

A Meta-Analysis on Adjustment Difficulties in Siblings of Children With and Without Autism Spectrum Disorder

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ABSTRACT:

This meta-analytic study aimed to examine how the presence of Autism Spectrum Disorder (ASD) in a child affects their sibling(s)'s social, emotional, and behavioral adjustment. Many studies have investigated how the presence of ASD in a child affects their sibling(s) in social, emotional, and behavioral adjustment, but their results are mixed. Moreover, the previous reviews did not address the personal characteristics that account for the differences in siblings' adjustment. This review focused on examining the three primary domains of adjustment and the moderating factors of their adjustment by applying a new meta-analytic approach of structural equation modeling (SEM) to provide a reliable statistical synthesis of effect sizes observed in existing individual studies. A total of 120 effect sizes – standardized differences between siblings of children with ASD and siblings of typically developing (TD) children—were calculated from 24 studies. The results revealed that siblings of children with ASD experienced more adjustment difficulties than siblings of TD children in all three domains. Furthermore, this study found that adjustment difficulties were more likely to be alleviated when siblings were older, female, and/or older than their sibling(s) with ASD. The findings provide important implications for support systems that can help siblings of children with ASD learn practical coping skills, develop a strong bond with family, and share their experiences with others in the community. Future research is needed to clarify other potential covariates that affect adjustment in siblings of children with ASD.

Keywords: siblings, autism, social adjustment, emotional adjustment, behavioral adjustment, structural equation modeling (SEM)

INTRODUCTION

Autism spectrum disorder (ASD) is a neurological and developmental disorder, characterized by severe deficits in social communication and interaction, restricted interests and repetitive behaviors, and symptoms that impede the person's everyday functioning (APA, 2013). The prevalence of ASD has been increasing steadily and also globally over the past 50 years (Elsabbagh et al., 2012; Poovathinal et al., 2018) - for 2014, 16.8 per 1,000 (1 in 59) children aged eight years in the United States (Bajo et al., 2018).

The challenges that a child with ASD experiences affect their family, school, and community in different ways (Gau et al., 2010; Lampert, 2007; Rao & Beidel, 2009; Weiss et al., 2014). While many studies have explored the social relationships that children with ASD build with their parents and peers (e.g., Kasari & Patterson, 2012; Kasari et al., 2014), others cast more light onto their siblings as they "adjust" their own relationships in a unique way (e.g., Knott et al., 1995; Mascha & Boucher, 2006; Rydberg, 2019). Typically developing (TD) children may endure the same or similar experiences of exclusion or neglect as their siblings with ASD (Burke, 2004), sharing the risk for maladjustment.

LITERATURE REVIEW

How does the presence of ASD in a child affect their sibling(s) in typical development? The literature is reporting somewhat different, even conflicting findings for this question. Some studies showed that siblings of children with ASD are at risk of adverse developmental outcomes and have more adjustment issues than their peers (e.g., Gold, 1993; Hastings, 2003a, 2003b; Kaminsky & Dewey, 2001; Pourbagheri et al., 2018; Shojaei et al., 2018; Verte et al., 2003). Others found no essential differences in their adjustment (e.g., Hastings & Petalas, 2014; Kaminsky & Dewey, 2002; Lampert, 2007; Malesa et al., 2013; Quintero & McIntyre, 2010; Rodrigue et al., 1993). Nevertheless, it is noteworthy that a few individual characteristics - i.e., age, gender, birth order, socio-economic status (SES), and family size - have been commonly identified as potential covariates for sibling adjustment (e.g., Gau et al., 2010; Gold, 1993; Herlihy et al., 2015; Lampert 2007; Toth et al., 2007). The following sections review current literature about the impacts of ASD on TD siblings focusing on the three primary domains of adjustment (social, emotional, and behavioral) and the moderating factors (covariates) of their adjustment.

Social Adjustment

Current findings on the social impact of ASD on TD siblings are inconsistent. Some studies showed that compared to siblings of TD children, siblings of children with ASD have more difficulties communicating and interacting with others socially (Dunn, 2007; Kaminsky & Dewey, 2001; Pourbagheri et al., 2018; Shojaei et al., 2018; Walton & Ingersoll, 2015) such as lack of affection, social interaction, conversation, relationships, and social appropriateness (Pilowsky et al., 2004). Other researchers also found that siblings of children with ASD are more likely to be introverted, internalized, or withdrawn (Barak-Levy et al., 2010; Hastings, 2003b; Macks & Reeve, 2007), which may explain low participation in social activities, problems with their peers, and poor academic performance (Barak-Levy et al., 2010). Although TD siblings acknowledge that more attention and support are needed for their sibling(s) with ASD, they expect their parents to treat them equally (Thibaut & Kelley, 1959; Kaminsky & Dewey, 2001).

On the other hand, it was found that siblings of children with ASD are socially competent and possess appropriate socialization skills (Gold, 1993; Kaminsky & Dewey, 2001; Pilowsky et al., 2004; Rodrigue et al., 1993; Verte, et al., 2003). They also have a more positive self-concept compared to siblings of TD children (Ferrari, 1984; Gold, 1993; Kaminsky & Dewey, 2002; Macks & Reeve, 2007; Rodrigue et al., 1993). Having multiple siblings facilitates consistent social development and healthy adjustment among siblings of children with ASD (Kaminsky & Dewey, 2002; McDonald et al., 2020; Verte, et al., 2003).

