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REVIEW ARTICLE

Is language ability associated with behaviors of concern in autism? A systematic review

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Abstract

This review systematically synthesized evidence on the association between structural language ability and behaviors of concern (BoC) in autism. Four databases were searched for studies that included >10 autistic participants, measures of structural language (content and/or form of language) and BoC, and an analysis of their association. BoCs included self-injurious behavior (SIB), aggression, tantrums, and externalizing behavior. Methodological quality of studies were assessed using the Newcastle Ottawa Scale. Forty-five publications ($n = 11{,}961$) were included. Forty studies were cross-sectional and five were prospective cohort studies. Over 70% of the studies investigating expressive language and SIB ($n = 10$), aggression ($n = 5$), tantrums ($n = 3$), and externalizing behavior ($n = 17$) reported an inverse association, where lower expressive language ability was associated with increased BoC. Eleven out of sixteen studies of combined expressive and receptive language reported an inverse relationship with SIB or aggression. All outcomes were rated as moderate to very low certainty of evidence. This review highlights evidence showing an inverse association between expressive or combined language ability and SIB, and externalizing behavior in autism. However, further high-quality studies that use standardized, consistent measures of language and behavior and investigate longitudinal associations are needed. Early detection and support for reduced structural language difficulties have substantial potential to assist in reducing BoC.

Lay Summary

The presence of language difficulties in autistic individuals have been thought to be linked with higher levels of behaviors of concern (BoC) (e.g., self-injurious behavior, aggression, tantrums). Our research found expressive language difficulties were most consistently linked with self-injurious and externalizing behaviors, however, receptive language difficulties did not have consistent links with any BoC. Information from this review can inform prioritization of interventions goals, support needs and prognostication.

KEYWORDS
autism spectrum disorder, behaviors of concern, structural language

INTRODUCTION

As a neurodevelopmental condition, autism spectrum disorder (autism) affects around 18 per 1000 children aged 8 years and under in the United States of America (Maenner et al., 2020). Autism has a parent-reported prevalence of 4.4% amongst 12 year old Australian children (May et al., 2020), and a cumulative incidence of 1.10% in 4 year olds (May & Williams, 2018). Autism is characterized by social-communication impairments, and stereotyped and repetitive behavior, and has environmental, genetic, and epigenetic causes (American Psychiatric Association, 2013). While not core diagnostic characteristics of autism, structural language difficulties and behaviors of concern (BoC) are commonly seen in autism. An association between language difficulties (i.e., difficulties with the content and form of language) and BoC in autistic individuals has been documented in the literature but the specific interactions across components of language and BoC are not well understood.

BEHAVIORS OF CONCERN

BoC including behaviors like aggression (such as verbal abuse, threats and physical violence), destructive behavior (such as breaking or destroying furniture and setting fires), disruptive behavior (such as repetitive screaming, smearing feces), and self-injurious behavior (SIB) (including self-biting, head banging) are sometimes observed in autism (National Collaborating Centre for Mental Health [UK], 2015). While not all individuals who display BoC are autistic, autism is an identified risk factor for BoC (McClintock et al., 2003). BoCs such as tantrums are the most commonly reported concerning behavior by parents after eating and sensory problems (Maskey et al., 2013). One in four autistic children have been reported to have clinically significant aggressive behavior (i.e., T-scores ≥70) on the *Child Behavior Checklist* (CBCL) (Hill et al., 2014). SIB is observed in 25%–44% of autistic people (Matson, Boisjoli, & Mahan, 2009), where the most common reported forms of self-injury are hand hitting, skin picking, and hitting oneself with an object (Laverty et al., 2020; Soke et al., 2016). This is understood to be a distinct symptom to self-harm in borderline personality disorder, where the most common form of self-harm is self-cutting (Buck et al., 2014; Lugnegård et al., 2011; Skokauskas & Gallagher, 2010; Steenfeldt-Kristensen et al., 2020).

BoCs contribute significantly to parental or caregiver stress (Davis & Carter, 2008; Miranda et al., 2019) and school provider burnout (Otero-López et al., 2009), which can reduce the quality of care and education provided to autistic children. BoCs are also associated with negative outcomes for the individual, including impaired social relationships (Luiselli, 2009), and adverse experiences, like use of physical restraints in medical settings (Gaskin et al., 2013; Taylor et al., 2011). In one study, the presence of aggressive behavior was associated with increased use of psychotropic drugs and melatonin, and more coexisting sleep, internalizing, and attention problems (Hill et al., 2014). BoCs can also impair safety, job retention, independent living and cohabiting in adulthood for autistic people (Luiselli, 2009).

There have been many hypotheses about why there is an increased prevalence of BoC in autism compared to non-autistic individuals. For example, the higher prevalence of mental health conditions in those diagnosed with autism has been proposed as one reason for higher rates of SIB, on the basis that a proportion of autistic individuals have co-occurring conditions that have a known association with SIB (Minshawi et al., 2014). Other explanations, such as release of frustration, expression of anger (Ho et al., 2012), and induced dissociation to avoid aversive emotion in situations of abuse or trauma, have been suggested as possible explanations for the BoC (Summers et al., 2017). SIB has also been considered as a form of restricted, repetitive behavior that is characteristic of autism (Iwata, Dorsey, et al., 1994; Iwata, Pace, et al., 1994).

LANGUAGE ABILITY IN AUTISM

Structural language difficulties are common in autistic individuals and are often detected early, in many cases prior to an autism diagnosis (Luyster et al., 2008; Mitchell et al., 2006). Language is made up of two modalities which include receptive language (understanding words, gestures, and symbols) and expressive language (using words, gestures, and symbols). Within these two modalities, there are three separate but overlapping domains: content (semantics), form (syntax, morphology, and phonology), and use (pragmatics) (Bloom & Lahey, 1978). While pragmatic language difficulties are universal in autism, there is variability in the content and form in language, hereafter referred to as structural language (Bloom & Lahey, 1978). A proportion of autistic children perform within the average range with their structural language skills (Brignell et al., 2018; Kwok et al., 2015), however, around 25 to 30% of autistic children are minimally verbal (Anderson et al., 2007; Kwok et al., 2015; Norrelgen et al., 2015; Rose et al., 2016; Tager-Flusberg & Kasari, 2013). About 30% of autistic people have a co-morbid diagnosis of intellectual disability, and concomitant receptive language impairment is not uncommon (Baio et al., 2018).

ASSOCIATION BETWEEN STRUCTURAL LANGUAGE ABILITY AND BOC

The association between structural language ability and BoC has been well documented in the literature within a range of populations including autism, intellectual

disability, and language impairment (Carpenter et al., 2002). Some explanations for the association include that BoC functions as a form of communication or occurs as a result of frustration when individuals are not able to express themselves adequately through language (Carpenter et al., 2002; Chiang, 2008). Another mechanism proposed is that BoC may serve as an adaptive cognitive tool that enhances the ability to recognize and regulate one's emotions that is not available to individuals with language impairments (Cole et al., 2010; Curtis et al., 2019).

A study found that 64% of individuals with a language impairment had associated externalizing behaviors (Conti-Ramsden & Botting, 2004). A meta-analysis conducted more recently in 2018 looked at the relationship between language disorders and problem behaviors in a general population of children and adolescents with and without autism or intellectual disability (Curtis et al., 2018). The authors concluded that delayed language development or disorders in children as compared to a typically developing population were associated with problem behaviors of moderate size ($g = 0.43$) and was statistically significant even after age was included as a moderator variable (Curtis et al., 2018). However, while the review considered the quality of language assessment and publication bias, it did not analyze the overall quality of included evidence (Curtis et al., 2018). In another study, with a population of those with intellectual disabilities, a risk marker for challenging behavior like self-injury was the presence of deficits in receptive and expressive communication (McClintock et al., 2003). Given the unique nature of autism, the association between structural language ability and BoCs requires more detailed investigation. Furthermore, what has been lacking in the research to date on this topic is a detailed examination of the specific modalities of language (e.g., receptive and expressive) and types of BoC (e.g., self-injury, aggression). A better understanding of which language difficulties are associated with which BoC can provide a more nuanced understanding around prioritization for intervention and more refined prognostication.

