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Functioning Among Typically Developing Siblings of Individuals with Autism Spectrum Disorder: A Meta-Analysis

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Abstract
The literature on typically developing siblings of individuals with autism spectrum disorder (ASD-Sibs) provides inconsistent results, with some studies reporting ASD-Sibs are more likely to have negative outcomes than comparison groups, and others reporting no significant differences. Therefore, the purpose of this study was to meta-analytically aggregate study effect sizes to more accurately calculate the degree to which ASD-Sibs function similarly or differently compared to siblings of people who do not have ASD. Studies were eligible for inclusion if they had a sample of ASD-Sibs older than 5; reported on emotional, psychological, behavioral, or social functioning; and provided information necessary for calculating relevant effect sizes. Results from 69 independent samples indicated that ASD-Sibs have significantly more negative outcomes than comparison groups overall ($g = -0.26$); specific areas of functioning in which ASD-Sibs fared worse include internalizing behavior problems, psychological functioning, beliefs, social functioning, and the sibling relationship, but no significant differences in adjustment, attention/hyperactivity, externalizing behavior problems, coping, or family functioning. Noteworthy sub-areas of functioning in which ASD-Sibs also fared worse included beliefs about disability ($g = -0.56$), anxiety symptoms ($g = -0.25$), and depression symptoms ($g = -0.36$). In terms of comparison group, ASD-Sibs had significantly lower functioning than siblings of individuals with other intellectual and developmental disabilities ($g = -0.31$), including Down syndrome ($g = -0.40$) and siblings of individuals without any disabilities ($g = -0.31$). Clinicians and service providers should work to ensure that ASD-Sibs are included in family interventions and support strategies, and researchers should further explore individual differences that may relate to enhanced or impaired functioning in ASD-Sibs.

Keywords Siblings · Autism spectrum disorder · Meta-analysis · Functioning

Introduction

The most recent prevalence rates in the United States estimate that 1 in 59 children has an ASD diagnosis (Centers for Disease Control and Prevention [CDC] 2018). Rates of ASD have continued to rise in the United States, more than doubling over a 10-year period from 2000 to 2010 (CDC 2018). Although the average number of children in families of individuals with ASD is unknown, extrapolation of data on family size (e.g., 78% of mothers have more than one child; Pew Research Center 2017) yields a conservative estimate of 2.7 million individuals with ASD in the United States with at least one sibling (ASD-Sibs). Regardless of the exact number of ASD-Sibs, understanding the experiences of this population is important, as the sibling relationship is typically the longest relationship a person will have in their lifetime and can have a substantial impact on emotional, behavioral, and psychological outcomes (Cicirelli 1995).

The majority of research on ASD-Sibs still falls under the category of *baby sib* studies, in which infant siblings of a child diagnosed with ASD are examined to determine potential early predictors of ASD. As younger ASD-Sibs

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are significantly more likely to receive an ASD diagnosis themselves than the general population (Szatmari et al. 2016), infant siblings can provide valuable insight into early markers and developmental patterns of ASD. Although some early studies examined family outcomes for individuals with ASD, interest in typically developing ASD-Sibs for their own experiences—rather than as comparators to the child with ASD or potentially developing an ASD themselves—has risen along with the diagnostic rates of ASD, though not necessarily at a commensurate rate. In particular, the systematic study of ASD-Sibs as distinct from siblings of people with other intellectual and developmental disabilities (IDD-Sibs) is a comparatively recent phenomenon. A review of ASD-Sib studies included only 12 articles published between 1997 and 2008, though the inclusion criteria focused on sibling functioning and did not include several studies of the sibling relationship (Meadan et al. 2010). However, the review did not limit the included studies to those that measured ASD-Sib outcomes in relation to a comparison group. The most commonly-cited review of siblings, Rossiter and Sharpe's (2001) meta-analysis, did not separate ASD-Sibs from other IDD-Sibs. The meta-analysis reported that IDD-Sibs exhibited a small but significant negative effect when compared to other populations (Rossiter and Sharpe 2001), a finding that has been frequently cited in support of further IDD-Sib research. However, many ASD-Sib studies report results that highlight how such siblings may experience different outcomes than even other IDD-Sibs (e.g., Hodapp and Urbano 2007).

Anecdotally, ASD-Sibs have reported feelings of anxiety, guilt, frustration, love, pride, and protection, among others (e.g., Petalas et al. 2009). The number of studies of typically developing ASD-Sibs has risen over the past decade, but the field still lacks consensus over the general outcomes for this population. In fact, many recent articles contain some variation on the phrase "findings are mixed" regarding social, emotional, and behavioral functioning among ASD-Sibs, especially as they compare to the general population, siblings of typically developing individuals, or siblings of individuals with other disabilities (e.g., Orsmond and Seltzer 2009; Tomeny et al. 2016; Verté et al. 2003). The present study aims to quantify, for the first time, the social, emotional, psychological, and behavioral functioning of ASD-Sibs, as compared to other nondisabled populations. Although the Simons Simplex Collection (Fischbach and Lord 2010) uses strict inclusion criteria for siblings, most ASD-Sib researchers implicitly define *typically developing siblings* as siblings without an ASD or any other IDD. For purposes of this study, we will use the same definition; any reference to ASD-Sibs refers to siblings of individuals with ASD who do not, themselves, have an ASD diagnosis or other intellectual or developmental disability.

One potential critique of ASD-Sib research in general is that it is a largely a-theoretical field. Although many ASD-Sib articles do not cite theoretical foundations, sibling research largely operates under family systems theory (Broderick 1993), a specification of Bronfenbrenner's ecological systems theory (Bronfenbrenner 1977). Systems theory posits that characteristics of individuals within the family, family relationships, and the family unit all interact and affect each family member. In the case of ASD-Sibs, family systems theory would suggest that having a brother or sister with ASD influences not only the functioning of the family as a whole, but each individual family member, including any siblings. Additionally, siblings are affected by such altered family functioning as well, including medical and therapeutic appointments, changes to the family routine, and potential differences in parenting practices related to the child with ASD (Meadan et al. 2010).

However, it is important to note that family systems theory does not mandate such impacts be negative. According to Bronfenbrenner, any change in the system can potentially affect the individual and, due to the specific characteristics of the individual, such effects may differ. An event (in this case, having a brother or sister with ASD) that affects Sibling A might have a completely different impact on Sibling B, even in the same family. Likewise, the impact on a sibling in Family A is unlikely to be the same as the impact on a sibling in Family B. Thus, while it is important to assess general patterns of outcomes among ASD-Sibs as a group, it is equally important to examine the potential differences based on possible related or moderating factors. More recently, Kovshoff et al. (2017) developed a more comprehensive framework in order to contextualize the complex nature of ASD-Sib experiences, encompassing such related or moderating factors. Although the present study primarily focuses on comparisons between ASD-Sibs and non-ASD sibs, it is worth keeping in mind the extensive framework in which such group differences may exist.

Existing Studies

Outcome Variables

ASD-Sibs researchers have studied a variety of outcomes for the given population. Although the most common are perhaps autistic symptoms themselves (e.g., Ozonoff et al. 2015), other studies of typically developing ASD-Sibs cover such varied factors as behavior problems, psychiatric symptoms, and social functioning, all aiming to answer the broad question of *How are ASD-Sibs doing*? Many of the outcomes measured by scientists are part of a complex network of behaviors and outcomes, phenomena that may operate quite differently for ASD-Sibs than they do for members of the general population.

Behavior Problems

As individuals with ASD are at risk for high levels of behavior problems (e.g., Eisenhower et al. 2005), many researchers have examined whether or not ASD-Sibs also have elevated levels of behavior problems. Although longitudinal studies of ASD-Sibs are rare, a few that have been conducted suggest higher levels of behavior problems among the individuals with ASD predict later increases in behavior problems among the ASD-Sibs (Hastings 2007). Not all behavior problems are created equal; however, most measures of behavior problems include total scales of overall behavior problems and split challenging behaviors into internalizing problems (e.g., worrying, withdrawal) and externalizing (e.g., aggression, delinquent behavior). Many studies use total behavior problems scores (e.g., Hastings 2003; Pilowsky et al. 2004), but differences in total scores between ASD-Sibs and comparison groups may reflect a cumulative effect (e.g., ASD-Sibs score slightly worse on each of the subscales, creating a significant cumulative difference in the total score), which washes out more subtle differences for specific types of behavior problems. Thus, the present study separates behavior problems by the most common types identified by modern measures—internalizing, externalizing, and attention/hyperactivity.

Although several well-validated instruments exist to measure type and severity of behavior problems (e.g., Child Behavior Checklist, Achenbach and Rescorla 2001; Strengths and Difficulties Questionnaire; Goodman et al. 2000), the line between *behavior problems* and *psychiatric symptoms*, particularly among children, is often muddled and imprecise. For the purposes of this study, the distinction lies in the measure used and the reported focus of the study.

Psychiatric Symptoms

As much of the modern sibling research grew out of a belief that siblings of individuals with IDD are a "population at risk" (San Martino and Newman 1974, p. 168), some researchers have tested the hypothesis that ASD-Sibs are at risk for psychiatric symptoms. Studies that rely on diagnostic rates have yielded mixed results; some have found that ASD-Sibs are no more likely to receive a clinical diagnosis than comparison groups (e.g., Pilowsky et al. 2004), whereas others have found that ASD-Sibs are more likely to have a psychiatric diagnosis (e.g., Jokiranta-Olkoniemi et al. 2016). Other studies have used scale measures, rather than dichotomous outcomes, to determine whether or not ASD-Sibs have increased levels of psychiatric symptoms, particularly anxiety and depression. In terms of specific diagnoses, some studies show that ASD-Sibs report similar levels of anxiety to comparison groups (e.g., Hallett et al. 2013; Tomeny et al. 2017), whereas others show that ASD-Sibs have higher scores on standardized anxiety scales (O'Neill and Murray 2016). Similarly, researchers have yet to reach a consensus on the likelihood of depressive symptoms among ASD-Sibs, with some findings showing significantly higher reports of depression (e.g., Hodapp and Urbano 2007; Lovell and Wetherell 2016), and others showing no differences in depressive symptoms between ASD-Sibs and comparison groups (e.g., Hallett et al. 2013; O'Neill and Murray 2016).

Social Functioning

As previously mentioned, although many researchers focus on measures of social functioning explicitly designed to indicate autistic symptoms among ASD-Sibs, others have focused on more broad definitions of sociability. Studies of social functioning in ASD-Sibs tend to operate under one of two hypotheses. The first hypothesis is that ASD-Sibs—either through sub-threshold autistic symptoms or through a lack of social interaction (and thus, social practice) with their brother or sister with ASD—will exhibit impaired social functioning (e.g., Pilowsky et al. 2004). The second hypothesis draws from a strengths-based approach and suggests that siblings of individuals with ASD are more likely to exhibit pro-social behavior and empathy, as their experiences at home prompt them to be more helpful and understanding of differences among others (e.g., Mascha and Boucher 2006). Although studies have examined several aspects of social functioning among ASD-Sibs, it is not yet known whether or not they exhibit either enhanced or impaired social skills overall.

Social functioning is comprised of many different skills and behaviors; self-report of social skills and emotions related to such skills may be very different from parent and teacher observations of positive social behavior. For example, empathy is defined by feelings, but measured by actions in parent and teacher reports (e.g., Lovett and Sheffield 2007). Helping behaviors can be labeled as *prosocial* in many cases, but could be indicative of parentification—that is, children taking on roles and caregiving behaviors typically filled by adults (Tomeny et al. 2016). Of course, no observed behaviors tell the full story of an individual's thoughts and motivations. Thus, measures of social functioning among ASD-Sibs is likely no less valid than similar measures of other populations. However, it is worth noting the unique considerations of ASD-Sibs that may relate to behaviors that teachers and parents observe as positive indicators of social functioning.

