Review Article

Educational, Behavioral, and Social Interventions for Autistic Children, with Particular Reference to those at the more Severe End of the Spectrum: An Overview of Systematic Reviews

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Abstract

This paper presents an analysis of the available systematic reviews of educational, behavioral, and social interventions for children on the autism spectrum. Forty-six out of 1299 full-text articles were assessed against eligibility criteria, with a further 16 articles excluded for different reasons. The quality of the remaining 30 reviews was variable, and a further five were excluded due to low methodological quality. In the remaining 25 systematic reviews, none of the intervention types was found superior to the others, and there is generally weak evidence for the effectiveness of the reviewed interventions in improving autism-related impairments. Applicability of this evidence on children with severe autism is generally questionable. While there have been repeated calls for more large-scale studies, specifically randomized controlled trials, we conclude that the environments in which interventions typically take place are not conducive to this. Instead, researchers are advised to adopt sensitive, evidence-based approaches that work well with small sample groups.

Keywords: Applied behavior analysis, children, communication, computer-based learning, education, early intensive behavioral intervention, interventions, picture exchange communication system, severe autism, social stories, treatment and education of autistic and communication physically challenged children

Introduction

This paper reports findings from an overview of systematic reviews concerning educational, behavioral, and social interventions for children with autism. In the context of our wider research, we were particularly interested in interventions for children at the more severe end of the spectrum, referred to as "level 3" in The American Psychiatric Association's Diagnostic and Statistical Manual of Mental Disorders (DSM-5, 2013).[1] DSM-5 describes individuals as requiring "very substantial support" because of their severe deficits in social communication and obsessed repetitive behavior that "markedly interfere with functioning in all spheres."

An initial scoping search of the literature using the OVIDSP and ERIC databases using the keywords "severe autism" and "systematic review" did not identify any systematic reviews specifically targeting severely disabled autistic children. There were, however, some studies which included, among others, participants at the more severe end of the spectrum.[2,3]

Therefore, it was decided to broaden the search to cover systematic reviews and meta-analyses that have investigated children within the whole spectrum of autism, then to look specifically for participants with severe disabilities within the selected studies of these reviews. This approach was expected to reveal the relevant body of evidence on the effectiveness of various interventions for children with autism in general, and those at the severe end of the spectrum in particular.

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A note on language

We acknowledge the recommendations by Kenny *et al*. regarding the use of language when writing about autism.[4] We have followed these in our own writing, however when referring to findings from systematic reviews we have adopted the relevant authors' terminology to avoid confusion.

Research questions

We conducted an overview and analysis of the available systematic reviews of educational, behavioral, and social approaches for autistic children to answer the following three questions:

1. How effective are the different educational and behavioral intervention in improving the difficulties of autistic children?
2. How strong is the evidence for the effectiveness of these interventions, or of one intervention compared to others?
3. How applicable is the available evidence to children at the more severe end of the spectrum?

Scope

The interventions which were included in this overview are the mainstream educational approaches which are listed below, regardless of the setting (school, care unit, or home) in which they are delivered:[5-7]

- Applied behavior analysis (ABA) and early intensive behavioral intervention (EIBI)
- Computer-based intervention (CBI) including computer-based learning (CBL)
- Picture exchange communication system (PECS)
- Social skills interventions including social stories
- Treatment and education of autistic and communication physically challenged children (TEACCH) program.

Methods

Criteria for considering reviews for inclusion

For a review to be included in this overview, it had to be a systematic review or meta-analysis of experiments or trials investigating the response of children with autism toward an educational, behavioral, or social intervention. Narrative reviews and opinion articles with no *a priori* design, or those containing an extensive search of literature were excluded, in line with the recommendation by Higgins and Green.[8] Pharmacological, nutritional, or clinical interventions were excluded also.

The procedure for inclusion was as follows:

1. The first author conducted database searches (see below for criteria)
2. From these results, the first author screened papers based on their titles and abstracts, for inclusion in the eligibility test
3. The first and the fifth author applied, independently, the eligibility criteria to the selected papers
4. Eligible reviews were assessed on methodological quality using the 11-item AMSTAR tool (NCCMT, 2011)[9]
5. Reviews with an AMSTAR score below five were deemed of too low quality and excluded from the final synthesis of evidence, in accordance with Cheung *et al*.[10]

Search Methods For Identification Of Reviews

A broad search strategy, with no time or language limits, with a combination of the following entries was conducted:

([AUTIS*] AND [CHILDREN] [REVIEW]) AND ([EDUCATION*] OR [PECS OR TEACCH OR ABA OR INTERVENTION]).

