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Intervention effects on spoken-language outcomes for children with autism: a systematic review and meta-analysis

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Abstract

Background Although spoken-language deficits are not core to an autism spectrum disorder (ASD) diagnosis, many children with ASD do present with delays in this area. Previous meta-analyses have assessed the effects of intervention on reducing autism symptomatology, but have not determined if intervention improves spoken language. This analysis examines the effects of early interventions on spoken-language in children with ASD.
Method A systematic review of 1756 studies of children with ASD who participated in early intervention resulted in the inclusion of 26 studies in the current review. These studies included 1738 participants with ASD who were, on average, 3.3 years old ($SD = 0.91$).
Results This random-effects meta-analysis of spoken-language outcomes for children with ASD who received early intervention as compared with usual treatments yielded a significant overall mean effect size of $g = 0.26$ (CI = 0.11 to 0.42). On average, children with ASD significantly increased their use of spoken-language following experimental early interventions. Treatments delivered simultaneously by a clinician and a parent resulted in greater gains in spoken-language than treatments delivered by a clinician or parent only. No other participant or study characteristics predicted individual-study effect sizes.
Conclusions Early intervention improves spoken-language outcomes for children with ASD, and the largest effects are found when both parent and clinician implement the intervention. Recommendations for practice include adding systematic parent training to interventions for spoken language to potentially improve outcomes. Future research should report standard language measures as well as child (cognitive ability and socio-economic status) and intervention characteristics to improve evidence related to the effects of interventions on spoken communication in children with ASD.

Keywords autism, early intervention, language, meta-analysis, parents, systematic review

Early intervention effects on spoken-language outcomes for children with autism: a systematic review and meta-analysis

Autism spectrum disorders (ASD) are defined by a significant delay in social communication and by the presence of repetitive behaviours and restricted

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interests (American Psychiatric Association, 2013). The core deficits of ASD, by definition, do not specify deficits in spoken-language development. Yet, 50% of children with ASD do not develop useful speech by age three (Tager-Flusberg and Kasari, 2013), and at least 30% of all children with ASD will remain minimally verbal following early interventions (Anderson *et al.*, 2007). It is critical to better understand the effects of early interventions that target spoken-language given that developing useful speech by age 5 is a significant predictor of long-term language and communication outcomes for children with ASD (Howlin, 2005; Tager-Flusberg and Kasari, 2013).

Predictors of long-term spoken-language outcomes for children with ASD include the following: initiating joint attention, receptive language, object interest and imitation skills (Smith *et al.*, 2007; Thurm *et al.*, 2006; Toth *et al.*, 2006; Watt *et al.*, 2006). Additionally, specifically teaching play and joint attention skills has resulted in better language outcomes (Kasari *et al.*, 2008). Given the range of skills associated with spoken-language development, the majority of interventions have taken one of two approaches to improving spoken-language: targeted or comprehensive. A *targeted* approach focusses on teaching pre-linguistic and communication skills. Interventions that specifically teach speech, receptive language, productive language, pre-linguistic communication or social use of communication may be considered targeted.

Comprehensive interventions teach a broad set of skills that are directly or indirectly related to spoken-language development. These may include cognitive skills, motor skills, self-help skills, play, imitation, receptive language and limited productive language skills. Comprehensive interventions tend to be more time intensive because of the increased number of goals and outcomes (Guifang *et al.*, 2004; Rogers & Vismara, 2008). For the current review, comprehensive interventions included any intervention package that focussed on spoken-language outcomes in addition to outcomes in at least one other domain. While the scope of the approaches differ, both comprehensive and targeted intervention packages often include parent training (Rogers & Vismara, 2008).

Parent training

A review of expressive language in children with language delays, who did not have ASD, identified limited evidence for spoken-language outcomes (Law *et al.*, 2004). However, there was no significant difference in total child language outcomes between parent-implemented and clinician-implemented interventions, suggesting that perhaps parent-delivered interventions may be just as effective as clinician-delivered interventions. A 2011 review of 18 studies (three of which included participants with ASD), found parent-training language interventions significantly improved expressive language, vocabulary and syntax for children with language impairment (Roberts & Kaiser, 2011). The findings of both meta-analyses of studies enrolling children with language impairment, support the effectiveness of parent-implemented treatments for improving spoken-language outcomes (Law *et al.*, 2004; Roberts & Kaiser, 2011).

Comprehensive early interventions for children with autism spectrum disorder

Meta-analyses of interventions for children with ASD indicate that there is some evidence that specific interventions improve language outcomes. A meta-analysis of 14 studies examining Early Intensive Behavioural Intervention (Reichow *et al.*, 2012) identified reductions in ASD symptomatology and improved overall language outcomes for children younger than 7 as compared with other treatments (Reichow & Wolery, 2009). Additionally, a recent Cochrane review of five studies identified Early Intensive Behavioural Intervention as an effective intervention programme for young children with ASD for improving communication as compared with children enrolled in a special education setting, although the studies in this review were of relatively low quality (Reichow *et al.*, 2012). Another review of 13 studies estimated that the Treatment and Education of Autistic and Related Communication Handicapped Children intervention programme had small ($g = 0.32$; −0.09–0.73), but non-significant, positive effects on verbal outcomes compared with usual treatments (Virues-Ortega *et al.*, 2013). Results in this review indicated that effects did not vary based on dose, length of intervention, age or setting. To date, the extent to which effects on spoken language vary based on different intervention components has not been explored.

The objective of the current study was to conduct a systematic review and meta-analysis to determine the effects of early interventions for children with ASD on spoken-language outcomes. This study addresses the following questions: (a) do early interventions improve spoken-language abilities in young children with ASD as compared with usual treatments? (b) does intervention dosage moderate the relationship between treatment and spoken-language outcomes? (c) does the addition of parent training moderate spoken-language outcomes? and (d) do the study characteristics including number of indicators of risk of bias, age of participants or type of intervention focus (targeted or comprehensive), moderate the relationship between interventions and spoken-language outcomes?

