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Parent-mediated communication interventions for improving the communication skills of preschool children with non-progressive motor disorders (Review)

Pennington L, Akor WA, Laws K, Goldbart J

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[Intervention Review]

Parent-mediated communication interventions for improving the communication skills of preschool children with non-progressive motor disorders

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ABSTRACT

Background

Children with motor disorders can have difficulties in producing accurate and consistent movements for speech, gesture or facial expression (or a combination of these), making their communication difficult to understand. Parents may be offered training to help recognise and interpret their child's signals and to stimulate their children's development of new communication skills.

Objectives

To assess the effectiveness of parent-mediated communication interventions, compared to no intervention, treatment as usual or clinician-mediated interventions, for improving the communication skills of preschool children up to five years of age who have non-progressive motor disorders.

Search methods

We searched CENTRAL, MEDLINE, Embase, CINAHL, PsycINFO, 12 other databases and three trials registers in July 2017. We also searched the reference lists of relevant papers and reviews, and contacted experts working in the field to find unpublished studies.

Selection criteria

We included studies that used randomised or quasi-randomised designs; compared a parent-mediated communication intervention with no treatment, treatment as usual or clinician-mediated therapy; and included children with non-progressive motor disorders up to five years of age.

Data collection and analysis

We used the standard methodological procedures expected by Cochrane.

Main results

This review included two randomised controlled trials involving 38 children (20 boys, 18 girls), aged 15 to 96 months, and their mothers. All children had developmental disabilities; 10 had motor disorders, but it was unclear if these motor disorders affected their gestural, vocal or verbal communication. Mothers attended eight group training sessions over 11 to 12 weeks and received two or three home visits. Outcomes were assessed immediately after training. We found no report of longer-term follow-up. One study took place at an intervention centre in Canada and the other in South Korea.

Both studies recruited small numbers of participants from single centres. Since it is not possible to blind participants attending or therapists providing training to group allocation, we considered both studies to be at high risk of performance bias. We also rated one study at high risk of attrition bias, and both studies at low risk of reporting bias.

There was very low-quality evidence for all outcomes assessed. There was no evidence of an effect of training for children's initiation of conversation or engagement in joint attention during interaction with their mothers. Mothers who received training became more responsive to their children's communication, but there were no differences in the extent to which they controlled conversation by directing their children. Missing data meant that we were unable to evaluate the effects of training on children's frequency of communication, frequency of spoken language in conversation, speech production, or receptive or expressive language development. There were no effects on maternal stress. We found no reports of the effects of parent training on children's use of individual communication skills, such as asking questions or providing information, on their generic participation or adverse outcomes. Neither did we find reports of mothers' satisfaction with treatment, its acceptability or their compliance with it.

Authors' conclusions

There is only limited, very low quality evidence that parent-mediated communication interventions may be associated with improvements in interaction between mothers and their preschool children who have motor disorders. The indirectness of the study samples and high risk of bias in the included the studies significantly limits our confidence in the evidence, as do issues with study design and lack of detail in results. It is not clear if training has been tested with children whose motor disorders limit the consistency and accuracy of movements underpinning spoken or gestural communication. Some speech and language therapists currently provide communication training for parents. Further research, with larger numbers of children whose movement disorders affect their speech and gestures, coupled with detailed reporting of children's baseline skills, is needed to test whether communication training for parents can help them to promote the communication development of their young children with movement disorders.

PLAIN LANGUAGE SUMMARY

Communication training for parents of preschool children with motor disorders

Background

Children with movement disorders, such as cerebral palsy, often have difficulty producing speech and gesture. This can make their communication difficult to understand. In the preschool years, speech and language therapy often involves training parents to recognise their child's communication signals and promote communication development.

Review question

Does communication training for parents (parent-mediated communication intervention) of preschool children with movement disorders improve the communication between children and parents? We were also interested in whether the training had any unintended consequences, whether it had an effect on parents' levels of stress and coping, and whether parents were satisfied with the training and complied with it.

Study characteristics

We searched for studies published up to July 2017. We found only two studies that reported the effects of parent communication training; one study took place at an intervention centre in Canada, the other in South Korea. The studies involved 38 children (20 boys, 18 girls), aged 15 to 96 months, and their mothers. Both studies compared parent communication training with no intervention for communication problems. Mothers attended eight group training sessions 11 to 12 weeks with two or three home visits. The studies involved children with a range of developmental difficulties; most had intellectual disability, 10 had movement disorders (cerebral palsy). However, the extent to which children's movement disorder affected their communication was not clear; all children appeared to have good use of their hands for gesture and pointing, and impairment of speech was not reported.

Results were assessed immediately after training. We found no report of results at a later date (longer-term follow-up).

Key results and the quality of the evidence

In the two small studies, it appears that mothers may have responded more frequently to their child's interaction following parent-mediated communication training. However, there was no associated reduction in mothers' directiveness (such as their use of commands) in conversation and no change in maternal stress. For the children, we found no evidence for change in children's initiation of conversation or of joint attention in interaction with others. Studies did not report any negative effects of training, mothers' adherence to guidance within the training or the acceptability of the programmes.

We were not able to evaluate the effects of parent-mediated communication intervention and frequency of children's communication, their use of spoken language in conversation with their parents, their speech production or their language development because the data were not available. We have no reports of children's development of individual communication skills, such as learning to ask questions, and no reports of defects of the intervention on their generic participation or harms arising from the intervention. Finally, we found no reports of maternal satisfaction with the treatment.

We judged the evidence from the included studies to be of very low quality because of issues with study design and a lack of detail in the results presented, and because it was not clear whether children's movement disorders affected their communication.

Research with larger numbers of families of children whose movement disorders affect their speech and gesture is needed, to test whether communication training for parents can help them to promote the communication development of their young children with movement disorders.

SUMMARY OF FINDINGS FOR THE MAIN COMPARISON [Explanation]

Parent-mediated communication intervention compared with no intervention for improving the communication skills of preschool children with non-progressive motor disorder

Patient or population: children up to 5 years of age with a communication difficulty associated with any non-progressive motor disorder acquired before 2 years of age
Settings: speech and language therapy clinic
Intervention: parent-mediated communication intervention on promoting communication development
Comparison: no intervention

Outcomes	Impact	Number of participants (studies)	Quality of the evidence (GRADE)	Comments
Children's ability to communicate effectively in everyday life: taking turns in conversation, initiating conversation and responding to others' conversational gambits Assessed with: observational scales of child interaction - Child Behavior Rating Scale (CBRS; Mahoney 1999b) (therapist rating of child engagement with parent in play in 7 domains (interest, attention to activity, persistence, initiation, affect, joint attention, cooperation). Items scored 1 to 5 on Likert scale; lower scores represent lower levels of coded behaviours) Follow-up: postintervention (time not specified)	1 study found no evidence of effect of parent-mediated intervention on children's initiation of conversation (MD −0.25 points, 95% CI −1.01 to 0.51; P = 0.54), or joint attention during interaction (MD 0.05 points, 95% CI −0.52 to 0.62; P = 0.87)	18 (1 study)	⊕◯◯◯ **Very low**[a,b,c]	The CBRS involved a subjective rating of child's engagement in interaction. For this review, we were interested in the initiation and joint attention domains of the CBRS, as these relate directly to communication
Adverse events	Not measured			
Child outcomes				

Speech and language function Assessed with: Sequenced Inventory of Communication Development (Hedrick 1975) (age-normed assessment (4-48 months); lower score represents lower levels of communication development) Follow-up: postintervention (time not specified)	1 study measured the effect of parent-mediated communication intervention on children's speech and language function but missing data prevented analysis	20 (1)	⊕◯◯◯ **Very low**[a,b]	1 included study reported a general communication ability measure
Children's generic participation: social	Not measured			
Parent outcomes				
Parents' communication and interaction strategies: directiveness and responsiveness Assessed with: Maternal Behavior Rating Scale (MBRS; Mahoney 1999a) (therapist rating of maternal behaviour on 12 items in 4 domains (responsiveness, affect, achievement orientation and directiveness). Items scored 1-5 on Likert scale; lower scores represent lower levels of coded behaviours) Follow-up: postintervention (time not specified)	1 study found increases in parents' responsiveness (MD 1.02 points, 95% CI 0.34 to 1.70; P = 0.01), but no effect on their directiveness (MD 0.49 points, 95% CI −0.02 to 1.00; P = 0.07)	18 (1 study)	⊕◯◯◯ **Very low**[a,b,c]	The MBRS involves a subjective rating of parent's engagement in, and control of, interaction with the child. For this review, we were interested in the MBRS domains of responsiveness and directiveness, as these relate directly to the communication opportunities provided to children by parents in conversation
Family stress and coping: parent stress Assessed with: Korean version of the Parent Stress Index (Shin 1997) (parent-reported measure. Total Stress score calculated	1 study found no effect of parent-mediated training in reducing parental stress related to their child's temperament (MD −6.50 points, 95% CI −11.64 to 1.36) , parent-child relationship (MD 0.	18 (1 study)	⊕◯◯◯ **Very low**[a,c]	1 included study reported outcomes regarding the potential impact of communication difficulties on parents' psychological well-being. Prior to intervention, parents in the training group ap-

from 20 items in each of 3 scales (child temperament, parent-child relationship, learning expectation). Each item rated on 5-point Likert scale to indicate the extent to which items disturbed the parent in last week; lower scores indicate lower levels of disturbance) Follow-up: postintervention (time not specified)	25 points, 95% CI −4.70 to 5.20; P = 0.92), or learning expectation (MD −0.58 points, 95% CI −2.87 to −1.71)		peared to have higher levels of stress regarding their relationship with their child and lower levels of stress regarding their child's temperament than parents in the control group
Satisfaction of child and family with treatment	Not measured		
Compliance with treatment	Not measured		

*The basis for the **assumed risk** (e.g. the median control group risk across studies) is provided in footnotes. The **corresponding risk** (and its 95% CI) is based on the assumed risk in the comparison group and the **relative effect** of the intervention (and its 95% CI)
CI: confidence interval; **CBRS:** Child Behavior Rating Scale; **MD:** mean difference

GRADE Working Group grades of evidence
High quality: we are very confident that the true effect lies close to that of the estimate of the effect.
Moderate quality: we are moderately confident in the effect estimate: the true effect is likely to be close to the estimate of the effect, but there is a possibility that it is substantially different.
Low quality: our confidence in the effect estimate is limited: the true effect may be substantially different from the estimate of the effect.
Very low quality: we have very little confidence in the effect estimate: the true effect is likely to be substantially different from the estimate of effect

[a]Downgraded by two levels due to risk of bias, and one level for indirectness. Risk of selection bias was unclear. No information was given on the allocation of participants to groups, although in one study, Kim 2005, parents and children in the parent-mediated intervention appeared to have higher levels of interaction difficulties than those in the control group prior to intervention. All children in the studies had developmental delays. Four children in Girolametto 1988 and six children in Kim 2005 had motor disorders (e.g. cerebral palsy). Participants in both studies were able to use their hands. It was not clear how far the results extended to children with motor disorder that impairs gesture and speech.