OBJECTIVES

We aimed to systematically identify, synthesize and assess the quality of the evidence examining the association between structural language ability and BoC in autism and provide the certainty of evidence for each outcome. We then aimed to assess the strength of the associations between specific modalities of language, expressive, receptive, and combined expressive and receptive, and specific types of BoC.

METHODS

The Preferred Reporting Items for Systematic Review and Meta-Analysis (PRISMA) (Page et al., 2021) and the Synthesis Without Meta-analysis (SWiM) (Campbell et al., 2020) reporting guidelines were used for this review. A protocol was developed a priori and published in the National Institute for Health Research International Prospective Register of Systematic Reviews (PROSPERO); registration number: CRD42020204192.

Eligibility criteria

Original peer-reviewed research and gray literature published in any language were eligible for inclusion. No study design or date limits were imposed on the search. Studies had to include a sample of greater than 10 participants (all ages included) with a clinical diagnosis of autism as defined by the International Classification of Diseases (ICD) or Diagnostic Statistical Manual (DSM) (American Psychiatric Association, 2013). We excluded studies that recruited participants that had co-occurring genetic or biological syndromes, like Neurofibromatosis Type 1 and Tuberous Sclerosis, secondary to an autism diagnosis. This is because these syndromes are distinct disorders even though individuals with these diagnoses can exhibit a similar clinical profile or symptoms to autistic people. Studies had to include a measure of structural receptive or expressive language (i.e., semantics, morphology, syntax). We did not include studies that reported on only written language/text (receptive and expressive). Both categorical and continuous data were included. Studies that focused on the form and content of language were included, but those that focused on measures of social communication or pragmatic language were excluded as difficulties in these areas are diagnostic features of autism.

The outcome of interest was BoC. This included behaviors which had the potential to put the individual and/or others at risk of harm, such as aggression, SIB, property destruction, and also behaviors that have been reported by parents as challenging, such as tantrums and disruptive behavior including inappropriate sexual behaviors, or urination or defecation in public (Dunlap et al., 1994). We also included behaviors categorized as externalizing behavior when we could not extract data on individual domains, such as self-injury or aggression. We have defined externalizing behavior to include a combination (or composite) of various behaviors, including that of hyperactivity and repetitive behavior (Achenbach & Edelbrock, 1978).

Search strategy

Four databases were searched up to the 15th of June 2022: Ovid MEDLINE, EMBASE (Ovid), PsycINFO (Ovid), and CINAHL (EBSCO). In addition, we reviewed reference lists of relevant studies and consulted with experts in the field to look for relevant studies. Search terms from databases are included in Appendix S1.

Study selection process

All studies were screened by title and abstract, and then by full text independently by two of three authors (Amanda Brignell, Charissa Chan, Wei Herng Wan). Any disagreements were resolved by consensus or seeking a fourth author's (Katrina Williams) opinion. If there were two or more papers that used overlapping study samples, only one paper was selected to represent the study based on the paper's sample size and quality of the analysis, including for example whether they had adjusted the analysis for covariates. The exception was if each paper of the same study presented unique data relevant to an exposure or outcome of interest.

Data extraction

Data extraction was performed independently by two authors (Amanda Brignell and Charissa Chan) from the included studies on each of the criteria presented in Appendix S2. For longitudinal studies, data was extracted at relevant time points that measured our exposure and outcome of interest. Any conflicts or disagreements were resolved by consensus or seeking a third author's (Tamara May) opinion.

Data management

To synthesize the studies, they were first grouped by language construct and then BoC. The three groups of language classification were: expressive language, receptive language, and combined expressive and receptive language. BoC were classified into four main categories by reviewing the questions used within the questionnaires administered to participants in the original study. These categories are SIB, aggression, tantrums, and externalizing behavior. Due to the variable constructs and definitions of externalizing behavior, these were subgrouped (see Table 2) to provide clarity as to what these behaviors were included. We classified expressive and receptive language ability separately because they have distinct characteristics and so that we could provide a more nuanced understanding of language associations with BoC.

Where studies had used more than one tool to measure the same construct, the tool that was reported in our study was chosen primarily based on the highest Newcastle Ottawa Scale (NOS) assessment, or secondarily if the tool was used more commonly by other studies to allow for greater between study comparisons.

Data analysis

We collated data on the effect estimates, direction of effect and p-values, and synthesized the results narratively as recommended by the Synthesis Without Meta-analysis (SWiM) reporting guidelines (Campbell et al., 2020). Substitutes (or calculations where possible) of effect sizes were included when these were not reported by the study. A correlation coefficient (r) of less than 0.40, and Cohen's d between 0.2 and 0.5 was considered a weak association, an r between 0.40 and 0.70 and d between 0.5 and 0.8 to be moderate, and an r greater than 0.70 and d greater than 0.8 to be a strong association (Schober et al., 2018). Precise p-values were recorded where available, however since they were often not reported in studies where the association was not significant, "$p > 0.05$" was recorded instead. If a crude statement of the presence of statistical significance was made then a conservative report of "$p < 0.05$" was recorded. Since the only data consistently reported across all studies was direction of effect, the results were synthesized by vote counting as recommended by the SWiM guidelines (Campbell et al., 2020; McKenzie & Brennan, 2019).

We planned to conduct a meta-analysis (see registered protocol ref) using correlations as the effect of interest, however due to the substantial heterogeneity in measures and test statistics reported by studies, meta-analyses were not performed. In addition, despite having contacted original study authors for results of interest that had been incompletely reported, some data were not available.

Study risk of bias assessment

As all included studies were non-randomized, observational studies, we used the Newcastle Ottawa Scale (NOS) to conduct a risk of bias assessment using a star scoring system (0–9), where more stars indicate higher quality (Wells et al., 2009). Risk of bias was assessed independently by two authors (Amanda Brignell and Charissa Chan). Any conflicts were resolved by consensus or seeking a third author's (Katrina Williams) assessment. The same criteria were used across studies regardless of if they were a cohort or cross-sectional study design. The criteria were based on the selection of cohorts, comparability of cohorts, and assessment of outcome (see Appendix S3). If results were missing, no star was awarded.

If caregivers of individuals with autism completed assessment measures for language or behavior, risk of bias was ascertained to be higher and hence no star was given, as this was considered similar to self-reporting. By contrast, if clinicians, researchers, or teachers had completed the measures, a star was given. However, if the same assessor completed both the exposure and outcome assessment tool, it was deemed that there was lack of blinding and hence no star for the outcome criterion was given. The criterion of comparability of cohorts was assessed by consideration of whether there had been adjustment for confounding variables. In longitudinal studies, we set a threshold of 12 months for duration of

follow up being sufficient for assessment of outcome criterion. Adequacy of follow up of cohorts were deemed sufficient if follow up rate was at least 80% or if description was provided on those lost.

Certainty of evidence

To review the certainty of evidence regarding the presence of an association, we adopted the Grading of Recommendations, Assessment, Development and Evaluation (GRADE) Working Group approach (see Appendix S4). Two authors (Amanda Brignell and Charissa Chan) rated the evidence across outcomes independently, and any disagreements were resolved by consensus or discussed with a third author (Katrina Williams). The domains that were considered to rate down certainty of the evidence were risk of bias, inconsistency, indirectness, imprecision, and publication bias, although imprecision could not be assessed since a meta-analysis was not conducted (Huguet et al., 2013; Schünemann et al., 2013). Certainty of the evidence was rated up if there was a moderate to large effect size, exposure-response gradient, and if plausible confounding was accounted for.

An overall GRADE certainty rating for each outcome was determined based on an average of ratings across studies.

RESULTS

Study selection

The search yielded 9237 records, in addition to 10 records identified through other sources. There were 2934 duplicates. 6138 records were excluded on title and abstract screening. The remaining 145 records were reviewed at full text level. Forty-two studies (45 records listed in Table 1) met inclusion criteria (see Figure 1 for PRISMA study flow diagram).

Study and participant characteristics

Of the 45 included publications (Table 1), 40 were cross sectional and five were longitudinal. There was a total of 11,910 participants, ranging from 15 to 1937 in each of the studies. Of these, 9657 (81%) were male. Twenty-eight studies were conducted in the USA, four in the United Kingdom, two in France, two in Italy, and one in each Australia, Canada, Greece, Japan, and The Netherlands. The mean age across all studies was 9 years 2 months (range: 1.3–52 years).