Methods

Eligibility

Specific inclusion criteria for selecting studies were based on participants, intervention, comparison group, outcomes and study design characteristics (Table 1). Criteria for participants included children with ASD younger than 8 who spoke English. Criteria for intervention components included any behavioural intervention that did not include a pharmacological component. Criteria for outcomes included spoken-language as measured by (1) standardised measures ($M = 100$; $SD = 15$), words produced (2) during an observation or (3) as reported by a parent or teacher. All studies that included a comparison group that did not receive intervention were included; both, randomised control trials and quasi-experimental studies were included. Pre-post designs and programme evaluations without a control group were excluded.

Sources

Peer-reviewed publications, dissertations, conference proceedings and reports published in English were selected for review. Eleven databases were searched to capture publications of all types: PsycINFO, PsycArticles, ProQuest Central, PAIS International, Linguistics and Language Behaviour Abstracts, International Bibliography of the Social Sciences, ERIC and multiple ProQuest Dissertations and Theses databases. References of included articles were reviewed for possible inclusion as well as forward searching via Google Scholar. An attempt to include a variety of 'grey' literature was performed by reviewing references of included studies, forward searching relevant references and by searching multiple databases for dissertations and theses and conference proceedings.

Search

The search was conducted on 31 December 2014. The Participants, Interventions, Comparison, Outcomes and Study Design search terms, outlined in Table 1 according to inclusion criteria, were selected to identify a wide range of studies that might

Table 1 Study inclusion and search strategy

	Criteria	Search terms
Participants	Autism spectrum disorder, speak English, younger than 8	Abstract: auti* OR ASD OR PDD OR Aspergers
Intervention	Behaviour/developmental, No pharma component	intervention OR therapy OR teach* OR treat*
Comparison	Usual treatments	assign* OR "control group" OR BAU OR "wait list" OR RCT OR random* OR quasi OR "treatment group" OR "intervention group" OR "group design" OR (before AND after) OR trial))
Outcomes	Spoken language	speech OR verbalizations OR communicat* OR articulation OR language OR expressive OR ESCS OR talk OR speak OR "social interact*" OR "social function*" OR "joint engagement" OR "joint attention"
Study design	Any group design study	
Other	English journals	la.exact("ENG")

be eligible for inclusion in the study (Impellizzeri & Bizzini, 2012). A total of 1809 studies were identified following a review of the references. All databases were searched concurrently within ProQuest, resulting in automatic removal of the initial set of duplications. An additional 53 duplicates were identified, resulting in 1756 unique studies screened for eligibility.

Variables

Inter-observer agreement

Two doctoral students coded all studies. Data were managed using the REDCap electronic capture tools (Harris *et al.*, 2009). Both coders rated each study that met inclusion criteria. Across all rated items for all studies ($n = 1188$), average agreement on categorical variables during the initial coding was 88% and agreement on all calculations was 96%. All disagreements were resolved through consensus coding and verified by examining the manuscript of the study. Disagreements were because of (1) miscalculations; (2) unidentified construct; or (3) disagreement on the interpretation of the definition. In cases where the disagreement was because of the definition, the coders discussed the definition and re-scored items independently.

Summary measures

All study variable definitions are summarised in Table 2. The post-intervention outcome difference between groups was estimated using the standardised effect size and standard error with the Hedges' *g* adjustment (Hedges and Olkin, 1985; Shadish *et al.*, 2008). A standardised effect size metric was selected to allow comparison across different measure types and scales that were used. The conservative Hedges' *g* adjustment was used because of the small sample size studies that were included.

Risk of bias

Risk of bias was rated on eight indicators recommended by Cochrane Collaboration (Table 3; Higgins *et al.*, 2011). Fidelity of implementation was rated as an additional risk factor; this rating was based on the overall fidelity score for the intervention and adequacy of the fidelity measure. Studies were rated as (a) high risk; (b) low; or (c) unclear risk based on each of the eight indicators that contributed to the total risk of bias score (range of possible total scores for bias was 0–8; Table 3).

Analytic strategies

A random-weights, average mean-difference effect size was used to estimate the overall effect and standard error of early interventions on spoken-language outcomes for young children with ASD. Analyses were conducted in R-Studio running R version 3.1.2 using the 'meta' package (R Core Team, 2014; Schwarzer, 2007). A random-effects model was selected because of the variability inherent in the ASD population. The random-effects model assumes a distribution of true between-study variability; thus, the results of this analysis could be generalised to the larger ASD population.

Heterogeneity in the sample was estimated and examined using three methods. The Q statistic determines the heterogeneity in the sample of studies. The τ^2 estimates the distributions of the average effects between studies. The I^2 statistic estimates the proportion of heterogeneity that was true between-study variability that could be explained with study characteristics.

Publication bias was analysed to determine the distribution and influence of small sample studies using three techniques. First, a funnel plot was used with visual analysis of the distribution of effects, followed by a trim and fill analysis to determine the influence of potentially missing studies (Stuck *et al.*, 1998; Duval and Tweedie, 2000). Finally, a linear regression was conducted to test the hypothesis of small-study bias (Egger *et al.*, 1997).

