



Empathy and Prosocial Behavior in Siblings of Children with Autism Spectrum Disorder: a Systematic Review

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Abstract

Siblings of children with ASD have, as a group, more autistic traits than typically developing individuals. A high prevalence of autistic traits may (or may not) include lower scores on measures of empathy and prosocial behavior compared with typically developing individuals. The purpose of this study was to conduct a systematic review of studies of empathy and prosocial behavior in siblings of individuals with ASD. We conducted a comprehensive literature search of studies published until March 2020. We identified 16 studies with a total of 1066 siblings that met inclusion criteria. Based on the review, we conclude that the current literature suggests that siblings of individuals with ASD exhibit normative empathic abilities and prosocial behavior.

Keywords Empathy · Prosocial behavior · Autism spectrum disorders · Siblings · Broader autism phenotype

Autism spectrum disorder (ASD) affects around 1.5% of children (Lyll et al., 2017). The main characteristics of ASD are impairments in social communication and interaction, and restrictive and repetitive behavior and interests (American Psychiatric Association, 2013). These characteristics are thought to be related to atypical development in domains like social cognition (Tager-Flusberg, 2007), executive functions (Hill, 2004), and temporal cognition (Boucher et al., 2007). In addition to atypical social and cognitive development, children with ASD often have concurrent mental health and/or behavioral problems, including increased risk of depression, anxiety, oppositional behavior, hyperactivity, and sleep problems (Carmassi et al., 2019; Pezzimenti et al., 2019; Simonoff et al., 2008).

According to family systems theory, all family members influence each other reciprocally (Dore, 2008). Around 80% of children have one or more siblings (Dirks et al.,

2015), and the sibling relationship is one of life's longest lasting relations. Typically developing (TD) children spend a lot of time with siblings, possibly even more than with parents (Buist et al., 2013; Dirks et al., 2015). Hence, the sibling relationship represents an important component of children's social, emotional, and cognitive development (Buist et al., 2013; Dirks et al., 2015). The combination of atypical development and concurrent mental health and behavioral problems that may accompany ASD can pose risks for siblings (Shivers et al., 2019). A meta-analysis of psychosocial functioning among siblings of children with ASD found that these siblings had significantly lower psychosocial functioning than siblings of children with other developmental disorders and TD controls, and were at higher risk of depression and anxiety (Shivers et al., 2019).

Factors that may influence the psychosocial functioning of siblings of children with ASD include differential treatment/attention from parents, behavioral problems displayed by the child with ASD (e.g., aggression toward siblings), and negative reactions from others (Haukeland et al., 2015; Tudor et al., 2018). Whereas sibling relationships typically include much social interaction, pretend play, competitiveness, warmth, and intimacy, this is not always the case when one sibling has ASD. For example, Kaminsky and Dewey (2001) found that relationships between children with ASD and their siblings were characterized by lower intimacy and warmth compared with other sibling relationships.

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For siblings of children with ASD, the risk of psychosocial difficulties is thought to be twofold (e.g., Orsmond & Seltzer, 2009). First, ASD is highly heritable, with heritability mostly due to common genetic variants (Gaugler et al., 2014; Tick et al., 2016). Thus, siblings of children with ASD, who on average share 50% of genes with the child with ASD, are at increased genetic risk of ASD or autistic traits. Many siblings of children with ASD have elevated levels of autistic traits without meeting the diagnostic criteria; this is often called the broader autism phenotype (Charman et al., 2017; Ingersoll & Wainer, 2014). The broader autism phenotype includes higher levels of autistic-type social difficulties and executive dysfunction among relatives of individuals with ASD, including siblings (see Ingersoll & Wainer, 2014 for review).

Much sibling research has focused on mental health, but less research has examined how growing up with a brother or sister with ASD affects siblings' social development. Older siblings often function as role models for younger siblings (Dirks et al., 2015). An important part of children's social and emotional development is the development of empathy and prosocial behavior. Empathy can be seen as a multidimensional construct comprising at least two dimensions; affective empathy and cognitive empathy. Affective empathy is defined as the emotional response triggered by seeing another person in emotional distress (Baron-Cohen & Wheelwright, 2004). In contrast, cognitive empathy is the cognitive understanding of other people's emotions, which is related to and partially overlapping with theory of mind (Baron-Cohen & Wheelwright, 2004). Prosocial behavior is defined as voluntary behavior aimed at benefiting or helping others (Eisenberg et al., 2007). Empathy, both the affective response and cognitive understanding, is closely related to prosocial behavior as an emotion that triggers prosocial behaviors (Eisenberg et al., 2007; Telle & Pfister, 2016; Zhao et al., 2019).

When an additional child is born into a family, parents have the opportunity to teach the older sibling about caretaking, sharing, and helping. Likewise, the younger child's empathy and prosocial development can be supported by scaffolding, modeling, and norm setting provided by the older sibling (Hughes et al., 2018). Parental efforts to teach their children empathy and prosocial behavior often include encouragement to display empathy and prosocial behavior towards siblings. Typically, this creates more teaching and learning opportunities for children with siblings compared with children without siblings (Hughes et al., 2018). However, several studies have demonstrated that the relationships between children with ASD and their TD siblings are characterized by less prosocial behavior compared with other sibling relationships (Kaminsky & Dewey, 2001; Knott et al., 1995, 2007).