The risk factors for social maladjustment include age, gender, birth order, and family size. That is, sisters of children with ASD have a higher level of social competence compared to brothers of children with ASD (Kaminsky & Dewey, 2002; Rao & Beidel, 2009; Verte et al., 2003). Older siblings are more likely to assist, advise, cooperate, and empathize with their sibling(s) with ASD (Brewton et al., 2012). Nevertheless, older male siblings are at higher risk for social challenges due to limited coping skills and low social competency (Hastings, 2003a; Macks & Reeve, 2007; Rodrigue et al., 1993; Verte et al., 2003). Pilowsky et al. (2004) also found that larger family size is associated with some developmental delays in siblings' socialization skills.

Emotional Adjustment

One's relationship with their sibling(s) is a major source of emotional development (Cummings & Davies, 1996).

There are contradictory findings and arguments about emotional adjustment among siblings of children with ASD. Several studies demonstrated that siblings of children with ASD are more likely to experience emotional difficulties such as depression, anxiety, withdrawal, aggression, and bad temper than siblings of TD children (Caliendo et al., 2020; Gau et al., 2010; Gold, 1993; Griffith et al., 2015; Lovell & Wetherell, 2016; Longobardi et al., 2019; Koukouriki & Soulis, 2020; Pourbagheri et al., 2018; Rodrigue et al., 1993; Ross & Cuskelly, 2006; Shojaei et al., 2018; Verte et al., 2003). Similar findings were reported for siblings of children with cognitive disabilities (Petalas et al., 2009).

On the contrary, other studies found no or fewer emotional issues among siblings of children with ASD than those of TD children (Jones et al., 2019; Kaminsky & Dewey, 2001; Hastings & Petalas, 2014; Rodgers et al., 2016). Pilowsky et al. (2004) noted that TD siblings are not necessarily vulnerable to emotional difficulties; rather, they are more empathic towards their sibling(s) with ASD. Similarly, both parent-reports and child self-reports in a recent study indicated no difference in internalizing symptoms (e.g., anxiety and depression) between siblings of ASD children and those of TD children (Rodgers et al., 2016).

Individual characteristics such as gender, birth order, and SES were identified as the moderating factors of emotional adjustment. That is, male siblings of children with ASD have a greater risk for emotional difficulties than female siblings of children with ASD (Hastings, 2003a; Macks & Reeve, 2007) and a higher level of depression than male siblings of TD children (Gold, 1993). Additionally, older siblings and those from lower SES are associated with more emotional issues among siblings of children with ASD (Macks & Reeve, 2007; Rodrigue et al., 1993).

Behavioral Adjustment

Siblings of children with ASD may be at risk for negative behavior problems such as aggression, hyperactivity, and demandingness (Gregory et al., 2020; Griffith et al., 2015; Macks & Reeve, 2007; Mascha & Boucher, 2006; Rodrigue et al., 1993; Ross & Cuskelly, 2006; Tomeny et al., 2012; Thijssen et al., 2017; Verte et al., 2003; Warren et al., 2011; Walton & Ingersoll, 2015). A few studies revealed a direct connection between the severity of ASD symptoms and TD siblings' difficulty in adjustment (Benson & Karlof, 2008; McHale et al., 2016; Rosello et al., 2020; Tsai et al., 2017). Many siblings of children with ASD reported that autistic behaviors (e.g., aggression,

tantrums, excessive anger, and explosiveness) affect their lives in one way or another (Mascha & Boucher, 2006; Sukhodolsky et al., 2021) and that aggression is the most common type of stressor (Ross & Cuskelly, 2006). Another stressor frequently mentioned by siblings of children with ASD is an embarrassment (Begum & Mamin, 2019; Mascha & Boucher, 2006; Lampert, 2007).

In contrast, some studies suggested that siblings of children with ASD are not vulnerable to behavioral difficulties and even adjust better (Kaminsky & Dewey, 2001, 2002; Hastings, 2007; Verte et al., 2003). Hastings (2007) found no significant difference in behavioral adjustment among siblings of children with ASD, Down Syndrome (DS), and Intellectual Disabilities (IDs). Shojaei et al. (2018) further confirmed no difference among siblings of children with ASD, IDs, and typical development.

Similarly to social and emotional adjustment, age, gender, birth order, SES, and family size were recognized as moderating behavioral adjustment in siblings of children with ASD. Male and younger siblings of children with ASD have more behavioral issues than female and older siblings (Hastings, 2003b; Macks & Reeve, 2007; Tomeny et al., 2012; Verte et al., 2003). No considerable difference exists in behavioral difficulties between older siblings of children with ASD and older siblings of TD children (Quintero & McIntyre, 2010), suggesting that getting older may help with behavioral adjustment. In addition, when siblings of children with ASD have a lower SES or more than one brother, they are more likely to have behavioral problems.

Purpose of the Study

A number of studies have investigated how the presence of ASD in a child affects their sibling(s) in social, emotional, and behavioral adjustment; as noted earlier, their results are mixed. Three systematic reviews have been completed (Aparicio & Mínguez, 2015; Meadan et al., 2010; Shivers et al., 2019), but questions remain unanswered that can be better addressed by new analytic techniques. For example, the most recent systematic review by Shivers et al. (2019) retained a sample of 69 studies applying a broad inclusion criterion. That is, the sample included not only studies on children of siblings with ASD but also studies on children of siblings with SD, IDs, or other developmental disabilities. Obviously, the finding of Shivers et al. (2019) is valuable in its own right, but the study ignored the possibility that the observed associations may differ meaningfully in sub-

populations (i.e., ASD, DS, ID, etc.). Also, the role of potential moderating factors was not fully addressed in the previous systematic reviews.

