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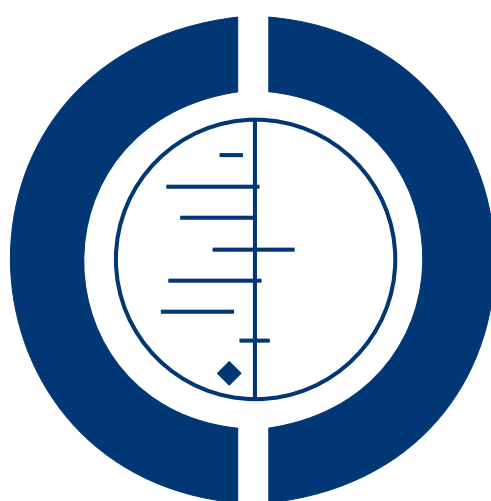
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Speech therapy for children with dysarthria acquired before three years of age (Review)

Pennington L, Miller N, Robson S



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Speech therapy for children with dysarthria acquired before three years of age (Review)
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TABLE OF CONTENTS

HEADER	1
ABSTRACT	1
PLAIN LANGUAGE SUMMARY	2
SUMMARY OF FINDINGS FOR THE MAIN COMPARISON	2
BACKGROUND	10
OBJECTIVES	11
METHODS	11
RESULTS	13
DISCUSSION	14
AUTHORS' CONCLUSIONS	14
ACKNOWLEDGEMENTS	14
REFERENCES	15
CHARACTERISTICS OF STUDIES	16
DATA AND ANALYSES	18
WHAT'S NEW	18
HISTORY	18
CONTRIBUTIONS OF AUTHORS	18
DECLARATIONS OF INTEREST	18
SOURCES OF SUPPORT	18

[Intervention Review]

Speech therapy for children with dysarthria acquired before three years of age

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ABSTRACT

Background

Children with motor impairments often have the motor speech disorder dysarthria, a condition which effects the tone, power and coordination of any or all of the muscles used for speech. Resulting speech difficulties can range from mild, with slightly slurred articulation and low-pitched voice, to profound, with an inability to produce any recognisable words. Children with dysarthria are often prescribed communication aids to supplement their natural forms of communication. However, there is variation in practice regarding the provision of therapy focusing on voice and speech production. Descriptive studies have suggested that therapy may improve speech, but its effectiveness has not been evaluated.

Objectives

To assess whether direct intervention aimed at improving the speech of children with dysarthria is more effective than no intervention at all.

To assess whether individual types of intervention are more effective than others in improving the speech intelligibility of children with dysarthria.

Search strategy

We searched CENTRAL, MEDLINE, EMBASE, CINAHL, LLBA, ERIC, PsychInfo, Web of Science, Scopus, UK National Research Register and Dissertation Abstracts up to April 2009, handsearched relevant journals published between 1980 and April 2009, and searched proceedings of relevant conferences between 1996-2009.

Selection criteria

We considered randomised controlled trials and studies using quasi-experimental designs in which children were allocated to groups using non-random methods.

Data collection and analysis

L Pennington conducted searches of all databases and conference reports. L Pennington, N Miller and S Robson handsearched journals. All searches included a reliability check in which a second review author independently checked a random sample comprising 15% of all identified reports. We planned that two review authors would independently assess the quality and extract data from eligible studies.

Main results

No randomised controlled trials or group studies were identified.

Authors' conclusions

We found no firm evidence of the effectiveness of speech and language therapy to improve the speech of children with early acquired dysarthria. No change in practice is warranted at the present time. Rigorous research is needed to investigate if the positive changes in children's speech observed in small descriptive studies are shown in randomised controlled trials. Research should examine change in children's speech production and intelligibility. It should also investigate the secondary education, health and social care outcomes of intervention, including children's interaction with family, friends and teachers, their participation in social and educational activities, and their quality of life. Cost and acceptability of interventions must also be investigated.

PLAIN LANGUAGE SUMMARY

Speech therapy for children with early acquired dysarthria

Dysarthria is a disorder which reduces the control of movements for speech. Children with dysarthria often have shallow, irregular breathing and speak on small, residual pockets of air. They have low pitched, harsh voices, nasalised speech and very poor articulation. Together, these difficulties make the children's speech difficult to understand. Dysarthria is caused by neurological impairment and can arise early in children's lives, from neurological damage sustained before, during or after birth, such as in cerebral palsy, or in early childhood through traumatic brain injury or neurological disease. Communication difficulties have a profound impact on children's development. They reduce the quality of life of children with cerebral palsy and place children at risk of social exclusion, educational failure and later unemployment. Speech and language therapy aims to help children to control the movements for breathing and speech and so become more intelligible. Small, observational studies have suggested that for some children therapy might have been associated with positive changes in intelligibility and clarity of children's voices. This review aimed to investigate if therapy is generally effective for children with dysarthria acquired early in life, and if certain types of therapy may be better than others. We found no randomised controlled trials or controlled group studies which investigate the effects of speech and language therapy to improve the speech of children with dysarthria acquired below three years of age. Rigorous research, using randomised controlled trials, is needed to evaluate if therapy can help children to increase the intelligibility of their speech and if enhanced intelligibility increases children's participation in social and educational activities and their quality of life.