These keywords were used as Medical Subject Headings, which is a comprehensive controlled vocabulary for the purpose of indexing journal articles and books. This broad search strategy was chosen in order not to miss any potentially relevant literature. Several databases of medical and educational literature were identified as possible sources of literature relevant to the aim of this review. These included the Cochrane Central Register of Controlled Trials, MEDLINE, EMBASE, PsycINFO, and ERIC. To ensure a comprehensive search, it was conducted through OVID (http://www.ovid.com/) and Web of Knowledge (http://www.isiknowledge.com) which cover all relevant databases.

For a review to be included, it had to be a systematic review or meta-analysis investigating an educational or teaching approach or behavioral intervention on participants <16 years of age with autism spectrum condition.

Data extraction and management

The data were extracted from the selected reviews by the first author of this paper, then cross-checked by to lessen bias and reduce inaccuracies. Any dispute about the numerical or qualitative data was resolved by re-examining the borderline articles. The data were then tabulated and listed alphabetically according to the type of intervention as so that it could be managed effectively. Reporting of the findings of this review adheres to the guidelines of the PRISMA statement.[11]

Assessment of methodological quality of reviews

The selected systematic reviews or meta-analyses were assessed for quality by two reviewers independently, using AMSTAR criteria. To guarantee that the two reviewers followed a similar routine to assess the quality of the selected reviews and to exclude the possibility that their agreements were due to chance, fixed-marginal multirater kappa was calculated.[12] Kappa was calculated using the Online Kappa calculator developed by Randolph.[13]

Data synthesis and evaluation of evidence

In this overview, the synthesis of evidence is based on the general conclusion of each review and the strength of evidence, based on the authors' own conclusions and cross-checked against the Grading of Recommendations Assessment, Development, and Evaluation (GRADE) system.[14] GRADE provides a framework to rate the evidence of the research reviewed (rather than the quality of the review itself).

Using GRADE, randomized controlled trials (RCTs) will be rated first as "HIGH" then downgraded to "INTERMEDIATE," "LOW," or "VERY LOW" if there are serious risks of bias, inconsistency, and indirectness of evidence, imprecision or publication bias.[14] Observational studies, on the other hand, are initially rated as "LOW," then can be upgraded if there are large effects. It must be noted that the GRADE system is a judgment on how much research is needed to be sure of the evidence in any outcome. Therefore, "VERY LOW" and "LOW" outcomes indicate that more research is needed in the areas under investigation. It could also indicate that a change of methodology may be necessary to raise the level of evidence.

Results

Results of the selection process

Initially, 1299 articles were identified through the database searches and another 31 articles through a manual search of references lists of systematic reviews. One hundred and eighty-seven articles were then removed because they were duplicates. The remaining 1143 articles were screened based on their abstracts, resulting in 1097 exclusions. The remaining 46 articles were screened based on their full text, leading to a further 16 exclusions.

Accordingly, 30 systematic reviews published since 2007 were deemed eligible for inclusion in this overview and subjected to the AMSTAR quality assessment with scores ranging from 3 to 10.

There was 75% agreement on AMSTAR quality between the two assessors ($\kappa = 0.7$). In cases of disagreement, the difference was no more than one AMSTAR point. Five reviews were then excluded from the final synthesis of evidence because they had an AMSTAR score below 5.

Description of included systematic reviews

Table 1 describes the characteristics of the 25 included systematic reviews and reports the number of selected studies and number of participants within each review.

Out of the 25 included reviews:

- Six examined the effectiveness of ABA and EIBI
- Four concentrated on PECS
- Four reviewed social skills interventions
- Three focused on CBL
- One review evaluated the evidence for TEACCH
- And the remaining seven reviews evaluated the evidence for multiple interventions.

There were 10 meta-analyses and 15 systematic reviews. Among the systematic reviews of single-subject studies, some have used a percentage of nonoverlapping data points or similar measures to provide a measure of intervention effectiveness.[39] One systematic review used both empirical results and expert opinions to reach a conclusion.[37]

The selected studies in these reviews vary according to each review's aims and objectives, as well as number of participants. The number of studies per review was five to 102, while the number of participants ranged from 26 to 2566. However, some studies have been selected by more than one review of the same intervention. For example, five studies selected by Hart and Banda's (2010)[26] ($n = 10$) overlap with Preston and Carter[27] ($n = 28$). This might be taken into consideration when one evaluates the weight of the evidence based on the number of selected studies and number of participants.