The purpose of this study is two-fold: to examine (1) whether children with ASD affect their TD sibling(s) on social, emotional, and behavioral adjustment and (2) to identify personal characteristics that account for the differences in siblings' adjustment. This study applied a new meta-analytic approach of structural equation modeling (SEM) to provide a reliable statistical synthesis of effect sizes observed in existing individual studies (Borenstein et al., 2009). Given that a few individual characteristics have been identified in the literature as risk factors for maladjustment, this study further examined if age, gender, and birth order moderate adjustment in siblings of children with ASD—i.e., if these variables enlarge or diminish the difference (if one exists) between siblings of children with ASD vs. siblings of TD children. Family size and SES were also considered but were ultimately excluded because there was not enough relevant data for a meta-analysis.

METHODS

Selection of Studies

Studies were searched by using Psychological Abstracts (PsycINFO), Education Resources Information Center

(ERIC), Wilson OmniFile Full Text, ProQuest (Dissertation Abstracts), and Google Scholar. Search keywords included “siblings” or “brothers and sisters,” “autism spectrum disorder,” “social adjustment,” “emotional adjustment,” and “behavioral adjustment.” Studies cited in the retrieved articles were also pooled together. The following inclusion criteria were applied: (a) a peer-reviewed or non-peer-reviewed journal article or dissertation written in English; (b) involves siblings of children with ASD (early childhood to 18 years old) and siblings of children without disabilities (early childhood to 18 years old); and (c) provides quantitative data available for computing an effect size(s) in social, emotional, and/or behavioral adjustment. Consequently, unlike Shivers et al. (2019), studies using siblings of children with DS, ID, or other developmental disabilities were excluded. Because the characteristics of ASD are generally homogeneous across different subpopulations and time periods, the study sample was not limited by participants' gender, ethnicity, language, or year of the study. All authors agreed upon the final selection of studies to be included in this review. The Preferred Reporting Items for Systematic Reviews and Meta-Analyses flowchart (PRISMA; Page et al., 2020) illustrates the sampling/screening process of the current meta-analysis (see Figure 1).

The following information was obtained from each study included in the final study sample: effect size –

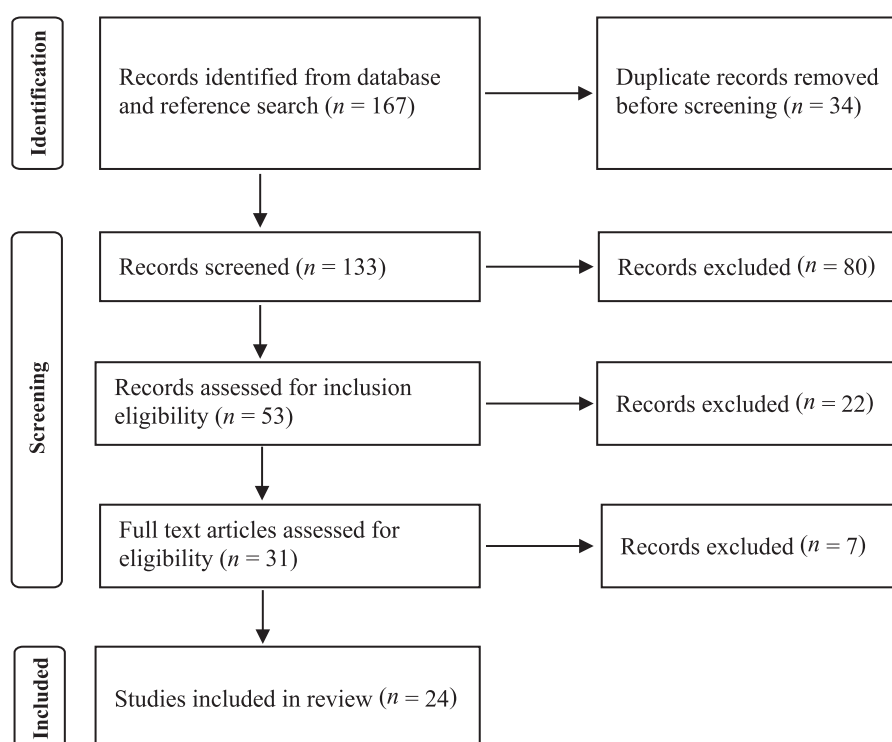


Fig. 1. Flowchart for the Identification of Studies (Adapted from PRISMA 2020 diagram)

the standardized difference between siblings of children with ASD and siblings of TD children (Cohen's d), the publication (type, year, country), demographics (target population, age, gender, birth order of siblings), and assessment instruments used.

Analytic Approach

SEM-based meta-analysis (Cheung, 2008, 2015, 2019) was particularly useful for the current investigation. That is, some of the studies in the sample provided no information about the covariates of siblings' age, gender, and birth order. If a conventional meta-analysis had been performed, those studies with missing data would have been excluded from the covariate (moderation) analysis thereby resulting in lower power and biased inferences. Alternatively, SEM-based meta-analysis efficiently handles missing data on covariates via full information maximum likelihood (FIML) estimation. This method produces smaller standard errors compared to listwise deletion (Cheung, 2008) and superior power to test small effect sizes compared to multiple imputations (Graham et al., 2007).

Analysis Procedure

The analysis proceeded in three steps using *Mplus* 8.0 (Muthén & Muthén, 1998–2017) and R package *metafor* 2.0 (Viechtbauer, 2017). The data and software codes used for the current meta-analysis will be available to readers upon request.

In the first step, random-effects SEM models, *without* covariates, were fitted separately for social, emotional, and behavioral adjustment, reasonably assuming that selected studies are a random sample of all existing studies in the literature (Borenstein et al., 2009). The model parameter of interest (intercept b_0) represented the weighted average difference in means between siblings of children with ASD and siblings of TD children.

In the next step, three covariates of siblings' age, gender, and birth order were introduced into the models. Given that the studies in the sample provided covariate information as aggregate statistics only (i.e., mean or percentage), study-level data were analyzed - mean age, % female, and % older. Now, the intercept b_0 indicated the marginally weighted average difference between the two sibling groups after controlling for the covariates. The new parameter, slope b_1 , represented the impacts of the covariates on the difference between two sibling groups - i.e., increase or decrease in the difference due to the covariate.

