabilities, it is of note to highlight results in Table 3 where the largest contributing effect size originated from studies comparing families of children with ASD to those with children with Down syndrome (also shown in Fig. 2). The large effect may be reflective of what researchers have termed the “Down syndrome advantage”, which highlights the behavioral phenotype of Down syndrome as being more social and associated with fewer behavioral challenges.
(Esbensen and Seltzer 2011; Seltzer et al. 2004). When looking at the individual results of the other disability categories, there is more variability. Although comparing groups based on a diagnosis is beneficial (Dykens and Hodapp 2001; Hodapp et al. 1998), comparisons may miss key variables associated with the behavioral phenotype such as diagnosis severity, if it is a “visible” diagnosis (e.g., ASD is an “invisible” diagnosis as compared to Down syndrome), the predictability of the course of the disability, and other associated medical complications (Gupta 2007).

As previously discussed, researchers have suggested that the core deficits associated with ASD such as impairments in social communication, or restrictive/repetitive behaviors are the most stressful for parents. To investigate specific behaviors through comparison studies, it would therefore be important to control for all other variables and to include comparison groups that share similar behaviors such as those diagnosed with OCD. Another alternative may be to compare families based on the behavioral phenotype of their child with ASD in a cross-sectional design such as that employed by Totsika et al. (2011). Therefore the question remaining for comparative researchers regarding families of children with ASD is no longer “are families with ASD more stressed than families without ASD” but “why are families under more stress and what are the specific moderators of stress that facilitate family resilience?”

In order to address “why” families are under more stress will require a better operational definition of stress that aims to unite theory and measurement, as well as a better way to compare specific behavioral phenotypes of children with disabilities. Many variables previously mentioned and some additional hypotheses for future investigation related to the differences between disabilities include (but are not limited to) the range, severity or frequency of challenging behaviors exhibited by children with ASD. In addition, the presence of other comorbid disorders, the “invisibility” status, and the prognosis and predictability of the course of the disability (including any associated strengths or weaknesses) are important to differentiate in future research. The availability of services and supports, the timing and process of diagnosis (e.g., receiving a medical vs. behavioral-based diagnosis), the attitude of professionals or the community in general about the disability, as well as other individual differences of parents and families are but a few suggestions for future studies.

In addition, resilience has emerged as a new topic of interest in family research as a means to emphasize healthy, positive family functioning in the face of chronic stress in an attempt to identify specific moderators and mediators of stress (Ylven et al. 2006). Research focused on resiliency aims to provide a more balanced description of the family experience of parenting stress. Many researchers suggest that the true experience of families should include an emphasis on family strengths and investigations of the positive impact of having a child with a disability can have on the family (e.g., Blacher and Baker 2007; Dykens 2006; Hastings et al. 2002; Hastings et al. 2005; Kayfitz et al. 2010; Scorgie and Sobsey 2000; Stainton and Besser 1998; Taunt and Hastings 2002). Since family functioning is a complex system and a diagnosis of disability can impact families in different ways (Seligman and Darling 2007; Sloman and Konstantareas 1990), it becomes important to identify and link positive factors (Gerstein et al. 2009) within the context of the broader construct of parenting stress. Introducing positive factors within a pre-existing framework of parenting stress will better our understanding of how stress can be moderated or mediated and thus how to facilitate positive family functioning.

Summary

Findings of this meta-analysis suggest that parents of children with ASD experience more parenting stress than those of children who have TD or another disability. However, it is important to highlight that the experience of parenting stress is not the sole experience for parents of children diagnosed with ASD. Although there has been a rich research tradition of cataloguing stress, there has been a paucity of research examining positive parental characteristics that may reduce the impact stress has on the family (Bayat 2007). As previously discussed, early interventions targeting the reduction of parenting stress also facilitate positive changes in the child’s ASD related behaviors. Therefore reducing parenting stress as an early intervention may facilitate familial functioning by restoring balance and thus moderating and reducing the effects of challenging behaviors. Reducing parenting stress may trigger an iterative process, whereby lessening challenging behaviors will minimize future experiences of parenting stress by providing parents with the skills necessary to overcome potential future challenging behaviors. It is important to remember that there is much more to the story of families of children with ASD beyond the experience of parenting stress and it is our responsibility as researchers to identify factors that facilitate family functioning and foster hope for the future.

Acknowledgments The authors would like to acknowledge funding received from the Social Sciences and Humanities Research Council. This paper was submitted as partial requirement for the first author’s Master’s degree. The first author would also like to thank Dr. Andy Field for his fantastic statistics website and resources.
References

References marked with an asterisk (*) indicate studies included in the meta-analysis.