Of the five studies that had longitudinal data of interest, the total cohort size was 866. These studies reported the associations between expressive language and SIB (Baghdadli et al., 2008), expressive language and aggression (Rodas et al., 2017), expressive language and externalizing behavior (Shattuck et al., 2007; Stringer et al., 2020), receptive language and SIB (Makrygianni & Reed, 2010), and combined language and SIB (Makrygianni & Reed, 2010).

Amongst all publications investigating the association between expressive language and BoC, 12 examined SIB, 11 examined aggression, four tantrums, and 20 externalizing behavior. Of those investigating receptive language, three examined SIB, four aggression, one tantrums, and four externalizing behavior. Of those investigating combined expressive and receptive language, seven examined SIB, eight examined aggression, and seven externalizing behavior.

Language and BoC measures varied across studies. In total there were 24 measures of language and 22 measures of BoC (see Table 1). The most common standardized measures of language used were the Vineland Adaptive Behavior Scale—Communication subscale (VABS-C) ($n = 11$ studies) and the Peabody Picture Vocabulary Test (PPVT; $n = 5$ studies). The most common standardized measure of BoC used was the Child Behavior Checklist (CBCL; $n = 7$ studies).

Risk of bias in studies

Included studies were rated between 1 and 6 stars out of a maximum of 9 stars on the Newcastle Ottawa Scale (Wells et al., 2009). In every study, most stars were given on the *Selection* criteria of the scale. Only 11 of the 38 studies adjusted for covariates to obtain a star under the *Comparability* criteria. Most longitudinal cohort studies scored at least one star under the *Outcome* criteria unless duration of follow up was less than 12 months. According to the Agency for Healthcare Research and Quality (AHRQ) standards (Viswanathan et al., 2012), because all studies had not scored stars consistently across all domains, they were rated moderate to high risk of bias. For details of risk of bias ratings see Appendix S3.

RESULTS OF SYNTHESES

Cross sectional studies

Expressive language and BoC

Eleven studies investigated the relationship between expressive language and SIB (Table 2). Of studies that reported an odds ratio, there was at least a three-fold decrease in SIB (OR range: 3.5–3.7) with higher expressive language ability. Nine of the 11 studies (77%) showed an inverse relationship between expressive language and SIB, and for 6 of the 10 the association was

statistically significant ($p < 0.05$ for all studies). Two studies showed a mixed picture with some associations being direct and others inverse, although effect sizes were not reported. Neither association was statistically significant ($p > 0.05$ for both studies).

Eleven studies analyzed the association between expressive language and aggression and in three of these the association was statistically significant. For the other studies, the associations were inconsistent with no clear magnitude and direction of the effect.

Only four studies investigated expressive language and tantrums. Three of the four showed an inverse direction of effect but none of these associations were statistically significant.

Receptive language and BoC

In total, nine studies investigated the relationship between receptive language and BoC (Table 3). Inconsistency was identified across these studies for the relationship between receptive language and the BoCs of SIB, aggression and externalizing behavior. Three studies analyzed the relationship between receptive language and SIB, of which Matson et al. (2009) reported a strong, negative relationship that was statistically significant.

Four studies investigated the association between receptive language and aggression, where half reported an inverse relationship that was statistically significant. One study reported a direct relationship where more receptive language ability was associated with more aggression, but the effect size was weak ($\beta = 0.005$) and the association was not statistically significant ($p > 0.05$).

The five constructs of externalizing behavior varied between the two studies that analyzed the relationship between receptive language and externalizing behavior. Three constructs found an inverse relationship, and one was not reported. Only one of the three constructs that found an inverse relationship was statistically significant and had a strong relationship ($\beta = -0.78$).

Only one study looked at the relationship between receptive language and tantrums. This study found a small, direct, not statistically significant relationship ($r_{pb} = 0.068$).

Combined expressive & receptive language and BoC

Seven studies analyzed the association between combined language and SIB, and five of the seven found an inverse relationship (Table 4). Amongst these five studies, reported effect sizes were weak, and only two found statistically significant relationships. One study reported a moderate, direct relationship that was statistically significant between combined language and SIB.

Eight studies analyzed the association between combined language and aggression. While five studies reported an inverse relationship, only one reported statistical significance ($p < 0.001$).

Of the seven studies that looked at the association between combined language and externalizing behavior, three reported a directly proportional relationship where more combined language ability was associated with more externalizing behavior. Only Matson et al. (2009) found the association to be statistically significant ($p < 0.01$).

For details of studies reporting data for each of the BoC areas, reference can be made to Tables 2–4.

Longitudinal studies

The five longitudinal studies reported on the associations between expressive language and SIB (Baghdadli et al., 2008), expressive language and aggression (Rodas et al., 2017), expressive language and externalizing behavior (Shattuck et al., 2007; Stringer et al., 2020), receptive language and SIB (Makrygianni & Reed, 2010), and combined language and SIB (Makrygianni & Reed, 2010). Three studies (Baghdadli et al., 2008; Makrygianni & Reed, 2010; Shattuck et al., 2007) used regression analysis and one (Rodas et al., 2017) used structural equation modeling, with language at baseline and BoC at follow up at least 6 months later. Only one study (Baghdadli et al., 2008) included covariates in the analyses.

Baghdadli et al. (2008) measured expressive language ability at baseline, and this was analyzed against two groups, one with a negative outcome of SIB (persistent, recurrent, or newly emerged) and one with a positive outcome (disappearance or never exhibited) over 3 years. The authors found an inverse, statistically significant relationship between expressive language ability and SIB (OR = 3.5, 95%CI 1.1–13.4). This study was rated four out of a maximum of nine stars for risk of bias on the NOS. Rodas et al. (2017) measured expressive language at baseline and analyzed this against the presence of aggression 6 months later. The authors found an inverse but not statistically significant relationship with an intercorrelation of −0.26. Rodas et al. (2017) was rated one star out of nine on the NOS. Shattuck et al. (2007) measured expressive language at baseline and analyzed this against a change in externalizing behavior (which comprised of aggression and inappropriate behavior) over 4 years and 6 months. They found that higher expressive language ability was associated with more externalizing behavior (unstandardized coefficient = 0.12), but this relationship was not statistically significant. On the other hand, Stringer et al. (2020) found an inverse relationship where less expressive language ability was associated with more externalizing behavior (which comprised of conduct

problems and tantrums) (intercept B = −1.64, slope B = −0.19) but was also not statistically significant. Both studies (Shattuck et al., 2007; Stringer et al., 2020) were rated six stars on the NOS, and which was the highest quality amongst all included studies.

Makrygianni and Reed (2010) measured receptive language at baseline and analyzed this against a change in SIB over 9 months, and found a weak directly proportional relationship where stronger receptive language ability was associated with more SIB (r = 0.086), although findings were not statistically significant. The authors found an inverse relationship between combined language and SIB (r = −0.007) that was also not statistically significant (Makrygianni & Reed, 2010). This study was rated three stars on the NOS.

Reporting biases

We were unable to complete analysis of potential publication or reporting bias due to the variability of measures between the studies. While studies that did not find statistical significance in the association of interest often did not report the effect sizes, we have still included these studies to ensure the presentation of results is comprehensive.

Certainty of evidence

We rated the association between expressive language and composite externalizing behavior to be moderate certainty of evidence. The associations that were rated as low certainty of evidence included expressive language and SIB, expressive language and aggression, expressive language and tantrums, receptive language and externalizing behavior, and combined language and SIB, combined language and aggression, and combined language and externalizing behavior. We found very low certainty of the evidence for receptive language and SIB, aggression, and tantrums. Details of risk of bias assessments are available in Appendix S4.

DISCUSSION

Language difficulties are often said to be associated with BoC, yet the details of these associations have not been well elucidated in autism. This review synthesized the current evidence on the association between different modalities of language (expressive, receptive, and combined) and types of BoC (SIB, aggression, tantrums and externalizing behavior) in autistic individuals. We adopted this approach because although BoCs often overlap clinically, they can exist in isolation or on a continuum with lower risk behaviors being replaced by higher risk ones. Expressive and receptive language difficulties can also be separately or simultaneously present in an individual, with the gap between the two language abilities potentially creating frustration for an individual, in turn resulting in BoC.

