A systematic review of speech, language, and communication interventions for children with Down syndrome from 0 to 6 years

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Abstract

Background: Speech and language acquisition can be a challenge for young children with Down syndrome, and while early intervention is important, we do not know what early interventions exist and how effective they may be. **Aims**: to systematically review existing early speech, language, and communication interventions for young children with Down Syndrome from birth up to 6 years, and to investigate their effectiveness in improving speech, language, and communication outcomes in children with Down syndrome. Other outcomes are changes in parental behaviour and their responsiveness **Methods**: We conducted a systematic search of relevant electronic databases to identify early intervention studies targeting speech, language and communication outcomes in children with Down syndrome published up to May 2020. Eleven studies which met the inclusion criteria were synthesised and appraised for quality using the PEDro-P scale. There was a total of 242 children. We identified three types of intervention: communication training and responsive teaching, early stimulation programme and dialectic-didactic approach. **Main contribution:** The findings from nine out of the 11 studies reported positive outcomes for children's language and communication up to 18 months following the intervention. All nine studies reported interventions which were co-delivered by parents and clinicians. However, there was also a de-accelerated growth in requesting behaviours in the intervention group reported by one study as well as a case of no improvement for the intervention group. Three studies provided some evidence of improvements to parent outcomes,

such as increased parental language input and increased responsiveness. However, there was a moderate to high risk of bias for all studies included. **Conclusions:** The findings from the review suggest that interventions which have high dosage, focus on language and communication training within a naturalistic setting and are co-delivered by parents and clinicians/researchers may have the potential to provide positive outcomes for children with Down syndrome between 0 and 6 years of age. Due to the limited number of studies, limited heterogenous data and the moderate to high risk of bias across studies, there is an urgent need for higher quality intervention studies in the field to build the evidence base.

What is already known on the subject?

Speech and language acquisition is usually delayed in children with Down syndrome, yet there are currently no standard interventions for children under 6. A number of research-based interventions exist in the literature, yet it is unknown how effective these are.

What this study adds

This is the first systematic review which specifically and exclusively focuses on parent and non-parent mediated speech, language, and communication interventions for children with Down syndrome between 0 and 6 years of age. It complements three existing recent reviews, each of which have a slightly different focus. O'Toole et al. (2018) focus only on parent-mediated interventions, excluding interventions not mediated by parents. Neil and Jones (2018) reviewed interventions including children and adults, and because of the very wide scope, there is no mention of what early interventions may be like or how effective these may be for young children with Down syndrome, and the authors did not assess risk of bias. Smith et al. (2020)

focus specifically on language interventions and exclude those focusing on speech articulation or pre-linguistic skills, and cover a wide age range (0-18). The findings from the review suggest that interventions which have high dosage, focus on language and communication training within a naturalistic setting and are co-delivered by parents and clinicians/researchers may have the potential to provide positive outcomes. We acknowledge that the current evidence-base comes from studies with moderate to high risk of bias hence our conclusions are not definitive.

Clinical implications of the study

Speech and language therapists will have synthesised information and a quick reference point on what type of interventions exist for children with Down syndrome under the age of 6, and evidence of which intervention approaches may be promising in terms of providing positive outcomes. However, it is acknowledged that, due to the limited number of studies and the moderate to high risk of bias inherent in the evidence, there is an urgent need for higher quality intervention studies in the field to build the evidence base.

Introduction

Speech and language development in children with Down syndrome

Down syndrome (DS) is a genetic condition caused by an additional copy of chromosome 21, which results in distinct facial features, health problems and learning disability (Chapman and Hesketh, 2000). It affects approximately 1 in 1000 births (Hughes-McCormack et al. 2020) and makes up the largest population of those identified with an intellectual disability. The average IQ of an individual with Down Syndrome is 50, with a range of 30 – 70 (Chapman and

Hesketh 2000). Communication, language, and memory are known to be adversely affected (Laws et al. 2000) and there is much variability within the condition (Davis-McFarland 2008).

Although children with Down Syndrome seem to go through the same stages of language development as typically developing children, the pace at which developmental milestones are reached is delayed (O'Toole and Chiat 2006). Children with Down Syndrome produce their first word around 21 months (Stoel-Gammon 2001), compared to typically developing children who usually produce their first word at around 12 months of age (Tomasello 2003). Expressive language tends to be more delayed than receptive language (Fidler and Nadel 2007, Mason-Apps et al. 2020). Social and non-verbal interaction using sign is also a relative strength for some individuals with Down Syndrome (Martin et al. 2009). Regarding language abilities, expressive and receptive grammar, verbal short-term memory, articulation, and phonology tend to be a relative weakness (Martin et al. 2009, Naess et al. 2015). Intelligibility is often adversely affected, as well as speech sound production and speech-motor planning (Buckley and Bird 2001) which can compromise joining in conversations and producing narratives (van Bysterveldt et al. 2014).

The gap in language outcomes between children with Down Syndrome and typically developing children widens as they reach school age. Some children with Down Syndrome enter school with a spoken vocabulary of approximately 300 words and communicate using 2-4 key word sentences (Buckley and Bird 2001). By 8-9 years of age, the average spoken vocabulary increases to around 450-500 words, ranging from 150-600 words, meaning that by 9 years of age some children with Down Syndrome have fewer than 200 spoken words (Buckley and Bird 2001). This may adversely affect later educational and social outcomes, as language ability at

school entry has been shown to predict later academic and psycho-social outcomes for typically developing children (Snowling et al. 2011).

Hearing loss is a significant contributor to the speech and language deficits experienced by children with Down Syndrome, with 40 to 80% of children with Down Syndrome having hearing impairments (Hans et al. 2010). Children with Down Syndrome who have moderate to severe hearing loss between the ages of 2 and 4 may go on to have more impaired receptive vocabularies, language comprehension and expression, and articulation difficulties compared to children with Down Syndrome who have hearing within the typical range (Laws and Hall 2014).

Existing interventions and previous reviews of interventions

Most individuals with Down syndrome have a speech, language and/or communication deficit that is prominent from an early age. Down Syndrome can be diagnosed prenatally (using amniocentesis and karyotyping of the foetal cell), or at birth, using diagnostic tests (Fidler 2005). The early identification allows for interventions to be administered from a very early age. And although early interventions targeting speech, language, communication outcomes for individuals with Down syndrome have been published as research papers, it is not clear if such interventions are effective and if these should be used in clinical practice. It is, therefore, of foremost importance to identify which early speech and language interventions exist (including clinician delivered, parent/carer-delivered or a mixture of clinician and parent/carer) and which ones may be effective in achieving better speech, language, and communication outcomes for young children with Down Syndrome. It is important to note that the speech, language, and communication difficulties of children with Down syndrome are on top of their learning disability, which has to be taken into account when developing appropriate interventions.

Early intervention (intervention in the pre-school years) has been shown to be effective for other clinical populations, such as autism spectrum disorders (ASD) (e.g., Dawson et al. 2010), and children with language delay (Buschmann et al. 2009). In recent years, systematic reviews have been used to assess the effectiveness of speech and language interventions for children with ASD (Hampton and Kaiser 2016) and developmental delay (Te Kaat-van den Os et al. 2017). By using this process, researchers can consider all research studies in a field, through a systematic and transparent process in selecting studies and considering the quality of the collective evidence (Marshall et al. 2015, Pring 2004). Furthermore, systematic reviews are important for supporting evidence-based practice.

To date, few systematic reviews have specifically focused on early speech and language interventions for young children with Down Syndrome. Neil and Jones (2018) focused on communication interventions for individuals with Down Syndrome which targeted speech, expressive syntax, phonology, and vocabulary, but included both children and adults. They concluded interventions employing behaviour analytic strategies offered promising results for improving communication and language outcomes. The review did not include a risk of bias analysis. O'Toole et al. (2018) completed a Cochrane systematic review focusing exclusively on parent-mediated interventions for communication and language in young children with Down syndrome including only randomised controlled trials (RCTs). The review concluded that there is insufficient evidence to establish the effectiveness of parent-mediated interventions. Another recent systematic review and meta-analysis by Smith et al. (2020) including eight studies (2020) focusing on language interventions for children and adolescents with Down syndrome aged between 0 and 18 years of age concluded that children with Down Syndrome have the potential

to respond positively to language intervention. The review included only experimental and quasi-experimental design studies which had a control group, and single case designs were excluded.

Additionally, an important aspect of social interaction is parental responsivity, and evidence suggests that children with Down syndrome had higher mental ability as measured by the Bayley Scale of Infant Development if their mothers had a more responsive interactive style (Mahoney et al. 1985). When discussing interventions for young children, caregivers' interactions with young children in promoting early language and communication development is crucial (Barton 2013), as the child's family and parents are best placed to support the child's development (O'Toole et al. 2018).