Family Functioning

Although many aspects of family functioning, such as parenting style, do not necessarily fall under the purview of sibling outcomes, as the behaviors and beliefs do not lie

solely within the siblings, other measured outcomes do involve other family members. Specifically, some researchers have examined the ASD-Sibs' perception of their relationship with their parents (e.g., Martins 2007), as well as the parents' perception of how having a brother or sister with ASD has affected the typically developing sibling (Kao et al. 2009). However, the majority of ASD-Sib research on family processes has focused on the relationship between the ASD-Sib and the individual with ASD.

Sibling Relationship

As social communication is one of the core diagnostic features of ASD, it is likely that ASD-Sibs are more likely than comparison groups to face challenges in interacting with their brother or sister with ASD. Additionally, individuals with ASD are more likely to exhibit behavior problems than typically developing individuals; such behavior problems could serve as a barrier to warmth in the sibling relationship. Like other outcomes, however, the sibling relationship can be conceptualized in many different ways. The most commonly used measures of sibling relationships measure both positive and negative factors. For example, the larger subscales of the Sibling Relationship Questionnaire (SRQ; Furman and Buhrmester 1985) include warmth (positive), conflict, and rivalry (both negative), and the more specific SRQ subscales include similarity, affection, nurturance (all three positive), and dominance (negative) both of and by the brother or sister. When it comes to siblings of individuals with ASD, however, it may not always be quite as clear as to whether outcomes are positive or negative. For instance, are scales meant to measure parental partiality—which would normally be considered a negative aspect of the relationship—properly scaled to account for the differing demands of parenting a child with disabilities? Does *teaching*—a scale labeled as positive on the sibling inventory of behavior (SIB; Schaefer and Edgerton 1979)—possibly indicate high levels of parentification? Additionally, as relationships between siblings in which one sibling has an IDD tend to be less egalitarian (Seltzer et al. 1991), some researchers have adapted other measures to try to better examine how ASD-Sibs feel about their brother or sister, adding items to assess guilt, overcompensation, and understanding (Hodapp and Urbano 2007; Martins 2007).

The relationship between individuals with ASD and their typically developing siblings is an important area of research, as a positive sibling relationship is related to more involvement later in life (Burke et al. 2016). As individuals with ASD may need more support in adulthood, having healthy family relationships can be beneficial both to the individual with ASD and the siblings.

Beliefs

Many studies examined ASD-Sibs' perception of different factors, including their beliefs about themselves (e.g., self-concept or self-esteem), beliefs about their brother or sister and the concept of disability, and beliefs about the world in general (e.g., optimism). Beliefs, particularly beliefs about disability in general and self-concept, have been the focus of many theses and dissertations (e.g., Lyons-Sjoström 2003; Pepa 2013), though some published studies have used self-concept scales of broader measures (e.g., strengths and difficulties questionnaire [SDQ]; Verté et al. 2003). Overall, beliefs of the ASD-Sibs is a considerably broad category, with comparatively few studies devoted to the topic and thus, no consensus in the field regarding general outcomes.

Coping

Research among parents and caregivers of individuals with ASD has shown that active, adaptive coping styles, such as problem solving, tend to be related to more positive outcomes, whereas passive, maladaptive coping styles, including denial and substance use, are related to more negative outcomes (e.g., Hastings et al. 2005). Similar to beliefs, many comparative studies on coping among ASD-Sibs were conducted as dissertation or thesis work (e.g., Mukherjee 2010), though one published study did examine problem- and emotion-focused coping styles between ASD-Sibs and siblings of individuals with Down syndrome (Orsmond and Seltzer 2007). Understanding potential differences in coping styles for ASD-Sibs has important implications for the development of targeted intervention strategies to help siblings manage stressors, both within the family and in general.

Adjustment

The concept of *adjustment* among ASD-Sibs is difficult to quantify. In many cases, researchers use the term *adjustment* to refer to more specific outcomes, such as behavior problems (e.g., Petalas et al. 2009) or psychiatric symptoms (e.g., Schwartz 2003). For other studies, however, *adjustment* refers to how ASD-Sibs are functioning in their environment, such as school or home. For example, the Weinberger adjustment inventory (WAI; Weinberger et al. 1987) measures a combination of self-esteem, social competence, and well-being to create a total score of overall socio-emotional adjustment, whereas the Behavior Assessment System for Children (BASC; Sandoval and Echandia 1994) includes a subscale for adaptive skills that measure how well individuals perform activities of daily living, communicate with others, and adapt to changes in their environment. Other measures, such as the stress response scale (Chandler 1983)

measure how children generally respond in the face of stress, a different outcome than adaptive coping mechanisms. Overall, *adjustment* is somewhat of an imprecise term within the sibling literature, but can be used to capture variance in sibling outcomes beyond more traditional measures of behavior problems or psychological symptoms.

Potential Moderators

In addition to the numerous outcome variables for ASD-Sibs, it is possible that certain variables may moderate such results. Of particular interest for the current study are the reporter and the comparison group. In regards to the reporter, Rossiter and Sharpe's meta-analysis of IDD-Sibs in general (2001) did not reveal overall negative effects for sibling self-report, yet parent reports showed a combined negative result. However, these studies included composite samples of siblings of individuals with any IDD; although ASD-Sibs were part of some samples, they were not analyzed separately. Of the studies that include both parent-report and self-report of ASD-Sib outcomes, few, if any, statistically compare the parent perspectives to that of the siblings. Therefore, combining separate instances of parent- and self-report remains the most viable option for understanding how outcomes for ASD-Sibs may differ by who is completing the study.

As for comparison groups, understandably, researchers cannot conduct true experiments on ASD-Sibs, as individuals cannot be randomly assigned to having a brother or sister with ASD or not. Therefore, understanding the ASD-Sib experience is often a product of comparing ASD-Sibs to other groups. The issue of whom to compare ASD-Sibs to is one that has previously been discussed in the disability family literature. Hodapp et al. (2005) raised the issue in a seminal editorial discussing the challenges of sibling research for families of anyone with an intellectual or developmental disability. To truly understand the ASD-Sib (or IDD-Sib) experience, results must be put in context of the outcomes and experiences of other individuals—but what is the most apt context for comparison? The most common comparison groups for ASD-Sib studies include the general population (e.g., normed samples from standardized measures; e.g., Hastings 2003), siblings of individuals without any disabilities (TD-Sibs; e.g., Kaminsky and Dewey 2002), siblings of individuals with physical illness or disabilities (PI-Sibs: e.g., Kao et al. 2009), and siblings of individuals with other intellectual or developmental disabilities (IDD-Sibs; e.g., De Caroli and Sagone 2013). Of the latter group, siblings of individuals with Down syndrome (DS-Sibs; e.g., Shivers et al. 2017) may be of particular interest, as families of individuals with DS often report more positive outcomes and fewer negative outcomes than families of individuals with other IDD, a phenomenon known as the *Down syndrome advantage* (Hodapp et al. 2001). Ultimately, each comparison group offers a different picture and different advantages and disadvantages for the attempted contextualization of the ASD-Sib experience.

Research Aims

Caregivers of individuals with ASD, particularly mothers, consistently report greater stress and worse psychological outcomes than other parents, including parents of children with other intellectual and developmental disabilities (e.g., Hayes and Watson 2013), but researchers are yet to reach a consensus on whether or not ASD-Sibs, as a whole, exhibit similarly persistent challenges. Given that the current body of research on ASD-Sibs is inconclusive regarding key areas of functioning, the present study aimed to address these mixed findings by using meta-analytic methods to combine results from studies of ASD-Sibs to examine the following question: *Do typically developing siblings of individuals with ASD have worse social, emotional, psychological, or behavioral outcomes than other nondisabled populations?* (Research Question 1)

Additionally, analyses were conducted to examine several moderators of the difference in overall functioning between ASD-Sibs and comparison groups, answering the following research questions:

Research Question 2: *How do outcomes for ASD-Sibs compare to specific comparison groups (e.g., TD-Sibs, IDD-Sibs)?*

Research Question 3: *Do ASD-Sibs report different outcomes for themselves than their parents do for them?*

Research Question 4: *Do report characteristics, methodological characteristics, or participant characteristics moderate the difference in overall functioning between ASD-Sibs and comparison groups?*

Method

The meta-analysis reporting standards (MARS; American Psychological Association 2009) were followed in the development, execution, and reporting of the present study. The protocol for this meta-analysis was not registered in a systematic review registry.

Study Selection

Relevant studies were collected through digital and manual searches. Three academic databases were used for the initial digital search of published articles: PsychInfo, EBSCOhost, and Web of Science. Consistent with the standard meta-analysis methodological practice of including effect sizes from unpublished reports to minimize issues surrounding

publication bias (i.e., inflated parameter estimates from only including effect sizes from published reports that tend to have larger effect sizes than unpublished reports; Lipsey and Wilson 2001), theses and dissertations were collected via ProQuest Dissertations and Theses Global. The search terms used were (*autis** OR *Asperger** OR *PDD-NOS*) AND (*sibling** OR *brother** OR *sister**). The digital search covered all journal articles, abstracts, theses, and dissertations published between January of 1960 and December of 2016, either online or in print. Only manuscripts available in English were included in the present analyses. A manual search of reference lists of the included studies was conducted, though it did not yield any manuscripts not already identified.

Figure 1 provides a flow diagram of sample identification, eligibility, and inclusion. The initial search terms were deliberately broad and led to several thousand results in total: 1463 from PsychInfo, 1345 from EBSCOhost, 1852 from Web of Science, and 334 theses and dissertations from ProQuest Dissertation and Theses Global. The titles and abstracts of all articles, theses, and dissertations were reviewed to determine if the studies included ASD-Sibs measured on aspects of psychosocial or behavioral outcomes. Of these results, 112 unique reports from published journals and 50 theses and dissertations were selected for further review, based on titles and abstracts.

For the 162 unique studies flagged for further attention by the researchers, eligibility for inclusion was determined by the following factors: (a) the measures included in the report assessed an emotional, behavioral, or psychological outcome for ASD-Sibs (because the focus of this meta-analysis was on typically-developing siblings, measures used to assess IDD, such as measures of cognitive ability or autistic symptoms, were excluded), (b) the report included information necessary for calculating relevant effect sizes, and (c) the ASD-Sibs in the sample were at least five years old. This age cutoff was chosen for two reasons: first, to help eliminate any *baby sib* studies examining ASD-Sibs for potential developmental markers of ASD and, second, to enhance the comparison validity of several outcome measures. Although certain areas of functioning, such as behavior problems, can be easily and validly assessed in preschool-age children, others, such as beliefs and coping, cannot. Therefore, a minimum age of five for all participants (i.e., the youngest child in the sample could be no younger than five) was selected to maximize both inclusion and validity of analyses.