SUMMARY OF FINDINGS FOR THE MAIN COMPARISON [Explanation]

Study	Participants	Intervention type	Intervention duration	Outcome measures	Outcomes	Timing of outcome measures	Methodological problems
Fischer-Brandies 1987	71 children with cerebral palsy, 4-14 years (mean 10 years), orofacial dysfunction.	Orofacial regulation therapy: wearing of removable plates for upper jaw, stimulators on palatal plate for tongue and upper lip plus motor speech therapy. 49 children also received physiotherapy (Vojta or Bobath or Castillo-Morales).	15 months	List of symptoms, rated as better or worse after treatment: abnormal tongue position; limited tongue mobility (single and multiple directions); type of tongue mobility problem (jerky, slow, vermicular); feeding (sipping, sucking, chewing, choking); drooling; abnormal tongue position 20/59; limited tongue mobility 33/56; palatal sound production; dental sound production.	Number of children showing improvement when symptoms rated as better or worse than at start of therapy by neuropaediatrician. Improvements observed (number showing improvement/number showing difficulties in area measured): abnormal tongue position 20/59; limited tongue mobility 33/56; jerky tongue movements 13/23, extremely slow tongue movements 10/21; sucking 15/31; sipping 23/30; chewing 21/37; severe drooling 28/40; labial sounds 24/38; palatal sounds 26/57; dental sounds 24/53. In 17 cases oral functions worsened after therapy.	Beginning and end of treatment; timing not specified.	Rater not blind to prior scores; no information on validity or reliability of outcome measures; ? isolated movement and speech sound production, no information on speech intelligibility; binary scale used in outcome measure (better/worse); no control group; before and after treatment measures only.
Fox 2005	5 children (2F, 3 M), aged 5;10 - 7;10 years with spastic type cerebral palsy	Lee Silverman Voice Therapy Loud.	4 weeks: 16 one hour sessions (4 times per week for 4 weeks) plus minimum 36 practice sessions between treat-	Acoustic measures: dB Sound Pressure Level (SPL), maximum phonation duration in seconds, harmonics to noise ratios (HNR) in	Change (as inferred from no overlapping data points) noted on all acoustic measures in maximum performance tests post ther-	Two weeks prior to treatment, two weeks post treatment and six weeks post treatment	

			<p>ment sessions. Four children received therapy, one child received no treatment.</p>	<p>dB, maximum and minimum pitch in HZ, pitch range in Hz, elicited in maximum performance tests, sustained vowels, sentence repetition and cartoon description.</p> <p>Perceptual measures: therapists' blinded preferences for recordings made at different times on overall loudness, loudness variability, overall pitch, pitch variability, articulatory precision, overall voice quality.</p>	<p>apy and at follow-up for three of the four children who received treatment. Trends noted in sustained phonation and sentence repetition for three children.</p> <p>No change or reducing scores for child who did not receive therapy.</p> <p>Therapists preferred overall loudness, loudness variability, pitch variability, articulatory precision, overall voice quality of post treatment recordings.</p>	
Fox 2008	<p>8 children (6F, 2 M) aged 6;01 -12;00 years with spastic type cerebral palsy</p>	<p>Lee Silverman Voice Therapy Loud.</p>	<p>4 weeks: 16 one hour sessions (4 times per week for 4 weeks)</p>	<p>Acoustic measures: dB SPL, jitter, HNR and duration of phonation in maximum performance tests and in sentence repetition.</p> <p>Perceptual ratings: children's parents rated voice quality using visual analogue scales.</p>	<p>Increase in vocal SPL in sustained vowels (F(2-12) = 5.14, p = 0.024) post therapy and follow-up; improvements in jitter (measure of voice quality) post therapy and at follow-up (F(2-12) = 5.27, p = 0.02); increase in SPL of spoken sentences after therapy (F(2-12) = 5.29, p = 0.02).</p> <p>Parents perceived their children's voices as "louder", less "nasal" and more "natural" after treatment.</p>	<p>Two weeks prior to treatment, two weeks post treatment and twelve weeks post treatment</p> <p>No blind rating of perceptual measures.</p>