Lastly, a funnel plot and fail-safe N were inspected for potential publication bias (Sterne & Egger, 2001; Sut-

ton, 2009). Publication bias may occur if certain types of studies are chosen over other types for a meta-analysis. Examples of publication bias include published studies rather than unpublished research; studies with significant results or large effect size(s); and familiar or easily accessible studies (Borenstein et al., 2009). Fail-safe N is the minimum number of additional studies with insignificant results that would be needed to change the conclusions of a meta-analysis (Fragkos et al., 2017). A small fail-safe N suggests that publication bias may affect the conclusions of the meta-analysis.

RESULTS

Characteristics of Study Sample

A total of 160 studies were identified from keyword searches and the references cited in the retrieved articles. Of those, 24 studies (15%) met the inclusion criteria and were included in the final study sample. The full references of the 24 studies are provided in the References section, each marked with an asterisk.

Table 1 summarizes the characteristics of the study sample. The studies were published between 1993 and 2020; with 55% in 2010 or later. Four studies (16.7%) were doctoral dissertations, and all others (83.3%) were peer-reviewed journal articles. More than half of the studies ($n = 10$, 58.3%) were conducted outside the United States: (a) three in Canada, (b) two in Iran, (c) two in Taiwan, (d) one in Belgium, (e) one in Israel, and (f) one in the United Kingdom. The studies used 24 different instruments—11 instruments were used for social adjustment, 11 for emotional adjustment, and 8 for behavioral adjustment (there were overlaps). The Child Behavior Checklist (CBCL; Achenbach, 1991) was the most frequently used instrument (34%), followed by the Difficulties Questionnaire (SDQ; Goodman, 1997) (16%).

A total of 120 effect sizes were calculated using the information reported in the 24 studies. Statistical power approached 1, given the weighted average effect size (-0.39), the average number of siblings per group (44.08), and the number of effect sizes analyzed (120).

Structural Equation Modeling (SEM) -Based Meta-Analysis

The weighted average difference in overall adjustment between siblings of children with ASD and siblings of TD children was -0.39 ($SE = 0.06$, $p < .01$) across the 24 studies in the sample. This difference indicated poorer overall adjustment among siblings of children with ASD, and such deficit was deemed moderate according

Table 1. Characteristics of Studies Included in SEM-based Meta-Analysis

Study	Publication type	Location	N		Mean age [range]		Gender (% female)		Birth order (% older)		Assessment instrument
			ASD	TD	ASD	TD	ASD	TD	ASD	TD	
Chien et al. (2017)	Journal	Taiwan	66	132	13.17 [8-19]	12.64 [8-19]	4.50	25.76	n/a	n/a	SAICA
Garon et al. (2009)	Journal	Canada	104	73	3.00 [n/a]	3.00 [n/a]	51.00	52.05	0	0	ADOS, ADI-R
Gau et al. (2010)	Journal	Taiwan	134	113	9.00 [3-12]	7.90 [3-12]	41.00	38.94	n/a	n/a	CBCL
Gold (1993)	Journal	Canada	22	34	13.52 [7-17]	12.78 [7-17]	50.00	50.00	45.45	67.65	CBCL, CDI
Kaminsky & Dewey (2002)	Journal	Canada	30	30	11.67 [8-18]	11.54 [8-18]	50.00	50.00	80.00	80.00	CBCL
Lampert (2007)	Dissertation	US	20	100	n/a [8-12]	n/a [8-12]	40.00	55.00	75.00	33.00	PBQ
Lovell & Wetherell (2016)	Journal	UK	20	18	12.60 [7-17]	11.00 [7-17]	45.00	55.56	n/a	n/a	CDI-2
Malesa et al. (2013)	Journal	US	36	23	5.29 [4-7]	5.47 [4-7]	n/a	n/a	0	0	SSRS
Pourbagheri et al. (2018)	Journal	Iran	58	58	6.77 [3-9]	6.20 [3-9]	36.20	50.00	n/a	n/a	SDQ
Rao & Beidel (2009)	Journal	US	7	8	10.60 [8-16]	11.50 [8-15]	57.10	25.00	n/a	n/a	CBCL
Rodgers et al. (2016)	Journal	US	42	42	9.52 [6-16]	9.86 [6-16]	42.90	100.00	n/a	n/a	BASC 2-PRS, BASC 2-SRP
Rodrigue et al. (1993)	Journal	US	19	20	10.22 [n/a]	9.45 [n/a]	52.60	60.00	n/a	n/a	CBCL
Rozga et al. (2011)	Journal	US	82	65	3.00 [n/a]	3.00 [n/a]	56.10	44.62	n/a	n/a	ADOS
Schwartz (2003)	Dissertation	US	28	16	11.60 [8-14]	11.60 [9-14]	71.40	68.75	75.00	68.75	CDI, CBCL