Results

Study selection

Of the 1809 abstracts and titles reviewed, 324 were selected for review at the full-text level for inclusion. Studies were excluded after reviewing the full text because of: not including participants with ASD or participants under age 8 ($n = 139$), not having a group experimental design ($n = 62$), not including a behavioural intervention ($n = 14$) or not including a measure of spoken-language ($n = 61$). Of the remaining studies, 26 met all inclusion criteria for the current analysis. Spoken-language measures across

Table 2 Variable definitions

		Definition
	Measure type	Observational: a frequency or ratio measure based on a structured observation Report: a parent or teacher report Standardised: a standardised measure
	Implementer	Parent: the parent was taught to implement the intervention Clinician: a clinician implemented all of the intervention. Parents may be updated on progress, but no specific training is offered to the parents Parent plus clinician: parents were trained to implement some or all of the same components that the clinicians implement. This did not include a few minutes of a clinician modelling the intervention during parent training.
	Intervention goal	Targeted: an intervention that specifically aims to improve language, communication or speech Comprehensive: an intervention that seeks to improve more than one area of development including language, cognition or autism symptomatology
Intervention component	Discrete trial	Named 'discrete trial', 'direct teaching' or 'applied behaviour analysis teaching methods'
	Shaping	Specifically names shaping as a teaching method
	Prompting	Specifically names a prompting procedure such as 'least-to-most', 'most-to-least' or 'prompt fading'
	Imitation	Specifically mentions direct teaching of imitation skills
	Receptive language	Specifically mentions direct teaching of receptive language skills such as picture identification or following directions
	Play based	The intervention is described as being play based or having a play component
	Expansions	Language expansions are specifically mentioned in the intervention procedure
	Modelling	Language modelling as a specific intervention component at the child's target level or specific vocabulary targets
	Responsiveness	Responding or responsiveness was specifically mentioned as a component
	Incidental teaching	Specifically mentions incidental teaching or describes naturally embedded trials
	Time delay	Specifically mentions time delay strategies or describes eliciting communication through pausing or waiting

studies primarily included measures of spoken vocabulary, and children across studies were primarily entering intervention using limited language equivalent to first or second phase of language learning (Tager-Flusberg *et al.*, 2009).

Study characteristics

The 26 studies were published between 1980 and 2014. The studies included 1738 total participants with ASD who were, on average 3.33 years old ($SD = .91$, range = 1.75–4.18). Of the 21 studies that reported cognitive scores, participants across studies had scores indicative of comorbid intellectual disabilities (>1.5 *SD* below the norm). Consistent with ASD incidence rates, children were primarily male (81%). Most of the studies were conducted in the USA (69%) and used a randomised control trial design (62%). Most studies reported receptive language ability or cognitive ability (69%); however, some reported scores included age equivalent scores, which cannot be meaningfully summarised across studies (Table 4). Additionally, the lack of variability in cognitive skills

Table 3 Risk of bias scoring overview

Item	Definition	Scoring		
		0: Low Risk	**0.5: Unclear risk**	**1: High risk**
Equivalence*	Group equivalence established through post-assignment statistical testing	The study adequately demonstrated no significant differences between groups at pre-test	{Not rated for this item}	The study demonstrated significant differences favouring one group at pre-test
Selection bias*	The groups were selected from the same pool of recruited participants	Participant selection was clearly described and there is no evidence of differing selection across groups	The selection process is not well described	The participants were selected from different samples within the population
Performance bias*	The assessment coders were blind to group assignment	Clear description of coders who were blind to treatment group	No clear description of the coders and their knowledge of treatment group	It was indicated that coders were aware of treatment group assignments
Detection bias*	The assessment administrators were blind to group assignment	Clear description of assessors who were blind to treatment group	No clear description of the assessors and their knowledge of treatment group	It was indicated that assessors were aware of treatment group assignments
ITT*	The study uses an intent to treat analysis by analysing all participants as randomised	The study clearly describes or labels the analysis as intent-to-treat	The study does not clearly describe how missing data are handled	The study reports list-wise deletion of missing data or participants
Fidelity	The study measures fidelity using a sensitive measure that captures all relevant intervention components and reports a high level of overall implementation fidelity	The study reports a measure that is likely sensitive to all of the intervention components and reports a range of fidelity scores above 85%	The study reports a measure of fidelity that does not clearly capture all of the intervention components, reports a range that falls below 85% or the measure of fidelity is not well described	The study reports an average fidelity below 85% or a range below 75%, the study states that it did not measure implementation fidelity, or only reports for secondary implementers
Other bias*	The study reports one of the following additional indicators of bias: (1) non-adjustment of baseline differences; (2) contamination of intervention; (3) Statistical errors; (4) Low reliability or validity; or (5) conflict of interest	The study does not report any evidence of the five additional risk factors	The study does not adequately describe the components related to five additional risk factors such as possible contamination of intervention is not described, lack of evidence to assess statistical errors or assessment validity is not reported	The study demonstrates significant baseline differences that were not adjusted for, or contamination of intervention is evident, or unit of analysis errors or statistical analysis errors are reported, or low reliability or validity is reported, or a conflict of interest is present

*Adapted from Cochrane Review Risk of Bias indicators (Higgins, Altman, Gotzsche, Juni, Moher, Oxman, et al., 2011).