Given the importance of children having the necessary language and communication skills needed for school readiness (and school officially starts between the ages of 4 and 6, depending on the country), it is essential to focus on interventions which target pre-school children, or children who have just started school, to learn which interventions exist, how effective they may be and how the language and communication skills of children with Down Syndrome can be better supported.

Aims and research questions

The aim of the current systematic review was to identify and critically appraise existing randomised controlled trials and quasi-experimental studies of speech, language and communication interventions for children with Down Syndrome aged 0-6 years, and to investigate their effectiveness in improving speech/language/communication outcomes for the children, as well as outcomes for the parents/caregivers in terms of parental responsiveness and

changes in communication. Interventions which target expressive and/or receptive speech, language, and communication outcomes, as well as those targeting pre-verbal communication skills, were included. Single case studies/series were excluded because there are multiple biases in these studies, which makes it hard to synthesise the different types of study designs.

The research questions are:

1. Which early speech, language and communication interventions exist for children with Down Syndrome aged 0-6 years of age?
2. Are speech, language, and communication interventions effective in improving receptive and/or expressive speech and/or language/communication skills of children with Down Syndrome aged 0 to 6?
3. Are there relevant secondary outcomes such as change in parental language input, communication behaviour and responsiveness?

Method

The current systematic review followed the guidance detailed in the Cochrane Handbook of Systematic Reviews (Higgins et al. 2020, Petticrew and Roberts 2008) and Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement (prisma-statement.org).

Search strategy

We developed the search strategy considering the Population, Intervention, Comparison, Outcomes (PICO) model (Booth and Fry-Smith 2004). The search terms encompassed three main concepts relating to Down syndrome, intervention, and speech/language/communication

outcomes. A comprehensive list of search terms was identified using the key words from relevant review papers and the co-authors' previous work in the field. We did not set a limit regarding publication date. The searches were originally conducted in December 2016 and were updated in May 2020. A list of 18 search terms was established though a process of discussion and modification. They included key words related to Down syndrome, language, speech, communication, intervention, signing, and synonyms (see Appendix 1).

Electronic searches

Separate searches were completed in four electronic databases (ERIC, LLBA, MedLine and PsychInfo) as suggested by a senior librarian at the University of [REMOVED FOR REVIEW] to identify studies published up to May 2020.

Searching other resources

A hand search was also conducted which involved: searching the reference lists and using the Cite Forward function for all included full-text papers, scanning reference lists of review papers and searching several relevant journals identified to be most likely to publish papers on speech and language intervention in children with Down Syndrome, including: *Down Syndrome Research and Practice, Speech and Language, Child Language, Teaching and Therapy, Early Child Development and Care, the International Journal of Language and Communication Disorders and the Journal of Speech, Language and Hearing Research*. We also contacted two authors who we believed had collected intervention data from children with Down syndrome but had not published these data separately. One author did not respond at all, the other one responded initially that they would try to find the data but then did not respond any further.

Inclusion criteria

The Population, Intervention, Comparison, Outcomes (PICO) model (Booth and Fry-Smith, 2004) was utilised to aid the development of inclusion criteria, with the addition of study design, as described below.

1) **Population:** children with a diagnosis of Down Syndrome, without additional diagnoses (e.g., ASD and aged from 0 to 6 years 11 months at the start of the study. Studies which included other participant groups (e.g., children with ASD or learning disabilities) were included, if separate results were available for the children with Down syndrome.
2) **Intervention:** an evaluation using randomised controlled trial (RCT) and quasi-experimental designs of an intervention focussing on improving receptive and/or expressive speech/language/communication outcomes in children with Down Syndrome. The intervention could be delivered by clinicians or parents, individually or as part of a group, and could target precursor skills (prelinguistic communication) or rudimentary communication skills, such as turn taking, or verbal communication.
3) **Comparison:** a control group, which could be no-treatment or usual care, or a different intervention, a group receiving a different dose of the active intervention, or chronological age/developmental age group comparison.
4) **Outcomes:** at least one of the immediate post-intervention outcomes would be a measure of expressive and/or receptive speech and/or language skills. This could be measured by standardised or criterion referenced assessments, author created measures, parental reports, spontaneous language samples or observations.

Exclusion criteria

Studies were excluded if: using a single case studies or multiple baseline design, the paper was not available in English, and/or the full text was not accessible.

Selection of studies

After removing any duplicates, the first author and a research assistant independently reviewed the titles and abstracts for all studies that were identified from the initial searches and rejected those studies that did not fulfil the inclusion criteria. Those which met the inclusion criteria, and any papers for which it was hard to decide whether to include/exclude based on the title and abstract, were reviewed in full by two of the co-authors independently. The results were then compared, and any disagreements were resolved through discussion. Details regarding the number of studies included at each stage of the process and reasons for exclusion are shown in Figure 1 following PRISMA guidelines (prisma-statement.org).

INSERT FIGURE 1 AROUND HERE

Quality appraisal

To assess the risk of bias and appraise the quality of included papers, we used the speechBITE Physiotherapy Evidence Database quality rating scale (PEDro-P: Perdices et al. 2009, http://www.speechbite.com). SpeechBITE is an online database set up to assist clinicians to identify the scientific quality of randomised and non-randomised control trials. The PEDro-P rating scale was appraised for reliability for speech and language therapy literature (Murray et al. 2013) and deemed reliable and consistent. It has been used in other systematic reviews of speech and language therapy interventions (Wren et al. 2018).

The second and last author completed the PEDro-P training and did the appraisal of the included studies independently, following the PEDro-P rating scale (Perdices et al. 2009), which is suitable for use to appraise the methodological quality of randomised and group comparison studies. Each criterion when met gets a score of 1, and there are 11 criteria. Criterion 1 relates to external validity and is not counted towards the total methodological rating score; thus, the maximum score is 10. Criteria 2 to 9 assess the internal validity and cover randomisation of participants to groups, concealed allocation, and similarity of groups at baseline, blinding of participants, clinicians and assessors, drop-out rates, and intention to treat analysis. Criteria 10 and 11 cover the interpretability of findings. A criterion was only considered to be met when the relevant information was clearly stated in the paper. When it was not easy to interpret information, a conservative approach was taken whereby a criterion was assessed as not passed and inferences were not made. Higher scores on PEDro-P were associated with higher methodological quality and greater internal validity. When raters' overall scores differed (and this happened for three studies), agreement was reached by checking the papers again for any criteria where there may have been disagreements and reaching consensus. See Table 3 in the Results section for PERro-P appraisal.

Data extraction and synthesis

Data extraction was carried out on the participant characteristics, design, location, setting, intervention (type, duration, intensity, and implementer), control condition, treatment fidelity, outcome measures used and response outcomes. Quantitative synthesis was not conducted due to heterogeneity of measures obtained through different study designs and insufficient data available to conduct a meta-analysis.

Results

As Figure 1 shows, the initial electronic search located 1,359 papers which were transported to a reference management programme (EndNote). 175 duplicates were removed which left a total of 1337 papers. After screening titles and abstracts, 358 full-text papers were read and screened against the pre-defined eligibility criteria. A total of 11 studies were identified as being eligible for the review and were subject to quality appraisal.

Description of included studies

Design, location and settings of studies

Four studies were described as RCTs (Girolametto et al. 1998, Karaaslan and Mahoney 2013, Yoder and Warren 2002, Yoder et al. 2014) and the remaining seven studies used quasi experimental designs/group comparison. Three of the studies were conducted in the USA (Weller and Mahoney 1983, Yoder and Warren 2002, Yoder et al. 2014), one in Canada (Girolametto et al. 1998), one in the United Kingdom (Bidder et al. 1975), four in Spain (Sanz Aparicio 1989, Sanz Aparicio and Balana 2002, Sanz Aparicio and Balana 2003, Sanz et al. 2011), one in Brazil (Andrade and Limongi 2007) and one in Turkey (Karaaslan and Mahoney 2013). The interventions were delivered in community settings and/or in the participants' homes.

Participants

The studies included a total of 242 children with Down Syndrome, with between 8 and 48 children with Down syndrome in each study. There were between 4 and 48 children in the intervention group and between 4 and 19 in the control groups. Several studies had two or more active intervention groups of children with Down syndrome. Only one study (Andrade and

Limongi 2007) included a control group of 4 typically developing children in addition to having a control group of children with Down syndrome. The children were aged between 0 and 55 months at the beginning of the interventions.

Tables 1 and 2 present the participant characteristics, and study design and outcomes of the 11 included studies.