Shojaee et al. (2018)	Journal	Iran	49	50	14.16 [10-18]	14.40 [10-18]	48.00	66.00	n/a	n/a	SDQ
Tomeny et al. (2012)	Journal	US	42	42	12.81 [6-18]	12.60 [6-18]	59.50	52.38	n/a	n/a	CSBQ
Toth et al. (2007)	Journal	US	42	20	1.69 [1.5-2.08]	1.87 [1.6-2.25]	50.00	35.00	0	55.00	ADOS, CSBS-DP, SEEC
Tremaine (2011)	Dissertation	US	14	14	4.57 [2-5.9]	4.50 [2-6]	50.00	50.00	0	0	SSIS
Verte et al. (2003)	Journal	Belgium	29	29	11.14 [6-16]	11.31 [6-16]	41.40	41.38	37.93	37.93	MESSY, CBCL
Walton & Ingersoll (2015)	Journal	US	69	93	10.43 [4-17]	10.99 [4-17]	52.20	52.69	50.72	65.59	SDQ, SRS
Warren et al. (2011)	Journal	US	39	21	5.28 [4-7]	5.36 [5-6]	43.60	33.33	0	0	SSRS, ADOS, SRS, CBCL
Wong (2007)	Dissertation	US	21	13	11.00 [8-15]	11.50 [8-14]	42.90	38.46	76.19	61.54	SSRS, SRS, CBCL
Yirmiya et al. (2006)	Journal	US	31	31	5.16 [n/a]	5.18 [n/a]	38.70	41.94	0	0	ESCS
Yoder et al. (2009)	Journal	Israel	43	24	2.81 [n/a]	2.87 [n/a]	44.20	29.17	n/a	n/a	RJA, SBC, WTC

Note. ASD = siblings of children with autism spectrum disorder, TD = siblings of typically developing children, ADI-R = Autism Diagnostic Interview-Revised (Lord et al., 1994), ADOS = Autism Diagnostic Observation Schedule (Lord et al., 2000), BAI = Beck Anxiety Inventory (Beck & Steer, 1990), BASC 2-PRS/SRP = Behavior Assessment System for Children-Parent Rating Scales/Self-Report of Personality (Reynolds & Kamphaus, 2004), BDI = Beck Depression Inventory (Beck et al., 1996), CBCL = Child Behavior Checklist (Achenbach, 1991), CDI = Children's Depression Inventory (Kovacs, 1992), CDI-2 = Children's Depression Inventory (Kovacs, 2011), CSBQ = Children's Social Behavior Questionnaire (Luteijn et al., 2000), CSBS-DP = Communication and Symbolic Behavior Scale-Developmental Profile (Wetherby & Prizant, 1998, 2002), ESCS = Early Social Communication Scales (Mundy et al., 1996; Seibert et al., 1982), MESSY = Matson Evaluation of Social Skills with Youngsters (Matson, 1988), PBQ = Prosocial Behavior Questionnaire (Weir, Stevenson, & Graham, 1980), RJA = Responding to Joint Attention (Deák et al., 2008; Presmanes et al., 2007), SAICA = Social Adjustment Inventory for Children and Adolescents (John et al., 1987), SBC = Social Behavior Checklist (Stone et al., 1990), SD = Semantic Differentials (Osgood et al., 1957), SDQ = Strengths and Difficulties Questionnaire (Goodman, 1997), SEEC = Vineland Social Emotional Early Childhood Scale (Sparrow et al., 1998), SRS = Social Responsiveness Scale (Constantino, 2002), SSIS = Social Skills Improvement System (Elliot & Gresham, 2007), SSRS = Social Skills Rating System (Gresham & Elliott, 1990), VABS = Vineland Adaptive Behavior Scales (Sparrow et al., 1984), WTC = Weighted Triadic Communication (Vaughan Van Hecke et al., 2007).

to Cohen's guideline (1992). As shown in Figure 2, the effect sizes varied considerably across the studies within each domain. The following sections report the results of SEM-based meta-analysis on social, emotional, and behavioral adjustment, both without and with accounting for the covariates of siblings' age, gender, and birth order.

Social Adjustment

Table 3 presents the analysis results for siblings' emotional adjustment. The weighted average difference in social adjustment was -0.26 ($SE = 0.07$, $p < .01$). This result indicated that siblings of children with ASD had greater social adjustment issues than siblings of TD children, although a such deficiency in the ASD group was small. The residual variance was also significant ($p < .01$), suggesting the necessity of covariates to explain the

variability of the effect sizes beyond study-level sampling error.

Siblings' age and birth order were found as significant moderating factors of their social adjustment. Note that either without or with a covariate, the estimate of the intercept parameter was negative. The estimate of the slope parameter for age was positive ($\hat{b}_1 = 0.04$, $SE = 0.01$, $p < .01$), therefore indicating that although siblings of children with ASD had more social problems than siblings of TD children, this difference diminished as siblings aged. Similarly, when siblings were older children in the family (i.e., older than their ASD or TD sibling), the difference in siblings' social adjustment was also lessened ($\hat{b}_1 = 0.01$, $SE = 0.00$, $p < .01$). However, siblings' gender was not related to the difference in social adjustment between siblings of children with ASD and those of TD children.