Table 4 Study description

		Age				Receptive			Cognitive				
Study	**N 1738**	**Mean**	**(SD) (range)**	**% Male**	**% Minority**	**Mean**	**(SD)**	**type**	**Mean**	**(SD)**	**type**	**Spoken-language measure type**	**Location**
Boyd et al. (2013)	183	4.01	(0.55)	83	54	28.47	(12.00)	Age	—	—	—	Standard	USA
Venker et al. (2012)	14	3.43	(0.86)	—	—	14.79	(7.04)	Standard	28.79	(13.80)	Age	Observation	USA
Salt et al., (2002)	17	3.41	(0.52)	82	—	—	—	—	44.21	(11.63)	Standard	Report	UK
Carter et al. (2011)	55	1.77	(0.22)	85	53	8.30	(4.96)	Age	—	(16.30)	Standard	Standard	USA
Tonge et al. (2014)	70	3.99	(0.33)	83	—	12.14	(18.51)	Raw	64.03	(27.96)	Standard	Standard	AU
Green et al., 2010	152	3.75	(2–5)	91	42	15.30	(9.75)	Raw	26.16	(9.75)	Standard	Standard	UK
Roberts et al. (2011)	84	3.57	(0.61)	91	—	—	—	—	62.32	(14.99)	Standard	Standard	AU
Rogers et al. (2012)	98	1.75	(0.86)	69	28	67.42	(72.03)	Standard	63.98	(16.50)	Standard	Standard	USA
Solomon et al. (2014)	112	4.18	(0.85)	82	25	51.81	(28.93)	Standard	33.88	(62.94)	Standard	Standard	USA
Siller, Hutman, & Sigman (2013)	65	4.75	(1.02)	91	72	17.02	(8)	Raw	25.64	(10.30)	Standard	Standard	USA
Bloch et al. (1980)	26	3.47	(0.58)	77	—	—	—	—	46.00	—	Standard	Observation	USA
Casenhiser et al. (2013)	51	3.71	(0.61)	—	—	—	—	—	35.29	(11.87)	Age	Observation	CN
Kasari et al. (2008)	57	3.55	(0.35)	79	35	19.87	(8.649)	Age	61.79	(18.84)	Standard	Standard	USA
Whalen et al. (2010)	47	—	[3-6]	—	—	—	—	—	—	—	—	Standard	USA
Hardan et al. (2014)	47	4.10	(1.55)	75	—	17.94	(6.866)	Raw	53.13	(15.65)	Standard	Observation	USA
Wetherby & Woods (2006)	35	2.60	(0.19)	83	37	—	—	—	—	—	Standard	Standard	USA
Schertz et al. (2013)	22	2.17	(0.31)	—	—	23.45	(6.73)	Raw	—	—	—	Standard	USA
Strain & Bovey (2011)	294	3.69	(0.37)	—	—	31.83	(8.156)	Raw	65.67	(8.13)	Standard	Standard	USA
Dawson et al. (2010)	45	1.95	(0.32)	71	27	21.15	(4.27)	—	60.25	(8.92)	Standard	Standard	USA
Drew et al. (2002)	24	1.91	(0.18)	79	—	52.50	(62.1)	Raw	77.05	(13.85)	Standard	Report	UK
Cohen et al. (2006)	39	2.64	(0.26)	83	—	52.19	(15.15)	Raw	60.53	(15.57)	Standard	Standard	USA
Vivanti et al. (2014)	57	3.42	(0.68)	88	—	40.57	(20.39)	Raw	51.12	(16.87)	Standard	Standard	USA
Goods et al. (2012)	11	3.38	(0.89)	—	51	11.48	(6.30)	Age	31.47	(12.64)	Standard	Standard	USA
Remington et al. (2007)	44	2.89	(0.32)	—	—	—	—	—	61.86	(16.53)	Standard	Standard	UK
Howard et al. (2005)	61	2.80	(1.01)	88	43	49.55	(16.26)	Standard	57.62	(16.06)	Standard	Standard	USA
Aldred, Green, & Adams (2004)	28	—	[2-5]	89	—	83.55	—	Raw	—	—	—	Report	UK

across studies limits our ability to use this as a moderator variable.

The interventions included direct teaching components (50%) and naturalistic teaching components (92%). Intervention implementers included (1) parents only (38%), (2) clinicians only (15%) or (3) both (47%). Most studies used a standardised measure of spoken-language (73%); the remaining studies measured spoken-language using observational (15%) or parent report (12%) measures. The specific intervention components that are described in each study are summarised in Table 5.

Risk of bias

Risk of bias was summarised as number of indicators of bias. The 26 included studies presented with an average four indicators of risk of bias (range: 0.5–7) out of eight possible indicators. The most common bias indicators were (a) a selection of comparison participants from a different source than intervention participants; (b) an analysis that did not use an intent-to-treat approach; (c) poorly measured or reported fidelity and (d) a quasi-experimental design (Table 6).

Publication bias

Visual analysis of the funnel plot for symmetry indicated little evidence of publication bias. One study (Aldred *et al.*, 2004), with a sample of only 28 participants and a very large effect size, $g = 1.57$, likely contributed to any implied bias in the plot. The trim and fill analysis indicated some small-study bias; six small studies were filled. However, the hypothesis of symmetry in the funnel plot could not be rejected in the Egger's linear regression test ($P = 0.662$). Thus, there was little evidence of small-study bias in the results and it is unlikely that publication bias influenced the results of the meta-analysis.

Synthesis of main effects

The random weights model estimated the overall effect of early interventions on spoken-language outcomes for young children with ASD as $g = 0.26$, 95% CI [0.11, 0.42; Fig. 1] or approximately four points on a standardised measure of language such as the *Preschool Language Scale- 5th edition* (Zimmerman *et al.*, 2011). The between-study variance was small ($\tau^2 = 0.083$), but this heterogeneity was significant [$Q = 59.08$ (25), $P = 0.0001$]. Of this heterogeneity, 57.7% was true variability (I^2) that can be explained by individual-study characteristics. Thus, moderator analyses were conducted to potentially explain some of the heterogeneity between studies.

Moderator analysis

Two meta-regression models and an analysis of variance (ANOVA) sub-group analysis were fit in an attempt to explain between-study variability within the overall analysis. During preliminary analyses, no systematic differences in outcomes between studies were found because of method of language outcome measures (standardised test, observational data or parent report) or year of publication. The first meta-regression included all 26 studies. The results indicated the total intervention dose ($\beta = 0.008$, SE = 0.010; total hours of intervention computed as length of treatment x hours per week), and number of indicators of bias ($\beta = 0.027$, SE = 0.027) did not significantly predict the magnitude of spoken-language outcomes. Because of the theoretically important impact that these variables might contribute to the overall model, they were retained as control variables in the second meta-regression. The results in the second meta-regression indicated the effect of targeted language interventions on spoken-language did not differ from the effect of comprehensive ASD interventions ($\beta = 0.029$, SE = 0.225). This analysis accounted for none of the heterogeneity between studies ($R^2 = 0.00\,\%$), further indicating that targeted and comprehensive interventions resulted in similar outcomes for spoken language.

The second meta-regression moderator analysis examined the impact of age of participants and included the same 26 studies and control variables. The null hypothesis could not be rejected: the effect of interventions on spoken-language for younger and older participants did not differ significantly ($\beta = -0.092$, SE = 0.096). This analysis accounted for none of the heterogeneity ($R^2 = 0.00\,\%$), indicating that interventions delivered at different ages resulted in similar outcomes.