1 Table 1: Participant characteristics

Study		N (male)		Chronological Age range (months)	Dev/Cog age or quotient at baseline (months), range or mean	Language age, or quotient at baseline (months), range or mean
Andrade & Limongi (2007)	Intervention	4 (2)		36-52	?	?
	Control (DS)	4 (2)		36-52	?	?
	Control (TD)	4 (2)		14-16	?	?
						Transcriptions of children's language and gestures
Sanz Aparicio (1989)	Intervention 1	4 (?)	4 (?)	0-2	?	64-68 DQ for language
	Intervention 2	4 (?)		0-2	?	64-68
	Intervention 1	4 (?)		3-5	?	61-64
	Intervention 2	4 (?)		3-5	?	61-64
	Intervention 1	4 (?)		6-8	?	54-59
	Intervention 2	4 (?)		6-8	?	54-59
	Intervention 1	4 (?)		9-11	?	54-56
	Intervention 2	4 (?)		9-11	?	54-56
	Intervention 1	4 (?)		12-17	?	53-55
	Intervention 2	4 (?)		12-17	?	53-55
	Intervention 1	4(?)		18-23	?	48-51
	Intervention 2			18-23	?	48-51
					Brunet-Lezine's First Childhood Scale	Language subtest of Brunet-Lezine
Sanz Aparicio & Balana (2002)	Intervention 1	12 (4)		0-1	?	?
	Intervention 2	12 (5)		3-4	?	?
	Intervention 3	12 (7)		6-7	?	?
					Brunet-Lezine's First Childhood Scale	Language subtest of Brunet-Lezine
Sanz Aparicio & Balana (2003)	Intervention 1	10(?)		0-2	?	?
	Intervention 2	10(?)		0-2	?	?
						Language subtest of Brunet-Lezine

				Brunet-Lezine's First Childhood Scale	
Bidder et al., (1975)	Intervention	8 (6)	12-33	9.5-22, mean 16.6	?
	Control	8 (6)	12-33	9.5-22, mean 14.8	?
				Griffiths test	Speech subscale
Girolametto et al. (1998)	Intervention	6 (5)	39.2	73.2	16.5mos
	Control	6 (5)	37.2	57.8	16.8mos
				Developmental Profile ll	CDI vocab age
Karaaslan & Mahoney (2013)	Intervention	7 (2)	55.1	20.4	18.6mos
	Control	8 (3)	44.1	19.8	17.1mos
				DD	ADSI
Sanz et al., (2011)	Intervention 1	10 (?)	5-8	78-85 DQ	?
	Intervention 2	10 (?)	5-8	78-85 DQ	?
				Brunet-Lezine's First Childhood Scale	Language subtest of Brunet-Lezine
Weller & Mahoney (1983)	Intervention 1-oral group	7 (3)	18-36	54.71 (standard score)	14.14 mos ELA, 14.29 RLA, 9.14 words spoken
	Intervention 2- total com	8 (5)	18-36	67.75 (standard score)	0 words signed 15.38 mos ELA, 17.25 RLA,14.63 words spoken, 0 words signed
Yoder et al. (2014)	Intervention	8 (?)	21*	14	7.5
	Control	9 (?)	22*	14	7
				BSID	CDI (median words)
Yoder & Warren (2002)	Intervention	16 (?)	22.5	12.7	1.37
	Control	19 (?)	21.6	12.1	0.94
				BSID	CDI (mean words)

Key: *median reported; ? - missing/not reported

Note: Abbreviations- mos-months; DS – Down syndrome; TD – typically developing children; BSID - Bayley Scale of Infant Development; DD - Denver Developmental Screening test-ll; ADSI - Ankara Developmental Screening Test-ll; ELA - Expressive Language Age; RLA – Receptive Language Age; EPBV- Environmental Pre-language Battery – verbal; CDI Communicative Development Inventories; total com-total communication, DQ-Developmental Quotient

1 Table 2: Study design and outcomes

Study	Design	Intervention/ Control	Duration (weeks)	Frequency	Length (minutes)	Measures	Child outcomes	Parent outcomes
Andrade & Limongi (2007)	Quasi-Experimental	Dialectic-didactic intervention Treatment as usual	40	Weekly (child) by researcher	?	Number of different words or signs	No significant differences between the DS groups for oral, gestural, or simultaneous oral/gestural communication	n/a
Sanz Aparicio (1989)	Quasi-Experimental	6 intervention groups (0, 3, 6, 9, 12, 18 mos), each group allocated to either modelled or written instructions	26-100	Twice weekly by clinician + 60 mins weekly at home by mothers	180	Language: Brunet-Lezine Scale	The children in all age groups whose parents had received modelled training scored better than those whose parents got only written instructions	n/a
Sanz Aparicio & Balana (2002)	Quasi-Experimental	3 intervention groups: Age 0 mos, Age 3 mos, Age 6 mos	52-78	?	150	Language: Brunet-Lezine	The children who started intervention at birth had better language scores than those who started later	n/a
Sanz Aparicio & Balana, 2003	Quasi-Experimental	2 intervention groups: Intervention 1 (modelled instructions) Intervention 2 (w	70-78	Twice weekly by clinician + 30mins weekly at home	150	Social development: Brunet-Lezine	The children whose parents received visual instructions (modelled)had significantly higher social	n/a

		ritten instructions)					developmental quotients	
Sanz et al., (2011)	Quasi-Experimental	Intervention 1 (social reward 1) Intervention 2 (social reward 2)	43	?	?	Language: Brunet-Lezine	Children who received both verbal approval+a gesture (e.g applause) did not significantly differ from those who received verbal approval only	n/a
Bidder et al., (1975)	Quasi-Experimental	Behaviour modification intervention Control	26	4 sessions weekly+4 sessions fortnightly+4 sessions monthly for mothers; 10 mins daily twice+ for children	135 (parent training) 10 (children)	Language: Griffiths Scale	The intervention group's language had advanced significantly more than the control group's	Mothers were asked for feedback but no formal assessment
Girolametto et al. (1998)	RCT	HPP Treatment as usual	13	Weekly (parents trained)	150 (parent training) 20	Child Vocab. CDI	HPP group used significantly more target words and more words overall than control. No significant difference for semi-structured interaction with clinician	Mothers in control group used fewer utterances than intervention group. Parents in the intervention used more target words
Karaaslan & Mahoney (2013)	RCT	RT Treatment as usual	24	Weekly	90-120	Language (DD-ll) Language-cognitive scale (ADSI) CBRS	Significant improvements across all development subscales (including language) than control group	Mothers in intervention showed more responsiveness and positive affect than control group (MBRS)

							Children in the intervention had higher attention and initiation than controls	
Weller & Mahoney, 1983	Quasi-Experimental	Intervention 1(oral language); Intervention 2 (total communication)	20	Weekly (for 4 weeks) for mothers + daily for children	60-120 (parents) 30 (children)	Receptive and expressive language, expressive and receptive vocabulary + signs (REEL, EPB)	No significant differences between the groups	n/a
Yoder & Warren (2002)	RCT	RE/PMT Treatment as usual	24	3-4 weekly (PMT) for children; Up to 12 RE for parents	20	Intentional pre-linguistic communication (commenting and requesting) Child Vocab (transcription)	DS children in control group had steeper growth curves. DS children in intervention group had de-accelerated growth in requests	Parent's responses to their child's communication
Yoder et al., (2014)	RCT	High dose (MCT) Low dose (MCT)	36	5 sessions/week PMT (high dose) Weekly PMT (low dose) 9 RE	60	Child Vocab CDI	Significantly more spoken words on CDI for those in higher dose group	n/a

Note: ? - information missing or unclear; HPP- Hanen Parent Programme; DD II- Denver Developmental Screening test-II; ADSI - Ankara Developmental Screening Test-II; CDI - Communicative Development Inventories; REEL – Receptive Expressive Emergent Language Scale; EPB – Environmental Pre-language Battery; RE – Responsive Education; PMT – Prelinguistic Milieu Teaching; Vocab – vocabulary; CBRS – Child Behaviour Rating Scale; MBRS – Maternal Behaviour Rating Scale; DS – Down syndrome; TD – typically developing children; mos - months

Description of interventions

The interventions targeted both verbal and non-verbal communication in children with Down syndrome. Interventions were either designed for, or adapted to be used with children with Down syndrome.

Communication training, responsive teaching, behaviour modification (including Hanen) (n=6)

A number of studies focused on training parents/caregivers to be more attuned to the child's communication needs and to respond in a way that is going to optimise the child's communication gains and the child's verbal or non-verbal communication.