From 483 studies in the included reviews, 99 (20%) were RCTs and 124 (26%) were Controlled Clinical Trials. Overall, the single-subject design was the preferred method of choice with $n = 260$ (54%).

Effects of interventions

Applied behavior analysis and early intensive behavioral intervention

Four systematic reviews concluded that EIBI produces large to moderate effects in interventions measuring changes in IQ levels.[15-18] Virués-Ortega reviewed single-subject studies and claimed that long-term interventions may lead to improvements in IQ level and language development in autistic children.[20] However, some expressed concerns over the quality of evidence[18] or limitations on the generalizability of the evidence.[17] Spreckley and Boyd concluded, after reviewing RCTs and Quasi RCTs, that there is not sufficient evidence for improved cognitive skills, adaptive behavior, expressive, and receptive language when comparing an experimental group with the standard care group.[19]

Computer-based intervention

Three reviews concerning the effectiveness of CBI intervention for people with autism suggest that CBI is a "promising practice" to improve emotional recognition and literacy skills.[22-23] However, Ramdoss *et al*. (2011) which addressed the use of CBI to teach communication skills warned that it "should not yet be considered a research-based approach" to teach communication skills to children with autism spectrum disorder (ASD).[23]

Picture exchange communication system

There was a wide range of findings in the four reviews analyzing the evidence for PECS effectiveness. Three reviews observed improved communication skills while two reviews described the effects as "small to moderate or promising."[25] Hart and Banda have not quantified the evidence in their review of single-subject studies but concluded that there was an increase in functional communication and speech.[26] In contrast, Preston and Carter who reviewed three RCTs with sixteen single-subject studies concluded that the data to support improvement in communication was limited. However, all noted that children could learn the use of PECS quite readily.[27]

Treatment and education of autistic and communication physically challenged children

Virues-Ortega *et al*. observed a small effect of TEACCH on perceptual, verbal, and cognitive skills, while the effect on adaptive behavior, including communication, was within the

Table 1: Characteristics, AMSAR scores and quality of evidence according to Grading of Recommendations Assessment, Development, and Evaluation of included reviews

Reference	Nature of the review	Study design(s)	*n*	Age of participants	Participants' diagnosis or categorization	Authors' conclusions on major outcomes	Authors' conclusion re severe autism	AMSTAR score	Quality of evidence according to GRADE
Eldevik *et al*., 2009[15]	An update of Reichow and Wolery (2009) Meta-analysis, with a focus on methodological improvement for EIBI	9 CCT	297	2.8-5.5	ASD with minimum mean of IQ 41 (SD 13) Some studies included children with IQ as low as 27.8 on average	EIBI produces large to moderate effects sizes for changes in IQ scores for children with ASD. Hence, EIBI should be an intervention of choice for children with ASD	Not reported	8	Low IQ
Howlin *et al*., 2009[16]	A systematic review to evaluate EIBI for children with autism	2 RCT 9 CCT (3 retrospective)	397	4.5-11.5	Participants classified as having autism, ASD, PDD or PDD-NOS	At group level, EIBI resulted in improved outcomes compared to the comparison group. Considerable variability in outcomes, with some evidence that initial IQ (not age) was related to progress	A group of low IQ >40 showed a slight increase in IQ following the intervention, but remained very much delayed	5	Low IQ
Peters-Scheffer *et al*., 2011[17]	Meta-analysis of EIBI based on ABA studies with pre-test post-test control group	11 CCT	344	<10	ASD or PDD-NOS	EIBI has a moderate to large effect in young children with autism in some outcomes, despite some limitations	Not reported	8	Low IQ, NV-IQ, Adaptive behavior, RL, EL Very low Daily living skills
Reichow *et al*., 2012[18]	Random effect meta-analysis for the effectiveness of EIBI in increasing the functional behaviors and skills	1 RCT 4 CCT	203	<6	ASD, PDD-NOS, Asperger, or atypical autism	There is some evidence that EIBI is an effective treatment for children with autism, however the quality of the evidence is of concern	Not reported	10	Low Adaptive behavior, symptom severity, problem behavior, IQ, EL, RL
Spreckley and Boyd, 2009[19]	Meta-analysis trials of ABI effectiveness	6 RCT 2 QRCT 1 follow up, 2 CCT	100	1.5-6	ASD or PDD	There is inadequate evidence that ABI has better outcomes than standard care for children with ASD	Not reported	8	Low Cognition, EL, RL, behavior
Virues-Ortega, 2010[20]	Meta-analysis	8 SS 14 CCT	836	2.2-5.5	All participants classified as having autism or PDD-NOS	Long-term comprehensive ABA leads to positive (medium to large) effects in terms of intellectual function, language development acquisition of daily living skills and social functioning in children with ASD	Not reported	8	Low IQ, NV-IQ, RL, EL, composite language, adaptation -communication, development, daily living skills, composite -adaptation, adaptation -socialisation

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Table 1: Contd...