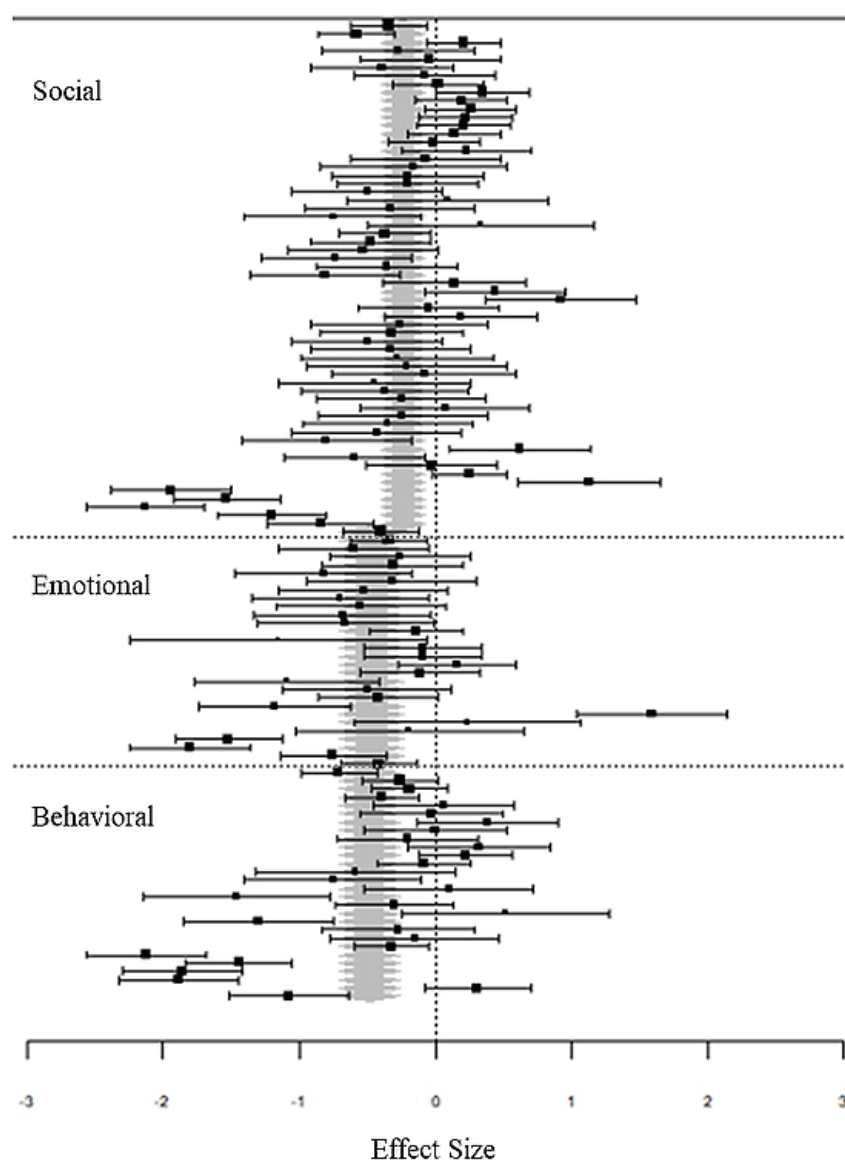


Fig. 2. Forest Plot of Effect Sizes for All 24 Studies

Emotional Adjustment

The weighted average difference in emotional adjustment was -0.57 ($SE = 0.10$, $p < .01$; see Table 3), indicating that siblings of children with ASD had more emotional problems than siblings of TD children, and this difference was moderate. Given that the residual variance was significant ($p < .01$), the covariates were entered into the model one-by-one to account for the variability in effect size.

Siblings' gender and birth order significantly moderated the difference in social adjustment between the two

sibling groups. The estimate of the slope parameter for gender (% female) was positive ($\hat{b}_1 = 0.03$, $SE = 0.01$, $p < .01$). This result indicated that although siblings of children with ASD experienced more emotional difficulties than siblings of TD children in general, the difference was smaller among female siblings. The estimate of the slope parameter for birth order (% older) was also positive ($\hat{b}_1 = 0.02$, $SE = 0.01$, $p < .05$). Similarly to the findings in social adjustment when siblings were older children in the family, the difference in their emotional adjustment

Table 2. SEM-based Meta-analysis Results for Social Adjustment

Covariate	Parameter	Estimate	SE	Lower	Upper	z	p
No	b_0	-0.26	0.07	-0.40	-0.12	-3.59	<.01**
	u	0.24	0.05	0.14	0.35	4.48	<.01**
Age	b_0	-0.61	0.13	-0.88	-0.35	-4.59	<.01**
	b_1	0.05	0.02	0.02	0.07	3.09	<.01**
	u	0.22	0.08	0.06	0.37	2.75	<.01**
Gender (% female)	b_0	-0.21	0.40	-1.00	0.58	-0.53	.60
	b_1	-0.00	0.01	-0.02	0.01	-0.13	.90
	u	0.27	0.09	0.09	0.44	3.03	<.01**
Birth order (% older)	b_0	-0.31	0.06	-0.43	-0.19	-5.17	<.01**
	b_1	0.01	0.00	0.00	0.01	3.92	<.01**
	u	0.02	0.02	-0.02	0.07	0.98	.33

Note. SE = standard error, Lower = lower limit of 95% confidence interval, Upper = upper limit of 95% confidence interval, b_0 = intercept, b_1 = slope for covariate, u = error variance.

* $p < .05$, ** $p < .01$.

Table 3. SEM-based Meta-analysis Results for Emotional Adjustment

Covariate	Parameter	Estimate	SE	Lower	Upper	z	p
No	b_0	-0.57	0.10	-0.76	-0.38	-5.87	<.01**
	u	0.16	0.06	0.04	0.29	2.64	<.01**
Age	b_0	-1.32	0.75	-2.78	0.14	-1.77	.08
	b_1	0.07	0.07	-0.06	0.20	1.10	.27
	u	0.15	0.04	0.06	0.23	3.29	<.01**
Gender (% female)	b_0	-1.91	0.49	-2.88	-0.94	-3.86	<.01**
	b_1	0.03	0.01	0.01	0.04	3.15	<.01**
	u	0.11	0.04	0.02	0.19	2.41	.02
Birth order (% older)	b_0	-1.45	0.53	-2.49	-0.41	-2.73	<.01**
	b_1	0.02	0.01	0.00	0.03	2.24	.03*
	u	0.02	0.02	-0.02	0.06	1.16	.25

Note. SE = standard error, Lower = lower limit of 95% confidence interval, Upper = upper limit of 95% confidence interval, b_0 = intercept, b_1 = slope for covariate, u = error variance.

* $p < .05$, ** $p < .01$.