Four studies (Girolametto et al. 1998, Karaaslan and Mahoney 2013, Yoder and Warren 2002, Yoder et al. 2014) used *communication training and responsive teaching* as the intervention method. This approach involves training parents/primary caregivers to enhance children's communication using one or more of the following techniques: Responsive Teaching (RT), Responsive Education (RE), Pre-linguistic Milieu Training (PMT) or Milieu Teaching (MT). Girolametto et al. (1998) trained parents with a view to increasing their children's expressive vocabulary, based on the principles of the Hanen Parent Programme (HPP; Manolson, 1992), a recognised intervention for children with developmental delays which aims to teach parents to better understand and recognise their child's communication attempts, to follow their child's lead and to respond appropriately to their child's actions (Fong et al. 2012). These have in common the fact that they aim to teach the child specific gestures, eye gaze and vocalisations. They focus on intentional communication (not necessarily words) and encourage the child to use

pointing to initiate or request an object/action or to respond to bids for joint attention. Responsive education is often combined with child treatment such as Pre-linguistic Milieu Training or Milieu Language Teaching (MLT). MLT aims to increase the frequency and complexity of verbal communication through modelling, mand-modelling time delay and incidental teaching (Fey et al. 2013). PMT targets children's non-verbal communication, focusing on child-centred play by adjusting the environment, following the child's lead and the use of prompts with a view to improving later speech and language production (Fey et al. 2013). Responsive Teaching (RT) encourages parents to increase their levels of responsiveness (e.g., turn-taking, joint attention), model developmentally appropriate communication, and discourages the use of directives. In the HPP programme, mothers were supported by being shown how to follow their child's lead, observe and listen, be face to face, wait, take turns etc with a view to maximising the modelling of target words, and how to use signs to accompany the words. A similar approach, called *behaviour modification*, was used in Bidder et al. (1975). The mothers were trained how to change their behaviour and modify their daily routines to support verbal communication with their child and how to involve other family members in the treatment plan. The training programme included mothers observing video tapes and demonstrations through an observation mirror. The programme was individually tailored for each child and emphasised the importance of early language development, activities which involve object manipulation, especially construction, developing the child's independence through self-feeding, toilet training, self-dressing etc. Weller and Mahoney (1983) used the *environmental language training programme,* a parent assisted programme which incorporates aspects of behaviour modification, Piagetian theory, concepts from the language acquisition literature and theories of communication development. The mothers were trained how to model language, record

behaviour changes, adapt teaching techniques based on the child's abilities. The mothers administered daily sessions to their children which included training in language comprehension, imitation, conversation, structured play, and generalization training.

Dialectic-didactic method (n=1)

Andrade and Limongi (2007) used the dialectic-didactic method, which is based on the clinical method proposed by Jean Piaget and focused on the building of knowledge through problem solving. The intervention was done by a researcher and materials included developmentally appropriate toys with the aim to improve the child's communication by prompting the child to use verbal communication through object and action naming.

Early Stimulation Programme (n=4)

Four studies conducted in Spain (Sanz Aparicio 1989, Sanz Aparicio and Balana 2002, Sanz Aparicio and Balana 2003, Sanz et al. 2011) state that they used an early stimulation programme aimed at infants with Down syndrome (from birth to 18 months). The papers mention using the following as intervention materials: Intervention programme for Down syndrome children (Hanson, 1979), the Early Stimulation Programme (UNICEF 1982) and the Bayley Child Development Scale (Bayley 1959). The papers do not provide any information as to how these materials (especially a standardised assessment such as the Bayley Scale) were used, or how the different intervention programmes were combined. The UNISEF (1982) programme does not seem to be available in English, and the reference to Hanson (1979) was unattainable. In the Sanz et al. (2011) paper, the early stimulation programme is described as targeting four main developmental areas: language, fine motor, gross motor, and social

development skills. Parents were also trained either by observing the clinician (modelled instruction), or by being given written instructions so that they could implement stimulation techniques at home.

Intervention procedures

Duration, intensity, and intervention implementer

The number of sessions per intervention varied and sessions sometimes only included parent training, sometimes only direct work with the children, and sometimes a combination of both. The total number of sessions ranged from 13 in Girolametto et al. (1998), all of which were parent training sessions, and up to 100 sessions in Sanz Apparicio (1989), all of which were sessions done directly with the child. The child focused session duration ranged from 10 mins each at least twice a day (Bidder et al. 1975), 20 minutes (Yoder and Warren 2002), 60 minutes in Yoder et al. (2014) to 90-120 in Karaaslan and Mahoney (2013). See Table 3 for further details.

The intervention duration ranged between 13 weeks (Girolametto et al. 1998) and 100 weeks in some of the *Early Stimulation Studies* (Sanz Aparicio 1989, Sanz Aparicio and Balana 2002, 2003), depending on when the children started their intervention. The intensity ranged between once per week (Girolametto et al. 1998, Karaaslan and Mahoney 2013, Yoder et al. 2014 - low intensity group) to 3-4 times per week (Yoder and Warren 2002), 5 times per week in the high intensity group in Yoder et al. (2014), to twice daily (Bidder et al. 1975).

Two interventions were delivered by the parent after receiving training from a clinician or researcher (Bidder et al.1975, Girolametto et al. 1998), one while a researcher worked with

the parent to help them use responsive intervention strategies (Karaaslan and Mahoney 2013) and one where the parents had training first and then direct support from a language trainer (Weller and Mahoney 1983). In Girolametto et al. (1998)'s study, a clinician delivered the Hanen Parent Programme to mothers in group sessions using discussions, video tapes, role play and coaching during home visits. In Bidder et al. (1975), the mothers had 12 sessions where they were taught behaviour modification techniques. In addition, there were also sessions for fathers and child minders. In Karaaslan and Mahoney's (2013) study, the trainer first explained how behaviour was linked to child development before describing strategies designed to elicit such behaviours (e.g., following the child's lead, responding positively and promptly to the child). Mothers were coached when trying to implement such strategies and integrate these into their daily routines. In Weller and Mahoney's study, mothers first received individualised instruction on how to implement the programme, and then received weekly instruction and assistance with delivering sessions, and feedback regarding their teaching performance during their daily lessons at home with their child.

In only one study, the researcher exclusively implemented the intervention without the parents getting involved (Andrade and Limongi 2007). In the remaining six interventions (Sanz Aparicio 1989, Sanz Aparicio and Balana 2002, 2003, Sanz et al. 2011, Yoder and Warren 2002, Yoder et al. 2014) the sessions were delivered by a clinician or researcher, in addition to the parent-delivered sessions at home. In the Yoder and Warren (2002) study, the parents were trained in responsive education (i.e., HPP), in small groups and 1:1 format. During the same six months, clinicians delivered Pre-linguistic Milieu Teaching directly to the children, three to four times per week. Clinicians aimed to promote play routines and the opportunities for children to

make requests for objects or actions. In the Yoder et al. (2014) study, parents completed nine 1-hour Responsive Education sessions at home and read the Hanen Parent Program book: *It Takes Two to Talk* (Pepper and Weitzman, 2004) in the first three months of intervention. Children received either Pre-linguistic Milieu Training or Milieu Teaching, depending on the number of spoken words at the baseline assessment. Children in the lower dose frequency received one 1-hour session each week and those in the higher dose frequency received five 1-hour sessions each week. In Sanz Aparicio (1989), Sanz Aparicio and Balana, (2002, 2003), the parents attended two group sessions, in which early stimulation was explained and then parents were given either written instructions or modelled instructions where they observed the clinicians working with their child in the clinic (Sanz Aparicio, 1989). As this earlier study showed that there was more improvement with modelled instructions compared to written instructions, in the remaining three studies parents observed the clinician working with their child and then implemented the programme at home (Sanz Aparicio and Balana 2002, 2003, Sanz et al. 2011).

Control Condition

Four studies used treatment as usual as the control group (Bidder et al. 1975, Girolametto et al. 1998, Karaaslan and Mahoney 2013, Yoder and Warren 2002). In the Bidder et al. (1975) study, the control group continued to receive usual care from health visitors and their general practitioner. In the Girolametto et al. (1998) study families in the control condition continued with their existing language interventions, which involved monthly consultations with a clinician, or liaison between the clinician and teachers and families. Parents in the control condition were not given the list of target words that were provided for the intervention group. In Karaaslan and Mahoney (2013), both the control and intervention groups continued to receive

early intervention services at their local special education centre. This took the form of group and/or individual education and teaching social skills using the Picture Exchange System (PECS). Both children in the intervention and control group in the study by Yoder and Warren (2002) continued with early intervention services from community providers.

The remaining studies used an active control. Yoder et al. (2014) compared the effects of dose frequency (i.e., lower, and higher dose frequency). Sanz Aparicio (1989) compared the effect of age when children started intervention, and whether the parents had observed the clinician on how to deliver the intervention or whether they had only received written instructions. In Sanz Aparicio and Balana (2002), the age at which children entered the intervention was compared, and Sanz Aparicio and Balana (2003) compared the effects of how training was provided for the parents (modelled versus written instruction). Sanz et al. (2011) compared the effect of different type of social reinforcement (verbal versus physical). Weller and Mahoney (1983) compared the effect of oral versus total communication. Andrade and Limongi (2007) reported that the control group had an intervention with different theoretical and methodological approach to the intervention group, but no details were given.