Individuals with ASD display lower scores on conventional measures of empathy and prosocial behavior (Harmsen, 2019; Russell et al., 2012; Song et al., 2019). Some studies

have found that individuals with ASD primarily display difficulties with cognitive empathy, whereas affective empathy appears to be comparable with that of TD individuals (e.g., Deschamps et al., 2014; Rogers et al., 2007). Lower scores on cognitive empathy could be due to differences in how individuals with ASD experience and display empathy (e.g., Fletcher-Watson & Bird, 2020). In addition, it seems that typically developing adults have difficulties with understanding the emotions displayed by individuals with ASD (Sheppard et al., 2016), suggesting a two-way, double-empathy problem (Milton, 2012). Notwithstanding these challenges sometimes experienced by social partners, evidence suggests that, in general, ASD is linked to difficulties with at least some domains of empathy and prosocial behavior (Harmsen, 2019; Song et al., 2019). In a general population sample, Zhao et al. (2019) found that empathy partially mediated the relationship between autistic traits and prosocial behavior. That is, more autistic traits were related to less empathy, which again was related to less prosocial behavior. Thus, the empathic and prosocial development of siblings of children with ASD may be affected in two ways: (1) through increased genetic risk of autistic traits with associated empathic and prosocial difficulties and (2) through more limited opportunities to learn and practice empathy and prosocial behavior in the sibling relationship with the child with ASD.

Alternatively, other research has shown that the experience of being a sibling of a brother or sister with a chronic disorder other than ASD can enhance empathy and prosocial behavior. For example, two studies compared siblings of children with physical disabilities (PD) to controls and found that siblings of children with PD scored significantly higher on measures of empathy and prosocial behavior (Perenc et al., 2015; Perenc & Pęczkowski, 2018). These findings contrast with the potential negative impacts such experiences can have on mental health and remind us that the assumption that siblings automatically will experience negative outcomes sometimes is wrong. Thus, there are at least two reasons why the topic of empathy and prosocial behavior deserves further study among siblings of children with ASD. First, such knowledge can provide important information about risk and protective factors in siblings, which can be targeted in interventions. Second, such knowledge can inform the field about how genetic risk of ASD and environmental risk of growing up with a child with ASD affects social developmental outcomes.

Three hypotheses can be presented regarding empathy and prosocial behavior in siblings of children with ASD. The first is that these siblings have impaired empathy and low levels of prosocial behavior due to shared genetic risk and/or the experience of living together with a child with ASD. The second is that these siblings display enhanced empathy and prosocial behavior due to the experience of living with a brother or sister with special needs. A third option is that the null hypothesis holds true, and these siblings display the same levels of

empathy and prosocial behavior as other children. The field has yet to establish which hypothesis is most supported. Therefore, our goal was to conduct a systematic literature review of studies of empathy and/or prosocial behavior in siblings of children with ASD inspired by the following research question: *Do siblings of children with ASD display lower, comparable, or higher levels of empathy and prosocial behavior compared to children who do not have siblings with ASD?* We chose typically developing children without a sibling with ASD as controls because this is the most natural comparison group when examining developmental outcomes in siblings of children with ASD. Further, we focus only on siblings aged 0 to 18 years, since adult sibling relationships are considerably different from childhood sibling relationships (e.g., Tomeny et al., 2017).

Method

We followed the Preferred Reporting Items for Systematic reviews and Meta-Analyses (the PRISMA statement), as far as the guidelines apply to a systematic review of cross-sectional studies (Moher et al., 2009).

Data Sources and Search Strategy

We consulted the following databases: PsycINFO, PubMed, and Web of Science. The search was conducted in March 2020 (ending 23rd March), using the following search string: *sibli* AND (autis* OR Asperger*) AND (prosocial* OR empath*)*. There were no limits on publication year. The initial search revealed 110 records. Fifty-one remained after limiting to English, peer-reviewed articles and removing duplicates (see Fig. 1). We also performed a comprehensive manual search by consulting previous reviews (Meadan et al., 2010; Shivers et al., 2019), reference lists of included papers, and papers known to the authors. Through the manual search, we identified six additional relevant papers.

Procedure and Study Selection

Inclusion and Exclusion Criteria

To be included, articles had to (a) be published in a peer-reviewed journal, (b) report original findings on prosocial behavior and/or empathy (i.e., include a specific measure of at least one of these constructs) in siblings (age ≤ 18 years) of individuals with ASD, and (c) include a comparison condition (i.e., a control group or comparison with established norms). Exclusionary criteria included articles published in languages other than English and qualitative studies.

Procedure

Two reviewers independently screened titles and abstracts of all 51 articles from the electronic search. After this screening, we then screened the full text of 19 articles to assess eligibility, of which ten were included in the final review (see Fig. 1). The inter-rater agreement for inclusion was very high (Cohen's kappa = .84). Disagreements were resolved through discussion. The six studies identified through the manual search were assessed for eligibility by one author and confirmed by another. Following the manual search, a grand total of 16 studies met the inclusion criteria and were included in the full review. Seven of the studies examined empathy and 10 of the studies examined prosocial behavior (i.e., one study examined both empathy and prosocial behavior).