Because of the comparatively small number of possible studies, all 162 potential manuscripts were reviewed by both the first and third authors for inclusion in the final sample. Of the original search results, 58.6% of articles were not eligible for inclusion. The most common reasons for exclusion were (a) lack of a relevant comparison group, (b) sample participants were too young (e.g., a *baby sibs* study examining risk of ASD in younger siblings of individuals with ASD), (c) irrelevant outcome measures (e.g., measures of cognitive ability or autistic symptoms), and (d) lack of reported statistical information necessary to calculate standardized mean difference scores (e.g., means without

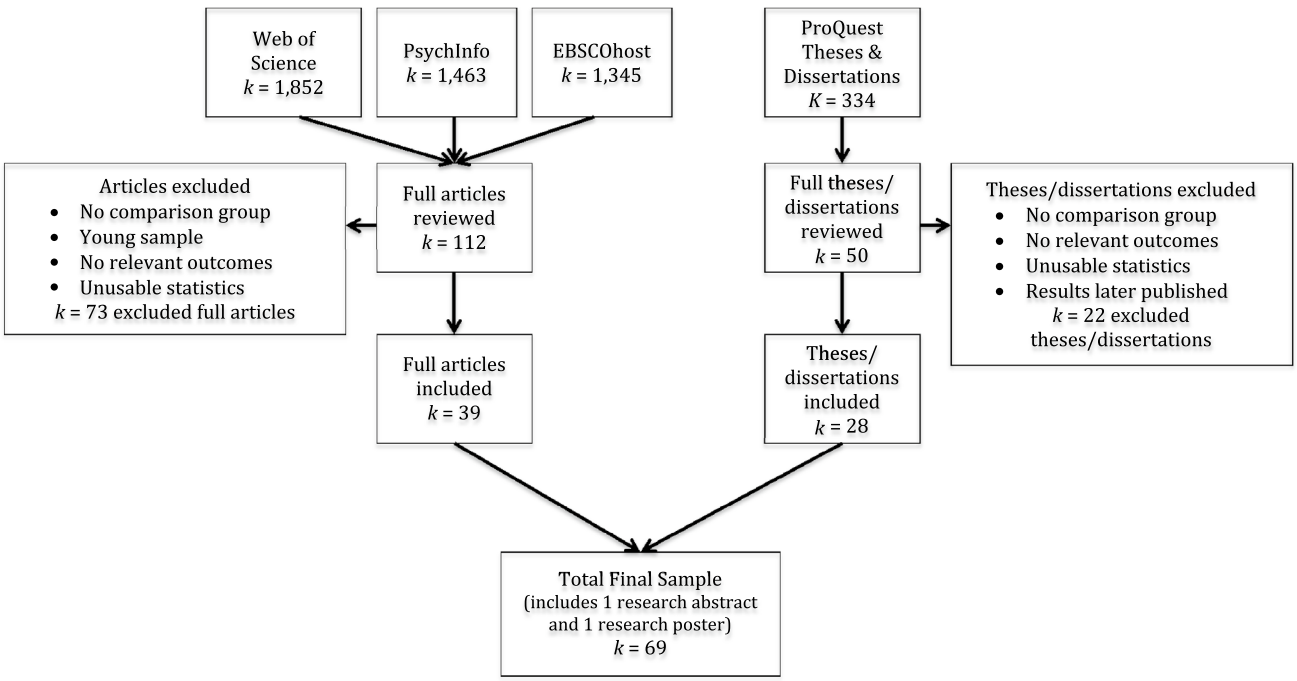


Fig. 1 Flow diagram of sample identification, eligibility, and inclusion

standard deviations; *F*-scores derived from analyses of three or more groups). Whenever possible, we employed standard transformation methods to compute standardized mean difference scores for situations in which they were not provided (Borenstein et al. 2009). Finally, several theses, dissertations, and conference abstracts were excluded, as the results were found in later published articles; therefore, only the peer-reviewed and published versions of such dissertation studies were used for analysis.

We found a substantial number of reports that did not include a comparison group in their own analyses, but implemented commonly used measures (e.g., the Strengths and Difficulties Questionnaire or the Child Behavior Checklist). Therefore, to maximize the number of included results, secondary searches were conducted to find independent reports with samples of comparable size and age range; that is, we searched for reports that included analyses on samples of similar age assessed using the same measure as the ASD-Sib reports. As the comparison groups from the already-included studies consisted of various categories (e.g., siblings of individuals with other IDD, siblings of individuals with no disabilities), the only inclusion criteria employed in selecting external comparison group studies were (a) the sample age range had to be similar to that of the ASD-Sib study and (b) the participants could not have any intellectual or developmental disabilities (i.e., the criterion that is generally used to identify ASD-Sib samples). This procedure led to a total of 14 reports included with matched comparison samples from separate samples. Relatedly, several of the reports used the original normed scores from the validation of the included measures (e.g., ASD-Sib scores on the Strengths and Difficulties Questionnaire compared to a normative sample from Great Britain on the Strengths and Difficulties Questionnaire; Meltzer et al. 2000). Multiple tests of potential differences in sample size revealed that comparing included samples to large normed samples did not differ by more than one one-hundredth of a score from samples of similar size to that of the included articles; thus, it was determined that including large, normed samples as comparison groups was acceptable. However, to maintain independence of samples, we only used each normative sample once. Ultimately, four reports on normative samples were included in the current study as comparison groups for included ASD-Sib samples without comparison groups. The current study included 39 published articles, 28 theses and dissertations, 1 abstract from conference proceedings, and 1 conference research poster.

Coding Procedures

All authors independently coded reports and consulted with one another in instances in which the appropriate code was not clear to reach consensus (Falconier et al. 2015). The first author reviewed all of the coding for accuracy for each report and the second author systematically reviewed each code across all samples to identify and verify anomalies. Heterogeneity test results indicated that there were no significant differences in effect sizes by coder ($Q[2] = 0.54$, $p = 0.762$).

The outcome variable *type of functioning* was coded into 8 original categories: *adjustment, behavior problems, beliefs, coping, family functioning, psychological functioning, sibling relationship*, and *social functioning*. Because of the wide range of outcomes covered by measures of behavior problems, the *behavior problems* category was further divided into *internalizing, externalizing, attention/hyperactivity*, and *total problems*. Brief descriptions of the topics of each category and measures used to assess said topics are found in Table 1.

As indicated in the introduction, there were several areas of potential overlap between measure subscales for *behavior problems* and *psychological functioning*. Therefore, additional analyses were conducted for *psychiatric symptoms* categories (i.e., *anxiety/depression symptoms, externalizing problem symptoms*, and *Attention deficit-hyperactivity disorder [ADHD] symptoms*), with subscales taken from both behavior problems measures and psychological outcomes measures. These *psychiatric symptoms* categories were used to examine whether or not differently focused measures of similar constructs (e.g., clinician diagnosis of anxiety and parent observation of behavior associated with anxiety) revealed similar patterns among ASD-Sibs. Measures included in the *psychiatric symptoms* categories are presented in Table 2.

The comparison group codes indicated to whom ASD-Sibs were compared. Each type of group was labeled, based on the report authors' distinction. For instance, some authors compared ASD-Sibs to siblings of individuals with specific diagnoses of IDD (e.g., Prader–Willi syndrome; O'Neill and Murray 2016), whereas others compared them to siblings of individuals with different diagnoses who were grouped together (e.g., life-limiting conditions; Fullerton et al. 2017). These specifications were then combined into five groups:

1. Intellectual and developmental disability siblings (IDD-Sibs): siblings of individuals with other intellectual or developmental disabilities.
2. Physical illness or disability siblings (PID-Sibs): siblings of individuals with physical chronic illness or disability.
3. Typically developing siblings (TD-Sibs): siblings of individuals without any physical or intellectual disabilities.
4. Typically developing individuals (TD-Individuals): individuals without any intellectual or developmental

Table 1 Constructs and measures for areas of functioning

Types of functioning	Constructs included	Measures included
Adjustment	Academic competence, stress level, activeness, school performance, adaptability, personal adjustment, parental concerns, dependence, impulsivity, passive-aggressive reactivity, repression, physical well-being, spiritual well-being, socio-emotional adjustment	Child Behavior Checklist (CBCL), Social Skills Rating System, Stress Response Scale, Weinberger Adjustment Inventory, Strengths and Difficulties Questionnaire (SDQ), bespoke surveys
Behavior problems—attention/hyperactivity	Attention problems, hyperactivity, inattention, thought problems	CBCL (parent-report, teacher-report, youth self-report), SDQ, Behavior Assessment System for Children Second Edition (BASC-2)
Behavior problems—externalizing	Externalizing behavior, conduct problems, aggression, delinquent behavior, disciplinary problems, rule breaking	CBCL (parent-report, teacher-report, youth self-report), SDQ, BASC-2, bespoke surveys
Behavior problems—internalizing	Internalizing behavior, anxious behavior, depression behavior, withdrawal, emotional problems, somatic complaints	CBCL (parent-report, teacher-report, youth self-report), SDQ, bespoke surveys
Behavior problems—total	Total behavior problems scales	CBCL (parent-report, teacher-report, youth self-report), SDQ, Social Skills Rating System, bespoke surveys
Beliefs	Attitudes toward disability, global self-concept, global self-esteem, social self-esteem, academic self-esteem, parental self-esteem, self-worth, optimism, pessimism, locus of control, guilt, life satisfaction, quality of life	Piers-Harris Children's Self-Concept Scale, Pictorial Scale of Perceived Competence, Perceived Competence Scale for Children, SDQ, Nowicki-Strickland Locus of Control Scale, Rosenberg Self-Esteem Scale, Culture-Free Self-Esteem Inventories 2, Children's Attributional Style Questionnaire, Benefit Finding Questionnaire, Caregiver Strain Questionnaire, Satisfaction with Life Scale, bespoke surveys
Coping	Problem-focused coping, active coping, suppression, planning, restraint, instrumental social support, adaptive emotion-focused coping, emotional social support, acceptance, religion, maladaptive emotion-focused coping, denial, mental disengagement, behavioral disengagement, substance use	COPE Inventory, Multidimentional Coping Inventory
Family functioning	Avoidance of parents, anxiety towards parents, perceived pressure from parents, positive affect, facilitation of independence, attitudinal independence, conflictual independence, emotional independence, functional independence	Parental Attachment Questionnaire, Experience in Close Relationships-Revised, Psychological Separation Inventory, Parental Attachment Questionnaire, Impact on Sibling Scale
Psychological functioning	Scale measures: Anxiety, generalized anxiety, separation anxiety, social anxiety, panic, OCD, depression, negative mood, negative self-esteem, ineffectiveness, interpersonal problems, functional problems, emotional problems Diagnoses: any psychiatric diagnosis, ADHD, oppositional defiant disorder, anxiety disorder, elimination disorder, tics disorder, depression, comorbid disorders, OCD, schizophrenia spectrum disorders, affective disorders, anxiety disorders, neurotic and personality disorders, alcohol and drug addiction or abuse, childhood onset disorders, learning disorders, emotional disorders, conduct and oppositional disorders, eating disorder	Center for Epidemiologic Studies Depression Scale (CES-D), Children's Depression Inventory-2, Revised Child Anxiety and Depression Scale, clinician report, Kiddie Schedule for Affective Disorders and Schizophrenia (K-SADS), International Classification of Diseases (ICD-10/ICD-9), Beck Depression Inventory, BASC-2, Revised Children's Manifest Anxiety Scale, Stress Appraisal Measure, Depression, Anxiety, and Stress Scale (DASS), Hospital Anxiety and Depression Scale

Table 1 (continued)

Types of functioning	Constructs included	Measures included
Sibling relationship	Companionship, empathy, teaching, positive involvement, rivalry, aggression, avoidance, anxiety, total relationship, hostility, warmth, conflict, closeness, relative status/power, similarity, admiration, affection, nurturance by sibling, nurturance of sibling, dominance by sibling, dominance of sibling, quarrelling, antagonism, competition, maternal partiality, paternal partiality, understanding, trust, fairness, respect, negative experiences, overcompensation, guilt, worry-current, worry-future, embarrassment	Sibling Inventory of Behavior, Lifespan Sibling Relationship Scale, Experience in Close Relationships-Revised, Sibling Relationship Questionnaire, Subjective Sibling Experiences, Positive Affect Index, Network of Relationships Inventory, bespoke surveys
Social functioning	Social problems, social competence, social communication, pro-social behavior, social skills, empathy, assertivity	CBCL, Pro-social Behavior Questionnaire, Concern for Others Scale, Vineland Adaptive Behavior Scale, Empathy Quotient, Social Skills Rating System, Matson Evaluation of Social Skills with Youngsters

"Bespoke surveys" refers to measures that were created for one specific study and have not been validated

disabilities, with no specification that they have brothers or sisters.