<p>Hartley 2003</p>	<p>4 boys with predominantly athetoid type cerebral palsy aged 10;05 - 13;00 years. Speech described as "borderline intelligible". All children used augmentative and alternative communication systems.</p>	<p>Subsystems approach. 2 blocks of therapy. 1st block concentrated on respiration and phonation. 2nd block focussed on articulation deficiencies noted during assessment</p>	<p>Two four week blocks of therapy. Duration and frequency of session were not specified.</p>	<p>Percentage intelligibility of single word speech on Children's Speech Intelligibility Measure (Wilcox and Morris, 1999) to one familiar and one unfamiliar listener per participant. Impairment scores on Robertson Dysarthria Profile (Robertson 1982)</p>	<p>Group comparison of intelligibility data across time. No difference in intelligibility across data collection points. Individual results for participants on Dysarthria profile showed positive change for one child.</p>	<p>Six weeks prior to therapy, one week prior to therapy, in the week between therapy blocks, one week after therapy completion, six weeks after therapy completion.</p>	<p>Results of four cases presented as a group for intelligibility investigation</p>
<p>Marchant 2008</p>	<p>One 13 year old girl with spastic type cerebral, hemiplegic palsy and severe spastic dysarthria. Hearing and vision within normal limits. Comprehension adequate for testing and therapy procedures</p>	<p>Two blocks of therapy. 1st block: phonetic placement, articulation therapy involving teaching of correct movement patterns for target speech sounds. 2nd block: relaxation of muscle groups using bio feedback from surface electromyography.</p>	<p>Two blocks of therapy each comprising ten sessions of 45 minutes over two weeks. Withdrawal of therapy for two weeks between therapy blocks.</p>	<p>Surface EMG amplitude of left and right obicularis oris and submental muscles; percentage single word, sentence and paragraph intelligibility; vowel formant frequencies; duration of alternative motion rates of repeated syllables; perceptual rating of voice characteristics using Duffy scale by two therapists blind to time of recording but not to aims of study; self-perception of speech impairment by participant.</p>	<p>Significant difference assumed if post therapy results were +/- 1 SD from pre therapy scores. Single word intelligibility improved after articulation therapy and improvement was maintained post EMG therapy. No change in intelligibility at sentence or paragraph level. Some change in motor control after EMG therapy: reduction in amplitude of non-speech movements and gap between syllables and increase in alternative motion rates. No change in participants' view of her speech disorder.</p>	<p>On each of three consecutive days before first block of therapy, on the day following first block of therapy, on the day following the second block of therapy.</p>	<p>No medium term or long term assessment of outcome. Therapists rating speech were not blind to aims of the study.</p>

<p>Pennington 2006</p>	<p>6 participants (4 girls, aged 10 -18 years) all of whom had cerebral palsy: four spastic type, one mixed type, one ataxic type. Hearing within normal limits. Two children with language delay, but comprehension adequate for simple verbal instructions; four children language comprehension within normal limits. All used speech to communicate. Dysarthria rated as mild to severe by local therapists.</p>	<p>Whole system approach, targeting control of breath supply for speech production and prosodic contrasts.</p>	<p>Individual therapy for 20-30 minutes. Five sessions per week for five weeks.</p>	<p>Percentage of single words (Children's Speech Intelligibility Measure) and connected speech (elicited in picture description) intelligible to three unfamiliar listeners. Listeners blind to time of recording. Semi-structured interview on acceptability of treatment.</p>	<p>Individual results presented for each participant. Four students increased single word intelligibility immediately after therapy, but gains in intelligibility were not maintained at follow-up. Two students did not increase intelligibility of single words. Increases in connected speech intelligibility were observed for three participants, gains were not maintained at follow-up. Three participants reported that the duration and intensity of the treatment were acceptable. Three participants reported that the therapy was too intensive and that either four weeks of therapy five times per week or three sessions per week for five weeks would be preferred.</p>	<p>One week prior to therapy, one week after therapy completion, six weeks after therapy completion</p> <p>No control group or maturational control.</p>
<p>Pennington 2009</p>	<p>16 participants, (9 girls age 12-18 years, mean = 14 years, SD = 2). 15 with cerebral palsy, one with Worster-Drought. Nine</p>	<p>Whole systems approach which focused on stabilising the students' respiratory and phonatory effort and control, speech rate and</p>	<p>Three individual sessions of 30 minutes each per week for six weeks.</p>	<p>Mean percentage intelligibility of single words (Children's Speech Intelligibility Measure) and connected speech to three</p>	<p>Group and individual results presented following treatment 15/16 children were more intelligible to familiar and/or unfamiliar listen-</p>	<p>Six weeks and one week before therapy, one week and six weeks after therapy completion.</p> <p>No treatment integrity checks; longer term effects of intervention were not evaluated.</p>