Data Extraction and Quality Assessment

The following data were extracted from the included studies: authors, publication year, country of origin, study design, participant characteristics (age and gender), recruitment method, type of comparison condition, measurements, and main results including effect sizes and *p*-values. If the article did not report effect size(s), we calculated Cohens *d* from data provided in the article using the following formula $(M_{\text{Group1}} - M_{\text{Group2}}) / SD_{\text{pooled}}$ (Cohen, 1992).

We conducted quality assessments using the Newcastle-Ottawa Scale (NOS) for case-control studies (Wells et al., 2019). This scale includes eight items that assess study quality within the domains of representativeness and sample selection, comparability of cases and controls, assessment procedures, and response rates. The maximum possible score is nine and the lowest is zero. The NOS case-control scale has shown fair to good interrater reliability, excellent test-retest reliability, and weak to moderate concurrent validity in previous studies (Moskalewicz & Oremus, 2020; Oremus et al., 2012). In our study, two reviewers independently rated quality. Agreement between raters was excellent, intra-class correlation (ICC; two-way random) = .88 (Cicchetti, 1994). Disagreements were resolved through discussion.

Results

See Tables 1 and 2 for an overview of the reviewed studies.

Descriptives

Participants

Data from a total of 1066 siblings of children with ASD were included across the studies. Sibling age ranged from 1 to 18 years. Samples ranged from 37.3% to 64.0% male.

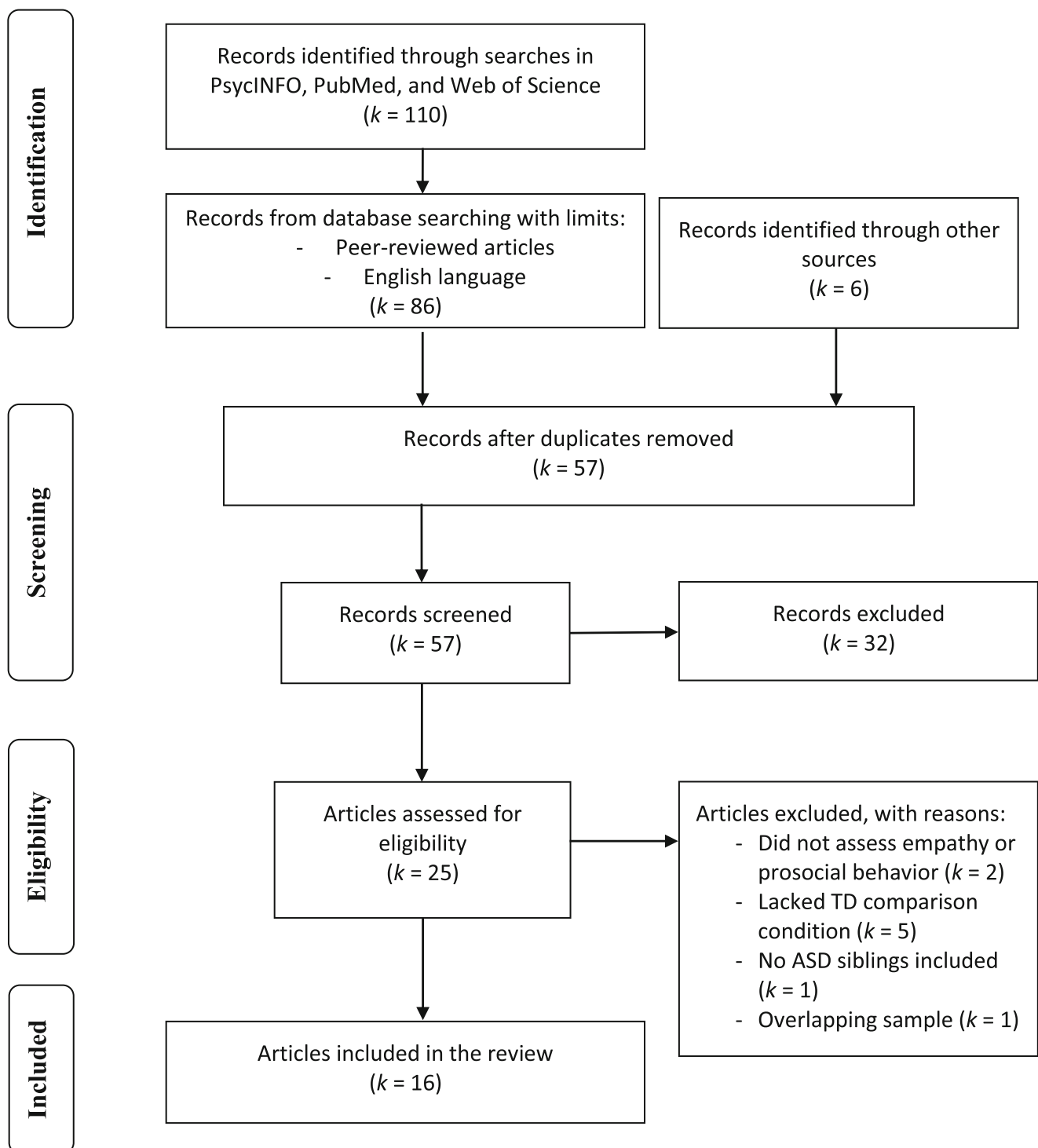


Fig. 1 Adapted PRISMA flow chart of the study selection process (Moher et al., 2009)

Measures and Informants

With two exceptions (McDonald et al., 2017; Tsang et al., 2016), the reviewed studies measured either prosocial behavior or empathy via only a single measure of the outcome of interest. McDonald et al. (2017) and Tsang et al. (2016) each

used both a behavioral measure and parent-report. The measures in McDonald et al. (2017) included assessment of both empathy and prosocial behavior.