The findings of our review varied depending on the different language modalities and types of BoCs investigated. We identified consistent inverse associations between expressive language and SIB, expressive language and composite externalizing behavior, and combined language and SIB. This was established by vote counting of the number of studies that had inverse associations. For these associations, higher language ability was associated with lower levels of BoC. However, the certainty of the evidence for all associations, except expressive language and externalizing behavior, which was rated moderate, was rated low to very low. This was mainly because the risk of bias of most studies was rated moderate to high.

Expressive language

Self-injurious behavior

Our findings for SIB are in keeping with a meta-analytic systematic review investigating language and BoC that included children with other neurodevelopmental disorders, such as intellectual disability, in addition to a subset of autistic individuals (McClintock et al., 2003). In that review, individuals with combined language difficulties were more likely to display self-injury, however whether included studies adjusted for covariates is unclear (McClintock et al., 2003). In our review, while 77% of studies showed an inverse relationship, only two (Baghdadli et al., 2008; Fradkin et al., 2019) of the 13 included studies that investigated SIB adjusted for covariates (autism severity, cognition, social skills, daily living skills, and sex), and only one of these found an inverse, significant relationship (Baghdadli et al., 2008). While we have identified the presence of an association, the paucity of covariate adjustment amongst our included studies means that the ability to draw a conclusion about the presence of a causative relationship between greater language difficulties and higher SIB is limited. This highlights the importance of accounting for important and clinically relevant covariates when studying associations in future research.

Composite externalizing behavior

The relationship between expressive language and composite externalizing behavior was also mostly consistent across studies, where 78% of studies found that greater expressive language ability was associated with less composite externalizing behavior. Our findings are consistent with a meta-analysis by Curtis et al. (2018), who found

TABLE 1 Included publications characteristics

Author, year	Study design (duration); location	Autism, *N*; male %	Age mean (SD), range y:mo	IQ (mean [SD], range)	Language measure			Behavior of concern measure	Covariates
					Expressive language	Receptive language	Combined language		
(Ando & Yoshimura, 1979)	Cross sectional; Japan	47, 83.0%	6–14	N.R.: all diagnosed mental retardation according to American Association on Mental Deficiency	Verbal ability scale (observed by researchers)			SIB scale (observed by researchers)	N.R.
(Baghdadli et al., 2003)[a]	Cross sectional; France	222, 82.4%	5(1:2), 2–7	DQ: 5.9% (<20), 70% (20–50), 20.3% (50–70), 4% not intellectually disabled	ADI-R Speech		VABS-C	SIB (observed by researchers)	N.R.
(Baghdadli et al., 2008)[a]	Cohort (3 years); France	222, 80.2% (n = 185 at follow up)	8(1:3), 5–10 (at follow up)	DQ (Vineland-DLS) >70: n = 10; DQ 50–70: n = 51, DQ 20–50: n = 147	'Speech' scale (rated by clinicians based on observation data and parent interview)			Adapted international classification of handicaps scoring system—SIB	Total CARS score; person-related cognition; object-related cognition; social skills; daily living skills
(Benninger, 2019)	Cross sectional; USA	396, 76.8%	2:8(0:7), 1:6–4	70.0(17.1), IQR 55–80.5	MSEL; Bayley-III			CBCL	Age, mother's race, mother's education level, cognitive development, autism severity
(Boonen et al., 2014)	Cross sectional; The Netherlands	206, 85.0%	9:11(1:10), 6–12	All IQ > 70			CCC-2: structural language subscales	SDQ	Age, sex, positive parenting, negative control, autism adapted parenting
(Brinkley et al., 2007)	Cross sectional; USA	275; 85.5% (n = 144 with language data)	10:7(4:5), 3–21	N.R.	ADI-R verbal communication item no. 19			ABC irritability	N.R.
(Charman et al., 2015)	Cross sectional; UK	42, 85.7%	10(2)	BAS-II Matrices T-score: 49.2 (12.7)		Combination: BPVS, CELF-CFD & TROG		SDQ	N.R.
(Chowdhury, 2012)	Cross sectional; USA	143, 76.2%	5:2(1:7), 3–11:7	N.R	ADI-R communication; CASL (core composite) or PLS-4 (total language) or Bayley-III (general language score)			Nisonger child behavior rating form	N.R.
(Clarke, 2021)	Cross sectional; USA	260; 80.4%	12:8(3:5), 4:5–20:4	75.7(28.3), 30–145	VABS-II Expressive	PPVT		ABC irritability subscale; RBS-R: self-injury subscale; VABS II externalizing subscale	Age, NVIQ, adapting skills
(Connolly, 2011)	Cross sectional; USA	85, 82.4%	9:4(2:2), 6–12	N.R.	ADOS levels (non-verbal, pre-verbal, phrase speech and verbal); CCC-2 GCC score			CBCL-teacher report; CBCL-parent report	N.R.
(De Giacomo et al., 2016)	Cross sectional; Italy	88, 92.0%	4:10, 2:4–11:10	IQ < 80 58.0%	ADI-R items B			ADI-R; ADOS items (ADI-R 81, 82; ADOS: E2 = aggression to others, and ADI-R: 83 ADOS:	N.R.

(Continues)

TABLE 1 (Continued)

Author, year	Study design (duration); location	Autism, *N*; male %	Age mean (SD), range y:mo	IQ (mean [SD], range)	Language measure			Behavior of concern measure	Covariates
					Expressive language	Receptive language	Combined language		
								D3 = aggression to self)	
(Dominick et al., 2007)	Cross sectional; USA	63, 87.3%[b]	7:7(2:6), 4:2–14:2	81.0 (18.9)	EVT	PPVT		Atypical behavior patterns questionnaire	N.R.
(Dreher, 2019)	Cross sectional; USA	24, 83.3%	11:5(2:2), 6:7–16:8	UNIT IQ 90.8(19.0), 45–115	CELF-5-core language			BPI-S: Aggression/ Destructive behavior subscale	CELF-5 core language, pragmatic language skills, age, IQ
(Duerden et al., 2012)	Cross sectional; Canada	250, 84.8%	7:5(3:2), 1:10–19:4	No SIB (n = 110): 96.5 (22.11), 42–149; SIB (n = 121): 88.7(25.5), 36–145			Combination: VABS-C, & OWLS: oral score	ADI-R self-injurious behavior: item 83	Atypical sensory processing; IQ; age; sameness, rituals and compulsion, social communication
(Estes et al., 2007)	Cross sectional; USA	57, 82.4%	6:1(0:2)	IQ < 70 (n = 22): NVIQ: 50.7 (9.7); IQ >70 (n = 35): NVIQ: 93 (11.6)			VABS-C (collected at age 6) and verbal IQ (differential abilities scales)	CBCL; ABC (collected at age 9)	N.R.
(Fok & Bal, 2019)	Cross sectional; USA	1937, 87.0%	10:1(3), 6–18	NVIQ (% <70): MV 85%, PS 41%, VF 9%	ADOS module			CBCL; ABC	NVIQ
(Fradkin et al., 2019)[c]	Cross sectional; USA	133, 80.5%	9:7(5:7)	IQ < 70 (n = 60): 16.7%	ADI-R item no. 30; CELF-4			ADI-R module: item no. 81, 82, 83	Sex
(Hartley et al., 2008)	Cross sectional; USA	169, 78.1%	3:6(0:11), 1:6–5:10	MSEL Visual Reception: non-verbal cognitive functioning: 1.89 (0.78)—DQ 54 (calculated)	MSEL: expressive language			CBCL	Nonverbal cognitive functioning, adaptive functioning
(Kanne & Mazurek, 2011)	Cross sectional; USA	1380, 86.6%	9:1(3:6), 4–17	84.7 (25.6)		PPVT		ADI-R	N.R.
(Kelley, 2006)	Cross sectional; USA	15, 66.7%	11.4 (2.1), 8.2–14.2	All IQ > 70	CELF-3	PPVT	VABS-C	BASC aggression	N.R.
(Kozlowski et al., 2012)	Cross sectional; USA	113, 86.7%	8:6(3:4), 4–16	All IQ > 70	ASD-DC			ASD-BPC	N.R.
(Lawrence, 2017)	Cross sectional; USA	145, 78.6%	9–21	IQ not reported. Vineland adaptive composite ranged 20–79 (social skills). All were in a residential facility for autism.			VABS-C	BPI-S; ABC-teacher form	Regression model included: step 1: age, sex, step 2: aggression, adaptive behavior composite, atypical sensory processing, irritability, stereotypies, and social communication, step 3: GI disorders, seizure disorders, and vitamin D deficiencies
(Lorang et al., 2022)	Cross sectional; USA	219, 73.5%	25:5(10:3), 11–53	All IQ < 70	ADI-R item no. 30 (classified verbal or minimally verbal)			Problem Behavior subscale of the scales of Independent Behavior—Revised (SIB-R);– self-injurious behavior-revised	N.R.