5. Normed comparison: normative samples from validated measures.

The normed comparison group was used for samples with reported results from measures with existing norms that did not include a comparison group. Norms for each measure were only used once to avoid violations of sample independence. Additionally, there were enough ASD-Sibs samples compared to samples of siblings of individuals with Down syndrome (DS-Sibs) to analyze the differences between ASD-Sibs and DS-Sibs separately to test the hypothesis of the *Down syndrome advantage*.

Each effect size was also coded based on who provided the data used to calculate the effect size. The majority of reporters were parents (i.e., mothers, fathers, or other guardians) or the siblings themselves (i.e., self-report), although a few samples included teacher-report. Because only two reports split parent-report results by mother-report and father-report (i.e., separate numeric results for mother reporters and father reporters), they were aggregated at the parental-report level (i.e., included in moderator analyses as *parent report*, along with all other reports that did not separate mother from father ratings in the original analyses).

Study quality was managed by coding various study design aspects to facilitate subsequent analysis of potential moderators (Card 2012). Study quality potential moderators were categorized as report characteristics and methodological characteristics. Report characteristics included report type (published peer-reviewed articles, unpublished conference abstracts and posters, and unpublished theses and dissertations) and report publication status (published or unpublished). Methodological characteristics included ASD-Sibs selection criterion (e.g., age, birth order, gender), sample design (cross sectional vs. longitudinal), whether or not study design tested an intervention, whether the comparison group was included or added by the researchers, and measure type (e.g., clinical assessment, interview questions, observation, questionnaire). We also quantitatively and categorically coded participant characteristics (e.g., age, gender, nationality, race, and ethnicity) of the ASD-Sib and comparison groups for moderator analysis. Multiple sample characteristics were also noted to aid in summarizing the samples (e.g., publication year, response rate, ASD severity; see Supplemental Table 1).

Statistical Methods

All analyses were completed with Comprehensive Meta Analysis Version 3 (Borenstein et al. 2014). Given the diversity in methodological characteristics among studies, random-effects models were used to weight the estimated

Table 2 Constructs and measures for psychiatric symptoms

Psychiatric symptoms	Subscales included	Included measures
ADHD symptoms	Attention problems, hyperactivity, inattention/hyperactivity, ADHD	SDQ, BASC-2, CBCL, Growing up with a Sibling with Autism: Adult Perspectives Survey, ICD-10/ICD-9, K-SADS
Anxiety/depression symptoms	Anxiety/depression, anxiety, depression, generalized anxiety, separation anxiety, social anxiety	CBCL, Youth Self-Report, BASC-2, Children's Depression Inventory Revised Child Anxiety and Depression Scale (RCADS), CES-D, Growing up with a Sibling with Autism: Adult Perspectives Survey, ICD-10/ICD-9, Revised Children's Manifest Anxiety Scale, Beck Anxiety Inventory, Beck Depression Inventory, Hospital Anxiety and Depression Scale (HADS), Center for Epidemiological Studies Depression Scale for Children, Multidimensional Anxiety Scale for Children (MASC), K-SADS, Depression, Anxiety, and Stress Scale (DASS)
Externalizing behavior symptoms	Externalizing problems, delinquent behavior, aggression, disciplinary problems, conduct problems, rule breaking, conduct and oppositional disorders	SDQ, CBCL, BASC-2, Growing up with a Sibling with Autism: Adult Perspectives Survey, ICD-10/ICD-9, K-SADS

standardized mean difference effect size for each comparison by the *inverse variance weight* (i.e., the inverse of the sum of the estimated within-sample variance and the estimated between-samples variance for all of the included samples; Borenstein et al. 2009). As mentioned in the introduction, many ASD-Sib studies utilized small samples of convenience; thus, we calculated Hedges' g values (i.e., unbiased sample estimate standardized mean difference effect sizes) for all comparisons to correct for small sample size bias (Hedges 1981). Samples analyzed in more than one report were grouped together to calculate a single effect size per sample, thus ensuring sample independence; instances in which multiple effect sizes from the same sample were analyzed together (e.g., mother-report of aggression and father-report of aggression to calculate parental-report of aggression, anxious behavior and depressive behavior to calculate internalizing behavior problems) were similarly averaged to calculate a single effect size per sample to insure sample independence (Lipsey and Wilson 2001).

The Hedges' g values for each comparison were aggregated to create a comprehensive estimate of the overall difference magnitude between ASD-Sibs and comparison groups across all included samples, along with measures of precision (SE, 95% CI) and significance (p; Borenstein et al. 2009). Odds ratios were also calculated to enhance the interpretability of the results; all odds ratio ranges were based on 95% confidence intervals. Moderator variable subgroup parameters were constructed such that samples would be distributed relatively similarly across subgroups. Group differences for moderating variables were testing using a mixed-effects model (fixed-effect model testing across subgroups and random-effects model testing within subgroups; Borenstein et al. 2009).

The following ranges are a guideline for interpreting standardized mean difference estimate magnitude: small ($0.20 \leq g < 0.50$), medium ($0.50 \leq g < 0.80$), and large ($g \geq 0.80$; Cohen 1988). Positive effect sizes indicate that, on average, ASD-Sibs demonstrated higher functioning than comparison groups, whereas negative effect sizes indicate that, on average, ASD-Sibs demonstrated lower functioning than comparison groups. The statistical power for the calculation of the estimated aggregated unbiased standardized mean difference (Hedges' g) in overall functioning between ASD-Sibs and comparison groups was 1.00.

Results

Included Samples Summary

Samples

A total of 69 independent samples (K) from 69 reports met inclusion criteria (see Fig. 1). In terms of sample independence, 1 of the 69 reports contained 2 independent samples (i.e., Jokiranta-Olkoniemi et al. 2016), increasing the number of independent samples by 1; conversely, 2 of the 69 reports were based on the same sample (i.e., Kaminsky and Dewey 2001, 2002; both reported on different outcomes for the same sample) and were consequently coded and analyzed as the same sample, reducing the number of independent samples by 1 (see Fig. 2). Because most of the reports contained multiple relevant effect sizes for the respective samples, a total of 836 effect sizes were coded across the 69 independent samples; analyses were conducted

by calculating a mean effect size for samples with multiple relevant effect sizes.

Included sample reports (publication year of published reports and completion year of unpublished reports) ranged from 1980 to 2017 (*Mdn* = 2010). In terms of included sample report publication status, 56.5% were published and 43.5% were unpublished dissertations, theses, abstracts, and research posters. Included samples were recruited from ASD organization websites; blogs; communities; healthcare systems; hospitals; mental health agencies; national databases and registries; national, state, and local organizations; preschool and early childhood centers; research centers; school systems; social networking sites; special recreation programs; state and local disability organizations; and university courses. Most of the samples were recruited from more than one source. The majority of reports (72.9%) did not indicate criteria for selecting the ASD-Sibs in relation to the person with ASD; the samples for which the criteria were reported included age (e.g., closest in age, age restriction), birth order (e.g., first-born siblings, older, younger), gender (e.g., brother, sister), involvement level (e.g., most involved), and volunteer (e.g., self-selection).

The vast majority of the data from included samples were obtained through cross-sectional research designs (95.7%) as opposed to longitudinal research designs (4.3%). The data from a few of the samples (4.3%) were collected from intervention research designs; only the pre-intervention effect sizes from these samples were coded. Most of the data were collected through questionnaire measures (89%), with the remainder of the data collected through clinical assessment and diagnosis, interview questions, and observation. The data were collected through self-report (46.1%), parent-report (43.8%), teacher-report (5.6%), medical records (3.4%), and researcher observation (1.1%).

The combined total sample size was 27,942, of which 6679 were ASD-Sibs and 21,263 were comparison group participants. The median ASD-Sibs sample size was 31 and the median comparison group sample size was 40; the difference in ASD-Sibs sample sizes and comparison group sample sizes ranged from 47 more ASD-Sibs to 7120 more comparison participants, with no median difference in sample size (*Mdn* = 0). See Supplemental Table 1 for additional descriptive sample characteristics information.

Participants

All of the ASD-Sibs samples consisted exclusively of ASD-Sibs; that is, no samples that included siblings of individuals with various intellectual and developmental disabilities were used for the present analyses. The comparison group samples consisted of siblings of individuals without any disability (47.0%; i.e., typically developing siblings or TD-Sibs), siblings of individuals with intellectual and developmental disabilities (31.3%; IDD-Sibs; e.g., Down syndrome, learning disability), siblings of individuals with physical illness or disability (6.0%; PID-Sibs; e.g., cancer, deafness, diabetes), siblings of individuals with other conditions (3.6%; i.e., behavioral problems, life-limiting conditions, mixed disabilities), and participants who were not in exclusively sibling samples (12.0%; i.e., normed comparisons, individuals with physical illness, and typically developing individuals or TD-Individuals). Additional demographic characteristics are presented in Table 3.

ASD-Sibs Overall Functioning

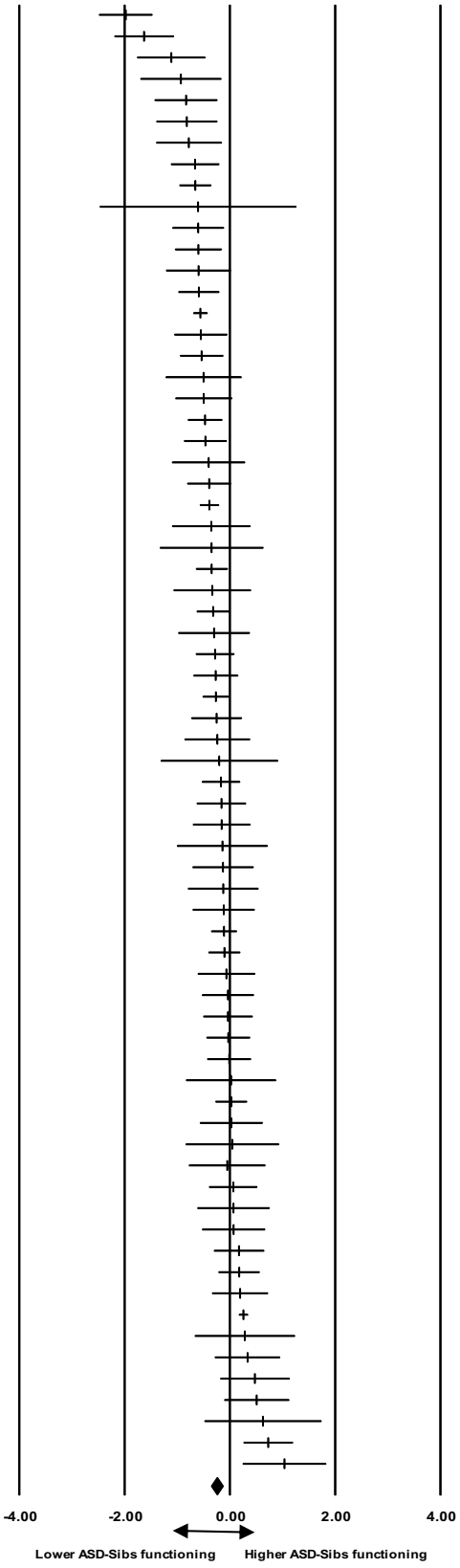
The estimated aggregated unbiased standardized mean difference (Hedges' *g*) in overall functioning between ASD-Sibs and comparison groups was −0.26 (SE = 0.06, 95% CI [−0.37, −0.15], $p \leq 0.001$, $K = 69$), indicating a small difference in terms of magnitude. The odds ratio equivalent of the estimated aggregated unbiased standardized mean difference in overall functioning between ASD-Sibs and comparison groups was 0.62 (95% CI [0.51, 0.76], $p < 0.001$); using the inverse of the odds ratio for the purpose of interpretation, the odds of ASD-Sibs overall functioning being lower than comparison groups were 1.3 to 2.0 times greater than the odds of comparison group overall functioning being lower than ASD-Sibs.