Reference	Nature of the review	Study design(s)	*n*	Age of participants	Participants' diagnosis or categorization	Authors' conclusions on major outcomes	Authors' conclusion re severe autism	AMSTAR score	Quality of evidence according to GRADE
Ramdoss *et al*., 2012[21]	Analysis of studies investigating CBI to improve emotion recognition of people with ASD	6 CCT 6 SS	330 (269 ASD)	4-52	At least 1 participant per study identified as having autism, Asperger or PDD- NOS. Severe autism was not reported	CBI is a promising practice to improve social and emotional skills for people with ASD. Preference and skills of students with ASD should be considered when designing CBI, as well as software customizability	Not reported	9.5	Very low Facial recognition, social skills
Ramdoss *et al*., 2011a[22]	Statistical analysis of quantitative research and summarization of other types of studies of CBI to improve literacy skills in students with ASD	11 SS 1 CCT	135	3-21	Mild to moderate autism symptoms. Only 2 studies included participants with severe autism	Preference and skills of students with ASD should be considered, along with software customizability when designing CBI. Overall, CBI is a promising practice to improve literacy skills for ASD students	Not reported	6.5	Very low RL, verbal expression, reading
Ramdoss *et al*., 2011b[23]	Analysis of studies involving CBI to teach communication skills to people with ASD	1 CCT 9 SS	70	3-14	3 studies included participants with severe autism	CBI promising but should not yet be considered a research-based approach to teaching communication skills	More research is needed	9	Very low Identification of words, decreasing echolalia
Flippin *et al*., 2010[24]	Meta-analysis of the literature on PECS in affecting communication and speech outcomes (single-subject and group studies)	4 RCT 1 CCT 6 SS	178	1-11	Participants should have ASD or PDD-NOS	PECS is promising but analysis revealed only small to moderate gains in communication which was demonstrated following training. Gains in speech were small to negative	Not reported	7	Low Gain in spoken words, gain in PECS exchange
Ganz *et al*., 2012[25]	Meta-analysis of studies for PECS relative to targeted functional communication and nontargeted associated outcomes (behavior, social skills and speech) for learners with autism	13 SS	26	3-17	Autism with intellectual disability or autism and multiple disabilities	PECS is a promising intervention method where functional communication outcomes associated with PECS were most impacted	Not reported	5.5	Low PECS exchange

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Reference	Nature of the review	Study design(s)	*n*	Age of participants	Participants' diagnosis or categorization	Authors' conclusions on major outcomes	Authors' conclusion re severe autism	AMSTAR score	Quality of evidence according to GRADE
Hart and Banda, 2010[26]	Systematic review of single- subject studies for effectiveness of PECS, its effects on speech and problem behaviors, generalization beyond training conditions, and social validly of the intervention	13 SS	36	3-40 (3-12 ASD)	ASD or autism and mental retardation	PECS yielded increase in functional communication in all, except 1 participant. Also it decreased problem behaviors and increased speech in some participants	Not reported	6	Very low Communication, increase speech, decreases problem behavior
Preston and Carter, 2009[27]	A descriptive review of (RCTs) to examine the empirical research on PECS using PND and PEM analysis	3 RCT 16 SS	456	1.6-3.3	ASD or PDD-NOS	Very limited data suggested some positive effect on both social communicative and challenging behaviors, while effects on speech development remain unclear	Not reported	6	Low Picture exchange, speech
Virues-Ortega *et al*., 2013[28]	Meta-analysis of pre-post and between groups studies of (TEACCH) in a variety of outcomes	7 CCT 6 SS	172	2.5-32.3	Autism or autism and PDD-NOS	Moderate to large gains in social behavior were observed. This provided a limited support for the TEACCH program as a comprehensive intervention perhaps due to the limited relative studies available	Not reported	8	Low Communication, social skills, adaptation composite, motor, cognitive
Karkhaneh, M. *et al*., 2010[29]	Qualitative analysis to synthesize all available controlled trials evaluating Social Stories for ASD SMD were calculated for key outcomes	4 RCT 2 CCT	135	4-14	Severity not reported in 2 studies. Moderate to low in 2 studies. Participants were able to read and communicate (verbal-non -verbal in 2 studies)	Social Stories may benefit children with HFA. The quality of the studies was questionable in terms of generalization and maintenance of the skills	Not reported	7	Very low Social skills
Kokina and Kern, 2010[30]	Meta-analysis of single- subject research to examine the use of social stories	18 SS	47	3-15	33 were diagnosed with autism, 4 with Asperger Syndrome and 10 with PDD-NOS The majority had high to average cognitive skills. A smaller group had cognitive delay	Social stories were more effective when addressing inappropriate behaviors than when teaching social skills. Participants' characteristics are important to be included in further similar studies	Not reported	6	Very low On several social skills outcomes