[b]Risk of bias. Girolametto 1988 did not state if outcome assessors were blind to group or timing of data collection.

[c]Risk of bias. Kim 2005 blinded outcome assessors to group and timing of data collection.

BACKGROUND

Description of the condition

Non-progressive motor disorders in childhood arise from a variety of conditions, including cerebral palsy, acquired brain injury, global developmental delay, Down's syndrome and genetic mutations. Exactly how many children are affected is currently unknown due to sparse population-level data. The most comprehensive data come from international surveillance of cerebral palsy. Cerebral palsy is defined as "a group of permanent disorders of the development of movement or posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain" (Rosenbaum 2007). Registries have shown that cerebral palsy affects two or three children per 1000 children in high-income countries (Cans 2008; Kirby 2011; Reid 2011). Prevalence is likely to be greater in low- and middle-income countries where health care is less abundant, but cerebral palsy rates in these regions have not yet been ascertained. One Dutch study estimated that, in children under 14 years of age, there are about 3.6 new cases of severe acquired brain injury per 100,000 children per annum (De Kloet 2013). However, it is unclear how many of these children have ongoing motor disorders. Again, the incidence may be greater in low- and middle-income countries with less access to maternal and child health services. Global developmental delay and Down's syndrome lead to low motor tone and slow acquisition of motor skills, and global developmental delay affects approximately 39 per 1000 children and Down's syndrome affects 1.4 per 1000 children (Boyle 2011; Parker 2010). Genetic mutations that cause motor disorders include the PRRT2 (proline-rich transmembrane protein 2) mutation and the GLUT-1 (glucose transporter type 1) syndrome (Blackburn 2012), but their prevalence is unclear. Many developmental disabilities, including those causing non-progressive motor disorders included in this review, are more common in boys and in families in who live in poverty (Boyle 2011). Disorders are diagnosed by paediatricians, paediatric neurologists and geneticists. Differential diagnosis may take some years due to the slowly evolving nature of some conditions.

Motor disorders impair the range, speed, strength and consistency of movements. When disorders affect the movements underpinning vocalisation, speech, gesture or facial expression (or a combination of these), parents and other carers find it difficult to recognise and interpret children's attempts to communicate, and this can lead to a breakdown in interaction (Hanzlik 1990; Light 1985; Pennington 2001). To promote effective interaction, parents may structure conversations around the children's communication signals that are easy to understand (Dunst 1985; Tannock 1992). However, this can lead to asymmetrical interaction, with parents introducing topics, asking forced, choice questions and then acknowledging their child's response. Such an uneven, parent-led pattern of conversation can make it difficult for children with motor disorders to learn new communication skills.

It is estimated that around 20% to 30% of children with cerebral palsy have no functional speech and a further 22% are able to speak but have intelligibility limitations due to their motor disorders (ACPR 2016; Nordberg 2013; Parkes 2010). Speech disorder is most likely to occur in bilateral spastic-type cerebral palsy (where muscle tone is increased on both sides of the body), in dyskinetic forms (where muscle tone fluctuates) and in ataxic forms (characterised by tremor and difficulties with muscular co-ordination) (Bax 2006; Parkes 2010). The prevalence of speech disorders in other conditions leading to non-progressive motor disorders is currently unknown.

Children with motor disorders who also have a cognitive impairment may take longer to reach milestones, such as intentionality and engaging in joint attention with another person, which are vital for interaction, and the development of linguistic understanding may be delayed. About 49% of children with cerebral palsy have an intellectual disability (IQ less than 70) and 28% have a severe intellectual disability (IQ less than 50) (Novak 2012). Current research suggests that receptive language is largely commensurate with cognitive development in cerebral palsy (Pirila 2007), but further epidemiological studies are needed to confirm this. Approximately 12% of children with cerebral palsy have hearing impairment and 35% have some visual impairment (ACPR 2016), both of which can influence communication development.

Communication difficulties have a profound impact on children's family, social and educational life. Children with communication and motor disorders are more at risk of lower quality of life and restricted social participation than their peers with and without motor disorders (Dickinson 2007; Fauconnier 2009). The impact of communication breakdown is felt throughout families, and parents report high levels of stress (Parkes 2011; Pousada 2013).

As differential diagnosis may not be possible in early childhood, and all motor disorders affecting speech and gesture can lead to intelligibility limitations, this review will be inclusive of all causes of non-progressive motor disorders in the preschool years. One exception to this is children with Down's syndrome, who will be considered in a separate review (O'Toole 2016). Thus, we will exclude studies examining only children with Down's syndrome, but will include studies in which Down's syndrome is one of a range of disorders causing motor impairment. Degenerative disorders, such as muscular dystrophies and metabolic disorders, may also be associated with motor impairment and may become apparent after a period of healthy development. As these disorders lead to a loss of skills rather than development following an atypical pattern, as is the case for children with non-progressive disorders, they were excluded from this review. Also, children with severe hearing or visual impairments, or both, have specific difficulties acquiring early interaction skills arising from their differences in processing communication signals, which are beyond the scope of this review.

Description of the intervention

Communication skills are developed in interaction, and children's most frequent communication partners are their parents, so therapy involves training parents to adapt their communication style. This is referred to as parent-mediated or indirect therapy. It aims to help parents of children with motor disorders to recognise and interpret their children's idiosyncratic attempts to communicate and to stimulate their child's development of new skills (e.g. Bruno 1998; Kaiser 1987; Kent-Walsh 2015; Mahoney 1988; Pepper 2004; Yoder 2002). Training is most often provided by speech and language therapists and other personnel with an interest in interaction (e.g. psychologists and early-years educators). It can be delivered to individuals or groups of parents and may take place in parents' homes or in health, education or social care settings.

Training often teaches parents about how communication develops, from preintentional communication through to non-verbal, intentional communication and then on to linguistic communication (Girolametto 1986; Hemmeter 1994; Pepper 2004). It covers the purposes for which communication is used, how communication involves communication partners taking turns in expressing and receiving signals (Hemmeter 1994; Mahoney 1988; Pepper 2004), and how messages are coconstructed by communication partners (Clarke 2017). Techniques to aid children's language and communication development are introduced, including how to create simulating environments, promote a need to communicate, allow sufficient time for children to join or start conversations, respond contingently to children's messages, and repeat language to aid association between spoken words and their referents. Parents are encouraged to apply this information to their interaction with their children (Fey 2006; Gibbard 2004; Girolametto 1996; Kaiser 2001; Mahoney 1988; Pepper 2004). Training often includes one-to-one coaching, whereby therapists watch the interaction between a parent and a child (either in real time or on video) and highlight which behaviours prompted the child to communicate so the parent can repeat these in daily interactions (Kaiser 1995; Kaiser 2003; McDuffie 2016; Pepper 2004). Coaching might also involve the parent watching the therapist modelling the interaction with the child (Kaiser 2003; Pepper 2004).

Young children who have severe speech impairment associated with their motor disorders may be introduced to augmentative and alternative communication (AAC) to supplement or replace their natural forms of communication. AAC includes signing, gesture and use of body movements (unaided AAC), or may introduce equipment such as objects to represent daily activities, photographs, pictures, symbols, speech generating devices and mobile technology communication applications (aided AAC). AAC provides access to a wider range of vocabulary and language but takes children and their parents time to learn. Conversation using AAC often involves the coconstruction of meaning by parents and children as children have access to a restricted vocabulary (Clarke 2017; Solomon-Rice 2011). Training is often provided for parents to teach them how to accommodate the use of the system in conversation and help their children to produce new vocabulary and language structures via the AAC system (Kent-Walsh 2015). Such training may be incorporated in the generic communication training described above (Pennington 2009), or be provided separately in programmes that focus specifically on AAC (Kent-Walsh 2015).

How the intervention might work

Parent communication training is based on the transactional theory of development, which hypothesises that children and their parents continuously adapt to each other's behaviours (Sameroff 2000). Children with motor disorders communicate using movements and vocalisations/words that may look and sound very different to those of their typically developing peers. Following the transactional hypothesis, helping parents to recognise and interpret their children's idiosyncratic communication behaviours should lead to parents' responding appropriately to children's messages. Such positive responses should, in turn, encourage children to repeat these behaviours and communicate more frequently. Teaching parents about how communication develops should enable them to apply this knowledge to continue to adapt their own communication patterns and provide ongoing support for their children's development from the beginnings of intentional communication to the understanding and use of multi-word utterances (Girolametto 1996). When children are just beginning to communicate intentionally, parents would follow their children's focus of attention and give their children more time to start interactions and produce messages. Children would be encouraged to jointly attend to an object/activity with their parents. Hearing their parents say the names of objects and actions during joint attention would help children to associate the spoken words with their referents to learn language. Over time, parents would introduce more varied play and communication opportunities and hence a need for children to communicate for a greater range of purposes, with a wider range of vocabulary and language structures. For children with limited spoken output, such vocabulary would be supported by the use of AAC; for example by parents pointing to symbols or pictures as well as speaking. In these approaches to therapy, there is an implicit or explicit assumption that changing parents' interaction style will have an effect on children's communication, expressive or receptive language (or both), and interaction (Pickstone 2009). The intervention may also serve to increase parents' confidence in their communication with their children, reduce parental stress as communication breakdowns become less frequent, and help children to interact successfully in a greater number of social activities and with a broader range of people.

Why it is important to do this review

The James Lind Alliance Childhood Disability Research Priority Setting Partnership rated the timing and intensities of interventions, and the effectiveness of communication interventions as the two most important areas for investigation (Morris 2015). Internationally, there has been a drive in research to develop early interventions to maximise the potential skill development associated with brain plasticity in infancy and the early years. Early communication intervention has often focused on training parents to provide parent-mediated intervention (Watson 2015). Previous Cochrane Reviews have considered parent-mediated intervention training programmes for children with autism (Oono 2013), and primary speech and language delay or disorder (Law 2003), and a current review is investigating for parent-mediated intervention children for with Down's syndrome (O'Toole 2016). However, the method of delivery of parent communication training, its contents, dosage and suitability for families of children with motor disorders have not been evaluated. A previous review considered speech and language therapy interventions to improve the communication skills of children with cerebral palsy and included parent-mediated interventions (Pennington 2003); its authors identified one randomised controlled trial (RCT) of a parent-mediated communication intervention. This review updates the section of the previous review that examined training delivered to parents of children with cerebral palsy (Pennington 2003), to identify new empirical data. It also considers interventions provided to parents of preschool children with other non-progressive motor disorders, as their communication development is similarly affected. By including all children with non-progressive motor disorders, we aimed to examine the generic effectiveness of parent-mediated training interventions in the preschool period, extending the utility of the review to service providers and policymakers.

OBJECTIVES

To assess the effectiveness of parent-mediated communication interventions, compared to no intervention, treatment as usual or clinician-mediated interventions, for improving the communication skills of preschool children up to five years of age who have non-progressive motor disorders.