Treatment Fidelity

Karaaslan and Mahoney (2013) reported 100% adherence to content and procedure for all sessions. Furthermore, all clinicians delivering the intervention were professionals educated to a doctoral level and completed 5 months of training on the intervention. Girolametto et al. (1998) evaluated the fidelity of parent implementation of the intervention, in terms of attendance and clinician ratings of parental adherence to the intervention. They reported that all mothers attended at least seven out of the nine sessions and completed all home visits. Clinicians

confirmed the mothers' use of target words and focused labelling during home visits. Yoder et al. (2014) reported intra-class correlation coefficients of .91 to .98 for absolute agreement on the number of correctly implemented teaching episodes, across both the higher and lower dose frequency groups. However, they did report that children in the lower dose frequency group experienced fewer cumulative teaching episodes than the higher dose group. Yoder and Warren (2002) reported no difference in the frequency and proportion of correct teaching between the lower and the higher dose frequency groups. The intervention lasted 9 months for both groups. Slightly more sessions were completed by the lower dose frequency group, but Fey et al. (2013) reported that the completed sessions were high for both groups and did not affect the analyses.

The remaining studies do not report any information on treatment fidelity.

Children's speech language and communication outcomes

Girolametto et al. (1998) used a parent-report measure of language, i.e., Mervis's adaptation of the Communicative Development Inventories (CDI) (Fenson et al. 1993). A direct assessment of language, i.e., Sequenced Inventory of Communication Development (SICD) (Hedrick et al. 1984) was completed at baseline but post intervention results were not reported. The use of target words was assessed during a free-play mother-child interaction.

Karaaslan and Mahoney (2013) used two developmental assessments to measure language: The Turkish version of the Denver Developmental Screening Test-II (Denver-II: Anlar and Yalaz, 1996) which covers four developmental domains: language, gross motor, fine motor and personal-social. It is unclear whether both expressive and receptive language were assessed. The second measure was the Ankara Developmental Screening Inventory (ADSI) (Savaşır et al.,

2005) which examines cognitive-language development, fine motor, gross motor, and social/emotional functioning, which is completed by parents or primary caregivers. It is also unclear if ADSI assesses expressive or receptive language, or both. A child behaviour rating scale (CBRS; Mahoney and Wheeden 1998) was used to assess children's interactive behaviour with their mothers in terms of attention and initiation.

The language/social subtest of the Brunet-Lezine First Childhood Scale (1971) was used in four studies (Sanz Aparicio 1989, Sanz Aparicio and Balana 2002, 2003, Sanz et al. 2011). It is unclear, however, which aspects of language are assessed.

Weller and Mahoney (1983) used several language outcome measures: Receptive Expressive Emergent Language Scale (REEL), the Environmental Prelanguage Battery (verbal subtests), parental report of number of spoken and signed, or only signed words.

Andrade and Limongi (2007) used spontaneous communication of oral language and coded linguistic concepts and gestural communication.

Bidder et al. (1975) used the language subtest of the Griffiths Scale (1954) and it is not specified which aspects of language were assessed. The authors refer to the subscale both as speech in one instance and language in another.

Yoder and Warren (2002) used transcriptions and observations to evaluate child language outcomes. Specifically, the Communication and Symbolic Behaviour Scales (CSBS: Wetherby et al. 1989), the Experimenter-Child Play Session (ECX: Yoder and Warren 1998) and the Parent-Child Session (PCX: Yoder and Warren 1998). Both the CSBS (specifically the Communication temptations and Book sharing sections) and the ECX measured child communication and lexical

density by adapting the environment to encourage child-initiated requests. The PCX measured child requests and comments through a series of three sections (structured play session, snack, and free play). All observations were transcribed and analysed using the Systematic Analysis of Language Transcripts (SALT) (Miller and Chapman 1993) to record the frequency of child-initiated comments, child-initiated requests, and lexical density.

Finally, Yoder et al. (2014) used the MacArthur-Bates Communication Development Inventories (MB-CDI: Fenson et al. 2003) which is a parental questionnaire, to measure expressive language (vocabulary).

Parental language, communication and response outcomes

In three of the 11 studies, parental outcomes were measured as well. Girolametto et al. (1998) examined maternal behaviour during a free play interaction, which was transcribed and analysed using the Systematic Analysis of Language Transcripts (SALT; Miller and Chapman 1993). The frequency of maternal utterances, complexity of the language (i.e., mean length of utterance) and the use of labelling (signed and spoken target words) was measured.

Karaaslan and Mahoney (2013) assessed maternal interaction behaviour through a mother-child free play interaction. The Maternal Behaviour Rating Scale (MBRS: Mahoney 1999) examined maternal responsiveness, affect, and achievement/directedness.

Yoder and Warren (2002) measured parental responsiveness during the parent-child interaction by counting the number and proportion of optimal parental responses to the children's communication acts. Optimal responses included complying with the child's communicative act,

immediate imitation of child vocalization and responding to the child's nonverbal communication (e.g., labelling an object the child points to).

Effect of interventions for the speech and language outcomes for children with Down syndrome

All 11 studies reported tests of statistical significance on at least one measure of language, verbal, or social communication post intervention. Post-intervention measures were administered at the end of the intervention with some studies reporting further follow ups (Sanz Aparicio 1989, Sanz Aparicio and Balana 2002, 2003). Two studies (Girolametto et al. 1998, Sanz et al. 2011) used non-parametric statistics. It was not clear whether parametric or non-parametric statistics was used in Bidder et al. (1975). The other eight studies used parametric statistics. Effect sizes were reported in three studies and were small to medium. Cohen's d effect sizes could not be computed for seven studies because they did not provide adequate descriptive statistics (Andrade and Limongi 2007, Bidder et al. 1975, Sanz Aparicio 1989, Sanz Aparicio and Balana 2002, 2003, Sanz et al. 2011, Yoder and Warren 2002)

Based on the p values and effect sizes reported (when provided) by the authors, seven studies showed significant positive effects (Bidder et al. 1975, Girolametto et al. 1998, Karaaslan and Mahoney 2013, Sanz Aparicio 1989, Sanz Aparicio and Balana 2002, 2003, Yoder et al. 2014). Bidder et al. (1975) reported that children in the intervention group obtained significantly higher language scores on the Griffiths Mental Development Scales by advancing their language skills at a mean rate of 6.56 months in comparison to the control group ($p<0.01$). Girolametto et al. (1998) reported that children in the intervention group produced significantly higher number of target words than the control group post intervention both in parental report ($U=7,0$, $p<0.05$),

and in free play (U=6.0, p<0.05), although only the intervention group had targeted words in their intervention. However, there were no statistically significant between group differences in vocabulary size as measured by the CDI, nor in the number of target words used by the two groups assessed by a clinician-administered semi-structured interview. Karaaslan and Mahoney (2013) reported a statistically significant difference between the treatment and control groups in post-test language quotient scores as measured by the Denver-II, in favour of the treatment group (F=6.50, p<0.05) with a small effect size (Hedge's g=0.42). Yoder et al. (2014) found that children with Down Syndrome in the higher frequency dose group showed significantly bigger growth in expressive vocabulary (as measured by the CDI) than those children in the lower frequency dose group (t=2.926, p<0.01) with a medium effect size (Hedge's g=0.55). Sanz Aparicio (1989), Sanz Aparicio and Balana (2002) reported that the earlier the children started intervention, the better their language outcomes on the Brunet-Lezine's First Childhood Scale (p<0.001, for both studies). The social development quotient was significantly higher for the children whose parents had visual training (i.e., they learnt how to stimulate their child by having directly observed the clinician as opposed having received only written instructions).

In the Yoder and Warren (2002) study, the children with Down syndrome who had received the intervention had a significantly lower growth curve in requesting behaviours (t for product term on slope = –2.18; p = .04; b = –.044) compared to the control group as measured during parent-child sessions, experimenter-child sessions and Communication and Symbolic Behaviour Scales (Wetherby and Prizant 1993).

No differences between intervention and control groups were reported in Andrade and Limongi (2007). Also, there were no differences in language outcomes between the two groups

(physical versus verbal reinforcement) in Sanz et al. (2011), nor were there any significant differences between the two intervention groups (oral versus total communication) in Weller and Mahoney (1983).

Effectiveness of intervention on parental/maternal language, or responsivity

Most studies included parent training, however, only three studies reported outcomes for parental responsiveness/communication/behaviour. Girolametto et al. (1998) reported significant differences between maternal utterances during mother-child interaction post-intervention, however, further investigation found that mothers in the control group showed a reduction in the number of utterances, whereas the mothers in the intervention group maintained their baseline level of utterances. Mothers in the intervention group also used more target labels than mothers in the control group, although there were no differences in linguistic complexity of maternal utterances. However, it is important to note that only mothers in the intervention group were aware of the target words.

Karaaslan and Mahoney (2013) found that mothers in the intervention group demonstrated significantly greater improvements in responsiveness, affect and achievement/directiveness as measured by the MBRS. Yoder and Warren (2002) reported that parents in the intervention group demonstrated a higher frequency and a higher proportion of responsiveness to child communicative acts as observed during the parent-child interaction compared to the parents of the children in the control group.

Risk of Bias- PEDro-P Appraisal

The risk of bias of the 11 included studies as assessed with the Pedro-P tool and is presented in Table 3.