(Continues)

TABLE 1 (Continued)

Author, year	Study design (duration); location	Autism, *N*; male %	Age mean (SD), range y:mo	IQ (mean [SD], range)	Language measure			Behavior of concern measure	Covariates
					Expressive language	Receptive language	Combined language		
(Maddox et al., 2018)	Secondary data analysis/cross sectional; USA	182, 94.5%	9:4(2:4), 6–15:7	104.0 (18.7)			VABS-C	BASC-2 aggression scale	Age, IQ, recruitment site, executive function, emotion regulation, social skills
(Makrygianni & Reed, 2010)	Cohort (9 months); Greece	86, 84.9%	6:7(3:1)[d]	64.9 (28.1)		PPVT	VABS-C	RBS-R – Self-injury	N.R.
(Maloy, 2021)	Cross sectional; USA	135, 64.4%	9:5(3:5), 2–17		CCC-2 (gross communication index)			ABC irritability subscale	N.R.
(Maskey et al., 2013)	Cross sectional; England	863, 86.2%	Median 3,7,5 (for autism, Asperger, ASD), 2–18	IQ < 70 (n = 693): 38.5%	Fluent speech vs. non-fluent (parent questionnaire)			Parent-reported frequency of ≥3 behaviors a week: tantrums, aggression, self-injurious behavior	N.R.
(Matson, Boisjoli, & Mahan, 2009) *Earlysteps database*	Cross sectional; USA	168, 75.6%	2:2(0:5), 1:5–3	N.R.		BDI-2: receptive communication component	BDI-2: expressive and receptive communication component	BISCUIT-part 3: Problem behavior (aggressive and SIB)	N.R.
(Matson et al., 2013)	Cross sectional; USA	109, 73.4%	8:2(3:6), 3–16	IQ < 70: 90.1%	ASD-DC: verbal communication			ASD-PBC[e]: challenging behaviors	N.R.
(Matson & Rivet, 2008)	Cross sectional: USA	148, 56.0%[b]	52:4(12:10), 21–88	Profound ID 76.5%, severe 12.4%, moderate 5.4%, mild 0.7%, unspecified 4.7%			VABS-C	ASD-BPA	VABS-C, VABS-DLS, VABS-socialization; ASD-BPA: aggression, disruptive behavior, SIB
(Mayes et al., 2017)	Secondary data analysis/cross sectional; USA	240, 78.8%	4:4(1:1), 1:4–6	95.0(26.5)	EIDP: expressive language			Pediatric behavior scale: tantrums	N.R.
(Mazurek et al., 2013)	Cross sectional; USA	1584, 83.5%	5:11(3:5), 2–17	76.3(23.9)	Verbal vs. non-verbal (observed by researchers)		VABS-C	Aggression vs. no aggression (observed by researchers)	N.R.
(Neuhaus et al., 2022)	Cross sectional; USA	145, 55.2%	12:5(3), 8–17	N.R.	CELF-4		VABS-C	CBCL aggressive	Age, race, household annual income, maternal education, paternal education
(Park et al., 2012)	Cross sectional; Australia	27, 81.5%	4:7(0:7), 3:6–5:11	WPPSI-III full scale IQ 76.1 (17.1)	VABS: expressive; TACL-III; SPELT-P2	VABS-: receptive		DBC-P: disruptive behavior (manipulates, abusive, irritable, kicks, hits, noisy, lies, lights fires)	Age, non-verbal cognitive functioning, sex
(Rattaz et al., 2015) *EpiTED cohort*	Cross sectional; France	97, 82%[b]	15(1:4)	Brunet-Lezine's oculomotor coordination subtest 74.1(60.1)			VABS-C	ABC	N.R.
(Rattaz et al., 2018) *EpiTED cohort*	Cross sectional; France	106, 84.9%	20:7(1:6)	DQ < 40: 64.7%, DQ < 55: 5.9%, DQ < 70: 3.9%, DQ ≥ 70: 25.5%	Speech scale (observed by researchers)			ABC	Best estimate DQ, CARS score, sleeping disorders, gastrointestinal disorders

(Continues)

TABLE 1 (Continued)

Author, year	Study design (duration); location	Autism, *N*; male %	Age mean (SD), range y:mo	IQ (mean [SD], range)	Language measure			Behavior of concern measure	Covariates
					Expressive language	Receptive language	Combined language		
(Richards et al., 2016)	Cross sectional; UK	149, 88.6%	10(4:11), 4–39	Not reported	Wessex scale (verbally/ partially verbal or nonverbal scale)			CBQ	N.R.
(Rodas et al., 2017)	Cohort (6 months); USA	159, 81.8%	5:6, 4–7[d]	Full scale IQ, all above 70: 93.5(13.4)	Combination: CCC syntax, CCC speech, and CASL syntax			CBCL aggressive	IQ, anxiety, hyperactivity/ inattention
(Salomone et al., 2019)	Cross sectional; Italy	82, 78.0%	3:9(0:10), 1–5	DQ (MA/CA = (23.6/44.93) 53 (calculated)	MacArthur-Bates communicative development inventories—expressive	MacArthur-Bates communicative development inventories—receptive		SDQ—conduct problems	Parent sex, parent age, SDQ hyperactivity/inattention; SDQ emotional symptoms
(Shattuck et al., 2007)	Cohort (4 years 6 months); USA	241, 75.5%	22(9:8), 10–52[d]	68.5% mental retardation (Wide range intelligence Test + Vineland screener)—not enough data for IQ	ADI-R verbal and non-verbal			Problem behavior scale—self-injurious behavior-revised	Age, sex, mental retardation status
(Sipes et al., 2011) *Earlysteps database*	Cross sectional; USA	774, 71.6%	2:2(0:5), 1–3[d]	N.R.	N.R.[f]			BISCUIT Part 3	Age, autism diagnostic group
(Stringer et al., 2020)	Cohort (11 years); UK	158, 83.5%	10–12[d]	N.R.	ADOS no phrase speech vs. fluent speech			SDQ conduct problems	VABS, Behaviors in Infancy (DISCO), ADOS G severity score, Maternal GHQ total, Parent education, Carstairs deprivation, school type
(Tevis et al., 2020) *Earlysteps database*	Cross sectional; USA	116, 77.6%	2:4	BDI-2 used to obtain developmental level but not reported in manuscript	BDI-2			BISCUIT Part 3	N.R.
(Uljarevic et al., 2018)	Secondary data analysis/ Cross sectional; USA	107, 83.2%	2:7(2:4), 1:7–3:10	62.6(18.0)			MSEL: verbal developmental quotient	CBCL-DP; ADOS-2: aggressive-disruptive	Chronological age; Nonverbal DQ; Vineland ABC; ADOS SA; ADOS RRB; ADOS total
(Williams et al., 2018)	Cross sectional; USA	346, 78.6%	12:11(3:4), 4–21	76.4(29)	Verbal vs. non-verbal (observed by researchers)			RBS-R: self-injury subscale; ABC irritability subscale; VABS-externalizing subscale	Age, NVIQ