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Table 1: Contd...

Reference	Nature of the review	Study design(s)	*n*	Age of participants	Participants' diagnosis or categorization	Authors' conclusions on major outcomes	Authors' conclusion re severe autism	AMSTAR score	Quality of evidence according to GRADE
Reichow *et al*., 2013[31]	A cochrane systematic review and meta-analysis of RCT examined the effectiveness of social skills. All studies selected were conducted in the US	5 RCT	196	6-21	ASD. The studies focused mainly on children aged 7 to 12, and the participants were all of average or above average intelligence	There is some evidence that social skills groups can improve social competence for some children and adolescents with ASD. More research is needed to draw more robust conclusions, especially with respect to improvements in quality of life	No RCT examined the interventions with lower functioning ASD	10	Low Social competence, social communication, emotion recognition, quality of life
Williams White *et al*., 2007[32]	To summarize the state of research in group-based SST programs for age-school children and adolescents	4 CCT 10 SS	141	6-35	All participants classified as having ASD	Group-based SST approaches may be a useful intervention for children with ASD, based on small initial efficacy studies. The field requires the development of manual-based curricula that can be evaluated in large RCT	Not reported	5.5	Very low Social skills
Lang *et al*., 2009[33]	Quantifiable review of the interventions (1998-2008) that focused on teaching play to children with autism by including range of conceptual and developmental models and were applied in different settings	14 SS 1 CCT	53	0-8	All participants had diagnosis of autism	Three factors (modeling, prompting, and naturalistic instruction) underlie successful play interventions for children with autism	Not reported	7	Low Functional and symbolic play
McDonald and Machalicek, 2013[34]	Examining the peer-reviewed interventions of mostly single subject studies (1980-2011) for adolescents with ASD	98 SS 4 CCT	154	12-21	Adolescents with ASD: Most had autism (without specification of severity). Other diagnoses were autism with/without intellectual disability, AS, PDD-NOS, high-functioning autism, moderate autism, autism and PDD- NOS 2 participants received multiple diagnoses and 3 had severe autism	Encouraging findings of the effectiveness of some approaches e.g., behavioral and CBL interventions. However, concerns regarding addressing such approaches to academic skills and in comparing their effectiveness across autism severity, age and interventions	Autism severity was rarely reported and most studies relied on previous diagnosis which may not provide a clear current assessment of autism severity	5	Very low Social skills, communication, challenging behavior, academic skills

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Table 1: Contd...

Reference	Nature of the review	Study design(s)	*n*	Age of participants	Participants' diagnosis or categorization	Authors' conclusions on major outcomes	Authors' conclusion re severe autism	AMSTAR score	Quality of evidence according to GRADE
Oono *et al*., 2013[35]	Meta-analysis of 10 RCT to assess the effectiveness of parent-mediated early interventions in terms of the benefits for children with ASD and their parents	17 RCT	919	1-7	Asperger's syndrome, PDD and PDD-NOS. One study with children with severe autism	There is a positive change in patterns of parent-child interaction and in child language comprehension, also a reduction in autism characteristics severity. However this evidence is uncertain	Reduction of severity of autism was taken as an outcome	9.5	Low Language - joint, communication, language - expression, language - comprehension, parent -child interaction
Ospina *et al*., 2008[36]	Observational and experimental studies were evaluated to summarize the evidence on the effectiveness of behavioral and developmental interventions	55 RCT 32 CCT 14 cohort studies	2566	76% (<6) 24% (6-12)	93% of the studies covered autistic children	There is some evidence for the effectiveness of parent-mediated interventions, particularly within Parent-child interaction and child language understanding also in reduction in autism severity. Evidence of whether such interventions may reduce parent stress is inconclusive	There is some evidence for the effectiveness of parent -mediated interventions in reduction in autism severity	9.5	Low Adaptive behavior, communication, EL, comprehensive language, daily living skills and intellectual function
Parsons *et al*., 2011[37]	To provide a synthesis of empirical research and expert evidence (2002-2008) to identify the best practice in educational provision for children with ASD	92 Not reported	NR		ASD	There is insufficiently strong evidence regarding the effectiveness of one type of intervention approach compared with another More research is needed on other types of educational interventions to establish what works best for young people with ASD	Not reported	8	Full characteristics of included studies were not reported
Schlosser and Wendt, 2008[38]	Review of 9 single- subject experimental designs and 2 group studies (1975-2007) to determine the effect of AAC interventions on speech production in children with autism or PDD-NOS	9 SS 2 CCT	125	3.1-12	ASD or PDD-NOS	Although AAC interventions may increase speech production, the modest gains observed require realistic expectations of predictive child characteristic's and skills	Not reported	7	Very low Speech: Vocalization and imitation of words