The estimated unbiased standardized mean difference in overall functioning between ASD-Sibs and comparison groups is summarized by sample as a forest plot in Fig. 2. The estimated between-sample variance for the true mean difference in overall functioning between ASD-Sibs and comparison groups (T^2) was 0.14 and the estimated standard deviation of the true mean difference in overall functioning between ASD-Sibs and comparison groups (T) was 0.37. The ratio of estimated true between-sample variance to observed between-sample variance (I^2) was 76.67%, signifying that if it were possible to eliminate the sampling error, the observed variance (i.e., the dispersion in the forest plot) would likely not be significantly reduced (Borenstein et al. 2017). An I^2 of 77% suggests limited overlap of sample estimate confidence intervals, such that the difference in overall functioning between ASD-Sibs and the comparison groups varied from one population to another. The prediction interval (a dispersion index of how extensively effect sizes vary across populations) was −1.00 to 0.49, indicating that the difference in overall functioning between ASD-Sibs and the comparison groups will occur between −1.00 and 0.49 in 95% of all populations (Borenstein et al. 2017).

Areas of Functioning

Analyses of differences by type of functioning (Table 4) indicated that ASD-Sibs had significantly more negative

Study name	Hedges's g	Standard error	Lower limit	Upper limit	p-Value	ASD-SIBS	Comparison
Eyuboglu, 2015	-1.98	0.26	-2.50	-1.46	0.000	41	43
Marciano, 2005	-1.63	0.29	-2.20	-1.06	0.000	31	30
Trubia, 2016	-1.11	0.34	-1.77	-0.46	0.001	31	31
Zomick, 2009	-0.93	0.40	-1.71	-0.15	0.019	14	13
Abdallah, 2015	-0.83	0.31	-1.44	-0.23	0.007		
Martins, 2007	-0.82	0.30	-1.41	-0.23	0.006	25	25
Schwartz, 2003	-0.78	0.32	-1.41	-0.15	0.016	28	16
Smith, 2006*	-0.66	0.24	-1.13	-0.19	0.006	40	44
Meyer, 2011*	-0.66	0.16	-0.97	-0.34	0.000	70	99
Rosa, 2016	-0.61	0.96	-2.48	1.27	0.527	24	22
Smith, 2000*	-0.60	0.25	-1.10	-0.10	0.018	31	53
Belkin, 2013	-0.60	0.23	-1.05	-0.15	0.009	31	54
Lovell, 2016	-0.60	0.32	-1.22	0.03	0.061	22	18
Pollard, 2013	-0.59	0.20	-0.99	-0.19	0.004	81	38
Jokiranta-Olkoniemi, 2016a	-0.56	0.07	-0.71	-0.42	0.000	3115	10235
Prystalski, 1997	-0.55	0.26	-1.07	-0.04	0.034	30	60
De Caroli, 2013	-0.54	0.21	-0.96	-0.12	0.012	46	94
Granat, 2012	-0.50	0.37	-1.23	0.23	0.180	13	17
Stampoltzis, 2014*	-0.50	0.28	-1.05	0.05	0.077	22	64
Miller, 2016	-0.47	0.17	-0.81	-0.13	0.006	79	60
Jokiranta-Olkoniemi, 2016b	-0.47	0.21	-0.88	-0.05	0.028	463	1540
Rodrigue, 1993	-0.40	0.36	-1.11	0.30	0.261	19	20
Tomeny, 2012	-0.39	0.22	-0.82	0.03	0.072	42	42
Hodapp, 2007	-0.39	0.10	-0.58	-0.20	0.000	176	284
Farber, 2010	-0.35	0.38	-1.10	0.40	0.358	14	13
Rao, 2009	-0.35	0.51	-1.34	0.65	0.495	7	8
Janecek, 2015	-0.34	0.16	-0.66	-0.03	0.031	140	342
Gold, 1993	-0.33	0.38	-1.08	0.41	0.380	11	17
Orsmond, 2007	-0.32	0.16	-0.64	-0.00	0.049	77	77
Wong, 2007	-0.30	0.35	-0.99	0.39	0.390	21	13
Chan, 2016*	-0.28	0.19	-0.66	0.09	0.143	116	116
Tomeny, 2017	-0.27	0.22	-0.70	0.17	0.226	45	37
Gau, 2010	-0.27	0.13	-0.53	-0.01	0.044	120	109
O'Neill, 2016	-0.25	0.25	-0.74	0.24	0.311	31	233
Shepard, 1992	-0.24	0.32	-0.87	0.39	0.457	19	19
Verté, 2003	-0.20	0.57	-1.32	0.92	0.727	29	29
O'Kelley, 2006	-0.17	0.19	-0.54	0.20	0.375	57	53
Bemister, 2012	-0.16	0.24	-0.64	0.32	0.508	31	79
Petalas, 2009	-0.16	0.28	-0.71	0.40	0.579	25	24
Huff, 2006	-0.14	0.45	-1.02	0.73	0.748	19	19
Quintero, 2010	-0.13	0.30	-0.72	0.46	0.667	20	23
Pepa, 2013	-0.13	0.35	-0.81	0.55	0.713	15	18
Warren, 2012	-0.12	0.31	-0.72	0.48	0.699	39	22
Hastings, 2014*	-0.11	0.13	-0.36	0.14	0.397	60	4228
Hallet, 2013	-0.10	0.16	-0.42	0.21	0.510	55	144
Ross, 2006*	-0.07	0.28	-0.62	0.49	0.816	25	24
Kaminsky, 2001 + 2002	-0.04	0.26	-0.54	0.47	0.886	30	30
Park, 2012	-0.04	0.24	-0.51	0.44	0.883	98	51
Rodgers, 2016	-0.03	0.22	-0.45	0.39	0.888	42	42
Kao, 2009	-0.01	0.22	-0.44	0.41	0.950	50	72
Mukherjee, 2010	0.02	0.44	-0.85	0.89	0.962	21	20
Walton, 2015	0.03	0.16	-0.28	0.34	0.871	69	93
Pope, 1987*	0.03	0.31	-0.58	0.63	0.928	17	64
Sanders, 1993	0.05	0.46	-0.85	0.94	0.919	18	37
Lyons-Sjostrom, 2003	-0.05	0.38	-0.79	0.69	0.892	14	43
Palafox, 2004*	0.06	0.24	-0.41	0.53	0.793	37	66
Moreno, 2010	0.07	0.36	-0.63	0.76	0.853	15	15
Pilowsky, 2004	0.07	0.31	-0.54	0.68	0.822	30	58
Fullerton, 2017	0.17	0.25	-0.32	0.66	0.490	32	32
Glasberg, 1998*	0.18	0.21	-0.23	0.58	0.391	63	38
Solarsh, 2016*	0.19	0.28	-0.35	0.74	0.483	53	50
Dempsey, 2012*	0.26	0.05	0.16	0.36	0.000	486	1753
Barak-Levy, 2010	0.29	0.49	-0.68	1.25	0.560	27	27
Lampert, 2007	0.33	0.32	-0.30	0.96	0.298	20	100
McCall, 2013	0.47	0.34	-0.19	1.14	0.164	20	20
Roeyers, 1995	0.51	0.32	-0.12	1.14	0.111	20	20
Bryce, 1983	0.63	0.57	-0.49	1.75	0.272	7	7
Surfas, 2005*	0.73	0.24	0.25	1.21	0.003	40	31
Berger, 1980*	1.04	0.41	0.23	1.84	0.012	20	45
	-0.26	0.06	-0.37	-0.15	0.000		



◂**Fig. 2** Forest plot of the aggregation of differences in overall functioning between ASD-Sibs and comparison groups by sample. Included samples are ordered from smallest effect size magnitude to largest effect size magnitude, and listed by report first author last name and publication year for published reports and completion year for unpublished reports. Samples analyzed in multiple reports are indicated by combining (+) the first author last name and year for the related reports. Reports with letters at the end indicate the presence of more than one independent samples included within the same report. Reports with an * did not contain a comparison group; comparison groups were identified and added for these reports. Antonopoulou et al. (2012), Carbone et al. (2014), Casey-Cappello (1997), Chalmers (2004), Dunn (2005), Hodgkinson (2010), Lardieri (1996), Munsie (1992), Sheehan (1989), and Sleeman et al. (2010) were used as external comparisons for the included reports indicated with an *. In addition to being presented numerically, sample point estimate Hedges' *g* difference scores and 95% confidence intervals (CI) are also presented graphically with vertical lines and horizontal lines respectively. The final row provides the random-effects model estimated aggregated unbiased standardized mean difference (Hedges' *g*) in overall functioning between ASD-Sibs and comparison groups (graphically indicated by the apex of the filled diamond) and associated 95% CI (graphically indicated by the width of the filled diamond). The double-arrow line represents the prediction interval (the dispersion index of the effect size ranges across populations)

beliefs (e.g., beliefs about themselves and beliefs about disability in general; $g = -0.25$, $p = 0.013$), more internalizing behavior problems ($g = -0.24$, $p = 0.004$), worse psychological functioning ($g = -0.28$, $p = 0.003$), worse sibling relationships ($g = -0.42$, $p < 0.001$), and poorer social functioning ($g = -0.23$, $p = 0.007$) than comparison groups; there were no significant differences in magnitude between the areas of functioning in which ASD-Sibs fared significantly worse than the comparison groups ($p = 0.359$). The odds of ASD-Sibs having more negative beliefs than comparison groups were to 1.1 to 2.3 times the odds of comparison groups having more negative beliefs than ASD-Sibs. Additional component analyses indicated that there were no differences in self-concept beliefs ($g = 0.02$, $p = 0.885$) yet significant moderate differences in disability attitudes and beliefs ($g = -0.56$, $p = 0.000$), indicating that the odds of ASD-Sibs having more negative attitudes and beliefs about disability were 1.7 to 4.6 times the odds of comparison groups having more negative attitudes and beliefs about disability.

The odds of ASD-Sibs having more internalizing behavior problems were 1.2 to 2.1 times the odds of comparison groups having more internalizing behavior problems. The odds of ASD-Sibs having lower psychological functioning were 1.2 to 2.4 times the odds of siblings of individuals without ASD having lower psychological functioning. The odds of ASD-Sibs having lower-quality relationships with their sibling(s) with ASD were 1.5 to 3.0 times the odds of comparison groups having lower-quality relationships with their sibling(s). The odds of ASD-Sibs having lower social functioning were 1.1 to 2.1 times the odds of comparison groups having lower social functioning. There were no significant differences between ASD-Sibs and the comparison groups in adjustment, attention and hyperactivity behavior problems, externalizing behavior problems, total behavior problems, coping, and family functioning. There were also no areas of functioning in which ASD-Sibs fared significantly better than comparison groups.

As mentioned in the method section, certain subcategories of functioning were measured in different ways—some with behavioral measures, other with more clinically focused methods. As assessed by *behavioral measures*, there were no differences between ASD-Sibs and comparison groups on combined anxiety and depression, ADHD symptoms, or externalizing behavior symptoms; however, as assessed by *psychiatric measures*, ASD-Sibs indicated higher levels of all three symptoms combined: anxiety and depression, ADHD symptoms, and externalizing behavior symptoms (see Table 4). Given the number of samples with effect sizes for psychiatric symptoms, cross-area analyses combining behavior problems areas (scales designed to measure behavioral symptoms, primarily through self-, parent-, and teacher-report) and psychological functioning (clinical measures and diagnostic categories primarily through trained clinical interview and observation) were conducted for ADHD symptoms, combined anxiety and depression symptoms, and externalizing behavior symptoms.