The random effects ANOVA model for the sub-group analysis of implementers (clinician only, parent only or parent plus clinician) summarises the

Table 5 Intervention characteristics

Study	Named intervention	Implementer: Parents, clinicians or Both	Length			Intervention goal	Best description of the intervention	Parent training	Direct teaching		
			Total weeks	Hours/ week	PT total hours				Discrete Trial	Shaping	Prompt fading
Boyd et al. (2013)	LEAP and TEACH	Clinicians	52	30	0	C	B		X		X
Venker et al. (2012)	Hanen MTW	Parents	10	2.15	24	T	N	X		X	
Salt et al. (2002)	Scottish EIP	Both	45	4	0	C	N	X			
Carter et al. (2011)	Hanen MTW	Parent	14	1	14	T	N	X			
Tonge et al. (2014)	PSwA	Parents	20	1.25	25	C	B	X		X	X
Green et al. (2010)	PACT	Parents	52	0.75	36	T	N	X	X	X	
Roberts et al. (2011)	Parent training model	Both	40	2	40	C	N	X			
Rogers et al. (2012)	ESDM	Parents	12	1	12	C	B	X		X	X
Solomon et al. (2014)	PLAY	Both	52	0.61	36	T	N	X			
Siller, Hutman, & Sigman (2013)	Focused playtime (FPI)	Both	12	1.5	18	T	N	X			
Bloch et al. (1980)	Speech remediation	Both	52	1	—	T	D	X	X	X	
Casenhiser, Shanker, & Stieben (2013)	MEHRIT	Both	52	2	104	C	N	X		X	
Kasari et al. (2008)	JASPER	Clinicians	6	2.5	0	T	B		X	X	
Whalen et al. (2010)	Teach Town basics	Clinicians	14	3.3	0	C	D		X	X	X
Hardan et al. (2014)	PRT parent training	Parent	12	2.33	12	T	N	X			

Table 5. *(Continued)*

Study	Named intervention	Implementer: Parents, clinicians or Both	Length			Intervention goal	Best description of the intervention	Parent training	Direct teaching		
			Total weeks	Hours/ week	PT total hours				Discrete Trial	Shaping	Prompt fading
Wetherby & Woods (2006)	ESI	Parent	52	2	104	T	N	X			
Schertz et al. (2013)	JAML	Parents	16	1	16	T	N	X			
Strain & Bovey (2011)	LEAP	Both	104	17	0	C	N	X			X
Dawson et al. (2010)	ESDM	Both	104	20	24	C	B	X		X	X
Drew et al. (2002)	Parent training	Parents	52	0.5	27	T	N	X			
Cohen et al. (2006)	EIBI	Both	52	40	70	C	D	X	X	X	X
Vivanti et al. (2014)	Group ESDM	Both	54	20	0	C	B	X			
Goods et al. (2012)	JASPER	Clinicians	12	1	0	T	N				
Remington et al. (2007)	EIBI	Both	52	25.6	—	C	D	X	X	X	X
Howard et al. (2005)	Behaviour-analytic	Both	63	30	21	C	D	X	X	X	
Aldred, Green, & Adams (2004)	PACT	Parent	52	0.5	26	T	N	X			

Table 5 Intervention characteristics

	Direct teaching			Naturalistic					
Study	**Imitation tasks**	**Matching tasks**	**Receptive language**	**Play-based**	**Expansions**	**Modelling language**	**Responsive**	**Incidental teaching**	**Time Delay**
Boyd et al. (2013)				×				×	
Venker et al. (2012)				×	×	×	×	×	×
Salt et al. (2002)				×					
Carter et al. (2011)				×			×		
Tonge et al. (2014)				×					
Green et al. (2010)				×	×		×	×	
Roberts et al. (2011)				×				×	
Rogers et al. (2012)				×		×	×	×	
Solomon et al. (2014)				×		×	×	×	×
Siller, Hutman, & Sigman (2013)				×		×	×		×
Bloch et al. (1980)						×	×		×
Casenhiser, Shanker, & Stieben (2013)				×				×	
Kasari et al. (2008)				×	×	×	×		
Whalen et al. (2010)	×	×	×						
Hardan et al. (2014)				×				×	

Table 5. *(Continued)*

	Direct teaching			Naturalistic					
Study	Imitation tasks	Matching tasks	Receptive language	Play-based	Expansions	Modelling language	Responsive	Incidental teaching	Time Delay
Wetherby & Woods (2006)				X		X	X	X	X
Schertz et al. (2013)				X	X	X	X		
Strain & Bovey (2011)				X				X	X
Dawson et al. (2010)				X			X	X	
Drew et al. (2002)				X		X	X		X
Cohen et al. (2006)	X		X					X	
Vivanti et al. (2014)		X	X	X			X	X	
Goods et al. (2012)				X					
Remington et al. (2007)									
Howard et al. (2005)								X	
Aldred, Green, & Adams (2004)				X		X	X	X	

[Note]T, Targeted intervention; C, Comprehensive intervention; D, Direct teaching; N, Naturalistic; B, Both naturalistic and direct teaching components. EIBI, Early Intensive Behavioural Intervention; EIP, Early Intervention Preschool; ESDM, Early Start Denver Model; JAML, Joint Attention Mediated Learning; JASPER, Joint Attention; Structured Play Engagement; and Regulation; LEAP, Learning Experiences and Alternative Program; MEHRIT, Milton and Ethel Harris Research Initiative Treatment; MTW, More Than Words; PACT, Pediatric Autism and Communication Therapy; PLAY, Play and Language for Autistic Youngsters; PRT, Pivotal Response Training; TEACCH, Treatment and Education of Autistic and Related Communication Handicapped Children.