We divided measure types into three main categories: (1) behavioral observation paradigms in which children witnessed an adult or child in distress and observers coded

Table 1 A summary of studies on empathy in siblings of children with ASD

Author(s) and publication year	Country of origin	Study design and comparison condition	Participants	Recruitment	Measure(s)	Main findings	Effect size for group differences (<i>p</i> -value)	Quality score
Campbell et al. (2015)	USA	Case-control study—comparison with siblings of typically developing children	<i>N</i> = 27 younger siblings of children with ASD (assessed at 22 months), 63% male	From a larger prospective study of children at risk for ASD (autism research facility)	Empathy: Behavioral coding of watching another person's distress	Siblings of children with ASD showed less empathic concern for a crying baby than controls, but there was no significant difference in the empathic concern for an experimenter hurting her finger	Cohens <i>d</i> (crying baby) = $-.92$ ($p = .037$) Cohens <i>d</i> (hurting finger) = $-.60$ (ns.)	4
Campbell et al. (2017)	USA	Longitudinal case-control study—comparison with siblings of typically developing children	<i>N</i> = 59 younger siblings of children with ASD (assessed at ages 22, 28, and 34 months), 55.9% male	Participants recruited for a prospective study of children at risk for ASD (autism research facility)	Empathy: Behavioral coding of watching another person's distress	Siblings of children with ASD showed less empathic concern for an experimenter hurting her finger at 22 months and 28 months compared with controls, but no difference at 34 months	Cohens <i>d</i> (22 months) = $-.58$ ($p < .001$) Cohens <i>d</i> (28 months) = $-.40$ ($p = .007$) Cohens <i>d</i> (34 months) = $.01$ (ns.)	6
Eyuboglu et al. (2018)	Turkey	Case-control study—comparison with typically developing controls	<i>N</i> = 41 siblings of children with ASD (Mean age = 12.2, range 8–18 years), 51% female	University outpatient clinic (Child and Adolescent Psychiatry)	Empathy: Unexpected Outcomes Test	Siblings of children with ASD scored lower on empathy compared with the control group	Cohens <i>d</i> = -1.78 ($p < .001$)	5
Hutman et al. (2010)	USA	Case-control study—comparison with typically developing controls	<i>N</i> = 88 younger siblings of children with ASD (seen at 12, 18, 24, and 30 months of age), 50% male	University autism clinic and organizations offering support to families with a child with autism	Empathy: Behavioral coding of watching another person's distress	No significant differences between siblings of children with ASD and controls at 18, 24, or 30 months of age in response to another person's distress.	NA	7
McDonald et al. (2017)	USA	Case-control study—comparison with siblings of typically developing children	<i>N</i> = 36 younger siblings of children with ASD (Mean age = 62.23 months, <i>SD</i> = 8.58, range 4 to 6 years), 64% male	Recruited as part of a longitudinal study of infants at high risk of ASD	Empathy and prosocial behavior: behavioral coding watching another person hurt themselves and EPRCQ ^a (parent-report)	No significant difference in observed empathy and prosocial behavior between siblings of children with ASD and controls. Siblings of children with ASD were rated higher than controls in empathy and prosocial behavior by parents	$\eta^2 = .16$ ($p = .003$)	4
Shivers (2019)	USA	Case-control study—comparison with siblings of typically developing children	<i>N</i> = 26 siblings of children with ASD (Mean age = 14.94, <i>SD</i> = 1.96, range 12–18), 53.8% female	Recruited through state and local organizations serving people with developmental disabilities and their families	Empathy: Interpersonal Reactivity Index (IRI) (self-report)	No significant difference between siblings of children with ASD and controls in overall empathy, but ASD siblings scored significantly higher on perspective taking compared with controls	Cohens <i>d</i> (empathy) = $.38$ (ns.) Cohens <i>d</i> (perspective taking) = $.55$ ($p = .028$)	4
Tsang et al. (2016)	USA	Case-control study—comparison	<i>N</i> = 24 siblings of children with ASD	Recruited through a university clinic for autism	Empathy: Behavioral coding of watching another person's	No significant difference in observed empathy or parent-reported cognitive and affective empathy between	Cohens <i>d</i> (observed) = $-.08$ (ns.) Cohens <i>d</i> (cognitive) = $.10$ (ns.)	5

Table 1 (continued)

Author(s) and publication year	Country of origin	Study design and comparison condition	Participants	Recruitment	Measure(s)	Main findings	Effect size for group differences (<i>p</i> -value)	Quality score
		with typically developing controls	(Mean age = 5.52, <i>SD</i> = 0.45), 37.5% male		distress and the Griffith Empathy Measure (GME; parent-report)	siblings of children with ASD and controls	Cohens <i>d</i> (affective) = .34 (ns.)	