	<p>children had spastic type cerebral palsy, two had dyskinetic type, four had mixed (spastic and dyskinetic) and one child had Worster Drought. The motor disorders of all children except the child with Worster Drought were bilateral. GMFCS ranged from 1 - 5 (median = 4). Dysarthria rated mild to severe by referring speech and language therapists. All children were able to comprehend simple instructions.</p>		<p>phrase length/syllables per breath.</p>	<p>familiar and three unfamiliar listeners. Listeners blind to time of recording for intelligibility measures. Questionnaire on the acceptability of therapy, using Likert scales.</p>	<p>ers in single word or connected speech. On average familiar listeners understood 14.7% more single words and 12.1% more words in connected speech after the therapy. Unfamiliar listeners understood 15.0% more single words and 15.9% more words in connected speech after therapy. All children reported that the therapy was acceptable and would recommend it to a friend.</p>		
<p>Puyuelo 2005</p>	<p>10 participants with cerebral palsy (3 girls), aged 3 years at the start of the study. Five children had athetoid type CP, four spastic type and one had ataxia. Children had "absence of articulated speech". Hearing and language comprehension within normal limits.</p>	<p>Intervention focused on increasing control of oral movement used in articulation, chewing and expiration. Second block of therapy focused on controlling exhalation for speech and coordination of exhalation and phonation; voice training; and prosody (intonation, pausing, rhythm and sound duration). In the second block of therapy advice was given to parents on</p>	<p>Two blocks of treatment. Each block comprised 11 months of twice weekly therapy, each session lasting 30 minutes.</p>	<p>Impairment scores on Spanish adaptation of Robertson Dysarthria Profile (Robertson 1982). Spectrographic analysis of a repeated sentence.</p>	<p>Group results presented. Following the first treatment only voice control increased. Following the second treatment scores increased for respiration, voice, articulation, intelligibility and prosody. Spectrographic analysis was also possible at the end of the second treatment, as children had developed some spoken output.</p>	<p>Before intervention, between first and second interventions, after intervention two. Exact timing of measures not specified.</p>	<p>No control group; long duration of treatment; no control of maturational effects; no blinding of assessor.</p>

		stimulating communication, and children engaged in story telling and recall to practice their speech skills with their parents. Whilst receiving the above therapies children also received Bobath neurodevelopmental treatment.				
Ray 2001	16 children aged 7 - 10 years (mean = 8) with mild to moderate spasticity associated with cerebral palsy. All children had scores within normal limits on Raven's Coloured Progressive Matrices, all had passes pure tone screening at 25dBHL bilaterally. Children had mild - moderate language delay but were able to understand simple instructions.	Orofacial myofunctional treatment, focusing on resting position of lips closed and tongue under hard palate, plus strength exercises for jaw, lips and tongue (exercises involving isolated movements not speech) and passive stretching of lips and tongue.	Treatment given five days per week for four months. Treatment sessions = 15 minutes individual therapy plus ten minutes group treatment. Parents were provided with exercises for children to complete at home	Four-point rating scale of function of lips, jaw and tongue, by one orthodontist and two speech language pathologists. Percentage errors on production of phonemes in 20 single words, as transcribed independently by two speech language pathologists. Percentage errors then converted to five-point scale.	Group difference in pre and post therapy scores for lip and tongue position and for percentage phonemes correct.	Pre and post therapy. Timings not specified. No blinding of assessors; no maturational or experimental control; no follow-up.
Robson 2009	Same as Pennington et al 2009	See Pennington et al 2009	See Pennington et al 2009	Perceptual measures: 16 therapists rated severity of voice impairment from recordings using validated four point scale.	Slight reduction in fundamental frequency, intensity and jitter of children's voices. Slight increase in speaking time between pauses. No change in perceived	See Pennington 2009 No long term follow-up.

Acoustic measures: HNR, RMS amplitude, shimmer APQ, jitter RAP, jitter PPQ, F0 mean, rate with pauses, rate without pauses, time with pauses and time without pauses.

severity of voice impairment.

BACKGROUND

Dysarthria denotes an articulatory disturbance which arises when neuromuscular impairment affects the tone, power and coordination of any or all of the muscles used for speech. The changes to tone, power and co-ordination influence the speed, range, strength and durability of movements, leading to loss or inaccuracy of articulatory movements. When this happens listeners perceive the distortion or omission of sounds and syllables and the alterations to voice quality characteristic of dysarthria. For example, changes to lip and tongue movement may cause 'tip' to be heard as 'sip', 'hip' or 'sieve'; 'beach' to be heard as 'eats'; 'decide' as 'sigh' or 'say'. Changes in tone, power and coordination affecting the larynx alter the quality of phonation (sound made when air passes through vibrating vocal folds) and the control of pitch and loudness. This may give an impression of loss of normal intonational rises and falls (sometimes termed monopitch) and blurring of contrasts between stressed and unstressed syllables (monoloudness). Lack of coordinated movement can lead to other alterations in the normal flow of speech, in the shape of perceived changes in rhythm. The speaker sounds as if they are stuttering or talking syllable by syllable. Voice may be quiet or there may be inappropriate swings in pitch and loudness. Such changes can also be associated with changes to respiratory function. The air needed to produce speech is insufficient, is poorly regulated and/or escapes too quickly. Apart from the consequences this has for phonation and articulation (as described above), it may also have a knock-on effect on the length of utterances a speaker can produce. Involvement of the soft palate typically leads to perceptions of excess nasality in a person's speech. Symptoms of dysarthria can range from mild slurring of speech sounds and slightly low pitch to complete inability to produce any intelligible words.