METHODS

Criteria for considering studies for this review

Types of studies

RCTs, including cluster-RCTs, and quasi-RCTs in which participants were allocated to intervention groups by methods that were not strictly random (e.g. date of birth).

Types of participants

Children up to five years of age who had a communication difficulty associated with any non-progressive motor disorder acquired before two years of age. We included children with additional intellectual impairments, including children with Down's syndrome, children whose vision was corrected by spectacles and children whose hearing was amplified by hearing aid(s) if they had identified motor difficulties. We included studies of children with intellectual/developmental disability, where at least one child in each group had additional motor disorders, as other participants would be likely to have delayed motor skills. Subgroup analysis for children with motor disorders was not mandatory. We excluded studies of only children with Down's syndrome, as they are being considered by another review (see O'Toole 2016). We also excluded children whose communication was primarily limited by a sensory impairment, as their communication development differs from children who can see and hear the world around them. We inferred motor disorder from descriptions of children's development and confirmed this with study authors, where possible. Where information about this was provided, children's communication difficulty was diagnosed by speech and language therapists or psychologists.

Parents of the children described above.

Types of interventions

We considered studies of training delivered to parents with the explicit aim of helping them to promote their child's communication development. Training could have been delivered to parents individually or in groups. Training could have been delivered by speech and language therapists, psychologists, early educators or others with specialist knowledge of communication. Training could have taken place in the home or in health, education or community support settings. Training programmes could have varied in dosage (intensity, frequency and duration). Training could have included communication via AAC as one mode of communication in a total communication approach or focus primarily on communication using AAC. We excluded facilitated communication, as it is not a valid form of AAC (Schlosser 2014).

Comparisons of interest were: parent-mediated intervention versus no intervention or waiting-list controls; parent-mediated intervention versus treatment as usual (e.g. multidisciplinary therapy groups providing motor, sensory and language stimulation); parent-mediated intervention versus clinician-mediated intervention.

Types of outcome measures

Primary outcomes

1. Children's ability to communicate effectively in everyday life. Outcomes included children's ability to:

i) take turns in conversation, initiating conversation and responding to others' conversational gambits;

ii) use communication for a wide range of purposes such as requesting attention, asking questions, answering questions, making comments and repairing conversation when they have not been understood; and

iii) use a range of modes of expression by vocalising, speaking, using gesture or using the AAC system.

2. Adverse events, including reductions in the frequency with which children communicate, or increases in negative behaviour. Outcomes could have been measured at the level of activity (i.e. the ability to execute a task), and at the level of participation (i.e. communication in life situations) (WHO 2001).

Measures could have included rating scales (e.g. Focus on the Outcomes of Communication Under Six (Thomas-Stonell 2010) and Therapy Outcome Measures (Enderby 2015)), communication assessments (e.g. Communication and Symbolic Behavior Scales (Wetherby 2002)), and observational coding schemes (e.g. frequency counts of children's initiations and responses in interaction).

Secondary outcomes

1. Child outcomes:

i) speech and language function, assessed using standardised measures of children's expressive and receptive language skills and speech production (e.g. Pre-school Language Scales (Zimmerman 2002); Communicative Development Inventory (CDI; Fenson 2006); Receptive-Expressive Emergent Language Scale-3 (REEL-3; Bzoch 1970)); non-standardised assessments of gestural ability; or production of messages using AAC on demand, measured using coding schemes developed for individual research studies that include validity and reliability data; and

ii) children's generic participation, assessed using validated measures (Assessment of Life Habits (Noreau 2007), and Children's Assessment of Participation and Enjoyment (King 2004)).

2. Parent outcomes:

i) parents' communication and interaction strategies, assessed using non-standardised measures (Responsive Augmentative and Alternative Communication Style (Broberg 2012); coding schemes that measured the frequency of parent communication behaviours (e.g. initiations of conversation; directives) developed for individual research studies that included validity and reliability data);

ii) family stress and coping (e.g. Questionnaire on Resources and Stress (Friedrich 1983) or Carer Strain Index (Robinson 1983));

iii) satisfaction of child and family with treatment (e.g. rating scales developed for individual studies, Patient Satisfaction Questionnaire Short Form (PSQ-18; Marshall 1994)); and

iv) compliance with treatment (e.g. number of sessions missed and reasons for this).

See Table 1 for methods stipulated in the protocol that were not required in this version of the review, but which will be applied in future versions.

Search methods for identification of studies

Electronic searches

In July 2017, we searched the electronic databases and trials registers listed below, from inception onwards.

1. Cochrane Central Register of Controlled Trials (CENTRAL; 2017, Issue 6) in the Cochrane Library, which includes the Cochrane Developmental, Psychosocial and Learning Problems Specialised Register (searched 26 July 2017).
2. MEDLINE Ovid (1946 to July week 2 2017).
3. MEDLINE In-Process & Other Non-Indexed Citations Ovid (searched 25 July 2017).
4. MEDLINE Epub Ahead of Print Ovid (searched 25 July 2017).
5. Embase Ovid (1974 to 2017 week 30).
6. CINAHL Plus EBSCOhost (Cumulative Index to Nursing and Allied Health Literature; 1937 to 26 July 2017).
7. PsycINFO Ovid (1806 to July week 3 2017).
8. Science Citation Index Web of Science (SCI; 1970 to 26 July 2017).
9. Conference Proceedings Citation Index - Science Web of Science (CPCI-S; 1970 to 26 July 2017).
10. Language and Linguistic Behaviour Abstracts ProQuest (LLBA; 1871 to 3 August 2017).
11. British Education Index EBSCOhost (BEI; 1929 to 26 July 2017).
12. ERIC EBSCOhost (Education Resources Information Center; 1966 to 26 July 2017).
13. *Cochrane Database of Systematic Reviews* (CDSR; 2017, Issue 7) part of the Cochrane Library (searched 26 July 2017).
14. Database of Abstracts of Reviews of Effects (DARE; 2015, Issue 2) part of the Cochrane Library (searched 26 July 2017).
15. LILACS (Latin American and Caribbean Health Science Information database; lilacs.bvsalud.org/en; searched 26 July 2017).
16. Rehabdata National Rehabilitation Information Center (naric.com; searched 27 July 2017).
17. SpeechBITE (speechbite.com; searched 27 July 2017).
18. ClinicalTrials.gov (clinicaltrials.gov; searched 27 July 2017).
19. UK Clinical Trials Gateway (www.ukctg.nihr.ac.uk/clinical-trials; searched 27 July 2017).

20. World Health Organization International Clinical Trials Registry Platform (WHO ICTRP; www.who.int/ictrp/en; searched 27 July 2017).

Search strategies for each source are in Appendix 1. We did not limit the search by the country in which the research was undertaken, the language in which the research was reported, year of publication or publication status. See Table 1 for methods stipulated in the protocol that were not required in this version of the review, but which will be applied in future versions.

Searching other resources

We handsearched the reference lists of relevant papers and reviews for studies not identified by the electronic searches. We approached authors working in the field to locate currently unpublished studies.

Data collection and analysis

We did not use all of the methods stipulated in our published protocol (Pennington 2017), either because these were not relevant (e.g. we did not encounter cluster-RCTs) or because they could not be deployed (e.g. subgroup analyses). Table 1 summarises the methods section of our protocol, which will be applied in updates of this review if possible. Here, we report only those methods deployed in this review.

Selection of studies

Two review authors (WA and LP) independently screened each title and abstract for eligibility against the inclusion criteria (see Criteria for considering studies for this review). In the event of a disagreement, resolution was reached in discussion with a third author (JG). When inclusion was uncertain, we obtained the full-text reports. Pairs of review authors (WA, JG, KL or LP) were randomly allocated to each report that appeared from the abstract to fit the inclusion criteria and independently reviewed each one to determine its inclusion. In the event of disagreement regarding inclusion, a third review author (WA, JG, KL or LP) reviewed the report independently and we reached consensus through discussion and by reassessing the inclusion criteria together.

We recorded the selection process in a PRISMA diagram (Moher 2009).

Data extraction and management

All review authors were involved in data extraction. Two of the four review authors (WA, JG, KL or LP) were randomly assigned to each report and independently extracted data, imputing it into Review Manager 5 (Review Manager 2014). We developed and piloted a data extraction form for the study to collect data on the following.

1. Country of origin.
2. Type of study: RCT; cluster-RCT; quasi-RCT.
3. Sample size: treatment and control groups; attrition.
4. Study population: parents (age, gender, relationship to child, educational level (high school, further education, higher education)); children (diagnosis of underlying disorder, type of motor disorder (spastic, dyskinetic, ataxic, hypotonic, mixed); age; gender; non-verbal cognitive development (standard scores, percentile rank); receptive language development (standard scores, percentile rank); modes of communication used (vocalisation, speech, gesture, facial expression, body movement, AAC); communicative functions used; number of intelligible words; gross motor function, classified using the Gross Motor Function Classification System (GMFCS; Palisano 2007); and upper limb function, categorised using the Manual Ability Classification System (MACS; Eliasson 2006), when possible.
5. Intervention: type of intervention; duration; frequency of sessions; group or individual; content of sessions; inclusion of coaching or didactic teaching only.
6. Comparator intervention: type of intervention; duration; frequency of sessions; group or individual; content of sessions; inclusion of coaching or didactic teaching only.
7. Intervention provider: speech and language therapist (or relevant term in country of origin); psychologist; teacher; other.
8. Fidelity of intervention: how this was assessed and by whom.
9. Outcome measures: parent outcomes; child outcomes; family outcomes.
10. Results: short term (zero to one month following intervention completion), medium term (two to five months after intervention) and long term (six or more months following intervention).
11. Adverse events.
12. Conflicts of interest, including declarations of conflicts of interest.

We resolved disagreements by discussion and by involving a third review author (WA, JG, KL or LP), when necessary.

Assessment of risk of bias in included studies

We rated the risk of bias of each included study using the Cochrane 'Risk of bias' tool (Higgins 2011a). Pairs of review authors (WA, JG, KL or LP) were randomly allocated to each study to extract data and rate risk of bias. Disagreements were resolved through discussion or by involving a third review author (WA, JG, KL or LP). We rated studies as having low, high or unclear risk of bias in: random sequence generation; allocation concealment; blinding of participants and personnel; blinding of outcome assessment; completeness of data collection; selective reporting; and other sources of bias. We applied the coding schedule in Appendix 2 for each source of bias.

Measures of treatment effect

Continuous data

The included studies reported only continuous data. These were analysed on the assumption that they came from a normally distributed population with no skew. The included studies used different types of measures to evaluate constructs and so we could not combine the data in a meta-analysis. See Table 1 for methods stipulated in the protocol that were not required in this version of the review, but which will be applied in future versions.

Unit of analysis issues

There were no unit-of-analysis issues as both included studies randomised individual children to one of two study arms. See Table 1 for methods stipulated in the protocol that were not required in this version of the review, but which will be applied in future versions.