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Table 1: Contd...

Reference	Nature of the review	Study design(s)	*n*	Age of participants	Participants' diagnosis or categorization	Authors' conclusions on major outcomes	Authors' conclusion re severe autism	AMSTAR score	Quality of evidence according to GRADE
Bellini and Akullian, 2007[39]	Meta-analysis of SS design studies to examine the effectiveness of video modeling and video self-modeling interventions for children with ASD	23 SS	80	3-20	ASD	Video modeling and VSM are effective interventions for addressing social communication skills in children and adolescents with ASD. Both maintained over time and transferred across persons and settings. Also, they can be implemented in a variety of settings	Not reported	6	Low Social and communication skills

AAC: Augmentative alternative communication, ABA: Applied behavioral analysis, ABI: Applied behavioral intervention, CBI: Computer-based intervention, CCT: Controlled clinical trial, EIBI: Early intensive behavioral intervention, EL: Expressive language, GRADE: Grading of Recommendations Assessment, Development, and Evaluation, HFA: High-functioning autism, IQ: Intelligence quotient, NOS: Not otherwise specified, NV: Nonverbal, PDD: Pervasive developmental disorder, PECS: Picture exchange communication system, QRCT: Quasi-randomized controlled trial, RCT: Randomized controlled trial, RL: Receptive language, SA: Severe autism, SD: Standard deviation, SS: Single-subject design, SST: Social skills training, TEACCH: Treatment and education of autistic and communication physically challenged children, VSM: Video self-modeling, ASD: Autism spectrum disorder, PND: Percentage of nonoverlapping data points, SMD: Standardized mean differences, CBL: Computer-based learning, AMSTAR: A MeaSurement tool to assess systematic reviews, PEM: Points exceeding the median

insignificant to minor range.[28] In comparison, there were moderate to large gains in social and maladaptive behavior. However, the authors point out that the limited support for the TEACCH programs in their meta-analysis could be due to the "limited pool" of studies available and therefore, their findings should be considered as "exploratory."

Social skills interventions including social stories

Four reviews examined the evidence of the effectiveness of interventions aimed at improving social skills using social stories,[29,30] and other social skills training (SST) techniques.[31,32] Social stories were found to be of value when given in a school setting and may be most effective when addressing inappropriate behavior.[30] The authors highlighted that the characteristics of participants and nature of settings are significant moderators to the effect of Social stories on the investigated outcome. However, the two reviews on Social Stories cautioned against a blanket generalization of their findings.

Further, a systemati review summarized the state of research in group-based SST and concluded that there was some evidence based on small initial efficacy studies that this intervention may be useful.[32] In addition, another review analyzed the evidence of interventions used to improve social skills and suggested that there is some evidence that social skills groups can improve social competence for some children and adolescents with ASD.[31] However, the generalizability of their findings is affected by the fact that the participants in the studies they reviewed have average or above average IQ. This was also the case in the claimed success of Social Stories.[30]

Multiple interventions

Among the 25 included reviews in this overview, there were six reviews which did not specify a particular intervention. Instead, they either targeted a specific population of people with autism, for example, adolescents,[34] or used different interventions through a specified mediator, for example, parents,[35] or analyzed interventions that may target a specific skill.[38] McDonald and Machalicek (2013) reviewed the effect of different interventions such as behavioral and technology-based interventions, social skills, and parent training interventions with adolescents with autism.[34]

In summary, reviews of multiple interventions recommended that further research is needed to identify the effectiveness of any one intervention compared to others. Parsons *et al*. reviewed the evidence for education based interventions for individuals with autism to inform best practice and concluded that there is insufficiently strong evidence to favor one particular intervention.[37] Ospina *et al*. concluded that no definitive behavioral intervention would improve all symptoms and advised that it is probably best to individualize each intervention and tailor it toward the specific needs of those involved.[36]

Discussion

Summary of main results

This overview attempted to synthesize the evidence of the effectiveness of mainstream educational and behavioral interventions for autistic children in general and those at the more severe end of the spectrum in particular. The selected systematic reviews listed in Table 1 investigated a variety of

interventions, and outcomes such as level of IQ, social skills, communication skills, and literacy.