Dysarthria in childhood is associated with congenital disorders such as cerebral palsy (Lepage 1998; Kennes 2002; Bax 2006; Odding 2006) and with acquired aetiologies such as brain tumours (van Mourik 1996; Cornwell 2003; Richter 2005) and traumatic brain injury (Chapman 2001; Netsell 2001; Cahill 2002). At present there is a dearth of information of the prevalence of dysarthria in children. In cerebral palsy, estimates of speech disorder in middle to late childhood range from 40% to approximately 50% (Kennes 2002; Bax 2006). However, precise prevalence figures are not known as previous research has used measures that combine speech and communication. Given that cerebral palsy occurs in approximately two per thousand live births, approximately one in a thousand may have dysarthria. How many additional children have dysarthria arising from other causes is not known. However, cerebral palsy and head injury remain two of the most common medical causes of referral to speech and language therapy (Petheram 2001). As the speech impairments are neurologically based they do not resolve. Intervention seeks to maximise children's speech performance, teaching them how to use different movements and lay down new motor programmes for those movements. The acquisition of new motor programmes requires

intensive practice (Schmidt 2005) involving considerable therapy input over long time periods. Dysarthria therapy, therefore, potentially carries considerable costs to health services even though the prevalence of the disorder in childhood may be small.

Therapy to reduce the motor speech impairments experienced by children, and the intelligibility limitations these impairments impose, has been advocated in textbooks on dysarthria (Love 1992; Hayden 1994; Strand 1995; Hodge 1999; Yorkston 1999). An approach that targets all subsystems of the vocal tract: breathing, nasal resonance, articulation and pitch control is commonly described, and is similar to intervention for adults with dysarthria acquired following neurological insults (e.g. a stroke). Treatment focusing on one or more subsystem in speech production may, for example, aim to help children control their breathing and maintain adequate pressure for speech across a phrase. This might involve teaching children how to start to speak at the beginning of exhalation and how to split utterances into smaller phases in which they can maintain adequate volume. Intervention also involves slowing children's speech rate, to allow more precise movement of muscles in the oral tract. Strand (Strand 1995) and Yorkston and colleagues (Yorkston 1999) also advocate increasing respiratory effort and making jaw movements bigger in speech to increase oral cavity volume, plus the use of speech and non-speech exercises to help close the airway to the nose during speech. Treatment for articulation has only been advised when other aspects of speech production have been or are being addressed, as "imprecise production of speech sounds (which is the most common perceptual characteristic of dysarthria) is not simply an oral articulatory problem, and is usually the result of laryngeal, velopharyngeal, respiratory and oral articulatory problems" (Strand 1995, p134). Thus, more precise articulation and improved intelligibility is thought to be achieved through developing control of breathing for speech, increasing background effort and slowing speech rate (Love 1992; Strand 1995; Yorkston 1999). Treatment for prosody (intonational contours of speech) and pitch control has been described (Yorkston 1999; Strand 1995). This comprises exercises to control the rate of words spoken and pauses used, increase volume and possibly the use of pitch change. As treatment of isolated oromotor movements has not been found to affect speech (Weismer 2006), all therapy is functional, being directed at speech production.

Although therapy for dysarthria in childhood has been described in textbooks its effects are currently unclear. Observational studies have suggested increases in intelligibility (Puyuelo 2005; Pennington 2006) and voice quality (Fox 2005) for some children following intervention focusing on breathing, voice and prosody. One investigation has been undertaken to review the general effectiveness of therapy (Yorkston 1996). However, this review was completed over a decade ago and was not undertaken systematically. Speech and language therapists, therefore, have little evidence on which to base treatment decisions. Some may provide dysarthria intervention as there is no evidence to suggest that the treatment

does not work or causes harm. Others may withhold treatment because there is no evidence showing its effectiveness.

Speech allows us to share complex thoughts and ideas quickly, and is the most highly prized form of human communication. Communication difficulties reduce the quality of life of children with cerebral palsy (Dickinson 2007) and children with speech and communication disorders are at risk of educational failure, social exclusion and later unemployment (ICAN 2007). Such problems not only have an obvious individual and family impact but also present considerable societal and economic consequences. To ensure that children have a clear means of communication augmentative and alternative communication (AAC) systems, such as symbol books and speech synthesizers, are often provided. However, many children still choose to communicate by speech. It is important to investigate if the speech of children with dysarthria can be improved since increased intelligibility will maximise the chances of communication success and may facilitate interaction in all areas of life. We aimed to conduct a systematic review of the studies of speech therapy for children who have acquired dysarthria early in life and to investigate the relative effectiveness of different types of treatment.

OBJECTIVES

1. To assess whether direct intervention aimed at improving the speech of children with dysarthria is more effective than no intervention at all.
2. To assess whether individual types of intervention are more effective than others in improving the speech intelligibility of children with dysarthria.

METHODS

Criteria for considering studies for this review

Types of studies

We looked for randomised controlled trials and studies using quasi-experimental designs in which children were allocated to groups using non-random methods.