Dealing with missing data

Where necessary, we requested data on participants from study authors, to confirm that at least one child in each group had a motor disorder. If information was not available, we classed the study as 'awaiting classification' and did not include it in this version of the review (e.g. Fey 2006).

Both included studies reported data on the number of families who dropped out of the study as well as the reasons for dropping out. We reported the number of participants included in the final analysis as a proportion of those participants who began the intervention (see Characteristics of included studies table). We concluded that the data were missing at random and analysed the remaining data, ignoring the missing data.

We requested missing outcome data from study authors, such pre- and postintervention group mean scores and standard deviations. If data were not available, we included the study in the review and reported, assessed and discussed the extent to which its findings affected the results of the review.

Assessment of heterogeneity

We were unable to assess heterogeneity due to a lack of outcome data available for one of the studies (Girolametto 1988). See Table 1 for methods stipulated in the protocol that were not required in this version of the review, but which will be applied in future versions.

Assessment of reporting biases

We were unable to assess the estimate of reporting biases due to the small number of studies included in this review. See Table 1 for methods stipulated in the protocol that were not required in this version of the review, but which will be applied in future versions.

Data synthesis

Data were unavailable for one study, Girolametto 1988, so we could not conduct a meta-analysis for any of the study outcomes. See Table 1 for methods stipulated in the protocol that were not required in this version of the review, but which will be applied in future versions.

'Summary of findings' tables

We randomly assigned two review authors (WA, JG, KL or LP) to each outcome. Using the GRADE approach (GRADEpro 2008), both review authors independently assessed the overall quality of the body of evidence for each outcome as high, moderate, low or very low quality, according to the presence of five criteria (risk of bias, imprecision, inconsistency, indirectness and publication bias). We resolved disagreements by involvement of a third review author (WA, JG, KL or LP). We reported these ratings, along with the number of studies and participants as well as the effect estimate, in Summary of findings for the main comparison, which we constructed using GRADEprofiler (GRADEpro GDT 2015), having imported data from Review Manager 2014. The table reported the evidence for the following outcomes assessed at postintervention for the comparison 'Parent-mediated communication intervention compared with no intervention': children's ability to communicate effectively in everyday life (taking turns in conversation, initiating conversation and responding to others' conversational gambits); adverse events; child speech and language function; child generic participation: social; parents' communication and interaction strategies: directiveness and responsiveness; family stress and coping: parent stress; satisfaction of child and family with treatment; and compliance with treatment.

Subgroup analysis and investigation of heterogeneity

As we were unable to perform a meta-analysis in this review, we could not undertake subgroup analysis. See Table 1 for details of planned subgroup analyses.

Sensitivity analysis

We were unable to conduct any sensitivity analysis for this version of the review. Table 1 sets out our plans for sensitivity analysis, which we will implement in future updates of this review, if possible.

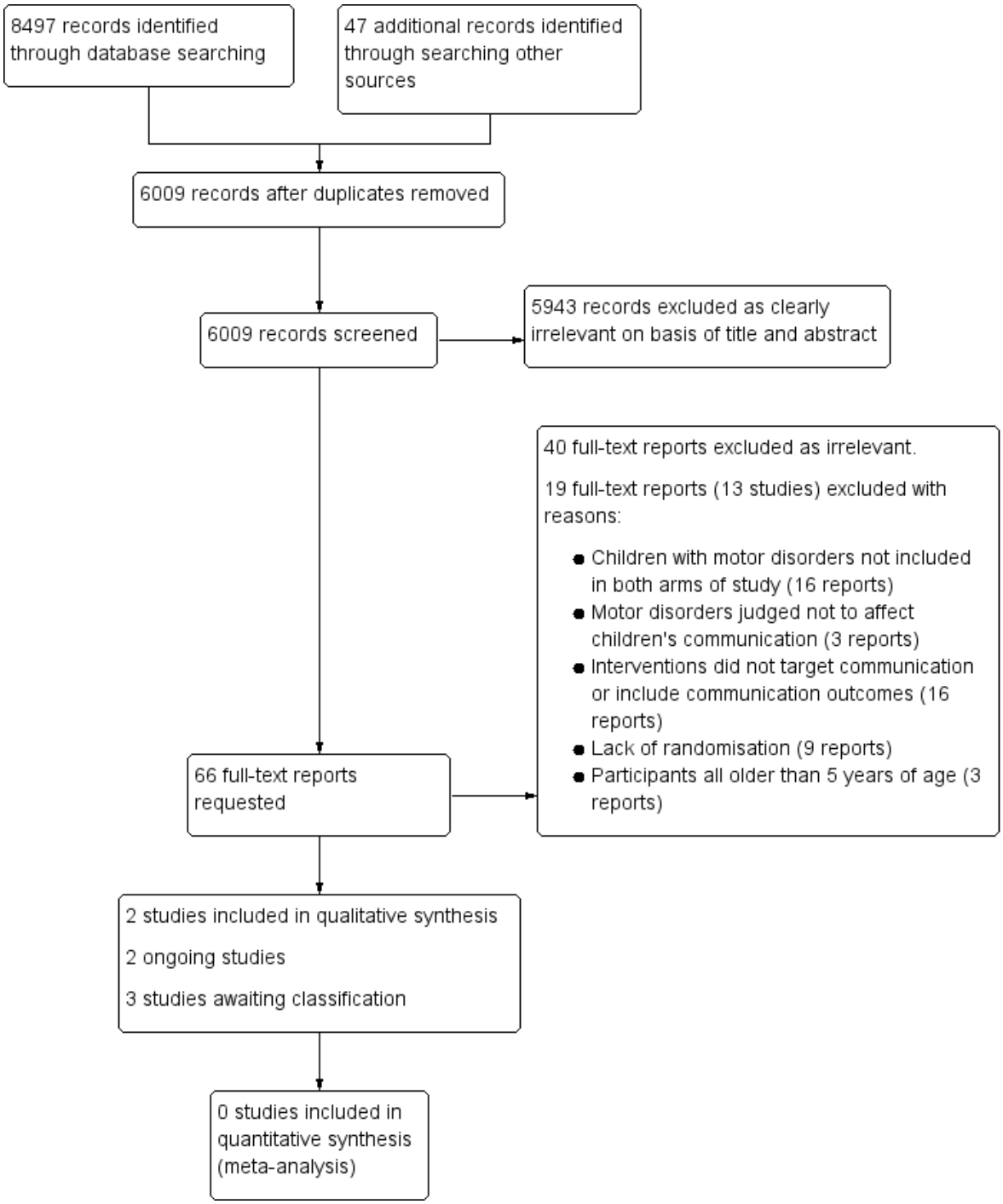
RESULTS

Description of studies

Results of the search

Our searches identified 8544 records, including six systematic reviews, whose references were handsearched (Baker 2012; Chorna 2017; Ketelaar 1998; Law 2003; Roberts 2014; Whittingham 2011). After removal of duplicates, two review authors (WA and LP) independently screened the titles and abstracts of 6009 records and subsequently excluded 5943 records on the basis of title and abstract. We retrieved the full-text reports for 63 of the remaining 66 records. We were unable to obtain the full-text reports of three records and classified these as 'awaiting classification' until we can obtain more information (Characteristics of studies awaiting classification table). We identified two studies from the 63 full-text reports that met our inclusion criteria (Included studies), and found two ongoing studies (Characteristics of ongoing studies table). Fifty-nine of the full-text papers were ineligible for the reasons given in Figure 1, which illustrates the flow of studies through the screening process. From these, we selected 19 excluded papers that at first appeared to meet the eligibility criteria, but on further inspection failed to meet one or more criteria (see Excluded studies).

Figure 1. Study flow diagram.



Included studies

This review included two RCTs, reported in one paper each (Girolametto 1988; Kim 2005). See Summary of findings for the main comparison.

Participants

One study had 18 child/mother participants, with 10 participants assigned to the intervention group and eight participants to the control group (Kim 2005). The other study had 20 child/mother participants where nine participants assigned to the intervention group, and 11 participants to the control group completed the study (Girolametto 1988). The total number of child participants from the two included studies was 38 (20 boys and 18 girls). Diagnoses included cerebral palsy (10 participants), Down's syndrome (11 participants), developmental delay (11 participants), intellectual disability (three participants), chromosomal abnormalities (two participants) and one participant with an unknown aetiology. Children ranged in age from 15 to 96 months (see Characteristics of included studies table). Both studies reported cognitive and communication developmental levels substantially lower than expected for their chronological ages for all study participants. Girolametto 1988 reported that all children had hearing within normal limits; Kim 2005 provided no information on children's hearing. All participants attended some special education and lived at home with their parents.

All parents who took part in the evaluation of the interventions were mothers, although fathers did attend the training in Girolametto 1988, but their communication behaviours were not measured. Mothers were similar in age and number of years spent in education across the studies. Mean maternal age was 34 years in both studies. Mean maternal years of education was 13 years in both studies. We retained reference to mothers throughout the review, as results pertain to mothers only.

Participants were recruited from single centres providing special education. There was no information on admission criteria for the services. Therefore, generalisability from the sample to the population of children with communication difficulties and non-progressive motor disorders was unknown. Neither study reported a sample size calculation or statistical power to detect a true difference between groups.

Setting

The two studies took place in Canada (Girolametto 1988), and South Korea (Kim 2005). Girolametto 1988 conducted training at an intervention centre. Kim 2005 did not specify where training was delivered. In both studies, therapists visited families at home to help parents implement the techniques they had learned in the group sessions.

Intervention content

The intervention provided in both studies was based on the theory that early parent-child interaction that incorporates joint attention, reciprocal turn taking and contingent responding by parents to children's communicative gambits promotes children's engagement in interaction and provide an ideal basis for learning the meaning of gestures and language (Wells 1981). The studies examined different training programmes: Girolametto 1988 provided It Takes Two to Talk (Manolson 1985; Pepper 2004); Kim 2005 delivered Relationship Focussed Intervention, adapted from the Family/Child Curriculum (Mahoney 1999a) and the Principle of Program Learning (Skinner 1958). In both programmes, parents attended group sessions in which they learned about early interaction and communication development, and the importance of following their children's lead and reducing their own directiveness. Sessions also covered how to create opportunities for interaction in play and daily activities. In addition to the group sessions in clinic, intervention also included home visits, in which therapists helped parents to apply principles they had learned in the group sessions in interaction with their children through one-to-one coaching. However, in both studies, details on individual session content and modes of delivery were lacking. It would not be possible to replicate the intervention from the information provided in the two reports. Neither was it possible to determine the extent to which training content overlapped between the two interventions.

Control content

Families allocated to control groups received no specified communication intervention during the studies. Children were either on the waiting list for intervention (Girolametto 1988), or attended the same early special education provision as those allocated to the intervention content, with no input related to responsive interaction (Kim 2005). Parent-child interaction was videorecorded at the same time points as families allocated to the intervention groups, but the studies provided no feedback on the interaction. We found no studies that compared parent-mediated intervention with clinician-mediated intervention in which therapists focused directly on children to teach them new communication behaviours.