^a Empathy and prosocial response subscale of the Conscience Questionnaire

their empathic and/or prosocial responses, (2) parent- or self-report questionnaires, and (3) observer-rated tests of empathic reasoning.

Studies ($k = 5$) based on behavioral measures used similar observation situations in which infants and toddlers were exposed to an adult pretending to hurt his/her finger or foot, or a crying baby (Campbell et al., 2015, 2017; Hutman et al., 2010; McDonald et al., 2017; Tsang et al., 2016). These paradigms were used to measure empathy. The studies used masked coders and interclass correlations between raters ranged from acceptable to excellent ($ICC = .73$ to $.95$).

Studies ($k = 12$) based on questionnaires often used the Strengths and Difficulties Questionnaire (SDQ; Goodman, 1997; $k = 10$), which includes a prosocial behavior subscale. The SDQ comprises a parent-report version for children aged 4 to 16 years, and a self-report version for children ≥ 11 years. Two studies utilized self-report (Hastings & Petalas, 2014; Tsai et al., 2016), while the others utilized parent-report. Questionnaires used to measure empathy included the Interpersonal Reactivity Index (self-report; Davis, 1983), Griffith Empathy Measure (parent-report; Dadds et al., 2008), and the Conscience Questionnaire (parent-report; Kochanska et al., 1994). Across studies, three used self-report, while the rest used parent-report.

Only one study was based on a test of empathy, as measured by the Unexpected Outcome Test (Eyuboglu et al., 2018). The Unexpected Outcome Test involves reasoning about the emotional states of others when these emotions are out of context (cognitive empathy). In the test, participants get described a situation where the emotional state of the protagonist is out of place (e.g., angry when going to a movie with a friend) and asked to provide additional information to solve the disparity (Dyck et al., 2001).

Comparison Conditions

Most studies used a control group comprising children who did not have siblings with ASD. Eight studies compared siblings to population-norms from a general child and adolescent population in the given country (i.e., SDQ norm data).

Empathy in Siblings of Children with ASD

Among the seven studies investigating empathy, four observed significantly lower empathy for siblings of children with ASD compared with controls, at least at some ages (Campbell et al., 2015, 2017; Eyuboglu et al., 2018). Three found no significant difference between siblings and controls (Park et al., 2012; Tsang et al., 2016), whereas two studies observed higher empathy in siblings in one or more domain(s) (McDonald et al., 2017; Shivers, 2019). Campbell et al. (2017) conducted a longitudinal study where significant differences between younger siblings of children with ASD and

Table 2 A summary of studies on prosocial behavior in siblings of children with ASD

Author(s) and publication year	Country of origin	Study design and comparison condition	Participants	Recruitment	Measures of prosocial behavior / empathy	Main findings	Effect size for group differences (<i>p</i> -value)	Quality score
Benson and Karlof (2008)	USA	Cross-sectional study—comparison with established norms	<i>N</i> = 72 siblings of children with ASD (Mean age = 7.1, range 3–11), 50% males	Ongoing longitudinal study of children receiving public school services for autism	Prosocial behavior: SDQ ^a (parent-report)	No significant difference in prosocial behavior between siblings of children with ASD and established norms	Cohens <i>d</i> = -.22 (ns.)	4
Chan and Lai (2016)	Hong Kong	Cross-sectional study—comparison with established norms	<i>N</i> = 116 siblings of children with ASD (Mean age = 10.4, range 6–18), 61% female	Recruited through a university-based child and adolescent psychiatric clinic and special schools for children with learning disability.	Prosocial behavior: SDQ (parent-report)	Siblings of children with ASD of both genders scored lower on prosocial behavior than established norms	Cohens <i>d</i> (males) = -.59 (<i>p</i> < .01) Cohens <i>d</i> (females) = -.86 (<i>p</i> < .001)	2
Griffith et al. (2014)	UK	Cross-sectional study—comparison with established norms	<i>N</i> = 168 siblings of children with ASD (Mean age = 10.54, <i>SD</i> = 3.4, range 4–17), 51% male	Data drawn from a larger study of families of children with ASD, recruited through national autism charity	Prosocial behavior: SDQ (parent-report)	Siblings of children with ASD scored lower on prosocial behavior than established norms, based on both mother- and father-report	Cohens <i>d</i> (mother-report) = -.33 (<i>p</i> < .001) Cohens <i>d</i> (father-report) = -.36 (<i>p</i> < .01)	2
Hastings (2003a)	UK	Cross-sectional study—comparison with established norms	<i>N</i> = 22 siblings of children with ASD (Mean age = 12.14, range 6–16), 50% male	Local school for children with autism	Prosocial behavior: SDQ (parent-report)	Siblings of children with ASD scored lower on prosocial behavior than established norms	Cohens <i>d</i> = -.62 (<i>p</i> < .05)	2
Hastings (2003b)	UK	Cross-sectional study—comparison with established norms	<i>N</i> = 78 siblings of children with ASD (Mean age = 6.70, range 4–16), 55% female	From a larger study of parents of children with ASD receiving early intensive behavioral intervention	Prosocial behavior: SDQ (parent-report)	No significant difference in prosocial behavior between siblings of children with ASD and established norms	Cohens <i>d</i> = -.24 (ns.)	2
Hastings and Petalas (2014)	UK	Cross-sectional study—comparison with established norms	<i>N</i> = 60 siblings of children with ASD (Age range: 11–17)	Data came from a larger study of families of a child with ASD	Prosocial behavior: SDQ (self-report)	No significant difference in prosocial behavior between siblings of children with ASD and established norms	Cohens <i>d</i> = -.19 (ns.)	2
McDonald et al. (2017)	USA	Case-control study—comparison with siblings of typically developing children	<i>N</i> = 36 younger siblings of children with ASD (Mean age = 62.23 months, <i>SD</i> = 8.58, range 4 to 6 years), 64% male	Recruited as part of a longitudinal study of infants at high risk of ASD	Empathy and prosocial behavior: behavioral coding watching another person hurting themselves and EPRCQ ^b (parent-report)	No significant difference in observed empathy and prosocial behavior between siblings of children with ASD and controls. Siblings of children with ASD were rated higher than controls in empathy and prosocial behavior by parents	$\eta^2 = .16$ (<i>p</i> = .003)	4
	UK						Cohens <i>d</i> = -.21 (ns.)	2