Table 6 Risk of Bias

Study	Design	Group equivalence	Selection bias	Performance bias	Detection bias	ITT	Fidelity	Other bias	Number of risk factors
Boyd et al. (2013)	Quasi	Equal	High risk	High risk	High risk	No	Low risk	unclear risk	5.5
Venker et al. (2012)	RCT	+ Control	Unclear risk	High risk	High risk	Yes	Unclear risk	High risk	5
Salt et al. (2002)	Quasi	+ Control	High risk	Unclear risk	Unclear risk	No	High risk	High risk	7
Carter et al. (2011)	RCT	Equal	Low risk	Unclear risk	Low risk	Yes	High risk	Low risk	1.5
Tonge et al. (2014)	RCT	+ Control	High risk	Low risk	Unclear risk	No	High risk	High risk	5.5
Green et al. (2010)	RCT	Equal	Low risk	Low risk	Low risk	Yes	Unclear risk	Low risk	1.5
Roberts et al. (2011)	Quasi	Equal	Low risk	Low risk	Low risk	No	High risk	Low risk	3
Rogers et al. (2012)	RCT	Equal	Low risk	Unclear risk	Low risk	Yes	High risk	Low risk	1.5
Solomon et al. (2014)	RCT	Equal	Low risk	Low risk	Low risk	Yes	Unclear risk	Low risk	0.5
Siller, Hutman, & Sigman (2013)	RCT	Equal	Low risk	Low risk	Low risk	Yes	Unclear risk	Low risk	0.5
Bloch et al. (1980)	Quasi	Unclear	High risk	Unclear risk	Unclear risk	Yes	High risk	Unclear risk	5
Casenhiser, Shanker, & Stieben (2013)	RCT	Equal	Low risk	Low risk	Low risk	No	High risk	Low risk	2
Kasari et al. (2008)	RCT	Equal	Low risk	Unclear risk	Low risk	Yes	Unclear risk	Low risk	1
Whalen et al. (2010)	RCT	Equal	Unclear risk	Unclear risk	Unclear risk	Yes	High risk	Unclear risk	3
Hardan et al. (2014)	RCT	Equal	Low risk	Low risk	Low risk	No	Unclear risk	Low risk	0.5
Wetherby & Woods (2006)	Quasi	Unclear	Unclear risk	Unclear risk	Unclear risk	No	High risk	High risk	6
Schertz et al. (2013)	Quasi	Matched	Low risk	High risk	Low risk	No	High risk	Low risk	4.5
Strain & Bovey (2011)	RCT	Equal	Low risk	Unclear risk	Unclear risk	No	High risk	Low risk	2
Dawson et al. (2010)	RCT	Equal	Low risk	Low risk	Unclear risk	No	High risk	Low risk	2.5
Drew et al. (2002)	RCT	+ Intervention	Low risk	Unclear risk	Unclear risk	Yes	High risk	Low risk	3
Cohen et al. (2006)	Quasi	Equal*	High risk	Low risk	Low risk	No	High risk	Low risk	4
Vivanti et al. (2014)	Quasi	Equal	High risk	High risk	Unclear risk	No	High risk	Low risk	5.5
Goods et al. (2012)	RCT	+ Intervention	Low risk	Low risk	Low risk	No	Unclear risk	Low risk	2.5
Remington et al. (2007)	Quasi	+ Control	High risk	Low risk	Low risk	No	Unclear risk	Unclear risk	5
Howard et al. (2005)	Quasi	Equal	High risk	Low risk	Low risk	No	Unclear risk	Low risk	3.5
Aldred, Green, & Adams (2004)	RCT	Equal	Low risk	Low risk	Low risk	Yes	Unclear risk	Low risk	0.5

*Minor differences in demographic data, no clear bias. RCT, randomised controlled trial. Quasi, Quasi-experimental.

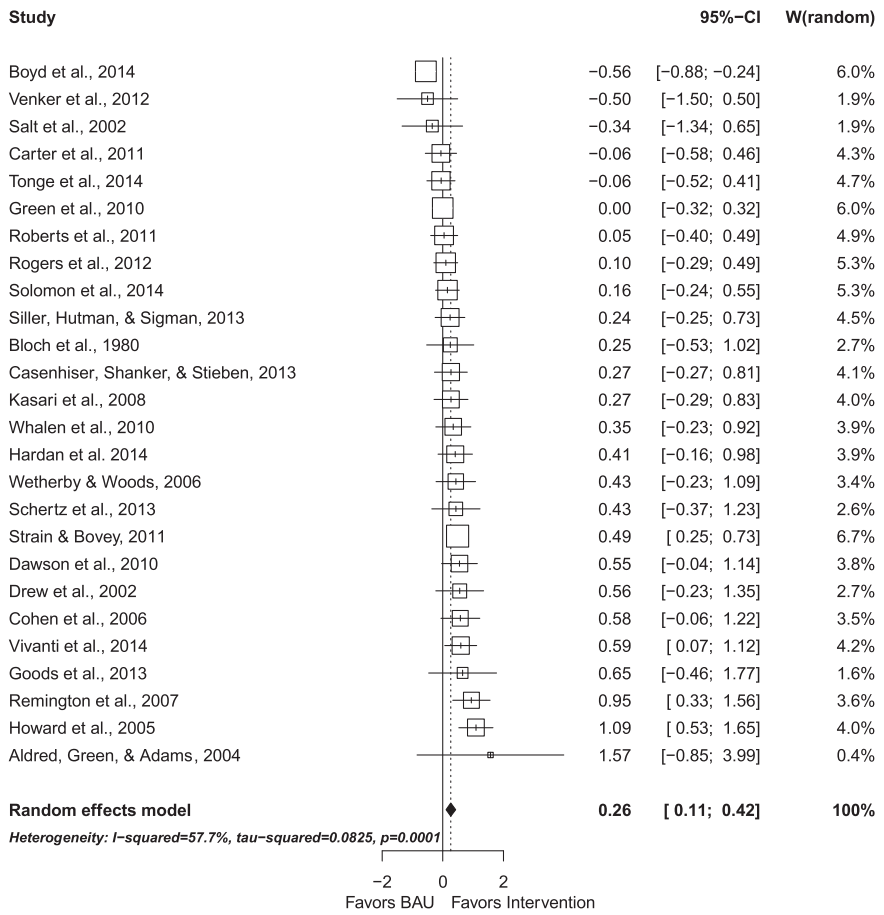

Figure 1 Effects of early autism interventions on spoken language.