Results showed that the six reviews evaluating the effects of ABA/EIBI generated similar conclusions, finding positive effects on the participants' level of IQ after the interventions.[15,27] However, all six reviews expressed serious concerns about the lack of RCTs to further support this trend. Other limitations are related to the quality of studies on EIBI because of a lack of randomized assignment of treatment in RCTs. A direct comparison between EIBI and other interventions is also lacking. Intervention duration and the amount of training of mediators for interventions are further sources of heterogeneity between the studies, making combining data from different studies in one conclusive outcome difficult.

The participants' age at which an intervention starts was found to be an important factor in determining the success of the intervention.[16] Other factors in the success of an intervention include the level of autism severity and the level of initial language skills (in particular, receptive language skills). However, there is controversy in the literature concerning what level of severity is more susceptible to intervention. Some argue that children with less severe symptoms will show better improvement in certain outcomes.[18] Others argue that there is more room for change if the individuals' levels of behavioral problems and autism symptoms are more evident.[16] Clearly, more research is needed with children with severe impairments to examine this argument further.

Further, IQ as a variable of choice in many studies has led to neglecting other variables such as social interaction. In some instances, an increase in IQ did not mean that the social functions of the participants have improved.[16,17] In addition, Eldevik *et al*. argue for more research to improve the adaptive behaviors of autistic children.[15] In summary, there is no evidence to suggest the superiority of one intervention over another.

Overall completeness and applicability of evidence

Many reviews raised concerns regarding experimental designs and robustness of the methodology used in investigating the effectiveness of interventions. Several argued that the most rigorous research design to investigate interventions may be RCTs in which both the control and intervention groups are allocated randomly and go through identical conditions apart from the intervention, and both the investigators and participants are unaware of which intervention was given.

There are inherent problems in implementing RCTs in autism research, however. For example, a double-blind design would be very difficult to achieve with educational interventions in school settings because participants would know the difference between their usual educational scheme and the interventional scheme. Further, randomization would be difficult to achieve with a small number of individuals with autism in each unit of study.

It is no surprise, therefore, that the types of studies included were mainly single-subject studies (where a participant's response to the intervention is compared to that participant's own baseline data). This design is widely used in psychology and educational research and other branches of applied social sciences. The single-subject design is sensitive to differences affecting the participant as it measures before and after changes in the same participant. This is in contrast to RCTs where a control group is composed of participants different from the treatment group.

Applicability of evidence to children at the severe end of the spectrum

There was a wide range of individuals with autism in the studies selected by these reviews. However, the severity of autism was rarely reported as shown in Table 1. Moreover, most studies failed to apply clear and concise criteria of how they described or assessed their participants' autism severity and they relied mostly on a previous diagnosis which may not take into consideration any improvement in the abilities of an individual since the first diagnosis was made, or the rich pattern of abilities and challenges often faced by autistic individuals.

In some reviews,[31] it was clear that the participants had high cognitive and verbal abilities and therefore the quality of evidence for the effectiveness of the intervention may need to be downgraded when used with children with more severe disabilities, as the indirectness rule of the GRADE system clearly suggests.

It is worth noting that the majority of studies are biased toward individuals with good cognitive, verbal and communication skills. Indeed, Reichow *et al*. who examined the empirical evidence of recently studied social skills interventions (SST) concluded that none of these studies examined the interventions with more severely disabled autistic individuals.[31] Two reviews,[35,36] though found some evidence that the involvement of parents, or more specifically parent-mediated intervention, can reduce the more severe symptoms of autism.

Merely, 9 out of the 25 included reviews reported that some children in their selected studies were at the more severe end of the autism spectrum. However, 4 of these 9 reviews had not reported the applicability of evidence for this group EIBI,[15] CBI,[22] PECS,[26] and Social Stories.[29] The five remaining reviews reported suitability of the interventions used in reducing autism severity in relation to their intervention, albeit with limited effectiveness. For example, Howlin *et al*. recognized a small increase in IQ following an EIBI intervention for a group of participants with previously low IQ.[16] Concerning CBI, Ramdoss *et al*. (2011) saw promise for teaching communication skills, but acknowledged that more research which includes participants with severe autism is needed.[23]

Overall, it is clear from the data provided by the included systematic reviews that there is little evidence for the effectiveness of interventions examined for severely disabled autistic children, due to both the small number of studies available and the unreliability or preliminary nature of the results.