Table 4. SEM-based Meta-analysis Results for Behavioral Adjustment

Covariate	Parameter	Estimate	SE	Lower	Upper	z	p
No	b_0	-0.48	0.13	-0.74	-0.22	-3.66	<.01**
	u	0.44	0.13	0.18	0.70	3.35	<.01**
Age	b_0	-1.08	0.42	-1.90	-0.26	-2.59	<.01**
	b_1	0.06	0.04	-0.01	0.14	1.67	.10
	u	0.39	0.11	0.19	0.60	3.76	<.01**
Gender (% female)	b_0	-0.83	0.62	-2.03	0.38	-1.34	.18
	b_1	0.01	0.01	-0.02	0.03	0.57	.57
	u	0.44	0.12	0.20	0.67	3.64	<.01**
Birth order (% older)	b_0	-0.36	0.17	-0.69	-0.02	-2.05	.04*
	b_1	0.00	0.00	0.00	0.01	0.92	.36
	u	0.17	0.13	-0.09	0.43	1.30	.20

Note. SE = standard error, Lower = lower limit of 95% confidence interval, Upper = upper limit of 95% confidence interval, b_0 = intercept, b_1 = slope for covariate, u = error variance.

* $p < .05$, ** $p < .01$.

decreased. However, the siblings' age was not a significant moderating factor in their emotional adjustment.

Behavioral Adjustment

Siblings of children with ASD also had more behavioral problems than siblings of TD children and such difference in behavioral adjustment was moderate ($\hat{b}_0 = -0.48$, $SE = 0.13$, $p < .01$; see Table 4). None of the siblings' age, gender, and birth order was significantly associated with the difference in behavioral adjustment between siblings of children with ASD and those of TD children.

Publication Bias

Figure 3 provides a funnel plot describing the relationship between the effect sizes and their standard errors.

If sampling error occurred randomly, effect sizes are expected to be distributed symmetrically (Borenstein et al., 2009). The 120 effect sizes calculated from the study sample showed a relatively symmetric dispersion around their average value, which confirmed the absence of publication bias in the current investigation. In addition, the estimated fail-safe N was 12,079, indicating that a very large number of additional studies with insignificant results are needed to change the current findings. Taken together, publication bias was not susceptible to this study.

In summary, this study found that children with ASD had a significant, negative impact on their sibling(s) in social, emotional, and behavioral adjustment. Siblings' age, gender, and birth order significantly moderated their social and emotional difficulties, while their behavioral

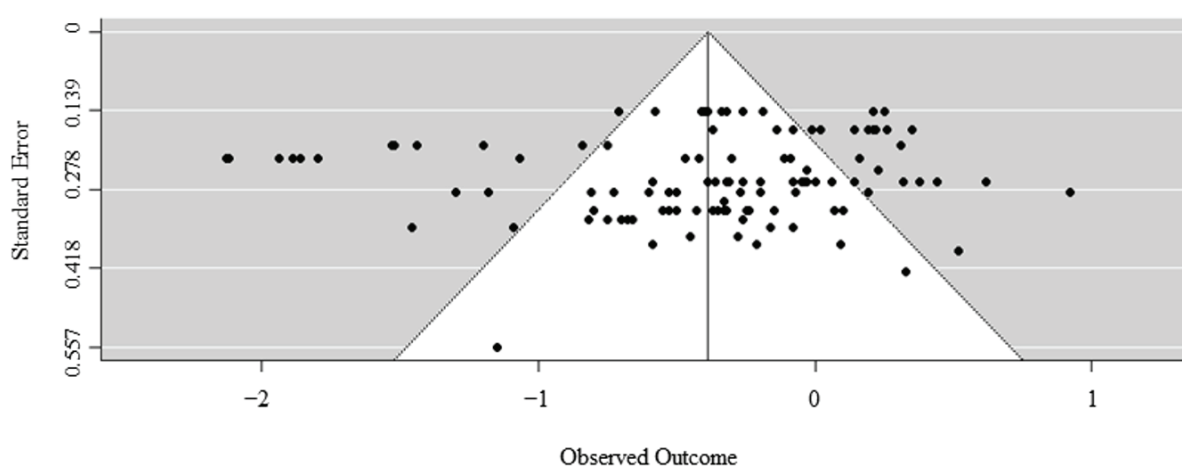


Fig. 3. Funnel Plot of Overall Impacts for All 24 Studies

adjustment was not affected by any of those individual characteristics. There was no evidence of publication bias in the analysis. The study findings and their implications are discussed in the following section.

DISCUSSION AND IMPLICATIONS

The purpose of this study is to (1) examine whether children with ASD affect their TD sibling(s) on social, emotional, and behavioral adjustment and (2) identify personal characteristics that account for the differences in siblings' adjustment. The existing research provided sufficient data for a comprehensive systematic review of these primary domains of development. As noted earlier, previous studies reported inconsistent and even conflicting findings on how the presence of ASD on a child affected their siblings' adjustment. Through SEM-based meta-analysis, this study confirmed significantly negative impacts on all three adjustment domains for siblings of children with ASD. The weighted average effect sizes were -0.26 , -0.57 , and -0.48 for social, emotional, and behavioral adjustment, respectively (-0.39 for overall adjustment). These estimates indicated that siblings of children with ASD had more adjustment difficulties than siblings of TD children. The deficit in the ASD group was relatively greater in the emotional and behavioral domains (medium) than in the social domain (small). Thus, more attention and support are needed for siblings of children with ASD, particularly to monitor and help their emotional and behavioral development.