outcomes within types of implementers (Fig. 2). There was a significant difference among the sub-groups [$Q = 59.08$ (25), $P < 0.001$]. None of the heterogeneity was explained within the parent-only group, the parent plus clinician group explained 36.4%, and 77.1% was explained by the clinician-only group. The sub-group analysis indicated a significantly better effect on language outcomes for parent plus clinician delivered interventions ($g = 0.42$) as compared with parent-only ($g = 0.11$) or clinician-only ($g = 0.08$) delivered interventions.

Discussion

The results of this meta-analysis demonstrate that, on average, early interventions for young children with ASD result in modest improvements in spoken language immediately following intervention, equivalent to children obtaining four additional points on a standardised language measure. Additionally, children who received intervention delivered by a parent plus clinician gained the equivalent of six more points than children receiving community interventions. This finding suggests that adding a parent-implemented component to clinician-delivered intervention further contributes to language gains. Changes in other features of intervention (i.e. total dosage of the intervention, targeted vs. comprehensive intervention approach) may not systematically contribute to language gains.

Although the results indicating average positive spoken-language outcomes across interventions appear promising, the magnitude of overall effect size is relatively small ($g = 0.26$). The effects may not be clinically meaningful considering that the average effect is within the standard error of measurement.

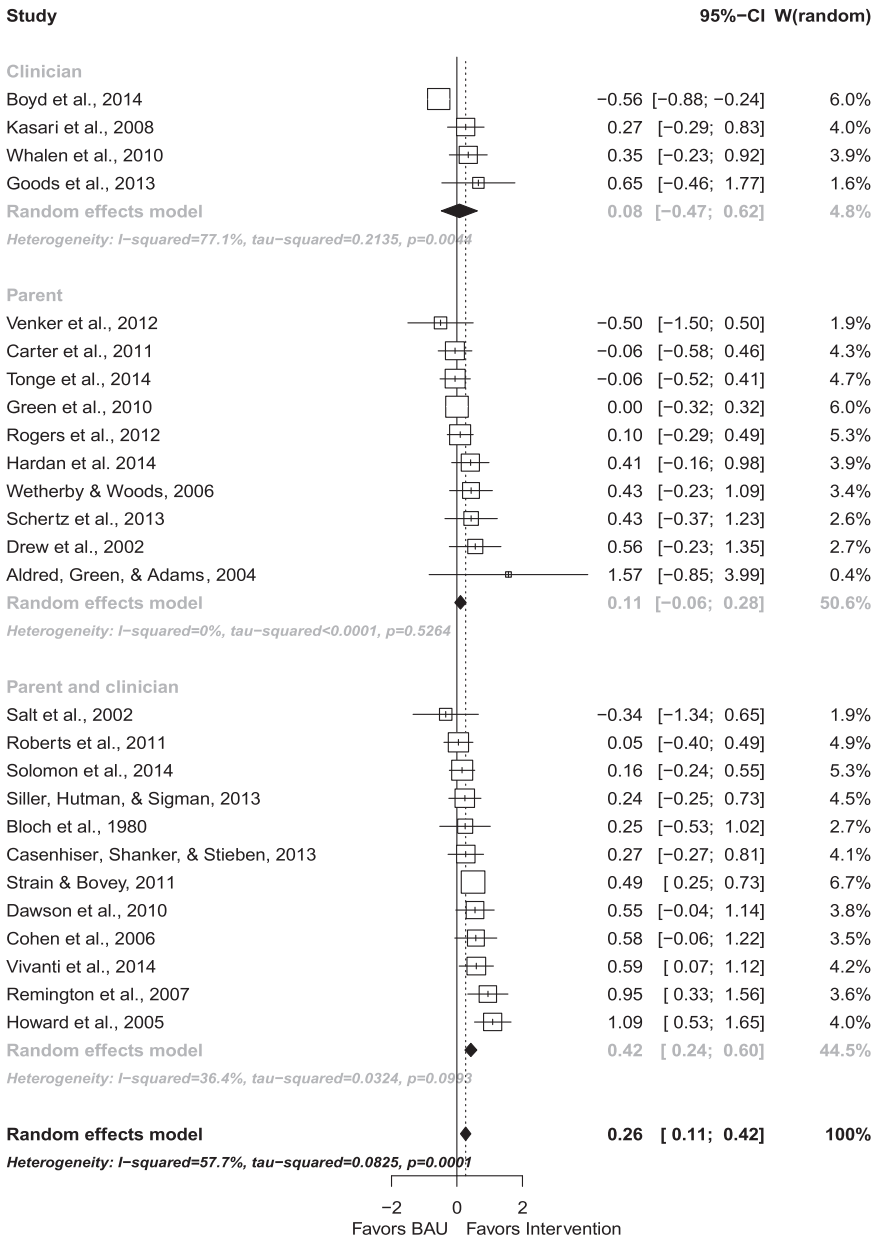

Figure 2 Effects of early autism interventions on spoken language by implementer.

Additionally, the improvement does not indicate movement beyond using single words, the second phase of language learning (Tager-Flusberg *et al.*, 2009). Because the results summarised in this meta-analysis were outcomes measured immediately after intervention, it is unclear if the difference between groups might have continued to increase over time or if the relatively modest overall effects would eventually diminish.

Intervention dosage and approach

Dosage did not predict better spoken-language outcomes. This finding was somewhat surprising

given the variability in dosage; however, these results should be considered within the context of variability of other intervention components (parent training and fidelity of treatment) across studies that may have mitigated the effect of any one component. Intervention type (targeted vs. comprehensive) also did not explain variation in the results; however, it was not possible to determine the exact dose of language-related intervention provided within comprehensive interventions. Further, true dosage was difficult to ascertain as it is unclear how many hours parents implemented the intervention with their children.