Abbreviations: ABC, aberrant behavior checklist; ADI-R, Autism diagnostic interview—revised; ADOS, autism diagnostic observation schedule (Module 1 = preverbal or single words; Module 2 = phrase speech; Module 3 = verbally fluent children and adolescents; Module 4 = verbally fluent adults); ADOS-RRB, ADOS—restricted, repetitive behaviors; ADOS-SA, ADOS-social activities; ASD, autism spectrum disorder; ASD-BPA, autism spectrum disorder—behavior problems for adults; ASD-BPC, ASD—behavior problems for children; ASD-DA, ASD—diagnosis for intellectually disabled adults; ASD-DC, ASD—diagnostic for children[g]; BAS-II, British Ability Scales–II; BASC, behavior assessment system for children; BPVS, British Picture Vocabular Scale; BDI-2, battelle developmental inventory[g], 2nd edition; CASL, comprehensive assessment of spoken language; CBCL, childhood behavior checklist; CBCL-DP, CBCL dysregulation profile; CBQ, challenging behavior questionnaire; CCC, children's communication checklist; DQ, developmental quotient; EIDP, early intervention developmental profile[g]; EVT, expressive vocabulary test; GCC, general communication composite; IQ, intellectual quotient; MSEL, Mullen Scales of Early Learning; MV, minimally verbal; N.R., not reported/ analyzed; NVIQ, non-verbal IQ; OWLS, oral and written language scale; PPVT, Peabody picture vocabulary test; PS, phrase speech; RBS-R, repetitive behaviors scale—revised; SIB, self-injurious behavior; SSiS, social skills improvement system rating scales; TROG, test of reception of grammar; UNIT, universal nonverbal intelligence test; VABS, Vineland Adaptive Behavior Scale; VABS-C, VABS—Communication; VABS-DLS, VABS—daily living skills; VF, verbally fluent.

[a]We attempted to contact the authors of Baghdadli et al.(Baghdadli et al., 2003) and Baghdadli et al.(Baghdadli et al., 2008) to determine if an overlapping sample was used. While we received no response, we have assumed that both papers are from the same study since the sample size and first author is the same and both relate to a large longitudinal study.

[b]Studies did not record all data of interest, approximates were made based on available data.

[c]Data was requested directly from author.

[d]For longitudinal cohort studies, baseline age reported.

[e]Matson 2013 refers to the ASD-PBC tool used however reference of tool cited is ASD-BPC, hence we assumed there was a typographical error.

[f]We attempted to contact the authors of Sipes et al. for more information about the language measure, however did not receive a response.

[g]These studies are less commonly known.

TABLE 2 Expressive language and behaviors of concern

Outcome	Author, year	*N*	Risk of bias: Selection	Risk of bias: Comparability	Risk of bias: Outcome	Risk of bias: Total (9)	Test statistic	Direction of effect	*p*-value	GRADE
Exposure: *Expressive language*										
Self-injurious behavior	Baghdadli et al., 2008	222	★	★	★★	4	OR = 3.5	↓	0.05	⊕⊕⊖⊖ Low
	Clarke, 2021	260	★★	–	–	2	β = −0.207	↓	0.002	
	Dominick et al., 2007	67	★★	–	–	2	r_{pb} = −0.309	↓	<0.05	
	Maskey et al., 2013	863	★★	–	–	2	X^2 = −17.4	↓	0.001	
	Rattaz et al., 2018	106	★★	–	–	2	OR = 3.7	↓	0.02	
	Richards et al., 2016	149	★	–	–	1	X^2 = 6.5	↓	0.006	
	Ando & Yoshimura, 1979	47	★★	–	–	2	OR = 3.71	↓	0.053	
	Chowdhury, 2012	143	★★★	–	–	3	r = −0.032	↓	>0.05	
	Williams et al., 2018	346	★★	–	–	2	$R^{2change}$ = 0.002	↓	0.478	
	De Giacomo et al., 2016	88	★★★	–	★	4	N.R.	↓/↑	0.48	
	Fradkin et al., 2019	133	★★★	★	–	4	N.R.	↓/↑	0.609	
Aggression	Dominick et al., 2007	67	★★	–	–	2	r_{pb} = −0.285	↓	<0.05	⊕⊕⊖⊖ Low
	Hartley et al., 2008	169	★★★	–	–	3	r = −0.19	↓	<0.05	
	Rodas et al., 2017	159	★	–	–	1	Intercorrelation = −0.26	↓	0.01	
	Ando & Yoshimura, 1979	47	★★	–	–	2	OR = −2.00	↓	0.36	
	Dreher, 2019	24	★★★	–	–	3	Variance estimate = 0.01	↓	0.54	
	De Giacomo et al., 2016	88	★★★	–	★	4	N.R.	N.R.	0.43	
	Kelley, 2006	15	★★	–	–	2	N.R.	N.R.	>0.05	
	Fradkin et al., 2019	133	★★★	★	–	4	N.R.	↓/↑	1.00	
	Mazurek et al., 2013	1584	★★	–	–	2	φ = −0.038	N.R.	0.13	
	Maskey et al., 2013	863	★★	–	–	2	X^2 = 0.003	↑	0.959	
Tantrums	Ando & Yoshimura, 1979	47	★★	–	–	2	OR = 1.43	↓	0.56	⊕⊕⊖⊖ Low
	Dominick et al., 2007	67	★★	–	–	2	r_{pb} = −0.029	↓	>0.05	
	Maskey et al., 2013	863	★★	–	–	2	X^2 = 3.8	↓	0.051	
	Mayes et al., 2017	240	★★	★	–	3	d = 0.4	↑	0.082	

(Continues)

TABLE 2 (Continued)

Exposure: ***Expressive language***

Outcome		Author, year	*N*	Risk of bias: Selection	Comparability	Outcome	Total (9)	Test statistic	Direction of effect	*p*-value	GRADE
Externalizing behavior	Code										
	1, 2	Benninger, 2019	396	★★	–	–	2	$\beta = 5.21$	↓	0.035	⊕⊕⊕⊖ Moderate
	1, 2	Hartley et al., 2008	169	★★★	–	–	3	$r = -0.32$	↓	<0.01	
	1, 2, 6, 8, 9	Lorang et al., 2022	2019	★★★	–	–	3	$r = 0.05$	↑	>0.05	
	1, 3	Connolly, 2011	85	★★★	–	★	4	$r = 0.001$	↓	>0.05	
	1, 3	Fok & Bal, 2019	1937	★★	–	–	2	$n_p^2 = 0.001$	↑	0.49	
	1, 3	Neuhaus et al., 2022	145	★★★	★	–	4	$\beta = 0.16$	↑	>0.05	
	1, 3, 5	Williams et al., 2018	346	★★	–	–	2	$R^{2change} = 0.213$	↓	<0.001	
	1, 3, 5	Clarke, 2021	260	★★	–	–	2	$\beta = -0.017$	↓	0.795	
	3, 5	Salomone et al., 2019	82	★★	–	–	2	$b = -0.001$	↓	>0.05	
	3, 5	Stringer et al., 2020	158	★★★	★	★★	6	Intercept $B = -1.64$, Slope $B = -0.19$	↓	>0.05	
	1, 5	De Giacomo et al., 2016	88	★★★	–	★	4	N.R. (negative)	↓	0.16	
	1, 3, 5, 6, 7	Fok & Bal, 2019	1937	★★	–	–	2	$n_p^2 = 0.012$	↓	<0.001	
	1, 3, 5, 6, 7	Maloy, 2021	135	★★	–	–	2	$\beta = -0.192$	↓	<0.001	
	1, 3, 5, 6, 7	Williams et al., 2018	346	★★	★	–		$R^{2change} = 0.018$	↓	0.041	
	1, 3, 5, 6, 7	Clarke, 2021	260	★★	–	–	2	$\beta = -0.078$	↓	0.252	
	1, 3, 5, 6, 7	Brinkley et al., 2007	275	★★★	–	–	3	$\rho = 0.09$	↑	N.R.	
	1, 3, 4, 5, 7	Park et al., 2012	27	★★★	★	–	4	$b = -0.36$	↓	>0.05	
	1, 8	Shattuck et al., 2007	406	★★★	–	★★★	6	Unstandardized coefficient = 0.12	↑	>0.05	
	1, 8	Sipes et al., 2011	774	★★	★	–	3	$\beta = -0.028$	↓	0.681	
	1, 8	Tevis et al., 2020	58	★★★	–	–	3	$t = 0.396$[a]	↓	>0.05	
	1, 8, 9	Kozlowski et al., 2012	113	★★	–	–	2	$\beta = 0.58$	↓	<0.001	
	1, 3, 5, 6, 7, 8, 9	Matson et al., 2013	109	★★	–	–	2	$\beta = -0.59$	↓	<0.01	
	1, 4, 6, 8, 9	Park et al., 2012	27	★★★	★	–	4	N.R.	N.R.	>0.05	

Note: ↓/negative effect size = more language ability, less behavior of concern; ↑/positive effect size = more language ability, more behavior of concern; ↓/↑: both negative and positive effect sizes reported in study; N.R.: not reported. *Code*: 1: aggression, 2: attention problems, 3: conduct—tantrums, bullies others, steals, lies, disobedient to authority, 4: hyperactivity, 5: tantrums, 6: SIB, 7: unstable mood, 8: inappropriate behavior, 9: repetitive behavior. Composite scores measuring externalizing behavior had also included repetitive behaviors (code 9), which is one of the diagnostic features of autism. These could not be removed from studies' measures.