This study also explored potential risk factors for maladjustment among siblings of children with ASD. Although a few previous studies reported that demographic factors were not related to siblings' adjustment (e.g., Kaminsky & Dewey, 2002; Pilowsky et al., 2004), this study found that in general, adjustment difficulties were more likely to be alleviated when siblings were female, older, and older than their sibling(s) with ASD. More specifically, among siblings of children with ASD, younger age was a risk factor for lower social competency; being male was a risk factor for emotional maladjustment; and being younger than their sibling(s) with ASD was a risk factor for both social and emotional difficulties. None of the siblings' age, gender, and birth order were related to the deficit in behavioral adjustment in this group. Hesse et al. (2013) noted that female siblings under the age of 18 are likely to mature faster than their male counterparts. Such discrepancy in maturity may be an important, yet unrevealed, factor related to dealing with the difficulties of having a younger sibling with ASD. Also, younger TD

siblings may "show up" more adjustment problems such as regressive and attention-seeking behaviors () because they perceive their parents' care is unfairly focused on their older sibling(s) with ASD. Some researchers have provided alternative accounts; that is, as the traits of genetic risk are shared among siblings, brothers and sisters of a child with ASD may have an increased risk for their adjustment (Petalas et al., 2012). Meyer et al. (2011) revealed that siblings' broader autism phenotype (BAP) was positively associated with adjustment difficulties, moderated by the severity of ASD. Further research is needed to comprehend the individual, biological, and environmental mechanisms behind siblings' adjustment as well as their interactions.

TD siblings of children with ASD may need help to cope with the social, emotional, and behavioral challenges they confront. Nevertheless, their needs may be easily neglected because parents' and professionals' care and support may be much more focused on their sibling(s) with ASD. The findings from this study suggest that siblings of children with ASD are vulnerable and more likely to have developmental issues compared to siblings of TD children and that there are some uncontrollable risk factors for their maladjustment. Therefore, practical support systems are recommended for siblings of children with ASD. In those systems, practitioners and professionals—e.g., teachers, behavior analysts, psychologists, therapists, and psychiatrists—should monitor the unique, developmental characteristics of siblings of children with ASD and provide them with personalized coping skills. More importantly, the entire family should be supported (Meadan et al., 2010). Brothers and sisters should be regarded as the integral target of family support at an earlier stage. Siblings of children with ASD would have fewer adjustment problems if early support and more formal social support are available to their families (Cebula, 2012; Hasting, 2003a, 2003b).

A few intervention strategies and activities can be considered along with the holistic support systems. For example, group treatments (e.g., playgroup, supporters, peers, and/or family) may help empower siblings' adjustment, resolve existing issues, and build a positive relationship with their sibling(s) with ASD (Berry, 2014; Hansford, 2013; Lock & Finstein, 2009). The group-mediated approach could help create a supportive community where siblings can receive support from others in similar situations and develop cognitive reframing and emotional flexibility (Berry, 2014). Another example is sibling-assisted aquatic programs that are designed to form a close bond between children with ASD and their

brothers and sisters (Chu & Pan, 2012), teaching skills for coping and playing with children with ASD, build a strong sibling relationship, and avoid unnecessary emotional challenges (Beyer, 2009; Carrillo, 2012). Recently, a robot-mediated intervention was introduced as a way to foster a collaborative play environment between children with ASD and their siblings (Huskens et al., 2015).

LIMITATIONS AND FUTURE DIRECTIONS

Although a few important findings and implications were discussed in this study which contributes to our understanding of siblings of children with ASD, the current investigation has three notable limitations that need to be addressed in future research. First, as in any meta-analytic research, the characteristics of siblings and families varied (i.e., heterogeneous) across the studies included in the sample. For instance, some siblings were receiving support from professionals at their school or home—e.g., 77% of participating families in Kaminsky and Dewey (2002), which might impact the current findings. Also, Rivers and Stoneman (2003) reported that marital stress caused negative relationships between children with ASD and their sibling(s). Negative sibling relationships can affect the sibling(s)' social, emotional, and behavioral difficulties. Thus, if available, siblings' and families' characteristics—such as the previous or current experience of support/intervention, knowledge of ASD, cultural/ethnic backgrounds, and relationship with other families of children with ASD—should be considered in future systematic reviews. Second, this study investigated the moderating role of siblings' age, gender, and birth order but other important covariates could not be examined due to limited data. Future research on potential covariates—including family size and SES, assessment type (e.g., self-appraisal vs. parents' or professionals' evaluation), assessment scales, and publication (e.g., type, year, country) - would help identify a more comprehensive list of factors that affect adjustment in siblings of children with ASD. Lastly, the current analysis did not account for subject-level heterogeneity in the covariates. As noted earlier, siblings' age, gender, and birth order were analyzed using aggregate data (mean or proportion) provided

by the studies in the sample. The relationship between effect estimates and average sibling characteristics across the studies might not be the same as that relationship within the studies, thereby increasing the risk for incorrect inferences about the covariates used (i.e., ecological fallacy). Besides, the current analysis did not control for study-level dependency in effect size. Although the effect sizes were in fact correlated between the social, emotional, and behavioral domains ($r = .45-.86$), the analysis was performed separately for each domain. A multivariate, subject-level data meta-analysis in future systematic reviews would provide greater statistical power, an ability to test subgroup differences (i.e., moderation) whilst avoiding ecology fallacy, as well as a more realistic, overarching picture of development than looking at its different domains individually (Cheung, 2013; Lyman & Kuderer, 2005; Reade et al., 2008).

CONCLUSION

Siblings of children with ASD are vulnerable to social, emotional, and behavioral difficulties. They are at greater risk especially if they are male, younger, and/or younger than their sibling(s) with ASD. Caregivers, practitioners, and professionals should pay more attention to their challenges and needs. Additionally, personalized, whole-family support systems should be developed and implemented to support siblings of children with ASD in learning effective coping skills, developing a strong bond with their family, sharing their experiences with others in the community, and asking for help. In this way, they will be able to overcome struggles and move forward to healthy adulthood.

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DECLARATION OF INTEREST STATEMENT

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