Results of cross-area analyses of differences in functioning by psychiatric symptoms (see Table 5) indicated higher levels among ASD-Sibs than comparison groups in ADHD symptoms ($g = -0.23$, $p = 0.020$, *inverse OR* 95% CI 1.07, 2.15) and combined anxiety and depression symptoms ($g = -0.25$, $p = 0.004$, *inverse OR* 95% CI 1.16, 2.13), and no difference in externalizing behavior symptoms ($g = -0.13$, $p = 0.085$, *inverse OR* 95% CI 0.97, 1.64). Although anxiety symptoms and depression symptoms were reported as combined in the internalized behavior problems area, they were reported separately in the psychological functioning area, making it possible to analyze them separately as well. *Anxiety symptoms* consisted of the following aggregated constructs: anxiety, anxiety disorders, generalized anxiety, panic, separation anxiety, and social anxiety. *Depression symptoms* consisted of the following aggregated constructs: affective disorders, depression, and depression symptoms such as negative mood and negative self-esteem. Results (see Table 5) indicated higher levels of anxiety symptoms ($g = -0.25$, $p < 0.001$) and depression symptoms ($g = -0.36$, $p < 0.001$) among ASD-Sibs than siblings of individuals without ASD. The inverse of the odds ratios was calculated for interpretation purposes and indicated that the odds of ASD-Sibs presenting with anxiety symptoms was 1.4 to 1.8 times the odds of siblings of individuals without ASD presenting with anxiety symptoms, and the odds of ASD-Sibs presenting with depression symptoms was 1.7 to 2.2 times

Table 3 Descriptive summary of sample participant characteristics

	k	%	*K*	*X*	SD	Min	Max
ASD-Sibs			69				
Average age	57	82.6		13.79	7.73	5.30	41.00
Average age SD	48	69.6		3.64	2.90	0.69	12.60
Gender (% female)	59	85.5		0.55	0.15	0.25	0.91
Ethnicity/race (% people of color)	31	44.9		0.26	0.29	0.00	1.00
Comparison group			69				
Average age	55	79.7		14.49	9.20	5.36	42.50
Average age SD	45	65.2		3.31	3.13	0.45	14.59
Gender (% female)	58	84.1		0.56	0.15	0.25	0.95
Ethnicity/race (% non-White)	31	44.9		0.28	0.31	0.00	1.00
Person with ASD			69				
Average age	44	63.8		12.63	7.25	4.35	34.88
Average age SD	39	56.5		4.31	3.04	1.12	13.00
Age difference with sibling	8	11.6		3.20	0.68	2.64	4.71
Gender (% female)	46	66.7		0.20	0.15	0.00	0.88
Gender differences (% same gender)	7	10.1		0.51	0.06	0.40	0.62
Intelligence quotient	7	10.1		84.71	27.48	40.00	118.00

k the number of samples for each sample participant characteristic category, *K* the number of relevant samples for each sample participant characteristic, *X* average, *SD* standard deviation, *Min* minimum reported value, *Max* maximum reported value

Table 4 Differences in functioning by area of functioning

Outcome	Summary Information					Heterogeneity		
	g	SE	95% CI	*p*	*k*	*Q*	*p*	*K*
Area of functioning						10.42	0.405	198
Adjustment	−0.18	0.13	[−0.44, 0.07]	0.162	12			
Behavior problems: attention/hyperactivity	−0.06	0.12	[−0.30, 0.17]	0.604	12			
Behavior problems: externalizing	−0.08	0.08	[−0.24, 0.08]	0.420	27			
Behavior problems: internalizing	**−0.29**	**0.08**	**[−0.41, −0.08]**	**0.004**	**26**			
Anxiety and depression	−0.23	0.15	[−0.53, 0.06]	0.121	6			
Behavior problems: total	−0.15	0.09	[−0.34, 0.03]	0.100	22			
Beliefs	**−0.25**	**0.10**	**[−0.45, −0.05]**	**0.013**	**22**	**10.63**	**0.001**	**19**
Disability attitudes and beliefs	**−0.56**	**0.14**	**[−0.82, −0.29]**	**0.000**	**4**			
Self-concept	0.02	0.11	[−0.20, 0.23]	0.885	15			
Coping†	−0.09	0.25	[−0.59, 0.41]	0.721	3			
Family functioning†	−0.18	0.21	[−0.60, 0.24]	0.403	4			
Psychological functioning†	**−0.29**	**0.10**	**[−0.47, −0.10]**	**0.003**	**21**	**32.28**	**0.000**	**24**
ADHD†	**−0.72**	**0.06**	**[−0.83, −0.61]**	**0.000**	**4**			
Anxiety and depression†	**−0.34**	**0.04**	**[−0.42, −0.27]**	**0.000**	**17**			
Externalizing behavior†	**−0.56**	**0.06**	**[−0.67, −0.45]**	**0.000**	**3**			
Sibling relationship†	**−0.42**	**0.10**	**[−0.61, −0.23]**	**0.000**	**21**			
Social functioning	**−0.23**	**0.08**	**[−0.39, −0.06]**	**0.007**	**28**			

Parameter estimates were calculated using a random effects model and heterogeneity *Q* tests for between-category differences were calculated using a mixed-effects model. Significant negative Hedges' *g* values indicate ASD-Sibs functioning was lower than comparison group functioning on average for the specified area of functioning. Bolded headers were significant at the $p \leq 0.05$ level for heterogeneity (i.e., between-category differences); bolded areas of functioning were statistically significant at the $p \leq 0.05$ level

g estimated aggregated unbiased standardized mean difference (Hedges' *g*), *SE* standard error, *95% CI* confidence interval, *p* level of statistical significance for the associated Hedges' *g* and heterogeneity *Q* test, *k* number of samples in each area of functioning, *K* the total number of samples included in analyses, *Q* the *Q*-value of the heterogeneity test for between-category differences

†Analyses with comparisons groups consisting exclusively of siblings of individuals without ASD

Table 5 Differences in functioning by psychiatric symptoms

Outcome	Summary information					Heterogeneity		
	g	SE	95% CI	*p*	*k*	*Q*	*p*	*K*
Behavior problems + psychological functioning						1.33	0.514	69
ADHD	**−0.23**	**0.10**	**[−0.42, −0.04]**	**0.020**	**16**			
Anxiety and depression	**−0.25**	**0.09**	**[−0.41, −0.08]**	**0.004**	**23**	3.57	0.059	27
Anxiety[†]	**−0.25**	**0.04**	**[−0.34, −0.17]**	**0.000**	**12**			
Depression[†]	**−0.36**	**0.03**	**[−0.42, −0.29]**	**0.000**	**15**			
Externalizing behavior	−0.13	0.07	[−0.27, 0.02]	0.085	30			

Parameter estimates were calculated using a random effects model and heterogeneity *Q* tests for between-category differences were calculated using a mixed-effects model. Significant negative Hedges' *g* values indicate ASD-Sibs functioning was lower than comparison group functioning on average for the specified area of functioning. Bolded categories of psychiatric symptoms were statistically significant at the $p \leq 0.05$ level

g estimated aggregated unbiased standardized mean difference (Hedges' *g*), *SE* standard error, *95% CI* confidence interval, *p* level of statistical significance for the associated Hedges' *g* and heterogeneity *Q* test, *k* number of samples in each area of functioning, *K* the total number of samples included in analyses, *Q* the *Q*-value of the heterogeneity test for between-category differences

[†]Analyses with comparisons groups consisting exclusively of siblings of individuals without ASD

the odds of siblings of individuals without ASD presenting with depression symptoms. The effect sizes for anxiety symptoms and depression symptoms were not significantly different from one another ($p = 0.059$).

Moderators

Analyses of differences in overall functioning based on comparison group (Table 6) indicated that ASD-Sibs fared

Table 6 Differences in overall functioning by comparison group

Outcome	Summary information					Heterogeneity		
	g	SE	95% CI	*p*	*k*	*Q*	*p*	*K*
Sibling versus non-sibling heterogeneity test						2.98	0.084	70
Sibling comparison groups	**−0.30**	**0.05**	**[−0.41, −0.19]**	**0.000**	**60**			
Non-sibling comparison groups	−0.06	0.13	[−0.31, 0.18]	0.611	10			
Comparison group heterogeneity test						10.18	0.117	80
Intellectual/developmental disabilities siblings (IDD-Sibs)[a]	**−0.31**	**0.12**	**[−0.55, −0.07]**	**0.011**	**12**			
Down syndrome siblings (DS-Sibs)	**−0.40**	**0.11**	**[−0.62, −0.18]**	**0.000**	**11**			
Physical illness siblings (PI-Sibs)[b]	−0.22	0.17	[−0.55, 0.11]	0.191	5			
Typically developing siblings (TD-Sibs)	**−0.31**	**0.07**	**[−0.44, −0.18]**	**0.000**	**39**			
Other impairments siblings[c]	0.26	0.24	[−0.21, 0.72]	0.275	3			
Normed	0.02	0.17	[−0.31, 0.35]	0.903	4			
Physical illness individuals + typically developing individuals (TD-Individuals)	−0.15	0.17	[−0.49, 0.18]	0.378	6			

Parameter estimates were calculated using a random effects model and heterogeneity *Q* tests for between-category differences were calculated using a mixed-effects model. Significant negative Hedges' *g* values indicate the level of overall functioning for ASD-Sibs was lower than that of the specified comparison group. Bolded headers were significant at the $p \leq 0.05$ level for heterogeneity (i.e., between-category differences); bolded comparison groups were statistically significant at the $p \leq 0.05$ level

g estimated aggregated unbiased standardized mean difference (Hedges' *g*), *SE* standard error, *95% CI* confidence interval, *p* level of statistical significance for the associated Hedges' *g* and heterogeneity *Q* test, *k* number of samples in each area of functioning, *K* the total number of samples included in analyses, *Q* the *Q*-value of the heterogeneity test for between-category differences

[a]Intellectual/developmental disabilities except ASD and Down syndrome (i.e., non-specific intellectual/developmental disabilities, developmental language disorder, learning disability, intellectual disability, Prader–Willi syndrome, traumatic brain injury)

[b]Non-specific physical illness, cancer, chronic illness, deafness, and diabetes

[c]Behavioral problems, life-limiting conditions, learning disability, speech impairment, or physical health impairment

significantly worse compared to comparison groups consisting of siblings ($g = -0.30$, $p < 0.001$); there were no significant differences between ASD-Sibs and comparison groups not consisting exclusively of siblings ($g = -0.06$, $p = 0.611$). Furthermore, ASD-Sibs overall functioning was significantly lower than that of siblings of individuals with Down Syndrome (DS-Sibs; $g = -0.40$, $p < 0.001$), siblings of individuals with other intellectual and developmental disabilities (IDD-Sibs; $g = -0.31$, $p = 0.011$), and siblings of individuals with typical development (TD-Sibs; $g = -0.31$, $p < 0.001$); there were no significant differences in magnitude between these comparison groups ($Q[2] = 0.50$, $K = 62$, $p = 0.777$). There were no differences in overall functioning between ASD-Sibs and the following comparison groups: siblings of individuals with physical illness or disability (PID-Sibs), siblings of individuals with other disabilities (i.e., behavioral problems, life-limiting conditions, learning disability, speech impairment, or physical health impairment), normed comparison samples, and individuals with physical illness or typical development. ASD-Sibs overall functioning was not significantly higher than that of any comparison group.

Self-report of functioning ($g = -0.22$, 95% CI [−0.35, −0.10], $p = 0.000$, $k = 41$), parent-report of functioning ($g = -0.25$, 95% CI [−0.39, −0.11], $p = 0.001$, $k = 39$), and medical records related to functioning ($g = -0.57$, 95% CI [−0.70, −0.43], $p = 0.000$, $k = 3$) were associated with significant differences in overall functioning between ASD-Sibs and comparison groups. Conversely, teacher-report of functioning ($g = -0.05$, 95% CI [−0.56, 0.46], $p = 0.851$, $k = 5$) was not associated with significant differences in overall functioning between ASD-Sibs and comparison groups.