Types of participants

Any child under 20 years of age who acquired dysarthria below three years of age. No exclusions were made on the basis of additional impairments (intellectual or sensory impairments, the presence of epilepsy) or prior receipt of speech and language therapy. We selected this age range because people who have identified special needs are entitled to statutory education provision up to 19 years of age in England, which could specify speech and language therapy. We excluded children who acquired dysarthria above three

years of age as they may differ from children with earlier acquired pathologies in terms of: their neural development, plasticity and recovery patterns; memories of fluent speech; retrieval of previously developed motor programmes; self image (seeing themselves as a fluent speaker rather than a person with a speech disorder) and patterns of communication development. Children with early acquired dysarthria may never have developed motor programmes for fluent speech or have memories of non-dysarthric speech and may not see themselves as an intelligible speaker. Furthermore, children with severe speech and motor impairments arising from congenital pathologies or those acquired in early infancy have highly unusual patterns of communication development. They take a mainly responsive role in communication and often fail to develop a full range of conversational skills (Pennington 1999). Interventions for children who acquire dysarthria at three years of age and above are the subjects of a separate review (Morgan 2008). We excluded children who did not have a definite diagnosis of dysarthria, with underlying neurological/neuromuscular pathology, and those who took part in studies that did not explicitly list dysarthria in their inclusion criteria. Thus, children who had other types of speech disorders, such as articulation problems without dysarthria, were not included in this review.

Types of interventions

Any therapy aimed at improving children's speech, whether provided individually or in groups, in the child's home, school or health service settings, except where it is provided as part of a holistic approach (e.g. as in conductive education where there are no specific speech interventions). Therapy can be provided directly by speech and language therapists (also known as speech-language pathologists, speech pathologists) or by other personnel under the direction of a speech and language therapist.

Types of outcome measures

Primary outcomes

Primary outcome measures relate to children's speech production: respiration, phonation, nasality, articulation, sound pressure level, intelligibility. These are classified as voice, articulation, fluency and rhythm of speech, production of notes and respiratory functions in the World Health Organisation International Classification of Functioning, Disability and Health (ICF). ICF activities of speaking, conversation, and discussion will also form primary outcome measures for this review. Measures used may be, for example: rating scales; oromotor skills tests; articulation tests; phonology tests; acoustic measures of pitch and loudness; physiological tests e.g. of respiration and nasal emission; intelligibility rates; coding schemes developed for individual research studies that include validity and reliability data.

Secondary outcomes

Satisfaction of participant and family with treatment; non-compliance with treatment; direct costs of treatment; adverse events, including time missed from education.

Search methods for identification of studies

Electronic searches

The following data bases were searched from 1980 or from inception up until the end of April 2009: the Cochrane Central Register of Controlled Trials (CENTRAL) published in The Cochrane Library (2007 Issue 3); MEDLINE; CINAHL, EMBASE; ERIC; Psych-INFO; Linguistics and Language Behaviour Abstracts (LLBA); Web of Science; Scopus; UK National Research Register; Dissertation Abstracts.

The search strategy below (developed from [Robinson 2002](#)) was used for MEDLINE and was modified for other databases.

1. dysarthria/rh, th [rehabilitation, therapy]
2. articulation disorders/rh,th [rehabilitation, therapy]
3. speech disorders/rh, th [rehabilitation, therapy]
4. voice disorders/rh, th [rehabilitation, therapy]
5. 1 or 2 or 3 or 4
6. child/ or adolescent/ or infant/ or child, preschool/
7. 5 and 6
8. randomized-controlled trial.pt.
9. controlled-clinical trial.pt.
10. randomized controlled trials/
11. random allocation/
12. double-blind method/
13. single-blind method/
14. or/8-13
15. animal/ not human/
16. 14 not 15
17. clinical trial.pt.
18. exp clinical trials/
19. (clinic\$ adj25 trial\$).tw.
20. ((singl\$ or doubl\$ or trebl\$ or tripl\$) adj (mask\$ or blind\$)).tw.
21. placebos/
22. placebo\$.tw.
23. random\$.tw.
24. research design/
25. (latin adj square).tw.
26. or/17-25
27. 26 not 15
28. 27 not 16
29. comparative study/
30. exp evaluation studies/
31. follow-up studies/
32. prospective studies/
33. (control\$ or prospectiv\$ or volunteer\$).tw.
34. cross-over studies/
35. or/29-35
36. 35 not 15
37. 36 not (16 or 28)
38. 16 or 28 or 37

Other searches

We handsearched the following journals from their inception

or from 1980 until end March 2009 (unless otherwise specified): American Journal of Speech-Language Pathology; Applied Psycholinguistics (1996 onwards); Augmentative and Alternative Communication; Child: Care, Health and Development and the Ambulatory Child; Child Language Teaching and Therapy; Developmental Medicine and Child Neurology; European Journal of Special Needs Education; Folia Phoniatrica; International Journal of Disability, Development and Education; International Journal of Language and Communication Disorders; International Journal of Rehabilitation Research; International Journal of Speech Pathology; Journal of Child Psychology and Psychiatry; Journal of Communication Disorders; Journal of Medical Speech-Language Pathology; Journal of Psycholinguistic Research; Journal of Special Education; Journal of Speech, Language and Hearing Research; Speech, Language and Hearing in Schools; Sprache Stimme Gehör. (The current titles are given for journals experiencing name changes since 1980.)