[a]We calculated result based on included study's data.

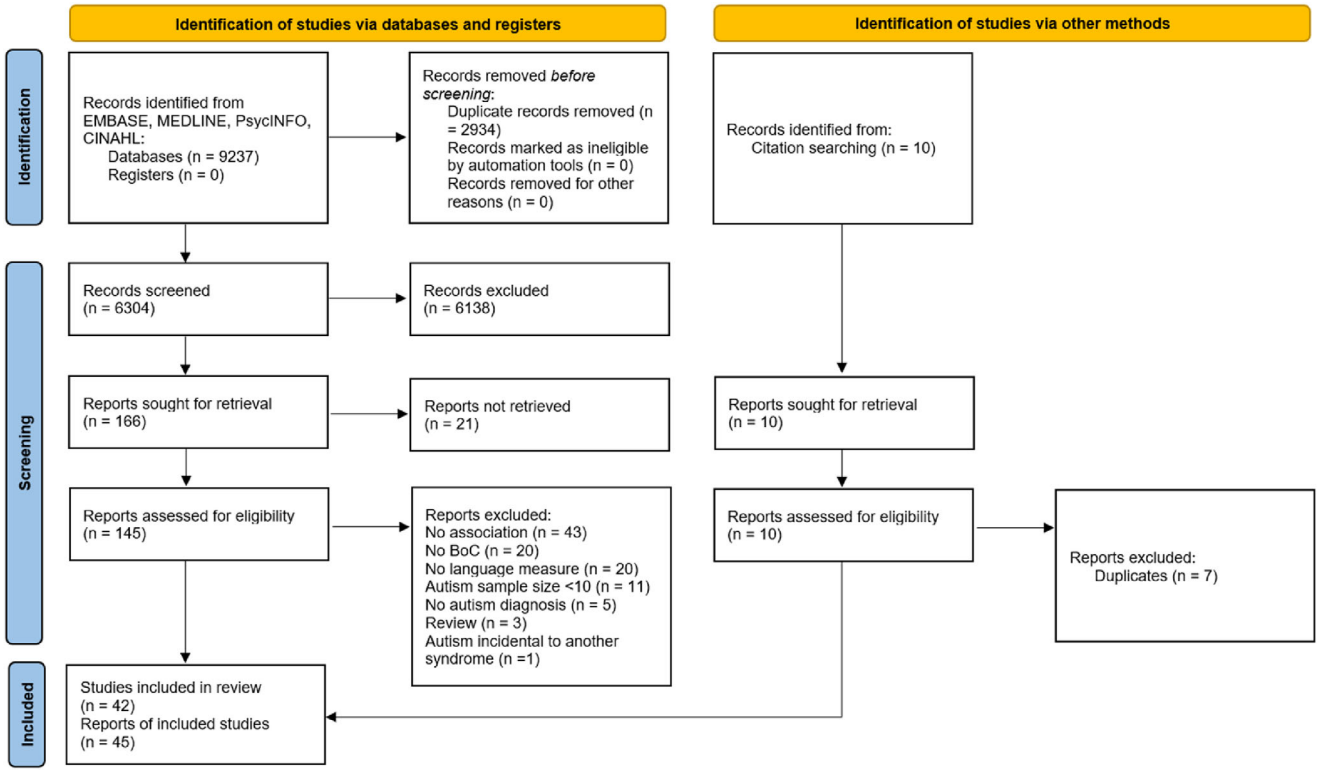

FIGURE 1 Study flow diagram

that children with language disorders display more problem behaviors compared with typically developing peers. In our review, five studies adjusted for covariates which included age, parental education, non-verbal IQ and sex (Neuhaus et al., 2022; Park et al., 2012; Sipes et al., 2011; Stringer et al., 2020; Williams et al., 2018). Given reported stronger associations between structural language difficulties and externalizing behavior amongst males (Hentges et al., 2021) and in children from higher socioeconomic backgrounds (Hentges et al., 2021; Madigan et al., 2019), it will be important for future studies to include these covariates to assess the presence and strength of associations. Our review found less consistency amongst the associations of expressive language and aggression or tantrums, in keeping with some previous studies looking at typically developing populations with mixed conclusions, some finding an association (Brownlie et al., 2004) and others not (McClintock et al., 2003).

Receptive language

There was low between-study consistency in the reported association between receptive language and any BoC. Although not definitive, it is possible that the association between receptive language and BoC is not as straightforward as expressive language, however, there were fewer studies that looked at the association between receptive language and BoC, and these were of low quality. One other possible explanation is that receptive language ability can be a sign of more general developmental difficulties, that may not be associated with frustrations that lead to BoC when receptive and expressive language abilities are commensurate.

Limitations of included evidence

This review included a large number of observational studies with a small median sample size of 147. Due to the substantial variability across study methods (e.g., types of measurement tools, statistical analyses, participants' age, presence of adjustment for covariates), we were not able to complete a statistical synthesis of results. Therefore, in the setting of conducting a systematic review without meta-analysis, we could not calculate an overall magnitude of effect of the associations of interest.

Other limitations of the evidence include inconsistent use of measures or terminology to assess to the same BoC. For example, the Aberrant Behavior Checklist includes items relevant to SIB, verbal and physical aggression, depressed and volatile mood, and temper tantrums which are termed "irritability," whereas the Autism Spectrum Disorder—Behavior Problems for Children tool describes the same items as "externalizing behavior." Similar inconsistencies occurred for language measures.

TABLE 3 Receptive language and behaviors of concern

Exposure: *Receptive language*											
				Risk of bias							
Outcome		**Author, year**	***N***	**Selection**	**Comparability**	**Outcome**	**Total (9)**	**Test statistic**	**Direction of effect**	***p*-value**	**GRADE**
Self-injurious behavior		Matson et al., 2009	168	★★	–	–	2	$r = 0.82$	↓	<0.05	⨁⊖⊖⊖ Very low
		Dominick et al., 2007	67	★★	–	–	2	$r_{pb} = -0.0768$	↓	>0.05	
		Makrygianni & Reed, 2010	86	★★★	–	–	3	$r = 0.086$	↑	>0.05	
Aggression		Dominick et al., 2007	67	★★	–	–	2	$r_{pb} = -0.286$	↓	<0.05	⨁⊖⊖⊖ Very low
		Matson et al., 2009	168	★★	–	–	2	$r = 0.32$	↓	<0.05	
		Kanne & Mazurek, 2011	1380	★★★	★	–	4	$\beta = 0.005$	↑	>0.05	
		Kelley, 2006	15	★★	–	–	2	N.R.	N.R.	>0.05	
Tantrums		Dominick et al., 2007	67	★★	–	–	2	$r_{pb} = 0.068$	↑	>0.05	⨁⊖⊖⊖ Very low
Externalizing behavior	Code										
	3, 5	Charman et al., 2015	42	★★	★	★	4	$r = -0.31$	↓	>0.05	⨁⨁⊖⊖ Low
	3, 5	Salomone et al., 2019	82	★★	–	–	2	$b = -0.001$	↓	>0.05	
	1, 3, 5, 6, 7	Clarke, 2021	260	★★	–	–	2	$\beta = 0.232$	↑	0.055	
	1, 3, 4, 5, 7	Park et al., 2012	27	★★★	★	–	4	N.R.	N.R.	>0.05	
	1, 4, 6, 8, 9	Park et al., 2012	27	★★★	★	–	4	$\beta = -0.78$	↓	<0.01	
	1, 3, 4, 5, 7	Park et al., 2012	27	★★★	★	–	4	N.R.	N.R.	>0.05	

Note: ↓/negative effect size = more language ability, less behavior of concern; ↑/positive effect size = more language ability, more behavior of concern; ↓/↑: both negative and positive effect sizes reported in study; N.R.: not reported. *Code*: 1: aggression, 2: attention problems, 3: conduct—tantrums, bullies others, steals, lies, disobedient to authority, 4: hyperactivity, 5: tantrums, 6: SIB, 7: unstable mood, 8: inappropriate behavior, 9: repetitive behavior. Composite scores measuring externalizing behavior had also included repetitive behaviors (code 9), which is one of the diagnostic features of autism. These could not be removed from studies' measures.