There were no significant differences in report characteristic variables (i.e., report publication status); see data censoring section for additional information. In terms of methodological characteristics, there were no significant differences in overall functioning between subgroups for study design or measurement type. Differences in overall functioning were significantly greater ($Q[1] = 4.73$, $p = 0.030$) among samples with an included comparison group ($g = -0.32$, 95% CI [−0.43, −0.21], $p < 0.001$, $k = 55$) than samples for which we added a comparison group ($g = -0.06$, 95% CI [−0.26, 0.15], $p = 0.569$, $k = 14$). In terms of participant characteristics, age, gender, and ethnicity and race did not moderate the difference in overall functioning between ASD-Sibs and comparison groups. Although ASD-Sibs were lower in overall functioning than comparison groups in both samples from the United States and samples from other countries, the magnitude among samples from other countries ($g = -0.38$, 95% CI [−0.54, −0.22], $p < 0.001$, $k = 30$) was significantly larger ($Q[1] = 3.83$, $p = 0.050$) than samples from the United States ($g = -0.17$, 95% CI [−0.31, −0.03], $p = 0.018$, $k = 38$).

See Supplemental Table 2 for complete moderator results.

Data Censoring

We conducted several tests for publication bias, a form of data censoring in which reports with large significant effect sizes are more likely to be published compared to reports with small non-significant effect sizes, increasing the risk of an overestimation of the true effect sizes (Lipsey and Wilson 2001). Because there is no single definitive test for publication bias, conducting multiple analyses of potentially missing samples allows for a cumulative assessment of the possible impact of publication bias. We conducted the following tests for publication bias: (a) report publication status moderator analysis, (b) inverted funnel plot of standard error by the estimated aggregated unbiased standardized mean difference (Hedges' g; Fig. 3), (c) fixed-effect model *trim and fill* test (Duval and Tweedie 2000), (d) inverse rank order correlation (Kendall's τ–b adjusted for ties; Begg and Mazumdar 1994), and (e) Egger test for publication bias (Egger et al. 1997). The results of each of the five publication bias tests suggested the absence of significant publication bias.

Discussion

The current study used meta-analytic methods to determine if ASD-Sibs differ from other populations in areas of social, emotional, behavioral, and psychological functioning. We examined 69 reports comprised of 69 samples of ASD-Sibs with over 800 individual comparisons. The overall results across all types of functioning show that ASD-Sibs have significantly worse outcomes than comparison groups, albeit small in magnitude. Individually, 20 of the included ASD-Sib samples had significantly worse outcomes than the comparison group; two had significantly better outcomes. The remaining comparisons were not individually significant. The confidence intervals around the comparisons varied, a finding that supports the common claim in ASD-Sib literature that "results are mixed" (e.g., Orsmond and Seltzer 2009; Tomeny et al. 2016). However, the number of aggregated results from included samples and comparisons allowed us to examine several different areas of functioning, as well as run moderation analyses by comparison group, reporter, and other key sample characteristics, all of which yielded a more comprehensive picture of the experiences of ASD-Sibs.

Areas of Functioning

Specific analyses of the different areas of functioning showed that ASD-Sibs had significantly worse internalizing behavior problems, more negative beliefs, worse psychological functioning, more negative sibling relationships, and

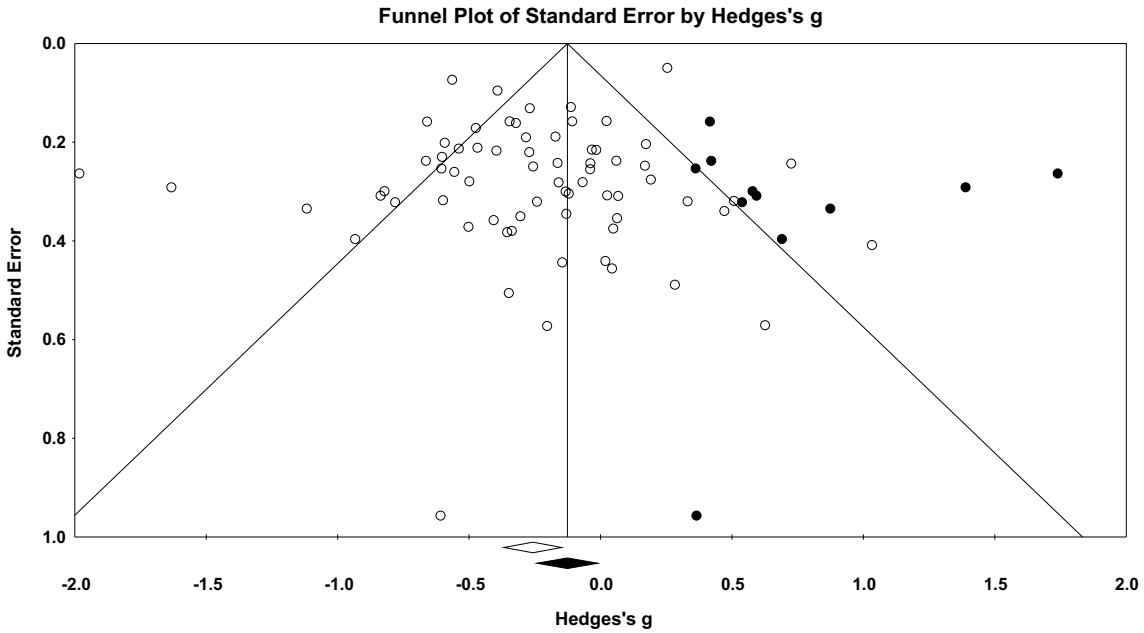


Fig. 3 Inverted funnel plot of standard error by represents the estimated aggregated unbiased standardized mean difference (Hedges' *g*) in overall functioning between ASD-SIBS and comparison groups. Each unfilled circle ($K=69$) represents an included sample ($k=69$), with the Hedges' *g* on the *X* axis and the standard error on the *Y* axis (with the direction reversed such that smaller error values are at the top and larger error values are at the bottom). Each filled circle ($k=11$) represents and imputed study from the *trim and fill* test for publication bias (Duval and Tweedie 2000). The unfilled diamond represents the estimated aggregated unbiased standardized mean difference in overall functioning between ASD-SIBS and comparison groups ($g=-0.26$, SE = 0.06, 95% CI [−0.37, −0.15], $p<0.001$, $K=69$). The filled diamond represents the adjusted Hedges' *g* based on the results of the *trim and fill* test for publication bias ($g=-0.13$, SE = 0.06, 95% CI [−0.24, −0.01], $p=0.033$, $K=80$); the vertical line is the adjusted Hedges' *g* point estimate for determining sample distribution symmetry. The apex of each diamond represents the point estimate and the width of each diamond represents the 95% confidence interval

greater impairment in social functioning. Each of these areas of functioning was tested in over 20 samples of ASD-Sibs; however, there were no differences between these types of functioning—that is, for example, ASD-Sibs were no worse off in terms of internalizing behavior problems than they were in terms of the sibling relationship. In contrast, there were no significant differences between ASD-Sibs and comparison groups in adjustment, coping, family functioning, attention/hyperactivity, externalizing, or total behavior problems. Although externalizing and total behavior problems were both represented in over 20 samples, adjustment and attention/hyperactivity were tested in 12 samples, and coping and family functioning were represented in fewer than 5 samples. The comparatively small number of samples, as well as the fairly broad definition of *adjustment*, may have contributed to the lack of significant findings for these areas of functioning.

The largest overall mean difference (though not significantly larger than other significant comparisons) was found for the sibling relationship. ASD-Sibs had significantly poorer relationships with their brother or sister with ASD than siblings of individuals without ASD. This result echoes the meta-analysis results of Rossiter and Sharpe (2001), who found impaired sibling relationships for siblings of individuals with any IDD, as compared to TD-Sibs. However, the consistency and magnitude of the current finding seems to support the hypothesis that the deficits in social communication found in individuals with ASD create challenges to the relationship between those individuals and their typically developing siblings (e.g., Rossiter and Sharpe 2001), especially considering that the comparison groups included siblings of individuals with other IDDs. Due to the typical lifelong nature of sibling relationships (e.g., Cicirelli 1995), as well as the potential protective nature of healthy sibling relationships against other negative developmental outcomes (e.g., Gass et al. 2007), the finding that ASD-Sibs are significantly more likely to have worse relationships with their brother or sister than comparison groups illustrates an important area for targeted strategies and interventions.

Additionally, ASD-Sibs were found to have significantly worse overall social functioning than comparison groups, a finding which seems to support the hypothesis that ASD-Sibs may be negatively impacted by either sub-clinical levels of ASD-like socio-communicative impairment or by the lack of social practice that children characteristically engage in with their siblings (e.g., Pilowsky et al. 2004). Although not

every first-degree relative of an individual with ASD qualifies for a classification under the broader autism phenotype (BAP), more research is needed to determine how behaviors related to the BAP may influence ASD-Sibs' social functioning. It is possible that family-wide BAP (i.e., broader autism characteristics in the parents or other family members) may also impede ASD-Sibs' development of social functioning. If this is the case, then factors such as total number of TD siblings and age difference among siblings may also play a role in how ASD-Sibs are able to learn social skills from their family members, and such factors deserve further consideration.

The finding that ASD-Sibs have significantly more negative beliefs than comparison groups is also worthy of further research. As currently conceptualized, *beliefs* covered self-concept, beliefs about disability, and overall worldview (e.g., general optimism), though the largest subset of measures in the category covered various aspects of self-concept, and ASD-Sibs did not report significantly more negative self-concept than comparison groups. In contrast, ASD-Sibs' beliefs about disability were significantly more negative than those of comparison groups. There were only four studies that covered beliefs about disability, but the magnitude of the aggregate results suggest that ASD-Sibs are more likely than comparison groups to have beliefs such as, "people with disabilities are aggressive, incomprehensible, or a limit to their families" (De Caroli and Sagone 2013, p. 1220). Previously anecdotal evidence has indicated that siblings of individuals with IDD are more accepting of differences and disability (e.g., Petalas et al. 2009), though few researchers have hypothesized about how the unique characteristics of ASD may affect ASD-Sibs' beliefs about disability in general.

Finally, due to the overlap in definitions, behavioral problems and psychological functioning were analyzed in multiple ways. When behavioral problem measures and psychological functioning measures were aggregated, ASD-Sibs were found to have more symptoms and behaviors related to anxiety/depression and ADHD than comparison groups; conversely, there were no differences in symptoms related to externalizing behavior. However, when separated out by clinical psychological measures and behavioral measures, only clinical psychological measures showed differences between ASD-Sibs and comparison groups, suggesting that ASD-Sibs are more likely to exhibit clinical symptoms of ADHD, anxiety/depression, and externalizing behavior than siblings of individuals without ASD. When further separated by clinical measure, ASD-Sibs had significantly higher likelihood of both anxiety and depression.

This difference in findings based on measurement type may be related to the range of responses possible. Whereas measures like the CBCL and SDQ use continuous scales, many of the measures of psychological functioning were dichotomously based on clinical diagnosis (i.e., participants either had a clinical diagnosis or they did not). Such reduced measurement variance could have disguised actual variability in symptom severity between ASD-Sibs and comparison groups, an interpretation supported by the fact that all individual categories of psychiatric symptoms showed significant differences when indicated by clinical measures, but none showed significant differences as measured by behavioral scales. However, both anxiety and depression (as measured by clinical diagnosis) and internalizing problems (as measured by behavior problems scales) were significantly worse for ASD-Sibs than for comparison groups, echoing large-sample findings from the literature (e.g., Jokiranta-Olkoniemi et al. 2016; Shivers et al. 2013). The robust findings for internalizing behaviors, as well as the lack of significant findings for other types of behavior problems, suggests that ASD-Sibs are not *acting out* or *poorly behaved*, but rather, potentially more anxious and depressed than comparison groups.