Duration of intervention

In one study, the intervention lasted for 11 weeks and consisted of eight group sessions and three individual home visits (Girolametto

1988). The other study had a 12-week intervention period, comprising eight, once-weekly group sessions of 1.5 to two hours followed by four weeks of once-weekly home visits, two of which were to provide instruction and the final two to provide feedback (Kim 2005).

Outcome measures

Although both studies measured parents' and children's interaction behaviours, they used different outcome measures, which precluded meta-analysis. Girolametto 1988 coded videorecorded parent-child interaction on a turn-by-turn basis to show the frequency with which parents and children took obligatory turns in conversation and whether these turns maintained a topic or redirected the conversation to a new topic. Kim 2005 rated parents' and children's communicative behaviours using subjective, four-point rating scales developed for previous research. Studies measured parent-child interaction from 10-minute observations of interaction. Girolametto 1988 also measured children's expressive and receptive communication using the standardised Sequenced Inventory of Communication Development (Hedrick 1975). Kim 2005 measured child behaviour using the Child Behavior Rating Scale (Mahoney 1998) and the Maternal Behavior Rating Scale (Mahoney 1999a), and parental stress using the Korean version (Shin 1997) of the the Parent Stress Index (Abidin 1995). This study did not included a measure of the child's comprehension. See Characteristics of included studies for further details of outcome measures.

Girolametto 1988 reported children's communication behaviours, including comprehension, as group means and ranges, and stated whether statistical comparisons were significant at P less than 0.05 or P less than 0.01 in one-tailed tests. The exact test statistics and P values were not reported, neither were confidence intervals (CIs). Change scores were available for individual children in the intervention group only. Contact with the study author confirmed that unpublished data were no longer available for the study (Girolametto 2018 (personal communication) in Girolametto 1988). Therefore, it was not possible to calculate estimates of effect size for any of the outcomes reported in this study. Kim 2005 reported mean scores and corresponding standard deviations for intervention and control groups at pre- and postintervention for all measures. Published data allowed the calculation of mean differences (MD) for postintervention scores with 95% CIs. All participants who completed the intervention and control content were included in the analyses in both included studies (38 child/parent participants). Studies measured only short-term effects (immediately after intervention). We rated the quality of evidence for all primary and secondary outcomes as very low, using the GRADE approach for assessing evidence quality (GRADEpro 2008). See Summary of findings for the main comparison.

Funding

One study was funded, in part, by a doctoral fellowship from the Social Sciences and Research Council of Canada (Girolametto 1988). The other study provided no information on funding (Kim 2005).

Excluded studies

Nineteen papers reported 13 studies of parent-mediated training; however, we excluded all on at least one criterion (Adamson 2010; Badr 2006; Barlow 2007; Buschmann 2008; Buschmann 2009; Cologon 2017; Fey 2006; Kaiser 2013; Karaaslan 2013; Kynø 2012; Milgrom 2004; Newnham 2009; Romski 2010; Seifer 1991; Tannock 1992; Warren 2008; Whitmore 2014; Woynaroski 2014; Yoder 2002). Reasons for exclusion included the following.

1. Children with motor disorders were not included in both arms of the study (16 papers).
2. Children had very mild motor disorders that did not affect their ability to sit unsupported, manipulate objects, produce gesture or spoken language. Their motor disorders were judged by study authors not to affect children's communication (three papers).
3. Interventions did not specifically target communication or include communication outcomes (16 papers).
4. Lack of randomisation (nine papers).
5. Participants were all older than five years of age (three papers).

The reasons for exclusion of individual studies are provided in the Characteristics of excluded studies table.

Studies awaiting classification

We were unable to obtain the full texts of three reports (Campbell 1977; Kogan 1978; Lee 2012). See Studies awaiting classification table for further details.

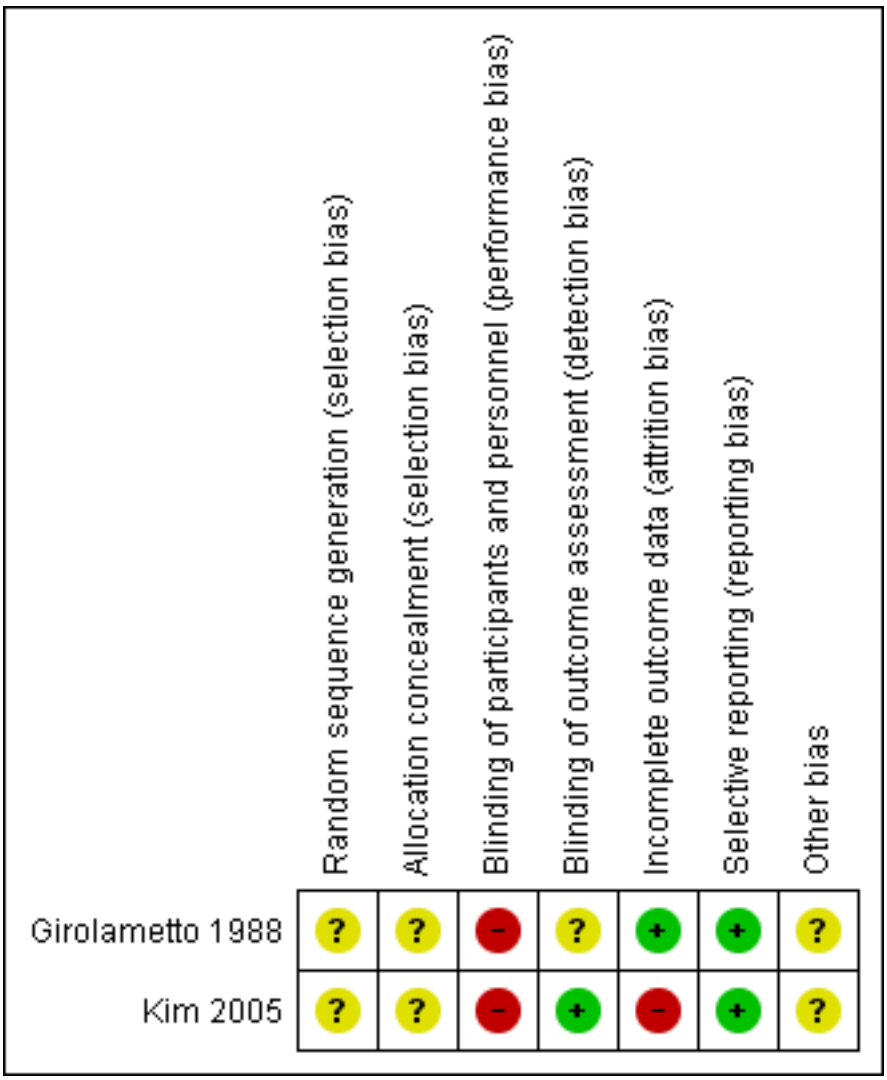
Ongoing studies

We identified two ongoing studies (ACTRN12616000653460; Eliasson 2016). See Characteristics of ongoing studies for further details.

Risk of bias in included studies

See Figure 2 for a summary of risk of bias.

Figure 2. Risk of bias summary: review authors' judgements about each risk of bias item for each included study.



Allocation

Neither study clearly described the allocation process with regard to sequence generation or allocation concealment. One study reported stratified randomisation based on aetiology and sex but provided no information on how this was achieved (Girolametto 1988). We judged the risk of bias for allocation as unclear for both studies (Girolametto 1988; Kim 2005).

Blinding

Due to the nature of the intervention, it was unlikely that parents and interventionists could be blinded to the treatment condition in either study, and therefore we considered both studies at high risk of performance bias (Girolametto 1988; Kim 2005).

One study undertook all outcome assessment blind to group allocation, so we judged this study at low risk of detection bias (Kim 2005). One study blinded outcome assessors to group allocation for speech and language function outcomes, but blinding was unclear for the main outcome measures taken from video-recorded parent-child interaction; hence, we judged this study at unclear risk of detection bias (Girolametto 1988).

Incomplete outcome data

One study was at low risk of attrition bias (Girolametto 1988). Kim 2005 was judged at high risk of attrition bias, as 3/13 children dropped out of intervention group (one family left the treatment centre and one missed three intervention sessions). Inclusion of these children could have impacted the estimates of treatment effectiveness.

Selective reporting

Both studies presented results based on all prespecified outcomes for all participants, so we judged them at low risk of reporting bias (Girolametto 1988; Kim 2005).

Other potential sources of bias

There were no other sources of bias.

Effects of interventions

See: **Summary of findings for the main comparison** Parent-mediated communication intervention compared with no intervention for improving the communication skills of preschool children with non-progressive motor disorder

Missing data in one study precluded testing of effect sizes (Girolametto 1988). Therefore, below, we present the outcomes for one study with 18 participants only (Kim 2005).

Primary outcomes

Children's ability to communicate effectively in everyday life

One study showed no evidence of an effect of parent-mediated training on children's initiations of conversation (MD −0.25 points, 95% CI −1.01 to 0.51; P = 0.54; Analysis 1.1), or their engagement in interaction measured through joint attention (MD 0.05 points, 95% CI −0.52 to 0.62; P = 0.87; Analysis 1.2), assessed at postintervention using the Child Behavior Rating Scale (CBRS; therapist rating of child engagement with parent in play in seven domains (interest, attention to activity, persistence, initiation, affect, joint attention, co-operation); items scored one to five on Likert scale; lower scores represent lower levels of coded behaviours) (Mahoney 1999b).

The study did not report children's use of communication (e.g. requesting attention, asking questions) or range of modes of expression (e.g. vocalisation).

Adverse events

The study did not report adverse events.

Secondary outcomes

Child outcomes: speech and language function

One study that measured children's expressive and receptive language development found that missing data precluded analysis of the effect of parent-mediated communication intervention on speech and language function (Girolametto 1988).

Child outcomes: children's generic participation

We found no studies that reported children's generic participation.

Parent outcomes: parents' communication and interaction strategies

Following parent-mediated communication intervention, mothers were more responsive to their children's communication (MD 1.02 points, 95% CI 0.34 to 1.70; P = 0.01; Analysis 2.1), as measured by the Mothers Behaviour rating Scale (MBRS; therapist rating of maternal behaviour on 12 items in four domains (responsiveness, affect, achievement orientation and directiveness); items scored one to five on Likert scale; lower scores represent lower levels of coded behaviours) (Mahoney 1999a). The study showed no evidence of an effect of parent-mediated communication intervention on mothers' directiveness in interaction (MD 0.49 points, 95% CI −0.02 to 1.00; P = 0.07; Analysis 2.2), also assessed at postintervention using the MBRS.

Parent outcomes: family stress and coping

The study showed no evidence of an effect of parent-mediated communication intervention on parental stress in terms of parents' views of their child's temperament (MD −6.50 points, 95% CI −11.64 to −1.36; Analysis 3.1), parent-child relationship (MD 0.25 points, 95% CI −4.70 to 5.20; P = 0.92; Analysis 3.2), or learning expectation (MD −0.58 points, 95% CI −2.87 to −1.71; Analysis 3.3), as measured at postintervention using the Korean version of the Parent Stress Index (20 items per scale rated on five-point Likert scale to indicate the extent to which items disturbed the parent in the last week; lower scores indicate lower levels of disturbance) (Abidin 1995; Shin 1997).