Table 2 (continued)

Author(s) and publication year	Country of origin	Study design and comparison condition	Participants	Recruitment	Measures of prosocial behavior / empathy	Main findings	Effect size for group differences (p -value)	Quality score
Petalas et al. (2009)		Cross-sectional study—comparison with established norms	$N = 25$ siblings of children with ASD (Mean age = 10.36, $SD = 3.38$), 52% female	Recruited through letters of invitation to mothers of children at special schools	Prosocial behavior: SDQ (parent-report)	No significant difference in prosocial behavior between siblings of children with ASD and established norms		
Tsai et al. (2016)—UK data	UK	Cross-sectional study—comparison with established norms	$N = 75$ siblings of children with ASD (Mean age = 12.7, $SD = 2.4$, range 8–18), 37.3% male	Recruited through schools, clinics, and online advertising	Prosocial behavior: SDQ (parent-report and self-report)	Siblings of children with ASD scored lower on mother-reported prosocial behavior than established norms No significant difference in prosocial behavior between sibling's self-report and established norms	Cohens d (mother-report) = 2 –.32 ($p < .008$) Cohens d (self-report) = –.21 (ns.)	
Tsai et al. (2016)—Taiwan data	Taiwan	Cross-sectional study—comparison with established norms	$N = 80$ siblings of children with ASD (Mean age = 12.7, $SD = 2.8$, range 7–18), 41% male	Recruited through schools, clinics, and online advertising	Prosocial behavior: SDQ (parent-report and self-report)	Siblings of children with ASD scored lower on prosocial behavior than established norms, based on both mother-report and self-report	Cohens d (mother-report) = 2 – 1.06 ($p < .01$) Cohens d (self-report) = – 1.02 ($p < .008$)	
Walton and Ingersoll (2015)	USA	Case-control study—comparison with siblings of typically developing children	$N = 69$ siblings of children with ASD (Mean age = 10.43, $SD = 3.57$), 47.8% males	Recruited through an online-research network for families of children with ASD	Prosocial behavior: SDQ (parent-report)	Siblings of children with ASD scored higher on mother-reported prosocial behavior than controls	Cohens $d = .35$ ($p = .031$)	4

^a Strengths and Difficulties Questionnaire, ^b empathy and prosocial response subscale of the Conscience Questionnaire

TD controls were found at 22 and 28 months of age, but this difference diminished by 34 months of age. When excluding siblings who go on to receive an ASD diagnosis early in life, there is little evidence of impaired empathy beyond 3 years of age in siblings of children with ASD (e.g., Campbell et al., 2017; Hutman et al., 2010; McDonald et al., 2017).

Prosocial Behavior in Siblings of Children with ASD

Of the 10 studies investigating prosocial behavior, four studies found less prosocial behavior in siblings of children with ASD compared with controls (Chan & Lai, 2016; Griffith et al., 2014; Hastings, 2003a; Tsai et al., 2016). Four studies found no difference between siblings and controls (Benson & Karlof, 2008; Hastings, 2003b; Hastings & Petalas, 2014; Petalas et al., 2009). Two studies found more prosocial behavior in siblings of children with ASD on at least one measure (McDonald et al., 2017; Walton & Ingersoll, 2015). McDonald et al. (2017) did not find a significant difference between siblings of children with ASD and controls on the behavioral measure of empathy and prosocial behavior, but parents of siblings reported significantly higher empathy and prosocial behavior than parents of controls.

Tsai et al. (2016) included siblings from the UK and Taiwan and compared siblings' SDQ scores with British and Chinese norms, respectively. Siblings of children with ASD from the UK scored significantly lower than population-norms on *parent*-reported prosocial behavior with no significant difference in *self*-reported prosocial behavior. In contrast, siblings of children with ASD from Taiwan scored significantly lower on *parent- and self*-reported prosocial behavior relative to norms. The UK sample effect size difference was small, whereas the Taiwan sample difference was large.