We checked published conference proceedings of the following organisations: European Academy of Child Development (1996 to 2008), International Society for Alternative and Augmentative Communication (1996 to 2008), American Speech and Hearing Association (1999 to 2008), Royal College of Speech and Language Therapists (1998 to 2009).

Reference lists of all studies selected for possible inclusion were checked for other possible eligible studies.

Studies reported in any language were eligible for inclusion.

Data collection and analysis

Selection of trials

One of the review authors (LP) independently screened for inclusion each title and abstract obtained from the database searches. Journals were handsearched by one of the three review authors. Fifteen percent of reports obtained in the searches were randomly selected and independently checked for inclusion eligibility by a second reviewer. Agreement between the reviewers on the reports included in the reliability check was 100%.

Data extraction

We planned that two of the three review authors (LP, SR, NM) would independently extract data into RevMan 4.2.

Data to be included:

Participants: age; gender; age of onset of disorder; diagnosis of underlying disorder; type of dysarthria; severity of dysarthria relating to respiration, phonation, nasality, articulation, sound pressure level, intelligibility.

Co-morbidity

Intervention: type of intervention; duration; frequency; provider: SLT/other.

Focus of intervention: respiration; phonation; nasality; articulation; sound pressure level; intelligibility.

Comparator intervention

type of intervention

duration

frequency

provider: SLT/other

focus of intervention: respiration; phonation; nasality; articulation; sound pressure level; intelligibility.

Quality assessment

We planned that the two review authors who extracted data on an individual study would also independently assess the study's methodological quality. Disagreements were to be resolved with the third review author. Agreement on methodology assessment was to be calculated using the Kappa statistic. Individual criteria were to be rated according to the Cochrane Handbook for Systematic Reviews of Interventions (Higgins 2006):

(A) adequate,

(B) component not reported or unclear,

(C) component reported but inadequate.

1. Method of allocation (assignment of participants to group)

(A) Well described randomised process.

(B) Allocation is not described or description leads to uncertainty in quality of allocation and possibility of bias.

(C) Non-random method (e.g. days of the week, alternate).

2. Allocation concealment

In the case of speech and language therapy interventions neither participant nor provider can be blind to the type of treatment given. Blinding in studies in this review was to refer to blinding of study research team and treatment provider to allocation process.

(A) Allocation was to be classed as adequately concealed if allocation was done using a centralised system independent of research team, use of pre-numbered opaque sealed envelopes, generation of allocation by computer by person not in charge of allocation.

(B) Methods of concealment not described or description does not allow bias to be ruled out.

(C) Providers of intervention undertake allocation or research team allocate participants and have access to participant characteristics.

3. Blinding of outcome assessors

(A) Reports state that assessors were blind to allocation.

(B) No information on blinding of assessors.

(C) Reports suggest that assessors are likely to know the group to which the participant was allocated (e.g. provided treatment, worked with person delivering treatment).

4. Loss to follow up

(A) Attrition is similar in both conditions and no greater than 25% of participants entering the trial.

(B) Loss of participants to follow up is not reported.

(C) Loss of participants to follow up is greater than 25% or is distributed unevenly across groups. Studies showing uneven loss to follow up will be considered separately in sensitivity analyses.

5. Intention to treat analysis

(A) All trial participants entered into the analysis in the group to which they were originally allocated.

(B) Intention to treat analysis not reported.

(C) Trial participants who did not complete their originally allo-

cated treatment removed from the analysis.

Data management

We planned to develop and pilot data extraction sheets, which would include a methodological assessment table for application of the codes above. We planned to enter extracted data into RevMan 4.2, and to contact authors of studies to request missing data.

Data synthesis

Continuous data

We planned to summarise similar outcome measures with continuous data using standardised mean differences.

Binary data

Binary data (e.g. reaching normal loudness: yes or no) may be used in early reports. We planned to calculate a standard estimation of the odds ratio for binary data, with a 95% confidence interval.

Heterogeneity

We planned to undertake meta-analysis of studies that investigated similar interventions, used similar outcome measures and included groups of participants who were clinically homogeneous.

We planned to assess possible inconsistency across studies using the I-squared (I²) statistic (Higgins 2003). For heterogeneous studies (Q-statistic = 0.1 and I² value of 25% or greater) we planned to conduct subgroup analysis only. We planned to undertake a narrative review of heterogeneous studies.

Subgroup analyses

Subgroup analyses were to be undertaken if studies fitting the criteria for meta-analysis could be grouped further according to participants' type of dysarthria, severity of dysarthria, age.

Sensitivity analyses

We planned to undertake sensitivity analyses to assess the robustness of review findings by investigating the impact of study quality: effects of randomisation; inadequate concealment; blinding of outcome assessors; unequal loss to follow up; failure to employ intention to treat design.

Assessment of bias

We aimed to investigate associations between effect size and study precision in terms of sample size using funnel plots.

RESULTS

Description of studies

See: [Characteristics of excluded studies](#).