Insert Table 3 about here

As shown in Table 3, out of the 11 included studies, only four demonstrated an adequate level of quality according to the PEDro-P criteria (Girolametto et al. 1998, Karaaslan and Mahoney 2013, Yoder and Warren 2002, and Yoder et al. 2014). By adequate level, we mean a score of 6 or above, in line with other reviews (Maher et al. 2003, Wren et al. 2018). The other seven studies lacked information about most of the following: clearly specified eligibility criteria; random allocation of participants and allocation concealment; blinding of participants and assessors; control groups being similar at baseline regarding key outcome measures; point measures and measures of variability for at least one outcome variable provided; all participants for whom outcome measures were available received the treatment or control condition as allocated. None of the studies reported any method of allocation concealment, hence they are at high risk of concealment bias. Only three studies reported a computer randomisation of participants. None of the studies provided any details of any methods to blind parents or personnel, hence they are all deemed at high risk of performance bias. It should be noted that, due to the nature of the interventions (where parents needed to be trained and assisted by clinicians), it would have been hard, if not impossible, for parents and clinicians to be blind.

The seven studies which scored lower than 6 did not provide usable outcome data because it was not possible to see the range of performance for speech/language/communication outcome measures across individuals in the groups.

Discussion

The aim of the systematic review was to investigate early speech and language interventions for children with Down Syndrome aged 0 to 6 specifically focusing on which interventions exist and whether they are effective for children with Down Syndrome. Eleven studies, four of which were randomised controlled studies, were identified as fulfilling the inclusion criteria and included in the review. It was not appropriate to complete a meta-analysis study due to the heterogeneity of outcomes and measures used, and information missing which did not allow to compute effect sizes for most of the studies. The results from the systematic review showed that the most common type of interventions are those which are based on social-interactionist theories of language acquisition, and suggest that children with Down syndrome under the age of 6 years at the start of the intervention can benefit from early intervention. There were also positive outcomes for the parents (where parent outcomes were measured), in that they increased their responsiveness, affect and non-directiveness in their interactions with their children. All studies had moderate to high risk of bias and any conclusions drawn are very preliminary and should be treated with caution given that no meta-analysis was performed, and results are based on individual RCTs and group comparison studies of small sample sizes, with effect size reported that have no power calculations.

Our first question asked which early interventions exist for children with Down Syndrome and the focus of these interventions. Broadly speaking, three different types of intervention were

identified in this review, and these will be discussed in turn. Communication training, response teaching and behaviour modification interventions were described in much more detail in the original studies than the other two types of intervention.

Communication training, response teaching and behaviour modification focused on training the parents (usually mothers) to adapt their communication style and the way they respond to their child in order to increase responsivity to their children's communication bids. Parental responsiveness is one of the most important social environmental factors which can influence children's language and general cognitive development both in typically developing children and in children with developmental disabilities (Landry et al. 2001). Parents in all six studies were given training on how to promote language learning, either via strengthening pre-linguistic precursors to language by encouraging joint attention, responding to their children's communication bids and promoting social communication skills, or via direct verbal communication training. Two studies specifically targeted children's expressive vocabulary (Girolametto et al. 1998, Yoder et al. 2014), which is often relatively weaker than receptive vocabulary in children with Down syndrome (Mason-Apps et al. 2020) by situating vocabulary learning within an interactionist, naturalistic context. In both studies there were positive outcomes in vocabulary growth following intervention (or in the case of Yoder et al., following a higher dosage), suggesting that it is possible, through early intervention, to accelerate lexical acquisition of children with Down syndrome. However, a flaw in the Girolametto et al. (1998) is the fact that only the parents in the intervention group knew what the target words were. The Karaaslan and Mahoney (2013) study did not specifically target vocabulary but used a relationship focused intervention which combines elements from the Hanen Parent Programme

with interaction strategies to train parents to promote the development of children's cognitive, communication and social emotional skills by encouraging contingent responding, reciprocity, joint attention, interaction for fun and non-directiveness (i.e., following the child's lead). Positive outcomes were reported in several aspects of the children's general development, including general language skills.

Yoder and Warren's (2002) study focused on developing children's prelinguistic communication (requesting and commenting) because these may elicit more language input from parents, and it is known that optimal parental responses mediate the association between intentional prelinguistic communication and later language (Yoder and Warren 1999) which in turn facilitates word learning. Also, they were interested in children's lexical density, which is the number of different symbols (i.e., sign and spoken words) children use to express different meanings. The children with Down Syndrome in this study decreased the number of requests they used following the intervention. The authors suggested this result may have been found due to the passive nature of children with Down Syndrome and their levels of hypotonicity as requesting requires gross motor skill development. The children with Down Syndrome tended to actively resist prompts to request during the training sessions. It should be noted that the children with Down syndrome were part of a larger group of children with intellectual disabilities and the intervention was targeting specific behaviours rather than specifically children with Down Syndrome. This highlights the importance of having interventions which are tailored to children with Down Syndrome, which are suitable to their behaviour phenotype and can be adapted to the capabilities and strengths of the child, so that we see accelerating rather than deaccelerating of abilities (Neil and Jones 2018).

Behaviour modification and environmental language training were used in two studies, both of which targeted verbal communication. The results were mixed, in that one study reported language gains for the intervention group (Bidder et al. 1975), where the other one (Weller and Mahoney 1983) reported no difference between an oral communication and the total communication groups, although there was an effect of time for both groups, indicating that both groups had made significant gains in language over 5 months. However, as there was no control group, it not possible to conclude that any language gains were due to the intervention.

Early Stimulation Programme: A cluster of studies (Sanz Aparicio 1989, Sanz Aparicio and Balana 2002, 2003, Sanz et al. 2011) refer to stimulation programmes and materials which are hardly described at all. These stimulation programmes targeted children's general development, including fine and gross motor skills, general cognitive skills, and language, but there is no explanation of what exactly the ingredients of the interventions were. The lack of details and information provided about the intervention programmes used severely limits replication of the studies and makes evaluation of the interventions very difficult.

Dialectic-didactic intervention was used in one study (Andrade and Limongi 2007) and focused on developing children's problem-solving abilities through play and promoting verbal communication, however details or examples of targets of the intervention are not provided. The reference for the intervention is a conference presentation and it is not available in English. The intervention did not seem to improve language or communication skills in the intervention group, however, the intervention group only had four participants. Also, it was stated that the participants in the control group were receiving intervention which had a different

methodological/theoretical focus, and it is not clear how this intervention would have affected the outcomes for the control group.

The evidence for positive parent outcomes following early intervention is more consistent. All studies which measured parent behaviour and responsiveness reported positive parent outcomes following the early intervention. Both Karaaslan and Mahoney (2013) and Yoder and Warren (2002) found parents becoming more responsive to child communications acts. Furthermore, Karaaslan and Mahoney (2013), also found improvements to maternal affect and decrease of directedness. Girolametto et al. (1998) reported that mothers in the intervention used labels more frequently although their language complexity did not change.

The implementers of the interventions varied and included speech and language therapists, highly trained researchers and parents or some combination of these (parents and researchers, or parents and clinicians). Parents were involved in delivering the intervention in all but one study. The results indicate that all of these can be successful in implementing an intervention and it is not possible to conclude that trained specialists may be more successful than non-specialist trainers such as parents or researchers.

There was large variability between studies with regard to dosage, hence it is not possible to derive common characteristics with regard to duration and frequency/intensity. It should be noted, however, that intensity was found to be important in Yoder et al. (2014) in that the children in the high intensity group who were receiving intervention 5 times a week had better vocabulary outcomes than those in the active control group who were receiving intervention once a week. It can thus be suggested that high frequency intervention may potentially lead to better outcomes.

Implications for future research

Quality of the evidence, limitations, and implications for future research

There is not sufficient evidence to suggest any particular early intervention. Although 11 studies were included in the review, seven of these did not meet the minimum criteria for quality using the PEDro-P appraisal tool. The collective evidence from the literature is underdeveloped and not sufficiently homogeneous for a meta-analysis. None of the studies addressed the issue of power. The sample sizes were generally small with some studies having only four children with Down Syndrome per group (Andrade and Limongi 2007). Even the studies which had a relatively large number of participants (Sanz Aparicio 1989, Sanz Aparicio and Balana 2002) and potentially more power suffered from severe limitations, such as inadequate information about the intervention methods, little or no description of the outcome measures, no treatment fidelity, no control groups which had the same baseline assessments as the intervention groups, and results were reported by only providing means without any standard deviations or ranges. The consequences of these severe limitations are such that they do not allow replication nor application in the field of clinical practice and hence the field of intervention research does not move forward.

The studies which scored the highest during quality appraisal were all randomised control trials and all used a similar intervention approach which is responsive education/focused stimulation. Interventions based on responsive education/focused stimulation techniques and the Hanen programme embedded within a social context and everyday activities have the potential to result in positive outcomes for children's language and communication. However, caution is needed, as one study reported a decrease in children's social communication skills following this

kind of intervention (Yoder and Warren 2002), and it was hypothesised that this was due to the fact the intervention was not tailored to the communication needs of children with Down syndrome. At this point in time, it is not possible to say whether one intervention is preferred over another. Future research should consider the longer-term effects of parental responsivity/communication style on child speech/language/communication outcomes.