Parent outcomes: satisfaction of child and family with treatment

We found no studies that measured satisfaction of child and family with treatment

Parent outcomes: compliance with treatment

We found no studies that measured compliance with treatment.

DISCUSSION

Summary of main results

We identified two small studies that met the inclusion criteria for this review (Criteria for considering studies for this review), but were only able to calculate effects sizes for Kim 2005 because of missing data in Girolametto 1988. The single study providing data for the review suggested that parent-mediated intervention may have positive effects on mothers' interaction style, helping them to increase their responsiveness to their children. However, we found no evidence of concomitant reductions in mothers' directiveness in conversation. Neither did we find changes in children's interaction patterns, such as increases in initiation of communication or engagement in joint attention for interaction, which would allow them to communicate effectively in everyday life.

We were not able to analyse the effects of parent-mediated communication intervention on children's frequency of communication, frequency of spoken language, speech, or receptive or expressive language development, due to missing data in the one study that measured these outcomes (Girolametto 1988). Neither study measured wider impacts of parent-mediated communication intervention in children's social participation or considered potential adverse events. One study measured parental stress (Kim 2005), and while there was no effect of parental training, differences between groups prior to therapy prevented firm conclusions being drawn about the impact of intervention on this outcome.

Studies measured communication in short play-based interactions of 10 minutes' duration immediately after training. We found no evidence to suggest that changes in interaction were maintained over time, generalised to daily interaction or stimulated children's longer-term communication development.

The studies used different tools to measure similar constructs. Examination of the full-text reports considered for the review showed that a wide range of tools has been used to evaluate parent-mediated communication interventions. Consensus is needed on the constructs that should be measured and the tools that should be used, with demonstration of their validity and reliability.

Overall completeness and applicability of evidence

Both studies recruited children with developmental disabilities and included some children who were identified as having motor disorders. All children had some degree of intellectual impairment. We found no studies that examined parent-mediated intervention delivered specifically to parents of children with motor disorders, or studies that included children with motor disorders who did not have intellectual impairment. Neither was it possible to disaggregate the results for children with motor disorders in this review. Because of a lack of detail on participants, it was not possible to ascertain how children in the studies communicated, and the impact of their motor disorders on the intelligibility of their communication signals. Therefore, we could not ascertain if parent-mediated intervention was effective for children with motor disorders whose communication signals were difficult to interpret because of a lack of consistency and precision in underlying movements.

The transactional theory of development suggests that communication patterns may change at any point in life as a result of interaction with others (Sameroff 2000). We focused on parent-mediated interventions delivered in the preschool years (up to five years of age), as at this point in children's lives their most frequent communication partner is usually their parent. Some of the children in the study by Kim 2005 were older than five years of age and results may suggest that parent-mediated communication intervention could be provided to parents of school-aged children.

Although some fathers attended training in the study by Girolametto 1988, we were able to estimate the effects of training mothers only. Therefore, results may not be generalisable to fathers or other primary carers.

All children included in the studies were receiving special education services. Consequently, results may not be applicable to families who are not receiving such services.

Parent-mediated interventions were developed in North America and have been applied further afield, in Europe (e.g. Badr 2006 and Karaaslan 2013) and Australasia (e.g. Whittingham 2011). The programmes assume that parents view active communication by children, in the form of conversation initiation, as positive and culturally acceptable. The study by Kim 2005 suggested parent-mediated communication intervention based on the transactional theory of communication development by Sameroff 2000 may be effective in other settings, although the high rate of attrition from parent-mediated communication interventions suggests that their acceptability requires further investigation.

Quality of the evidence

We judged the studies included in this review to be of very low quality using GRADEpro 2008. Sample sizes were small and lacked justification. Studies were at risk of allocation bias; neither provided information on how allocation was achieved or maintained during recruitment. Furthermore, in the study by Kim 2005, parents allocated to the two groups appeared to differ prior to therapy, particularly on parental stress measures. Before intervention, mothers in the intervention group had a higher mean level of stress related to their relationship with their child than mothers in the control group. Postintervention, mean scores reduced to levels similar to the control group. Analysing postintervention scores disregards individual change from baseline. Children with motor disorders who receive intervention to promote their communication vary widely in their communication and language development at the start of therapy (e.g. the frequency with which they initiate interaction and the number of words they produce)

and the rate at which they learn new skills. Similarly, their parents may differ in interaction characteristics and measures of well-being. Therefore, it is important to consider differences both between groups and within groups over time when estimating an intervention's effect. Unfortunately, the data presented in the studies in this review did not allow us to do so, which may have inflated some results (e.g. parents' responsiveness) and negated others (e.g. parent stress).

As with all parent-training interventions, it is not possible to blind participants and personnel providing the intervention to group allocation, so risk of performance bias was also high for both studies. Both studies achieved blinding of outcome assessors, although we rated one study at unclear risk of detection bias as the text did not explicitly state that outcome assessors were blind to group allocation for all outcomes (Girolametto 1988). We judged one study at high risk of attrition bias, as the reasons for loss to follow-up could be related to treatment satisfaction for two out of the three children lost in the parent-mediated intervention group (Kim 2005). All participants who completed the study were included in the analysis of all outcome measures, thereby reducing the risk of reporting biases.

Potential biases in the review process

Terminology to describe developmental disabilities has changed since the 1980s. It is possible that our search did not capture all studies of parent-mediated communication interventions that included children with motor disorders. We excluded studies that involved children with developmental delay and intellectual disability where there was no reference to motor disorder, motor skills or diagnoses that included motor disorders. However, some children in these excluded studies may have had difficulties producing precise and consistent movements in communication as communication was not specified.

Agreements and disagreements with other studies or reviews

A previous version of this review covered parent-mediated intervention and speech and language therapy provided directly to children with cerebral palsy aged up to 19 years and included all study designs (Pennington 2003). That review identified four studies of parent-mediated intervention, involving 34 children aged eight months to 17 years, none of which were RCTs or quasi-RCTs. Those studies suggested similar improvements in children's communication to those observed in the present study with children taking more turns in conversation and becoming more engaged in interaction.

AUTHORS' CONCLUSIONS

Implications for practice

Some speech and language therapists currently provide individual or group communication training for parents, although the content and intensity of such training varies across providers (Watson 2015). Conclusive evidence of the effectiveness of parent-mediated communication interventions for children with motor disorders has not been demonstrated by this review.

Implications for research

Children with motor disorders may have difficulties in producing consistent and precise movements for gestural, vocal and verbal communication, which may render their attempts to communicate unintelligible. This review shows that further research is needed to test the effectiveness of parent-mediated communication interventions for children whose motor disorders impairs their expressive communication. Children with motor disorder may or may not have intellectual impairment. It is important to differentiate between these two groups as rates of change may differ. Studies that focus on children with intellectual/developmental disability should include a subgroup analysis of participants who also have motor disorders because of the complexity of their expressive communication.

To enable comparison across studies, and amalgamation of data in future systematic reviews, there is an urgent need for researchers to agree a core set of descriptor and outcome measures. Difficulties in individual developmental domains should be identified and described in sufficient detail to allow readers to ascertain the potential impact of the domain on the intervention outcome.

To test the theory of transactional communication development, outcomes should consider parents' interaction style, showing how they provide opportunities for children to join and control conversation; children's communication, measuring the frequency with which they take turns in conversation, the intentions they express (communicative functions) and the complexity of their communication (e.g. length of turn, number of different words/referents, phrase structure); acceptability of interventions; and impact on parental stress as both a potential benefit but also a potential adverse event.

Outcomes should be measured in the medium term (six months) to capture transactional changes in parents' and children's conversation behaviours, and the longer (e.g. one year) to evaluate if training facilitates ongoing, positive change to children's communication development trajectories.

Studies should recruit children who receive a variety of different services via a range of different sources to ensure generalisation of findings.

The studies included in this review involved small samples, limiting their ability to detect true differences and generalisability. Future research must provide sample size calculations to show that

it is capable of detecting differences between groups. Research reporting must also be improved, particularly in the area of participant identification, recruitment, and selection and allocation to demonstrate risk of selection bias. Application of CONSORT 2010 may help in this regard. This review was hampered by poor descriptions of the interventions provided. Future research must ensure adequate descriptions of interventions through adoption of the template for intervention description and replication (TIDieR) guidelines (Hoffmann 2014), to allow testing of adherence and development of a core set of active ingredients.

As stated above (Overall completeness and applicability of evidence), parent-mediated interventions have been adopted in cultures that expect children to be equal, independent, communication partners in conversations with parents. Future research should explore their acceptability and effectiveness in other cultures.

The group of communication partners who receive training should also be considered. The studies included in this review are not unusual in involving only mothers in their evaluations of treatment effect, as they are often children's primary carers. However, fathers, other family members and paid carers may also spend considerable time with children and influence their development. Future research should also consider the effects of training on their interaction and influence.

Finally, the economic impact of communication training provision and its outcomes should be evaluated in any future trial.

ACKNOWLEDGEMENTS

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* *Indicates the major publication for the study*

CHARACTERISTICS OF STUDIES

Characteristics of included studies *[ordered by study ID]*

Girolametto 1988

Methods	Parallel-group randomised controlled trial
Participants	**Setting:** Toronto, Canada **Children** **Sample size:** 20. Intervention = 9 (5 boys, 4 girls); control = 11 (5 boys, 6 girls) **Completers:** 20/22 children completed the study (intervention = 9/11; control = 11/11) **Mean age:** intervention = 3.3 (SD 1.2) years; control = 3.1 (SD 1.2) years **Diagnoses:** 11 = Down's syndrome; 4 = cerebral palsy; 2 = chromosomal abnormalities; 3 = unknown aetiology **Developmental measures** 1. Global developmental age (Griffiths Mental Development Scales, Griffiths 1970): intervention = 16.4 (SD 5.8) months; control = 18.5 (SD 6.8) months 2. Receptive language age (SICD): intervention = 18.4 (SD 7.2) months; control = 19.6 (SD 6.8) months 3. Expressive language age (SICD): intervention = 15.1 (SD 7.1); control = 14.4 (SD 4.5) months 4. All children had good head control and use of hands **Parents** **Sample size:** 20 mothers; intervention = 9; control = 11 **Mean age:** intervention = 35.6 (SD 4.6) years; control = 35.1 (SD 6.3) years **Mean years of education:** intervention = 13 (SD 2.4) years; control = 13.9 (SD 2.7) years **Employment:** not reported
Interventions	**Intervention:** It Takes Two to Talk, the Hanen Early Language Parent Programme (Manolson 1985) **Method:** group teaching, video-coaching, feedback **Contents:** observing child, following child's lead, contingent responding, creating opportunities for communication. Training provided by certified speech-language pathologist and 2 parent assistants **Dosage:** 1 full-day workshop and 1 session per week for 11 weeks consisting of 8 × 3-hour group sessions and 3 individual home visits **Control:** waiting list
Outcomes	**Timing of outcome measurement:** pretest and post-test, exact timing not specified **Child outcomes** 1. Discourse analysis of parent-child interaction: turn taking, contingent responsiveness, topic control, topic maintenance, responsiveness 2. Communication development: SICD (Hedrick 1975) **Parent outcomes** 1. Discourse analysis of parent-child interaction: turn taking, contingent responsiveness, topic control, topic maintenance, responsiveness All primary outcomes