Quality Assessment and Risk of Bias

Our quality assessment (Newcastle-Ottawa Scale) showed considerable variability in quality among studies. The scores ranged from 2 to 7 points on the 0-to-9 point scale. We scored studies of empathy higher ($k = 7$, $M = 5.00$, $SD = 1.15$) than studies of prosocial behavior ($k = 10$, $M = 2.60$, $SD = 0.97$). The studies focused on prosocial behavior rarely secured comparability between cases and controls (i.e., the use of population-norms prevented comparison of samples on demographic variables). Further, studies of prosocial behavior used almost exclusively self-report measures, lacked independent validation of cases (i.e., validation of ASD diagnosis in brother/sister), and did not obtain representative samples (i.e., random samples or all cases in a defined catchment area). More of the empathy studies included independent validation of ASD diagnoses, behavioral measures of outcomes, and comparability between cases and controls (i.e., covariates included in the analyses or groups matched on background measures).

However, none of the empathy studies were rated to recruit representative samples of cases, and none reported response rates or non-responder characteristics.

Discussion

The purpose of this review was to collate research examining whether siblings of children with ASD display lower, comparable, or heightened levels of empathy and prosocial behavior when compared with children who do not have siblings with ASD. Authors of previous studies have repeatedly noted the considerable heterogeneity among studies of siblings of children with ASD and the inconclusiveness due to the mixed findings (e.g., Meadan et al., 2010; Shivers et al., 2019; Tomeny et al., 2012). This review supports this message with our findings of considerable heterogeneity in methodology, samples, and results of studies on empathy and prosocial behavior in siblings of children with ASD (Meadan et al., 2010; Shivers et al., 2019).

With regard to empathy, the reviewed studies suggest that siblings of children with ASD likely have normative empathy development, particularly as empathy evolves beyond the toddler years. Only one study found lower empathy in siblings over age 3 years (Eyuboglu et al., 2018). However, the extent to which the Unexpected Outcomes Test (UOT), as featured in Eyuboglu et al. (2018), measures empathy can be questioned because the UOT is cognitively oriented toward reasoning and knowledge about emotions, and the original authors describe it as a subtest of emotion recognition (Dyck, 2012; Dyck et al., 2001). Whereas some authors consider emotion recognition and understanding to be the same as cognitive empathy (e.g., Westby & Robinson, 2014), a consensus definition of empathy does not exist (Fletcher-Watson & Bird, 2020) and the UOT may neglect to detect components of empathy. The other measures used to assess empathy in the reviewed studies were seemingly more established empathy measures. Specifically, the behavioral measures are similar to or based on measures used in other developmental research (e.g., Nichols et al., 2015; Sigman et al., 1992; Zahn-Waxler et al., 1992), and the remaining parent- and self-report measures have been reported to be reliable and valid (Dadds et al., 2008; Gilet et al., 2013; Kochanska et al., 1994).

Some studies have demonstrated heightened empathy in siblings of children with ASD (e.g., McDonald et al., 2017; Shivers, 2019), suggesting that some siblings may benefit from their experiences of having a brother or sister with ASD, and develop understanding and compassion. Intact empathic abilities in siblings of children with ASD suggest that the presence of more autistic traits on a group level does not necessarily include deficits in empathy like those characteristic of ASD (Harmsen, 2019; Song et al., 2019). However, future studies should examine whether levels of autistic traits

and empathy are related in siblings. As noted by Shivers (2019), diagnostic criteria and the tools used to assess for ASD do not always include empathy deficits, but rather focus on general social skills and social responsiveness. Further, high levels of autistic traits in one area (e.g., executive dysfunction) do not always imply high levels of autistic traits in another area (e.g., difficulties with empathy). Thus, autistic traits among siblings of children with ASD are not necessarily incompatible with intact empathic abilities.

A recent area of research important to the understanding of empathy in individuals with ASD and their siblings is alexithymia. Alexithymia, which includes difficulties with interpreting one's own emotions, is often observed in individuals with ASD (Kinnaird et al., 2019). Moreover, higher levels of alexithymia are associated with lower levels of empathy (Grynberg et al., 2010). Similarly, a recent study found that alexithymia was associated with empathy, but autistic traits were a stronger predictor (Shah et al., 2019). Together, these studies underscore the need to consider both the levels of alexithymia and autistic traits in future investigations of empathy in siblings of children with ASD.

It has been proposed that siblings of children with ASD may develop more patience, understanding of individual needs, and perspective-taking abilities due to their everyday experiences with their brother or sister with ASD (e.g., Fjermestad et al., 2019; Shivers, 2019). A largely unexplored area of research is to what extent such experiences may buffer against increased genetic risk. In future studies, it would be interesting to examine how empathy, prosocial behavior, autistic traits, alexithymia, and polygenetic risk of ASD interact among siblings of children with ASD.

With regard to prosocial behavior, the results suggest considerable heterogeneity among siblings of children with ASD. Some studies indicate less prosocial behavior, others normative levels of prosocial behavior, and a few studies suggest higher levels of prosocial behavior in siblings of children with ASD. Based on the reviewed studies, no consistent developmental pattern was observed; the findings are mixed independent of which age group was investigated. It is likely that these highly inconsistent findings reflect the general heterogeneity of traits among siblings of children with ASD. For example, one study found that at the mean group level, siblings of individuals with ASD have intermediate levels of ASD symptoms, higher than TD controls but lower than individuals with ASD (Ruzich et al., 2016). Nevertheless, the authors also found that the sibling group could be divided into two subgroups: (1) one group with few autistic traits that was similar to TD controls and (2) a group with many autistic traits that was more similar to a group of individuals diagnosed with ASD. Thus, it could be that studies showing lower levels of prosocial behavior in siblings of children with ASD have an overrepresentation of the second subgroup, whereas studies showing higher levels of prosocial behavior among siblings

of children with ASD have an overrepresentation of the first subgroup. Additional studies are needed to confirm or disconfirm this potential explanation.