Another potential limitation is that some studies used a language outcome measure based on parent report measures. Very young children often find it difficult to cooperate when formal assessments are administered, or if they need to interact with strangers, which may lead to the assessment being stopped and data not to be collected. Hence parental reports of language, such as the Mac Arthur-Bates Communicative Development Inventory (CDI) (Fenson et al. 2003) are widely used because parents have extensive experience with their children in a variety of naturalistic settings (Feldman et al. 2005). The CDI has a reasonable predictive and concurrent validity. For example, children's scores on the CDI at ages 2 and 3 correlate significantly positively with standardised receptive language measures (Feldman et al. 2005). The concurrent validity of the CDI was established for children with Down syndrome in a study by Miller et al. (1995). The measures of children's language obtained on the CDI and laboratory (standardised) measures of language correlated strongly (between 0.70 and 0.82). However, due to the possible inherent bias in parent report, especially when parents were also administering the intervention, it may be more scientifically reliable if children were assessed using a combination of standardised assessments with well-established parental questionnaires. A combination of measures will allow for more reliable information to be gathered and should give a more accurate view of the child's development and relative changes.

It is also very important that when language/communication measures are reported, it is made clear whether expressive or receptive language is being assessed. Key statistics, such as the mean and standard deviation should also be reported to allow for meta-analysis to be completed. Sometimes language/communication measures used are poorly described, which makes it difficult to combine in a meta-analysis. Furthermore, if a measure was taken at baseline, it needs to be reported post-intervention as well. For example, Girolametto et al. (1998) administered the SICD at baseline but did not report the results post-intervention.

Although positive outcomes have been reported up to 18 months post intervention in a few studies (Sanz Aparicio 1989, Sanz Aparicio and Balana 2002), and there was some evidence that the earlier a child started an intervention the better their language outcomes, none of the studies included longitudinal (over several years) follow up assessments. The follow up was only up to the age of 24 months. Hence it is unclear whether any benefits of the interventions are maintained longer term. Future research should endeavour to include longitudinal follow up assessments to consider the long-term impact of early intervention.

Dosage and duration of interventions varied, and some studies did not report frequency or length of intervention sessions. The number of sessions varied hugely from 13 to over 100 sessions where an intervention lasted for 18 months. Only one study compared high versus low dosage and found evidence in favour of high dosage (Yoder et al. 2014). Due to the lack of other relevant data in the remaining studies, it is not possible to draw conclusions about optimum dosage and frequency.

Conclusions

The common component of all but one of the interventions was parental training and an expectation for parents to implement parts or the whole of the intervention at home by teaching their child, responding contingently, adapting their behaviour to match the child's communication level and needs. Three studies also reported positive changes in parental input and responsivity. Eight studies reported positive language outcomes for the children following intervention. In two out of the 11 studies, the intervention did not seem to improve children's language/communication outcomes, and in one study the children with Down syndrome deaccelerated their social communication skills post-intervention, possibly because the intervention was not specifically tailored for children with Down syndrome. Seven of the 11 studies were low in quality which makes it impossible to replicate these and to fully appraise. Although we can tentatively conclude that early interventions for children with Down syndrome may have positive outcomes, this review highlights the need for higher quality studies.

Declaration of interest: The authors report no conflicts of interest.

Data availability statement

Data sharing is not applicable to this article as no new data were created or analyzed in this study. The data come from published articles which are all listed in the Reference list below.

References

ANDRADE, R. V. & LIMONGI, S. 2007. The emergence of expressive communication in the child with Down syndrome. *Pro-fono: revista de atualizacao cientifica,* 19**,** 387-392.

ANLAR, B. & YALAZ, K. 1996. Denver II Developmental Screening Test Manual: adaptation and standardization for Turkish children. *Ankara: Meteksan*.

BARTON, E.E., & FETTING, A. 2013. Parent-implemented interventions for young children with disabilities: a review of fidelity features. Journal of Early Intervention, 35, 2, 194-210

BIDDER, R. T., Bryant, G. & GRAY, O. P. 1975. Benefits to Down's syndrome children through training their mothers. *Archives of Disease in Childhood*, 50, 383-386

BOOTH, A., & Fry-Smith, A. 2004. Developing the research question. *Etext on Health technology Assessment (HTA) Information Resources*.

BRUNET, O. & LEZINE, I. 1971. Escala para medir el desarrolla psicomotor de la primera infancia. MEPSA, Madrid.

BUCKLEY, S. & BIRD, G. 2001. *Speech and language development for infants with Down syndrome (0-5 years)*, Down Syndrome Educational Trust.

BURGOYNE, K., DUFF, F. J., CLARKE, P. J., BUCKLEY, S., SNOWLING, M. J. & HULME, C. 2012. Efficacy of a reading and language intervention for children with Down syndrome: a randomized controlled trial. *The Journal of Child Psychology and Psychiatry,* 53**,** 1044-1053.

BUSCHMANN, A., JOOSS, B., RUPP, A., FELDHUSEN, F., PIETZ, J. & PHILIPPI, H. 2009. Parent based language intervention for 2-year-old children with specific expressive language delay: a randomised controlled trial. *Archives of Disease in Childhood,* 94**,** 110-116.

CHAPMAN, R. S. & HESKETH, L. J. 2000. Behavioural phenotype of individuals with Down syndrome. *Developmental Disabilities Research Review,* 6, 84-95.

DAVIS-MCFARLAND, E. 2008. Family and cultural Issues in a school swallowing and Feeding program. *Language, Speech, and Hearing Services in Schools,* 39**,** 199-213.

DAWSON, G., ROGERS, S., MUNSON, J., SMITH, M., WINTER, J., GREENSON, J., DONALDSON, A. & VARLEY, J. 2010. Randomized, controlled trial of an intervention for toddlers with autism: the Early Start Denver Model. *Pediatrics,* 125**,** 17-23.

FENSON, L., DALE, P., REZNICK, J., THAL, D., BATES, E., HARTUNG, J. & REILLY, J. 2003. *MacArthur communicative developmental inventories: User's guide and technical manual* (Baltimore, MD:Paul H. Brookes).

FENSON, L., DALE, P., REZNICK, J., THAL, D., BATES, E., HARTUNG, J., PETHICK, S. & REILLY, J. 1993. *MacArthur Communicative Development Inventories (CDI),* (San Diego, CA Singular Publishing Group).

FEY, M. E., YODER, P. J., WARREN, S. F. & BREDIN-OJA, S. L. 2013. Is more better? Milieu Communication Teaching in toddlers with intellectual disabilities. *Journal of Speech, Language, and Hearing Research,* 56**,** 679-693.

FIDLER, D. J. 2005. The emerging Down syndrome behavioural phenotype in early childhood: Implications for practice. *Infants & Young Children,* 18**,** 86-103.

FIDLER, D. J. & NADEL, L. 2007. Education and children with Down syndrome: Neuroscience, development, and intervention. *Mental retardation and Developmental Disabilities Research Reviews,* 13**,** 262-271.

FONG, N. W. Y., HO, S. K. Y., SO, B. J. W. & LIAN, W. B. 2012. Evaluation of the Hanen It Takes Two to Talk Intervention Programme. *Proceedings of Singapore Healthcare,* 21**,** 251-256.

GIROLAMETTO, L., WEITZMAN, E. & CLEMENTS-BAARTMAN, J. 1998. Vocabulary intervention for children with Down syndrome: Parent training using focused stimulation. *Infant-Toddler Intervention,* 8**,** 109-125.

GRIFFITHS, R. 1970. *The abilities of young children. A comprehensive system of mental measurement for the first eight years of life*. Child Development Research Centre, London

HAMPTON, L. H. & KAISER, A. P. 2016. Intervention effects on spoken-language outcomes for children with autism: a systematic review and meta-analysis. *Journal of Intellectual Disability Research* 60**,** 444-63.

HANS, P., ENGLAND, R., PROWSE, S., YOUNG, E. & SHEEHAN, P. 2010. UK and Ireland experience of cochlear implants in children with Down syndrome. *International Journal of Pediatric Otorhinolaryngology,* 74**,** 260-264.

HEDRICK, D. L., PRATHER, E. M. & TOBIN, A. R. 1984. *Sequenced Inventory of Communication Development*, (Seattle: University of Washington Press).

HIGGINS, J., THOMAS, J., CHANDLER, J., CUMPSTON, M., LI, T., PAGE, M. & WELCH, V. E. 2020. *Cochrane Handbook for systematic reviews of interventions version 6.0, 2019* [Online]. Available: www.training.cochrane.org/handbook [Accessed].