TABLE 4 Combined expressive and receptive language and behaviors of concern

Exposure: *Combined expressive & receptive language*											
				Risk of bias							
Outcome		**Author, year**	**N**	**Selection**	**Comparability**	**Outcome**	**Total (9)**	**Test statistic**	**Direction of effect**	***p*-value**	**GRADE**
Self-injurious behavior		Lawrence, 2017	145	★★	–	–	2	$r = -0.345$	↓	<0.01	⨁⨁⊖⊖ Low
		Rattaz et al., 2015	97	★★★	–	–	3	OR = 0.8	↓	<0.04	
		Duerden et al., 2012	250	★★	★	–	3	$\beta = -0.087$	↓	0.41	
		Makrygianni & Reed, 2010	86	★★★	–	–	3	$r = -0.007$	↓	>0.05	
		Matson & Rivet, 2008	298	★★	–	–	2	$\beta = -0.09$	↓	>0.05	
		Baghdadli et al., 2003	222	★	★	★	3	N.R.	N.R.	>0.05	
		Matson et al., 2009	168	★★	–	–	2	$r = 0.42$	↑	<0.05	
Aggression		Mazurek et al., 2013	1584	★★★	–	–	3	$d = -0.18$	↓	<0.001	⨁⨁⊖⊖ Low
		Estes et al., 2007	57	★★★	–	–	3	N.R.	↓	>0.05	
		Maddox et al., 2018	182	★★	★	–	3	Adjusted $R^2 = 0.018$	↓	>0.05	
		Matson & Rivet, 2008	298	★★	–	–	2	$\beta = -0.21$	↓	>0.05	
		Uljarevic et al., 2018	107	★★	–	–	2	$r = -0.030$	↓	>0.05	
		Kelley, 2006	15	★★	–	–	2	N.R.	N.R.	>0.05	
		Lawrence, 2017	145	★★	–	–	2	$r = 0.062$	↑	>0.05	
		Matson et al., 2009	168	★★	–	–	2	$r = 0.94$	↑	<0.05	
Externalizing behavior	Code										
	3, 4, 5	Boonen et al., 2014	206	★★	★	–	3	$\beta = 0.02$	N.R.	0.778	⨁⨁⊖⊖ Low
	1, 2, 7	Uljarevic et al., 2018	107	★★	–	–	2	$r = 0.176$	↑	>0.05	
	1, 3	Neuhaus et al., 2022	145	★★	★	–	3	$\beta = -0.34$	↓	>0.05	
	1, 3, 5, 6, 7	Estes et al., 2007	57	★★★	–	–	3	N.R.	↓	<0.05	
	1, 3, 5, 6, 7	Lawrence, 2017	145	★★	–	–	2	$r = 0.013$	↑	>0.05	
	1, 3, 6, 8, 9	Matson et al., 2009	168	★★	–	–	2	$R_c = 0.56$	↑	<0.01	
	8, 9	Matson & Rivet, 2008	298	★★	–	–	2	$\beta = -0.06$	↓	>0.05	

Note: ↓/negative effect size = more language ability, less behavior of concern; ↑/positive effect size = more language ability, more behavior of concern; ↓/↑: both negative and positive effect sizes reported in study; N.R.: not reported. ***Code***: 1: aggression, 2: attention problems, 3: conduct — tantrums, bullies others, steals, lies, disobedient to authority, 4: hyperactivity, 5: tantrums, 6: SIB, 7: unstable mood, 8: inappropriate behavior, 9: repetitive behavior. Composite scores measuring externalizing behavior had also included repetitive behaviors (code 9), which is one of the diagnostic features of autism. These could not be removed from studies' measure.

Additionally, some studies used crude measures of language and BoC. For instance, they reported on the presence or absence of structural language difficulties or whether the Autistic individual had BoC based on the caregiver's response to one or two questions. Few studies used standardized clinician-administered measures. We grouped studies based on the measures used to assess language and behavior to see if the quality and comprehensiveness of the measure made a difference to the findings, however we did not see patterns in the findings based on the measure type. Furthermore, the type of measure used was accounted for in our risk of bias analysis and assessments of certainty of evidence.

Externalizing behavior includes multiple types of behavior (aggression, tantrums, SIB, inappropriate behavior amongst others), and some tools from the included evidence provide a composite score of these BoCs. As such, it was not possible to separate each behavior construct. Other studies have suggested that the association between expressive language and externalizing behavior may be driven predominantly by SIB (McClintock et al., 2003), which is consistent with our findings on the relationship between SIB and expressive language. Alternatively, another consideration is that expressive language ability has associations with other BoCs, such as aggression, tantrums, or socially inappropriate behavior that are more nuanced and yet to be elucidated, particularly given a number of studies used blunt measures of language and BoC. Similarly, expressive and receptive language were not always available as separate constructs, and it is not surprising that an overall language score may underestimate a true association of one key language type.

The small number of studies that had included covariates in their analyses meant it was not possible to draw conclusions about the contribution of language to BoC relative to other important factors such as autism symptoms and intellectual disability. Similarly, few studies had investigated the associations longitudinally so information about the change in language and temporal association with a change in behavior could not be reported. This review is therefore also limited in drawing conclusions around causality. Generalisability of findings to adults is also limited, as the mean age of participants from the included evidence was 9.2 years and only 12 studies included adult (>18 years) participants. Finally, authors of some studies failed to report findings that were not statistically significant which introduces the potential for reporting bias.

Strengths and limitations of the review

We followed best practice guidelines such as PRISMA (Page et al., 2021) and SWiM (Campbell et al., 2020), in addition to searching a range of databases to identify studies. Data was screened and extracted by two people to minimize the chance of errors. We also extracted types of language and behavior from the evidence in identified studies separately which provides more specificity of associations. We completed assessments of risk of bias and judged the overall certainty of evidence so that the findings can be interpreted in the context of study quality of type of evidence. However, we were unable to complete a meta-analysis as planned in the protocol, as described above which means an overall finding and effect size could not be reported.

Implications of results

While findings of an association between language and BoCs in autism have long been reported and are not unexpected, this review provided novel findings about the association between different modalities of language and BoCs. It would be expected that BoC results at least in part from expressive language difficulties rather than vice versa even though causation cannot be established. As such, this review highlights the importance of identifying expressive language difficulties early. If language abilities can be improved through targeted interventions, supports and environmental modification, it may be expected there will be secondary benefits for behavior. This in turn could improve functioning and participation (Anderson et al., 2007; Baghdadli et al., 2007; Hudry et al., 2010) and reduce harm and stress to the individual and their family.

With more consistent use of high quality, detailed and standardized measures of language and BoC, analyses that adjust for important factors that may influence the association, and data of a temporal association between language gain and BoC reduction, we will have high-quality evidence that can inform intervention and support, and inform preventive approaches. Service embedded trials should be the next step to provide the type of evidence that is needed about the effectiveness of improving communication, using language, augmentative and alternative communication or other approaches, prevent BoCs emerging and reduce their persistence.

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DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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