A recent study found that more problem behavior in the child with ASD was related to a better sibling relationship (Tudor et al., 2018). The authors suggest that high levels of problem behavior displayed by the child with ASD are easily observable, which in turn makes the care needs of the child with ASD more obvious and promotes prosocial behavior from siblings. Alternatively, siblings of children with milder challenges may be less likely to engage in such scaffolding. Given the complex dynamics in families of children with ASD, future studies should investigate if characteristics of the family system serve as possible mediators or moderators of the relations between being a sibling of a child with ASD and empathy and prosocial behavior. Unsurprisingly, autistic traits in TD siblings have also been found to predict prosocial behavior and should be considered in analyses involving this domain (Petalas et al., 2012).

Unlike the empathy measures, the prosocial behavior measures were more consistent across studies. All but one study used the SDQ prosocial subscale (Goodman, 1997). Hence, differences in prosocial behavior across studies cannot be attributed to the use of different measures. This strengthens the notion of true heterogeneity among siblings and suggests that factors beyond simply being a sibling of a child with ASD are at play.

Notwithstanding, the SDQ (Goodman, 1997) includes a 5-item subscale measuring relatively simple forms of prosocial behavior: (1) consideration of other people's feelings, (2) sharing with other children, (3) helping if someone's hurt, (4) being kind to younger children, and (5) volunteering to help others. Some of these behaviors (e.g., consideration of others' feelings) can tap into other concepts, such as empathy, and these statements do not cover more complex forms of prosocial behavior (e.g., giving someone a compliment to make them feel good; cooperating with others; engaging in volunteerism). The psychometric properties of the SDQ are considered acceptable in terms of test-retest reliability, internal consistency, and validity (Stone et al., 2010). However, the convergent validity of the SDQ has largely been established by (a) positive correlations between the problem-oriented scales and measures of psychopathology and (b) negative correlations between the prosocial behavior subscale and measures of psychopathology (see Stone et al., 2010 for review). Although negative correlations with psychopathology suggest the prosocial subscale is a strength-based subscale, the extent to which this establishes the ability of the subscale's scores to adequately assess prosocial behavior is unclear. Because 9 out of 10 studies of prosocial behavior in siblings of children with ASD used the SDQ, future research would benefit from using multiple informants and assessing the convergent validity of the prosocial subscale when compared with

other measures of prosocial behavior to provide points of comparison and allow for a more comprehensive assessment of the construct.

Moreover, our ability to make strong conclusions is tempered by methodological limitations across the reviewed studies. Our quality assessment indicates a need for studies with more comparable control groups, representative samples of cases, and more use of independent validation (i.e., beyond self-report) of the ASD diagnosis, as well as more comprehensive, and preferably behavioral, measures of empathy and prosocial behavior.

Researchers have developed and tested the efficacy of a number of different interventions targeting psychosocial adjustment in siblings of children with developmental disabilities (Haukeland et al., 2020; Jones et al., 2020). Our findings suggest that empathy and prosocial skills most likely do not need to be the primary foci of these interventions. For parents, these findings can be reassuring; siblings may be at general risk of psychosocial maladjustment but they seem to display as much empathy and prosocial behavior as their peers without a brother or sister with ASD. Furthermore, siblings' strengths in empathy and prosocial behavior can be beneficial for the entire family and contribute to more harmonious family dynamics. A theme for further research may be how siblings' empathy and prosocial behavior contribute to family dynamics and other family members' well-being.

Our review has limitations. First, we did not search for or included so-called grey literature, like theses and dissertations. We did not search for unpublished studies, so it is possible that studies with null findings have been neglected. Second, some of the reviewed studies of prosocial behavior did not explicitly aim to investigate prosocial behavior in siblings but used measures that included prosocial behavior as a subscale of broader measures of mental health or empathy. This means that few of the articles mentioned prosocial behavior in their title, abstract, or keywords, and thus, only four were identified through the initial systematic search whereas six were identified through the manual search (see Fig. 1). We performed a comprehensive manual search consulting previous reviews, reference lists of included papers, and papers known to the authors. Nevertheless, relevant papers could have been missed, and the conclusions in our review should be interpreted considering this possibility. Third, the applicability of the Newcastle-Ottawa quality assessment scale has been disputed (Stang, 2010); however, the scale is widely used and has demonstrated adequate psychometric properties (Moskalewicz & Oremus, 2020; Oremus et al., 2012). Hence, the use of a standardized quality assessment specifically developed for case-control studies, which in the current study showed excellent reliability, could also be considered a strength.

To conclude, considering how the genetic and environmental risks of being a sibling of a child with ASD may affect

sibling development (Ingersoll & Wainer, 2014; Shivers, 2019), empathy and prosocial behavior are important domains that researchers, clinicians, and the affected families and siblings need to better understand. This review suggests that, overall, most siblings of children with ASD have intact empathy and prosocial behavior.

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Declarations

Conflict of Interest The authors declare no competing interests.

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