HUGHES-MCCORMACK, L. A., MCGOWAN, R., PELL, J., MACKAY, D., HENDERSON, A., O'LEARY, L. & COOPER, S.-A. 2020. Birth incidence, deaths and hospitalisations of children and young people with Down syndrome, 1990–2015: birth cohort study. *BMJ open,* 10**,** e033770.

LANDRY S. H., SMITH, K.E., SWANK, P. R., ASSEL, M. A., & VELLET, S. (2001). Does early responsive parenting have a special importance for children's development or is consistency across early childhood necessary? *Developmental Psychology,* 37, 387-403

LAWS, G. & HALL, A. 2014. Early hearing loss and language abilities in children with Down syndrome. *International Journal of Language & Communication Disorders,* 49**,** 333-342.

LAWS, G., BYRNE, A. & BUCKLEY, S. 2000. Language and memory development in children with Down syndrome at mainstream schools and special schools: a comparison. *Educational Psychology,* 20**,** 447-457.

MAHER, C. G., SHERRINGTON, C., HERBERT, R. D., MOSELEY, A. M., and ELKINS, M., 2003. Reliability of the PEDro scale for rating quality of randomized controlled trials. *Physical Therapy,* 83, 713–721.

MAHONEY, G. D. 1999. Maternal Behaviour Rating Scale. (Tallmadge, OH: Family Child Learning Centre).

MAHONEY, G. & WHEEDEN, C. 1998. The effects of teacher style on the interactive engagement of preschool-aged children with disabilities. *Journal of Developmental and Learning Disorders,* 2**,** 293-315.

MAHONEY, G.J., FINGER, I. & POWELL, A. 1985. Relationship of maternal behavioural style to the developmental of organically impaired mentally retarded infants. American Journal of Mental Deficiency, 90, 3, 296-302.

MAHONEY, G. & WHEEDEN, C. 1998. The effects of teacher style on the interactive engagement of preschool-aged children with disabilities. *Journal of Developmental and Learning Disorders,* 2**,** 293-315.

MANOLSON, A. 1992. *It Takes Two to Talk. A Parent's Guide to Helping Children Communicate,* (Hanen Centre).

MARSHALL, J., GOLDBART, J., PICKSTONE, C. & ROULSTONE, S. 2015. Application of systematic reviews in speech-and-language therapy. *International Journal of Language & Communication Disorders***,** 1-12.

MARTIN, G. E., KLUSEK, J., ESTIGARRIBIA, B. & ROBERTS, J. E. 2009. Language characteristics of individuals with Down syndrome. *Topics in Language Disorders,* 29**,** 112-132.

MASON-APPS, E., STOJANOVIK, V., HOUSTON-RPICE, C., SEAGER, E., & BUCKLEY, S. 2020. Do infants with Down syndrome show an early receptive language advantage? *Journal of Speech, Language and Hearing Research*, 63, 585-598

MILLER, J. & CHAPMAN, R. 1993. SALT: *Systematic Analysis of Language Transcripts*. (Madison, WI: University of Wisconsin).

MOSELEY, A. M., ELKINS, M. R., VAN DER WEES, P. J. & PINHEIRO, M. B. 2019. Using research to guide practice: The Physiotherapy Evidence Database (PEDro). *Brazilian Journal of Physical Therapy***,** 384-391.

MURREY, E., POWEL, E., TOGHER, L., MCCABE, P., MUNRO, N., & SMITH, K. 2013. The reliability of methodological ratings for speechBITE using the PEDro-P scale. *International Journal of Language and Communication Disorders,* 48, 297-306

NAESS, K.-A. B., LERVAG, A., LYSTER, S.-A. H. & HULME, C. 2015. Longitudinal relationships between language and verbal short-term memory skills in children with Down syndrome. *Journal of Experimental Child Psychology,* 135**,** 43-55.

NEIL, N. & JONES, E. A. 2018. Communication intervention for individuals with Down syndrome: Systematic review and meta-analysis. *Developmental Neurorehabilitation,* 21**,** 1-12.

O'TOOLE, C., LEE, A. S. Y., GIBBON, F. E., VAN BYSTERVELDT, A. K. & HART, N. J. 2018. Parent-mediated interventions for promoting communication and language development in young children with Down syndrome. *Cochrane Database of Systematic Reviews (10).*

O'TOOLE, C. & CHIAT, S. 2006. Symbolic Functioning and Language Development in Children with Down Syndrome. *International Journal of Language & Communication Disorders,* 41**,** 155-171.

PEPPER, J. & WEITZMAN, E. 2004. *It takes two to talk: A practical guide for parents of children with language delays,* (The Hanen Centre).

PERDICES, M., SAVAGE, S., TATE, R., MCDONALD, S. & TOGHER, L. 2009. *Rater's manual for between-group studies (RCTS and NonRCTs): introduction to the Physiotherapy Evidence Database (PEDro) Scale for rating methodological Quality, Adapted for PsycBITE (Pedro-P),* (Sydney, NSW, Australia: University of Sydney).

PETTICREW, M. & ROBERTS, H. 2008. *Systematic reviews in the social sciences: A practical guide,* (John Wiley & Sons).

PRING, T. 2004. Ask a silly question: two decades of troublesome trials. *International Journal of Language & Communication Disorders,* 39**,** 285-302.

SACHSE, S. & VON SUCHODOLETZ, W. 2008. Early identification of language delay by direct language assessment or parent report? *Journal of Developmental & Behavioral Pediatrics,* 29**,** 34-41.

SANZ APARICIO, M. T. 1989. Effects of observation training of parents in the early stimulation of trisomy-21 babies. *Early Child Development and Care,* 41, 89-101

SANZ APARICIO M. T. & BALANA, M. 2002. Early language stimulation of Down's syndrome babies: a study on the optimum age to begin. *Early Child Development and Care*, 172, 651-656

SANZ APARICIO M. T. & BALANA, M. 2003. Social early stimulation of trisomy –21 babies. *Early Child Development and Care*, 173, 5, 557-561

SANZ, T., MENENDEZ, J. & ROSIQUE, T. 2011. Study of different social rewards used in Down's syndrome children's early stimulation. *Early Child Development and Care,* 181. 4. 487-492

SAVAŞıR, I., SEZGIN, N. & EROL, N. 2005. *Ankara Developmental Screening Inventory Manual*, (Ankara).

SMITH, E., HOKSTAD, S. & NÆSS, K.-A. B. 2020. Children with Down syndrome can benefit from language interventions; Results from a systematic review and meta-analysis. *Journal of Communication Disorders***,** 105992.

SNOWLING, M. J., HULME, C., BAILEY, A. M., STOTHARD, S. E. & LINDSAY, G. 2011. Better Communication Research Project: Language and Literacy Attainment of Pupils during Early Years and through KS2--Does Teacher Assessment at Five Provide a Valid Measure of Children's Current and Future Educational Attainments? Research Report. DFE-RR172a. ERIC.

STOEL-GAMMON, C. 2001. Down syndrome phonology: Developmental patterns and intervention strategies. *Down Syndrome Research and Practice,* 7**,** 93-100.

TE KAAT-VAN DEN OS, D. J., JONGMANS, M. J., VOLMAN, M. & LAUTESLAGER, P. E. 2017. Parent-implemented language interventions for children with a developmental delay: A systematic review. *Journal of Policy and Practice in Intellectual Disabilities,* 14**,** 129-137.

TOMASELLO, M. 2003.*Constructing a Language: A Usage-Based Account of Language Acquisition* (Cambridge, MA).

VAN BYSTERVELDT, A., GILLON, G. & FOSTER-COHEN, S. 2014. A phonological awareness intervention case study of a child with Down syndrome. *Speech, Language and Hearing,* 17**,** 25-36.

WELLER, E.L. & MAHONEY, G. J. 1983. A comparison of oral and total communication modalities on the language training of young mentally handicapped children. Communication Modalities, 18, 103-110

WREN, Y., HARDING, S., GOLDBART, J. & ROULSTONE, S. 2018. A systematic review and classification of interventions for speech-sound disorder in preschool children. *International Journal of Language and Communication Disorders,* 53, 3, 446-467

YODER, P., WOYNAROSKI, T., FEY, M. & WARREN, S. 2014. Effects of Dose Frequency of Early Communication Intervention in Young Children with and without Down Syndrome. *American Journal on Intellectual and Developmental Disabilities,* 119**,** 17-32.

YODER, P. J. & WARREN, S. F. 2002. Effects of prelinguistic milieu teaching and parent responsivity education on dyads involving children with intellectual disabilities. *Journal of Speech, Language, and Hearing Research,* 45**,** 1158-1174.

YODER, P. J. & WARREN, S.F. 1999. Maternal responsivity mediates the relationship between prelinguistic intentional communication and later language. *Journal of Early Intervention,* 22, 126-136

YODER, P. J. & WARREN, S. F. 1998. Maternal responsivity predicts the prelinguistic communication intervention that facilitates generalized intentional communication. *Journal of Speech, Language, and Hearing Research,* 41**,** 1207-1219.