Girolametto 1988 (*Continued*)

Notes	**Study start and end dates:** not known **Conflicts of interest:** not stated

Risk of bias

Bias	Authors' judgement	Support for judgement
Random sequence generation (selection bias)	Unclear risk	Not described
Allocation concealment (selection bias)	Unclear risk	Not described
Blinding of participants and personnel (performance bias) All outcomes	High risk	Parents and interventionists could not be blinded to treatment
Blinding of outcome assessment (detection bias) All outcomes	Unclear risk	Communication development assessment measure completed by assessor blind to treatment and control group. Blinding of coders in discourse analysis measures not stated
Incomplete outcome data (attrition bias) All outcomes	Low risk	2 participants lost in intervention group; 1 due to difficulty attending training, no reason stated for the other
Selective reporting (reporting bias)	Low risk	All expected outcomes reported
Other bias	Unclear risk	None identified

Kim 2005

Methods	Parallel-group randomised controlled trial
Participants	**Setting:** South Korea **Children** **Sample size:** 18 (10 boys, 8 girls); intervention = 10; control = 8 **Completers:** 18/23 children completed the study (intervention = 10/13; control = 8/10) **Mean age:** intervention = 6.2 (SD 1.7) years; control = 6.1 (1.8) years **Diagnoses:** mental retardation or developmental disorders by psychiatrist or psychologist. Met DSM-IV criteria for mental retardation, motor skills disorder or pervasive developmental disorders **Developmental measures** 1. Vineland Social Maturity age: intervention = 3.9 (SD 2.5) years; control = 3.9 (SD 2.3) years 2. Cognitive age (Carolina Curriculum, Johnson-Martin 2004): intervention = 31.8 (SD 17.8) months; control = 32.9 (SD 19.5) months

	3. Communication age (Carolina Curriculum, Johnson-Martin 2004): intervention = 25.1 (SD 14.8) months; control = 28.5 (SD 18.4) months 4. Social adaptation age (Carolina Curriculum, Johnson-Martin 2004): intervention = 36.4 (SD 19.7) months; control 37.9 (SD 22.4) months 5. Fine motor age in months (Carolina Curriculum, Johnson-Martin 2004): intervention = 39.9 (SD 22.3) months; control 37.9 (SD 24.0) months 6. Gross motor age in months (Carolina Curriculum, Johnson-Martin 2004): intervention = 37.8 (SD 23.3) months; control 38.4 (SD 24.0) months **Parents** **Sample size:** 18 mothers; intervention = 10; control = 8 **Mean age:** intervention = 34.0 (SD 2.9) years; control = 34.0 (SD 5.9) years **Mean years of education:** intervention = 13.6 (SD 2.1) years; control = 12.6 (2.3) years **Employment:** 3 mothers employed (intervention = 1; control = 2)
Interventions	**Intervention:** Relationship Focussed Intervention, adapted from the Family/Child Curriculum (Mahoney 1999a) and the Principle of Program Learning (Skinner 1958) **Method:** classroom-based instruction; home-based instruction; feedback and evaluation **Content:** responsive interaction; level of developmental functioning; interacting with children in play; turn taking; following the child's lead; increasing responsiveness; decreasing directiveness; child development; feedback about parent-child interaction in their daily routine; implementing responsive interaction strategies in the daily routine **Dosage:** 1.5-2 hours per week, every week for 3 months **Control:** no parent intervention. Children attended special education, as per intervention group
Outcomes	**Timing of outcome measurement:** pretest and post-test, exact timing not specified **Child outcomes** 1. Child Behavior Rating Scale (Mahoney 1999b), rating attention to activity, persistence, initiation, joint attention and co-operation **Parent outcomes** 1. Maternal Behavior Rating Scale (Mahoney 1999a) rating responsiveness, affect, achievement orientation and directiveness 2. Korean version of the Parent Stress Index (Abidin 1995) All primary outcomes
Notes	**Study start and end dates:** not known **Conflicts of interest:** none identified

Risk of bias

Bias	**Authors' judgement**	**Support for judgement**
Random sequence generation (selection bias)	Unclear risk	Not stated
Allocation concealment (selection bias)	Unclear risk	Not stated

Kim 2005 (*Continued*)

Blinding of participants and personnel (performance bias) All outcomes	High risk	Parents and interventionists could not be blinded to the intervention
Blinding of outcome assessment (detection bias) All outcomes	Low risk	2 raters, blinded to the intervention condition, coded videotaped observations
Incomplete outcome data (attrition bias) All outcomes	High risk	18/23 children completed the study; 3/13 children dropped out of intervention group (1 hospitalised, 1 left centre, 1 missed 3 intervention sessions), 2/10 children in control group did not complete postintervention measures (1 family refused to be videorecorded, 1 hospitalised)
Selective reporting (reporting bias)	Low risk	All expected outcomes reported
Other bias	Unclear risk	No other sources of bias identified

DSM-IV: Diagnostic and Statistical Manual of Mental Disorders (APA 2000); SD: standard deviation; SICD: Sequenced Inventory of Communication Development.

Characteristics of excluded studies *[ordered by study ID]*

Study	Reason for exclusion
Adamson 2010	No reference to motor impairment. Cerebral palsy was listed among the aetiologies, but no other reference to motor impairment or physical disability. Part of the same study as Romski 2010.
Badr 2006	Participants not reported as having communication difficulties
Barlow 2007	Non-experimental design. Did not target communication development
Buschmann 2008	Focused on evaluation of a diagnostic workup, not interventions to improve language and communication in children
Buschmann 2009	Children did not have motor disorders
Cologon 2017	Children did not have motor disorders; both arms of the study received parent-mediated training, hence no non-parent-mediated comparator
Fey 2006	Children had very mild motor disorders that did not affect their ability to sit unsupported, to manipulate objects, to produce gesture or spoken language

(*Continued*)

Kaiser 2013	Children did not have motor disorders
Karaaslan 2013	Children did not have motor disorders
Kynø 2012	Intervention not aimed at improving communication
Milgrom 2004	Did not examine parent-mediated intervention to improve communication. Not an RCT
Newnham 2009	Intervention not aimed at improving communication
Romski 2010	Inadequate information on presence or absence of motor disorder among participants. Compared 3 parent-mediated interventions and had no control without parent-mediated intervention
Seifer 1991	Intervention not specifically for children with communication difficulties. Study had a young age group, for which specific communication difficulties may not be identified
Tannock 1992	Only 1 child in 1 arm of study had a motor disorder
Warren 2008	From same study as Fey 2006. Children had very mild motor disorders that did not affect their ability to sit unsupported, to manipulate objects, to produce gesture or spoken language
Whitmore 2014	From the same study as Romski 2010; looked at secondary motor outcomes
Woynaroski 2014	From same study as Fey 2006. Children had very mild motor disorders that did not affect their ability to sit unsupported, to manipulate objects, to produce gesture or spoken language
Yoder 2002	Children with intellectual disability, 1 participant had 'mild cerebral palsy'

RCT: randomised controlled trial.

Characteristics of studies awaiting assessment *[ordered by study ID]*

Campbell 1977

Methods	No information available
Participants	No information available
Interventions	No information available
Outcomes	No information available
Notes	Title only, full text yet to be made available

Kogan 1978

Methods	Study design unknown
Participants	Developmentally delayed children aged 3-5 years
Interventions	Parent instruction programme
Outcomes	Changes in parent-child interactions, decrease areas of conflict, and increase self-esteem of both parent and child
Notes	Abstract only, full text yet to be made available

Lee 2012

Methods	Study design unknown
Participants	Children aged 1.5-5 years with developmental delay
Interventions	A home programme intervention
Outcomes	Comprehensive Developmental Inventory for Infants and Toddlers (CDIIT) assessments, including cognition, receptive language, expressive language, gross motor, fine motor, social, and self-care domains, at baseline and after 8 weeks (postintervention)
Notes	Abstract only, yet to receive any response from contacted authors

Characteristics of ongoing studies *[ordered by study ID]*

ACTRN12616000653460

Trial name or title	Community-based parent delivered early detection and intervention program for infants at high risk of cerebral palsy in a low-resource setting: a randomised controlled trial
Methods	Randomised controlled trial
Participants	**Inclusion criteria:** children with cerebral palsy or at risk of cerebral palsy, scored by a certified General Movements Assessors. Aged 12-40 weeks' corrected age **Exclusion criteria:** no developmental concerns at 12-40 weeks' chronological age **Sample size:** 142 **Recruitment:** through developmental healthcare professionals and community workers
Interventions	**Intervention:** multidisciplinary, family-centred intervention delivered peer-to-peer for 15 fortnightly visits and then delivered by carer to infant. The intervention is based on games and strategies based on the Creative Curriculum Learning Games (Sparling 2008) **Control:** health advice during 15 fortnightly visits
Outcomes	**Primary outcomes** 1. Infant functional abilities measured using the mobility domain of Pediatric Evaluation of Disability

ACTRN12616000653460 (*Continued*)

	Inventory - Computer Adaptive Test (PEDI-CAT) 2. Carers' mental health assessed using Depression, Anxiety, Stress Scale - Short Form (DASS) **Secondary outcomes** 1. Infant cognitive development (Bayley Scales of Infant Development III) 2. Infant nutritional status (weight, body mass index, height/length, head circumference, mean upper arm circumference) 3. Health resource use 4. Infant motor skills (Peabody Developmental Motor Scales) 5. Infant neurological status (Hammersmith Infant Neurological Examination) 6. Quality and extent of stimulation at home (HOME Inventory) 7. Near vision detection scale and Canadian occupational performance measure **Timing of outcome measurement:** baseline and postintervention (18 months' chronological age)
Starting date	**Date registered:** 19 May 2016 **Actual start of recruitment:** 13 March 2017 **Status:** recruiting
Contact information	Dr Katherine Benfer. **Email:** k.benfer@uq.edu.au
Notes	**Public title:** LEAP-CP: Learning through everyday activities with parents for infants at high risk of cerebral palsy in a low-income country **ANZCTR identifier:** ACTRN12616000653460 **Funding source:** Endeavour Queen Elizabeth II Diamond Jubilee Scholarship **Conflict of interest:** none identified