Quality of evidence

Only systematic reviews and meta-analyses with an AMSTAR score of five or higher were included in this synthesis of evidence. This method of establishing the evidence from high-quality reviews and studies is standard practice in systematic reviewing. It is also advised to use a grading system to judge the quality of evidence, as applied in the current overview. The quality of evidence from reviews based on studies using RCTs is judged to be stronger than other methods of investigations, such as observational research. It can therefore be postulated that the conclusion of Spreckley and Boyd[19] that there is inadequate evidence that ABA has better outcomes than standard care for children with ASD carries more weight than Virués-Ortega conclusion that long-term comprehensive ABA leads to positive (medium to large) effects in a range of outcomes in children with ASD.[20] This is despite the latter having based their conclusion on a larger sample (n = 836 vs. n = 100). The reason for this is that Spreckley and Boyd based their evaluation on a range of studies including six RCTs while the lack of a control group in Virues-Ortega's study was evident. However, the quality of evidence in all research on ABA and EIBI was generally considered low, indicating that further research is needed to have greater confidence in the estimate of effect.

Potentials biases of the overview process

A potential bias is that some of the secondary outcomes in the selected original articles of the systematic reviews overviewed may have been masked by the tendency of the reviewers to report the major outcomes. Further, it should be pointed out that this overview included only publications which were peer-reviewed systematic reviews or meta-analyses. Reviews in the grey literature and government reports were omitted. We believe that by focusing on peer-reviewed material only, the quality of the material considered and thereby the quality of this overview has been maximized.

Conclusions and implications for research

Although social difficulties are a central feature of the diagnostic approaches to autism of both DSM and WHO, the emphasis on the effectiveness of social and behavioral interventions has only gathered momentum in the last decade or so. One factor that might explain the increase in research and application of social-based intervention is the development of theories to explain autism further. For example, in a synthesis framework published in Dialogues in Clinical Neuroscience the argument was that many of the social-based approaches have attempted to "marry specific psychological theories with studies of brain and behaviour."[40] The authors of this synthesis gave an example of Theory of Mind and the need to find a neurological explanation for it, but also mention the other dominating theories of deficits in executive function and weak central coherence.

One of the challenges that this overview revealed is that more RCTs need to be carried out to improve the quality of evidence. However, as argued earlier, since randomizing and blinding of interventions is difficult in educational and behavioral research, the next-best alternative to RCTs is the single-study design, which may in fact be more sympathetic to the needs and preferences of participants and the environment in which research is taking place.

It is essential to improve the quality of evidence of autism intervention research in general and systematic reviews in particular, to inform the research community. There is a particular need to study children at the more severe end of the spectrum. One way of doing this is to further research interventions that show promising results in improving some outcomes for this group of children since the quality of evidence is still very low. Likewise, the quality of data available on speech development resulting from the application of PECS is weak too, albeit promising.

This overview also indicates that interventions such as social stories, which are based on the assumption that children have high levels of receptive language skills, may not be appropriate for children with severe autism due to their limited, or delayed language skills. Instead, interventions such as PECS and CBI may be the tools of choice for this group of children because these interventions are based on visual stimuli.

Other factors such as the duration of the intervention, age, and level of severity of the participants should be considered in research since they have been recognized as effective variables in the evaluation of an intervention's success.

Conclusion

In summary, this overview of educational, behavioral, and social interventions for children with autism, with a particular focus on the severe end of the spectrum, found that:

- None of the intervention types is superior to the others
- There is generally weak evidence for the effectiveness of the reviewed interventions in improving symptoms of autism
- Applicability of this evidence on children with severe autism is generally questionable.

Clearly, there is a gap in the research investigating the responses of children with severe autism to educational and behavioral interventions. Indeed, the limited research in this area is illustrated by the prevalence of "VERY LOW" and "LOW" classifications for most reviews, as applied by the GRADE evaluation system [Table 1]. It suggests an urgent need for research with severely disabled autistic children.

Most importantly, there does not appear to be a definitive intervention that would improve all symptoms of autism. Any intervention that wants to be effective needs to be customized to the specific circumstances, needs and abilities of the individual.

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