Moderator Analyses

Analyses determined that ASD-Sibs had significantly worse functioning than siblings of individuals with IDD other than ASD (IDD-Sibs) and Down syndrome (DS-Sibs), as well as siblings of individuals with no disabilities (TD-Sibs). Although there were no differences between ASD-Sibs and any other type of comparison group (i.e., comparison groups not limited to siblings; siblings of individuals with physical illness, physical disability, or other impairment), each of these comparison group categories were limited to six or fewer studies, potentially limiting the power to detect significant differences. The current findings support the so-called *Down syndrome advantage* (Hodapp et al. 2001), in that ASD-Sibs were more likely to have negative functioning than DS-Sibs. However, the significant differences between ASD-Sibs and both IDD-Sibs and TD-Sibs may indicate that systematic processes in families of individuals with ASD facilitate different outcomes for the typically developing siblings in the family. This reflects the consistent findings among parents and other caregivers of individuals with ASD who report significantly more stress overall than other parents, including those of children with other IDDs (Hayes and Watson 2013). The challenges unique to ASD-Sibs, as compared to siblings of individuals without ASD, deserve further consideration to determine what family- and individual-level factors may be driving these negative outcomes, as well as which characteristics can act as protective factors.

Many researchers have speculated on the relative validity of parent-report vs. self-report of sibling outcomes (e.g., Guite et al. 2004). Single-sample studies have shown generally low levels of agreement between parent-report and self-report on the same constructs (see Rossiter and Sharpe

2001 for review). Our analyses, however, found statistically identical results by parent-report and self-report. This result highlights the importance of meta-analysis; whereas individual parent–child dyads may differ in their respective perceptions of sibling outcomes, overall, both ASD-Sibs and parents seem to identify the same level of negative sibling outcomes. Although care should still be taken when deciding on measure and reporter, including considerations of age and variable, the current results suggest that both parents and ASD-Sibs are valid sources of information for sibling outcomes.

The lack of difference between published and unpublished reports is worth noting. Publication bias in meta-analysis—the risk of inflated parameter estimates by only including data from published reports which tend to have larger effects than unpublished reports—increases both sampling error and result bias, as well as decreases both the accuracy and generalizability of study findings (Lipsey and Wilson 2001). The current study, despite including comparable numbers of published ($k = 39$) and unpublished ($k = 30$) reports, found no difference in aggregate effect size. The majority of unpublished studies were theses or dissertations; we only found one dissertation that was later published. The lack of transition from dissertations to published work has many potential causes. First, although exact rates are difficult to calculate, many dissertations are never published. Second, multiple dissertations included bespoke measures–measures that were created specifically for the given study-that were not validated or previously used in research (e.g., Janecek 2015). If the researchers were unable to provide sufficient psychometric data for reviewers, the study may not be accepted for publication. Finally, although the number of ASD-Sib studies has risen over the past decade, the publication of sibling-focused research still lags behind the amount of studies focusing on the individual with ASD or their caregivers. We hope that the current study will provide a solid, cumulative foundation on which future sibling researchers can base further analyses, contributing to the collective knowledge of ASD-Sib experiences.

Two moderators were found to be significant: sample comparison group (added vs. included) and nationality (United States vs. other countries). A total of 14 studies in the current analyses did not include a comparison group, so separate studies with samples of roughly the same size and age range were used. However, due to the idiosyncratic nature of reporting in several of the studies (e.g., separating results by gender, Solarsh 2016; reporting of total scores vs. subscales; Stampoltzis et al. 2014), many of the selected comparison studies used more targeted populations or situations (e.g., children with rheumatoid arthritis, Meltzer 1987; examining outcomes for children in the face of parental conflict; Amaya-Hodges 2012). Studies designed to examine such populations of concern may recruit samples of individuals who function differently than individuals recruited to serve as a comparison sample. The finding that samples from the United States yielded lower overall differences than samples from other studies is perhaps less explicable. There were not enough studies from any single country outside the United States to analyze separate comparisons, so more research is needed to determine the cultural factors that may impact ASD-Sib outcomes around the world.

Methodological Issues

As with all meta-analyses, the present study is limited by the quality of the studies included. Of particular salience to the current analyses are (a) the variety of measures used, (b) the interpretation of said measures, (c) inconsistent statistical reporting, and (d) inconsistent demographic descriptions.

First, the included studies used a wide variety of measures. Although certain measures, such as the CBCL and the SRQ were used in multiple studies, there were also multiple measures that were used for only a single study, both existing validated measures (e.g., Beck Depression Inventory; used by Martins 2007) and bespoke measures created for the given study (e.g., Bemister 2012). Such lack of replication, though unfortunately common in behavior science, nonetheless introduces additional measurement error to the analyses. Second, although we understand researchers' interest in a broad spectrum of potential outcomes for ASD-Sibs, the lack of consistent conceptualization of outcomes can be confusing for other researchers, as well as clinicians and families. For instance, whereas some researchers described results from the CBCL as *behavioral problems* (e.g., Gau et al. 2010), other researchers described them as *adjustment* (e.g., Barak-Levy et al. 2010). When trying to understand the entire ASD-Sib experience, it is important to report and interpret results in a way that allows for future integration of said results with additional studies. We attempted to create a template for classifying and describing sibling outcomes based on the measures used (see Table 1), but we acknowledge that several outcomes do not fit neatly into the categories we identified. Still, using a consistent identifiable system for reporting and interpreting ASD-Sib outcomes can help further cohere this growing and important field.

The third limitation of the current study is that posed by inconsistent statistical reporting. We noticed a pattern of studies missing standard effect size data (e.g., means presented without associated standard deviations; p values presented as levels of significance, such as $p > 0.05$ or $p \leq 0.05$, without the exact p value; one-way ANOVAs instead of a t test) that presented challenges for calculating, or in some instances imputing, the effect sizes necessary for sample aggregation. When examining group differences, reporting

means, standard deviations, and exact *p* values is important for transparency and interpretation of results.

Finally, the moderation and meta-regression analyses were limited by the inconsistent reporting of demographics. Many studies did not report any breakdown of age, gender, or ethnicity, limiting the number of variables that could be included in the meta-regression models. Previous studies have shown significant differences in ASD-Sibs functioning by age and gender (e.g., Hodapp et al. 2010; Orsmond and Seltzer 2000), suggesting that the overall impacts of demographics deserve further consideration. Although the meta-regression still contained a fair number of studies, the analysis of gender influences would be better served by reporting separate means for males and females. Additionally, many ASD-Sib studies include samples covering a wide age range (e.g., Demspey et al. 2012; Surfas 2005); thus, the mean age may not be an accurate reflection of the age of the participants. Notwithstanding the challenges of recruiting ASD-Sib samples, we encourage researchers to separately examine various developmental stages. In regards to ethnicity, the regression analyses used percentage of the sample that was People of Color as the predictor variable. This is obviously an incomplete calculation of racial/ethnic variability. Thus, the lack of findings should be interpreted with caution.

In addition to demographics, several other characteristics of the individual with ASD, including method of diagnosis (e.g., self-report, clinical diagnosis by the research team, etc.), severity of ASD, gender, age, and presence of behavior problems were unable to be included in moderator or meta-regression analyses due to underreporting across included samples. Information about the brother or sister with ASD, however, is considerably easy to collect. Even if direct analyses on gender combination (e.g., same gender or different gender sibling dyads) and age difference between siblings are not reported, including age and gender of the individual with ASD in the report would enhance interpretability and future meta-analytic reviews.

Future Directions

The current findings have broad-ranging implications for research, practice, and policy. As the first quantitative meta-analysis examining social, behavioral, emotional, and psychological outcomes among ASD-Sibs, the present study provides an important aggregation of 69 individual studies of such outcomes. Because of the variety of measures used, sample sizes, and findings reported by these individual studies, the current, comprehensive results provide important information for ASD-Sibs, parents, and clinicians. Although there are considerable individual differences among ASD-Sibs, the current findings suggest that, overall, ASD-Sibs have significantly more negative outcomes than comparison groups, including siblings of individuals with other intellectual and developmental disabilities. This finding that ASD-Sibs seemingly face challenges greater than typically developing individuals in families without ASD, similar to findings of parents and caregivers of individuals with ASD (e.g., Hayes and Watson 2013), suggests that the unique difficulties faced by families of individuals with ASD do, in fact, extend to individual outcomes among siblings. Therefore, policymakers and clinicians should make a concerted effort to expand support efforts to include all members of the family, including typically developing siblings.

For researchers, the current study has multiple implications. First, we provide aggregate evidence for the negative outcomes faced by ASD-Sibs, including a breakdown of areas of functioning. We certainly do not discourage replication or further exploration of group differences, but, using the current results as a foundation, future studies can include additional analyses to better understand the numerous factors that contribute to individual differences in outcomes. Many studies have already done such analyses, with particular focus on behavior problems of the individual with ASD (e.g., Hastings 2007; Shivers et al. 2013). In addition to studying family factors, however, more research should be devoted to understanding the impact of individual-level factors, such as support networks and coping strategies, to provide support for future interventions targeting multiple factors. Kovshoff et al. (2017) provide a framework for such future research that combines elements of various existing developmental and family theories into the *Siblings Embedded Systems Framework*. Many of the findings of the current study can be incorporated into this framework, including sibling beliefs as a part of the ASD-Sibs' "personal interpretation of events," and social functioning as a measure of peer relationships (Kovshoff et al. 2017, p. 39). We encourage future sibling researchers to continue to build on the empirical and theoretical work cited here. Given the development of family-wide supports is the ultimate goal of many ASD-Sib researchers, by understanding not only the outcomes unique to ASD-Sibs, but the numerous factors that impact such outcomes, the research community can work with families and service providers to create effective strategies that improve outcomes for siblings and families most at risk.

Importantly, however, when reporting results of future studies of ASD-Sibs, we call for an improvement in the inclusion of demographic information and relevant statistical output. When reporting group differences, researchers should include both the mean and standard deviation of continuous variables. When reporting sample characteristics, breakdown of sibling age, gender, and ethnicity, as well as brother or sister with ASD age, gender, and ethnicity should be included. To better understand the effects of age and gender, future studies can (a) attempt to recruit a more developmentally focused sample (e.g., young children, adolescents), and (b) report separate results for males and

females. Additionally, more focus needs to be given to adult siblings to better understand the experiences of ASD-Sibs in adulthood and later life. Finally, we strongly encourage researchers, research organizations, and funding agencies, to support longitudinal studies of sibling and family experiences among families of individuals with ASD. A common rationale for ASD-Sib research is the importance of the sibling relationship for individuals with IDD in adulthood (e.g., Tomeny et al. 2016); therefore, longitudinal analyses are necessary to better understand how ASD-Sib experiences in childhood and adolescence translate over time.

Finally, it is essential to consider clinical implications of the current study. The results show a small but significant difference between ASD-Sibs and comparison groups, suggesting that ASD-Sibs may, on average, need more support than other siblings, particularly in terms of the sibling relationship, social functioning, and psychological functioning. However, there is still considerable variability among sibling outcomes. Therefore, parents, teachers, and clinicians should look into the specific circumstances of siblings' lives to determine what kinds of supports, if any, are most appropriate across the lifespan.

Summary

The current study is the first quantitative aggregation of comparisons between ASD-Sibs and other populations, covering social, emotional, behavioral, and psychological functioning. Overall results show that ASD-Sibs are more likely to exhibit impaired functioning than comparison groups, particularly in terms of internalizing behavior problems, psychological functioning, social functioning, beliefs, and the sibling relationship. These results expand on the findings from Rossiter and Sharpe (2001), which showed significantly negative outcomes for siblings of individuals with IDD. Due to the large number of individuals with ASD (CDC 2018), the focused study of ASD-Sibs, rather than siblings of individuals with any IDD, is of increasing importance. The current review and quantitative aggregation of comparative ASD-Sib research have important implications for both further research and the development of targeted supports and strategies to support healthy outcomes for ASD-Sibs and their families.

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Compliance with Ethical Standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical Approval This article does not contain any studies with human participants or animals performed by any of the authors.

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

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