We found a total of 1156 abstracts, 1146 of which did not fit all criteria for inclusion in this review. Full texts of the remaining ten papers were considered for potential inclusion (Fischer-Brandies 1987; Ray 2001; Hartley 2003; Fox 2005; Puyuelo 2005; Pennington 2006; Fox 2008; Marchant 2008; Robson 2009; Pennington 2009). All were excluded on the grounds that they were observational studies. Thus, no papers were identified as fit-

ting the inclusion criteria for this review. Agreement between the reviewers on exclusion was 100%.

To show the developing evidence for dysarthria intervention for this clinical group we have described the studies Table 1 and present a summary of their findings here. Most observational studies investigated interventions designed to control respiratory effort and breath support for speech (Hartley 2003; Fox 2005; Puyuelo 2005; Pennington 2006; Fox 2008; Robson 2009; Pennington 2009). Those that included multiple data collection points pre and post therapy and blinded outcome assessment provide support for the potential effectiveness of this type of intervention, with increases in speech intelligibility and improvements in acoustic measures associated with voice quality being observed (Pennington 2006; Fox 2008; Robson 2009; Pennington 2009). Three studies involved nonspeech exercises (Fischer-Brandies 1987; Ray 2001; Puyuelo 2005) and indicated no improvement or were unable to do so because of methodological flaws in the study design (e.g. lack of blinding of assessors, indefinite intervention and measurement). Marchant 2008's single case experimental design showed no effect on intelligibility of either articulation-based therapy or surface electromyography to reduce orofacial spasticity.

Risk of bias in included studies

No controlled studies were identified for this review.

Effects of interventions

See: [Summary of findings for the main comparison Excluded, observational study findings](#)

No controlled studies were identified for this review.

DISCUSSION

Children with early acquired dysarthria have reduced quality of life and are at risk of social exclusion, failure in education and later unemployment. In addition, there can be psychosocial, family and societal economic consequences. Children with dysarthria are often prescribed AAC systems to supplement their natural modes of communication but children still prefer to communicate by speech wherever possible. Pre-trial observational studies have suggested that interventions teaching children to produce slow, loud speech may be associated with increases in speech intelligibility, voice quality and clarity. However, in this review we identified no randomised controlled group studies of interventions to improve the speech of children with dysarthria acquired below three years of age. Rigorous research is needed to investigate if the interventions described in observational studies, and advocated in dysarthria treatment texts, are generally effective in increasing the

intelligibility of children's speech and improving children's voice quality and clarity, as such changes have the potential to increase children's social and educational outcomes. Evidence would be best generated through randomised controlled trials. The observational studies identified in this review provide the data needed for the design and development of such trials. To generate evidence of treatment effectiveness future trials should investigate change in speech impairment and levels of conversation activity and participation. They must also test generalisation and duration of effects. Trials should therefore include: acoustic measures of voice production in single word speech, conversational speech and maximum performance speech tasks; change in speech intelligibility in single words and conversational speech to familiar and unfamiliar listeners; change in the short and medium term (e.g. one month and three months after treatment); change in performance in conversational activity and participation; participants' own perceptions of change and speech adequacy/acceptability.

AUTHORS' CONCLUSIONS

Implications for practice

No changes in practice are currently warranted.

Implications for research

Observational studies suggest that interventions teaching children with dysarthria to produce slow, loud speech may be associated with increases in speech intelligibility, voice quality and clarity. Rigorous research, in the form of randomised controlled trials, is needed to test the general effectiveness of speech and language therapy for children with dysarthria. Such research should evaluate changes in speech impairment and function, by measuring speech intelligibility, voice quality and clarity. As intelligible communication allows children to engage with the world around them it is important that future research also investigates the impact of intervention on children's activity and participation. This should include the extent and success of children's communication with friends, family, teachers and strangers; their engagement in social and educational activities; and their quality of life. The costs of intervention and the acceptability of therapy to children and their parents must also be examined.

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those of the NHS, the National Institute for Health Research or the Department of Health

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* Indicates the major publication for the study

CHARACTERISTICS OF STUDIES

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

Fischer-Brandies 1987	Observational study
Fox 2005	Observational study
Fox 2008	Observational study
Hartley 2003	Observational study
Marchant 2008	Observational study
Pennington 2006	Observational study
Pennington 2008	Observational study. Preliminary report, more detailed information given on same study in Pennington 2009.
Pennington 2009	Observational study
Puyuelo 2005	Observational study
Ray 2001	Observational study
Robson 2009	Observational study

DATA AND ANALYSES

This review has no analyses.

WHAT'S NEW

Last assessed as up-to-date: 17 May 2009.

18 May 2009	Feedback has been incorporated	Converted to new review format.
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HISTORY

Protocol first published: Issue 1, 2008

Review first published: Issue 4, 2009

5 October 2007	New citation required and major changes	Substantive amendment
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CONTRIBUTIONS OF AUTHORS

Lindsay Pennington and Nick Miller designed the study. Lindsay Pennington created the first draft of the review.

DECLARATIONS OF INTEREST

None known.

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- New Source of support, Not specified.

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