Parent training

For children with ASD, interventions implemented by both parents and clinicians together may be especially important for three reasons. Firstly, parents may benefit from the clinician modelling the intervention with their children. Learning from models is an adult learning strategy that may result in better parent fidelity of intervention and ultimately a higher dose of intervention outside clinical environments (e.g. Dunst, Trivette, & Hamby, 2007; Roberts *et al.*, 2014). Secondly, children with ASD may benefit when there is a consistent language teaching strategy across communication partners that supports generalisation of skills. Children with ASD often require systematic teaching across communication partners and settings to generalise skills (Plaisted, 2001). Additionally, parent buy-in might be greater and parent stress might be reduced when the clinician and parent are co-interventionists and parents can observe improvements in their child while the clinician implements the intervention (Kaiser et al., in press; Kaiser and Roberts, 2013). Reductions in parent stress may result in positive child outcomes through direct and indirect paths. Greater parent buy-in is likely to increase parent involvement in behavioural interventions, and therefore may result in greater dose delivered by the parent (Carr *et al.*, 2015; Kasari & Sigman, 1997; Solish and Perry, 2008).

Limitations

This review has several limitations. Firstly, a meta-analysis is limited by descriptive and outcome data available in the individual studies selected for review. The literature review did not include studies that did not report spoken-language outcomes. In the excluded studies, it is unknown if measures of spoken-language were not included because they were not conceptually important in the study or if the measures were collected but not reported because of lack of significant outcomes. Although little small-study bias was indicated in the analysis, the review may have missed studies with smaller samples because of publication bias (Ferguson and Brannick, 2012).

Secondly, variable and eclectic treatments are commonly delivered to children with ASD in the community: therefore, the comparison samples most likely vary across studies (Stahmer, 2007). Reporting the number of hours of intervention received by children in the control group and characterising the quality of the community intervention are critical for interpreting the results of experimental intervention vs. control studies. Children in the intervention groups may have benefited from the combination of experimental and community treatments or may have received more total hours of intervention than children in the control groups.

Thirdly, spoken vocabulary was the primary outcome measure across studies. Vocabulary does not account for other important features of spoken language such as grammar, phonology and rate of social communication. Although other features of language use may be accounted for in standardised measures of language, the early items of these measures are primarily vocabulary based (Tager-Flusberg *et al.* 2009). Future research should make a better effort to include multiple measures of use and complexity of spoken language in addition to vocabulary to best represent spoken-language growth in this population.

Finally, the findings of this meta-analysis should be interpreted cautiously because many studies did not measure child characteristics at the beginning of intervention (e.g. receptive language, speech skills, play skills, cognition or socio-economic status) or important aspects of intervention delivery (e.g. fidelity, parent generalised use of the intervention and clinician level of expertise), resulting in a limited set of moderators for investigation. In this set of studies, there was limited reporting and low variability in key constructs such as intellectual disability (ID). This meta-analysis is not able to speak to the impact of ID on spoken-language outcomes in children with ASD

because of the low variability in ID across studies. A more complete description of the specific phenotypic characteristics of the participants with ASD is critical for better understanding, which intervention strategies are most effective for each ASD profile (Blacher & Christensen, 2011). Making information about secondary outcome variables, fidelity measures, clinician expertise and training, standard scores for use in moderator analyses and dosage of specific intervention components available in online appendices or other resources linked to publications is important for future studies and would allow for more complete meta-analyses. The current analysis is limited by the limitations of the studies that were included, and the explanations of variability and heterogeneity within studies are limited. Thus, the results of this review must be considered in the context of the available measures of child-level characteristics and the limited descriptions of the interventions.

Recommendations

Practice

Three recommendations for practice can be offered as a result of this review. Firstly, both targeted and comprehensive approaches to early interventions may increase spoken-language of children with ASD. Secondly, early interventions should include parent training in addition to direct interventions by a clinician. Although the results of this meta-analysis indicated immediate improved outcomes for interventions implemented by the clinician plus parent, at least one study has indicated that children may continue to improve over time when parents are trained (Kaiser & Roberts, 2011). Thirdly, because of the overall modest outcomes for spoken language, long-term intervention may be needed for children with ASD to become fluent language users. Adaptive interventions that monitor progress and tailor the intervention to fit the child's response to treatment should be considered, especially for children who begin with minimal verbal skills (Kasari et al, 2014).

Future research

More consistent and systematic reporting of measures in early intervention studies is needed. When age-equivalent scores are reported, standardised scores should also be reported or made available to allow for moderator analyses in future meta-analytic reviews. Additionally, studies that include parents should describe how and what the parent was taught, the fidelity of the parent training, process and fidelity of, the parent's implementation of the intervention with the child. Measures of parent buy-in of the parent–clinician relationship should be reported to advance understanding of factors affecting parent-clinician collaborative intervention delivery (Carr *et al.*, 2015). Training and background of the interventionists are also important. The specific credentials of clinicians (e.g. speech language pathologists, behaviour analysts or research staff), level of education and training and the fidelity interventionist implementation should be reported. Finally, for future meta-analysts to be most successful, sub-groups within individual studies should be reported to best determine for whom, and under what conditions interventions are effective. For example, studies should include sub-groups of participants with and without ID to best understand the role of ID on spoken-language outcomes within the ASD population.

Conclusions

Early interventions for children with ASD resulted in improved spoken-language outcomes as compared with outcomes for children who received usual community treatments. The average gains of $g = .26$ (four standardised points) may not be sufficient to substantially improve functional language use for all children with ASD. Given the heterogeneity of children with ASD and the observed variability in outcomes, it is important to continue efforts to determine which children benefit most and least from early language interventions and to tailor interventions to specific characteristics of children within this population.

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