Eliasson 2016

Trial name or title	Efficacy of the small step program in a randomised controlled trial for infants below age 12 months with clinical signs of cerebral palsy
Methods	Parallel-group randomised controlled trial
Participants	**Inclusion criteria:** 3- to 8-month old children at risk for cerebral palsy due to early neonatal events affecting the brain **Exclusion criteria:** participants with unstable health, uncontrolled epilepsy, progressive disorders or diagnosis with a specific syndrome **Sample size:** 30 (15 in each arm) **Recruitment:** hospital-based recruitment. Participants will be recruited based on neurological signs of cerebral palsy, with a formal diagnosis made at 2 years
Interventions	**Intervention:** small step programme, an intervention targeting hand use, mobility and communication, delivered by parents who will be coached weekly by therapists. Training and intervention goals will be individualised to the child's need **Control:** usual care, the hospital follow-up programme, not standardised, but includes instructions to parents regarding home training
Outcomes	**Primary outcome** 1. Peabody Developmental Motor Scale (second edition, Folio 2000)

Eliasson 2016 (*Continued*)

	Timing of outcome measurement: baseline, 14 weeks' and 35 weeks' postintervention, plus additional measures at 7 and 21 weeks' postintervention and at 2 years' adjusted age **Secondary outcomes** 1. Child development 2. Specific brain pathology 3. Parents' perspective of the programme **Timing of outcome measurement:** baseline, 14 weeks', 35 weeks' and 2 years' postintervention
Starting date	**Study start date:** January 2014 **Status:** recruiting
Contact information	Linda Holmstsröm, PhD. **Email:** Linda.Holmstrom@ki.se Ann-Christin Eliasson, PhD. **Email:** ann-christin.eliasson@ki.se
Notes	**ClinicalTrials.gov identifier:** NCT02166801 **Funding source:** Swedish Research Council (grant number 521-2013-3096), Special grants supporting Stockholm City Council (LS 1411-1372), Stiftelsen Frimurare-Barnhuset in Stockholm, Foundation Sunnerdahls Handikappfond, Olle Engkvist Byggmästare, Promobilia, Norrbacka-Eugenia Stiftelsen, Stiftelsen and Sven Jerrings Fond **Official title:** A small-step program for development of new treatment principles for children with cerebral palsy and other neurodevelopmental disorders

DATA AND ANALYSES

Comparison 1. Parent-mediated intervention versus no intervention: child communicative participation outcomes

Outcome or subgroup title	No. of studies	No. of participants	Statistical method	Effect size
1 Initiating conversation	1		Mean Difference (IV, Random, 95% CI)	Totals not selected
2 Joint attention	1		Mean Difference (IV, Random, 95% CI)	Totals not selected

Comparison 2. Parent-mediated intervention versus no intervention: parents' communication and interaction strategies

Outcome or subgroup title	No. of studies	No. of participants	Statistical method	Effect size
1 Maternal Behavior Rating Scale (MBRS) Responsiveness	1		Mean Difference (IV, Random, 95% CI)	Totals not selected
2 MBRS Directiveness	1		Mean Difference (IV, Random, 95% CI)	Totals not selected

Comparison 3. Parent-mediated intervention versus no intervention: family stress and coping

Outcome or subgroup title	No. of studies	No. of participants	Statistical method	Effect size
1 Parent Stress Index: child temperament	1		Mean Difference (IV, Random, 95% CI)	Totals not selected
2 Parent Stress Index: parent-child relationship	1		Mean Difference (IV, Random, 95% CI)	Totals not selected
3 Parent Stress Index: learning expectation	1		Mean Difference (IV, Random, 95% CI)	Totals not selected

ADDITIONAL TABLES

Table 1. Methods stipulated in the protocol that were not required in this version of the review, but which will be applied in future versions

Types of outcome measures	We will compare baseline measures with outcomes grouped into the following time points: short term (0-1 month following intervention completion), medium term (2-5 months after intervention) and long term (≥ 6 months following intervention) We will combine results from studies where tools measure the same outcome using the same type of data (e.g. frequency of child communication behaviours; standard scores on child language measures)
Electronic searches	We will seek translations of papers published in languages other than English, when necessary
Measures of treatment effect	**Binary data** For binary data, we will calculate an OR with a 95% CI.
	Continuous We will report the effect size as an MD with 95% CI if studies have use the same continuous outcome measure. For studies that evaluate the same construct using different continuous outcome measures that share the same method of administration (e.g. questionnaires; frequency counts of behaviours measured in direct observation), we will summarise results using the SMD with 95% CI
Unit of analysis issues	**Cluster-randomised trials** We will use the *Cochrane Handbook of Systematic Reviews of Interventions* for advice on analysis of cluster-randomised trials (Higgins 2011b).
	Cross-over trials In cross-over trials, we will include data from the first period only, so as not to include data from the same participant twice
	Studies with multiple treatment groups If a study investigates multiple treatment groups, we will make single pairwise comparisons by combining data from all eligible parent-mediated intervention groups and comparing these with data combined from all eligible control groups
Assessment of heterogeneity	We will assess heterogeneity in clinical characteristics of study samples (e.g. parents' years in education; ratio of mothers to fathers in group composition; children's age; level of communication development; use of AAC) using the Chi^2 test to assess if statistical heterogeneity is likely to be due to chance alone. We will use the I^2 test and Tau^2 to describe the variation in effect estimates that is due to heterogeneity rather than sampling error (Higgins 2002).
Assessment of reporting bias	If we identify > 10 studies that fit the inclusion criteria, we will use funnel plots of effect estimates to assess the possibility of publication bias on primary outcomes, and we will use Egger's test to test for funnel plot

Table 1. Methods stipulated in the protocol that were not required in this version of the review, but which will be applied in future versions (*Continued*)

	asymmetry (Egger 1997).
Data synthesis	We will undertake meta-analysis using RevMan 5 applying a random-effects model, if two or more studies report interventions that are similar in terms of topic, delivery methods and dosage (duration, frequency and intensity of sessions), and include similar participants (parents and children) and use similar outcome measures (Review Manager 2014).
Subgroup analysis and investigation of heterogeneity	If we identify multiple studies with sufficiently similar participants, interventions and outcome measures, we will conduct the following subgroup analyses to explore possible sources of heterogeneity 1. In the presence of severe or profound intellectual or receptive language impairment (impairment in either function ≥ -1.9 standard deviations versus non-verbal or receptive language score < -2 standard deviations) 2. In parental education (high school versus further or higher education) 3. In dosage of intervention (frequency and duration of sessions) and 4. Between specific 'named' interventions (e.g. Hanen programmes (see, for example, Pepper 2004) or Enhanced Milieu Teaching (see, for example, Hemmeter 1994)).
Sensitivity analysis	We will use the 'Risk of bias' assessment to inform sensitivity analyses. As it is difficult to blind parents and training providers to the type of intervention, sensitivity analyses will use data from risk of bias arising from random allocation generation, allocation concealment, loss to follow-up and incomplete reporting of outcomes. We will remove studies judged to have a high risk of bias in these areas to determine their effect on the pooled estimate. We will undertake a sensitivity analysis of binary outcomes if data are considered missing at random, adopting both a best-case and worst-case scenario in which, for example, children in the experimental group are imputed to have a good outcome (best case) and poor outcome (worst outcome)

ACC: augmentative and alternative communication; CI: confidence interval; MD: mean difference; OR: odds ratio; SMD: standardised mean difference.

CONTRIBUTIONS OF AUTHORS

LP and JG conceived and designed the review.

LP and WA screened titles and abstracts. JG was involved in disagreement resolution.

All authors considered full-text reports for inclusion in the review, extracted data from the included studies, and assessed the risk of bias and quality of the evidence. In the event of disagreement regarding inclusion, a third review author (WA, JG, KL or LP) reviewed the report independently, and we reached consensus through discussion and by reassessing the study against the inclusion criteria together.

LP and WA wrote the review with drafts reviewed by KL and JG.

LP has overall responsibility for the review.

DECLARATIONS OF INTEREST

LP is a Speech and Language Therapist and Senior Lecturer at the Institute of Health and Society, Newcastle University, UK. LP has led an early phase trial of parent-mediated intervention for children with cerebral palsy. LP's institution received funding from the National Institute for Health Research (NIHR) Cochrane Incentive Awards to complete this review and from Sparks, The Children's Medical Research Charity, to develop a smart phone app for use in parent-mediated therapy (14NCL01). The latter study does not fit the criteria for this review, as assessed by KL and JG, and is not included. LP received payment of accommodation fees by Cerebral Palsy Alliance to present at a symposium on Early Intervention at the International Cerebral Palsy Conference, 31 May to 1 June 2016, Stockholm, Sweden.

WA is currently a PhD Student at Newcastle University.

KL is an Advanced Speech and Language Therapist, at the Northumberland Tyne and Wear NHS Foundation Trust, UK. KL's previous employer (City Hospitals Sunderland) received a set fee to release KL for 30 days (January to June 2016), as an internship with Dr Pennington (Health Education North and NIHR Clinical Academic Training Programme).

JG is Faculty Head of Research and Knowledge Exchange (Health, Psychology and Social Care Faculty) at Manchester Metropolitan University, UK. JG currently receives funding (14/70/153) from the NIHR Health Services and Delivery Research (HS&DR) Programme, to conduct the following research: "Identifying appropriate symbol communication aids for children who are non-speaking: enhancing clinical decision making." The project, which commenced in February 2016 and will run for three years, addresses the tangentially-related research area of clinical decision-making in augmentative and alternative communication. JG's role is primarily in systematic reviews to inform the project and, as such, JG does not perceive any conflict of interest. JG also declares that she received funding for external evaluation of the 'ENABLIN+' project, aimed at staff training for working with children with complex and intense support needs, Contract 541981-LLP-1-2013-1-BE-LEONARDO-LMP, and that this does not bring any conflict of interest. JG's institution was paid by the International Society for Augmentative and Alternative Communication, Norway, for two lectures on early communication and evidence-based practice in the spring of 2016. JG receives royalties for Coupe-O'Kane and Goldbart (1997) "Communication Before Speech." JG declares the book focuses on children with profound disabilities rather than motor disorders and addresses assessment and generic strategies rather than specific interventions.

SOURCES OF SUPPORT

Internal sources

- Newcastle University, UK.

Salary for Lindsay Pennington

- Manchester Metropolitan University, UK.

Salary for Juliet Goldbart

External sources

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DIFFERENCES BETWEEN PROTOCOL AND REVIEW

1. Types of participants

i) We included only studies that had a least one child with motor disorders in each group whose motor disorders may have influenced their communication, to increase similarity between groups in individual studies.

2. Electronic searches

i) We did not search EU Clinical Trials Register (www.clinicaltrialsregister.eu/ctr-search/search) because it does not register psychotherapeutic trials. We were not able to search NIH Clinical Research Center (www.cc.nih.gov/home/clinicalstudies.html) because the website returned the message "Site unavailable."

3. Summary of findings for the main comparison

i) We produced one 'Summary of findings' table for our main comparison, rather than one table for outcome, as stated in our protocol (Pennington 2017)