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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

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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.
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<th>Timing of measures</th>
<th>Methodological problems</th>
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<td>Fischer-Brandies 1987</td>
<td>71 children with cerebral palsy, 4-14 years (mean 10 years), orofacial dysfunction.</td>
<td>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).</td>
<td>15 months</td>
<td>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; labial sound production; palatal sound production; dental sound production.</td>
<td>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.</td>
<td>Beginning and end of treatment; timing not specified.</td>
<td>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.</td>
</tr>
<tr>
<td>Fox 2005</td>
<td>5 children (2F, 3 M), aged 5;10 - 7;10 years with spastic type cerebral palsy</td>
<td>Lee Silverman Voice Therapy Loud.</td>
<td>4 weeks: 16 one hour sessions (4 times per week for 4 weeks) plus minimum 36 practice sessions between treatment</td>
<td>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 therapy.</td>
<td>Two weeks prior to treatment, two weeks post treatment and six weeks post treatment</td>
<td>Two weeks prior to treatment, two weeks post treatment and six weeks post treatment</td>
<td>Two weeks prior to treatment, two weeks post treatment and six weeks post treatment</td>
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<tr>
<td>Study</td>
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<tr>
<td>Fox 2008</td>
<td>8 children (6F, 2 M)</td>
<td>4 weeks</td>
<td>16 one hour sessions (4 times per week for 4 weeks)</td>
<td>dB, maximum and minimum pitch in Hz, pitch range in Hz, elicited in maximum performance tests, sustained vowels, sentence repetition and cartoon description.</td>
<td>Increase in vocal SPL in sustained vowels ($F(2-12) = 5.14, p = 0.024$) post therapy and follow-up</td>
<td>Two weeks prior to treatment, two weeks post treatment</td>
<td>No blind rating of perceptual measures.</td>
</tr>
</tbody>
</table>
Hartley 2003

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.

Subsystems approach. 2 blocks of therapy. 1st block concentrated on respiration and phonation. 2nd block focused on articulation deficiencies noted during assessment.

Two four week blocks of therapy. Duration and frequency of sessions not specified. 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. Impairments scores on Robertson Dysarthria Profile (Robertson 1982) used for comparison of single word intelligibility.

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.

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.

Results of four cases presented as a group for intelligibility investigation.

Marchant 2008

One 13 year old girl with spastic type cerebral palsy, hemiplegic palsy and severe spastic dysarthria. Hearing and vision within normal limits. Comprehension and intelligibility of conversation within normal range.

Two blocks of therapy. 1st block: phonetic placement, articulation therapy involving teaching of correct movement patterns for targets sounds. 2nd block: relaxation of muscle groups using biofeedback from surface electromyography.

Two blocks of therapy each comprising ten sessions of 45 minutes over two weeks. Withdrawal of therapy for two weeks between therapy blocks. Surface EMG amplitude of left and right orbicularis oris and submentalis muscles; percentage single word, sentence and paragraph intelligibility.

Significant difference assumed if post therapy results were +/-1 SD from pre therapy scores. Single word intelligibility scores showed improvement. Percentages of syllables, words and sentences correct post therapy. No change in participants’ view of her speech disorder.

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, six weeks after therapy completion.

No medium or long term assessment of outcome. Therapists rating speech were not blind to aims of the study.

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

Copyright © 2009 The Cochrane Collaboration. Published by John Wiley & Sons, Ltd.
<p>| Pennington 2006 | 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. Whole system approach, targeting control of breath supply for speech production and prosodic contrasts. Individual therapy for 20-30 minutes. Five sessions per week for five weeks. 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. 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. One week prior to therapy, one week after therapy completion, six weeks after therapy completion. No control group or maturational control. |
| Pennington 2009 | 16 participants, (9 girls age 12-18 years, mean = 14 years, SD = 2), 15 with cerebral palsy, one with Worster-Drought. Nine Whole system approach which focused on stabilising the students’ respiratory and phonatory effort and control, speech rate and Three individual sessions of 30 minutes each per week for six weeks. Mean percentage intelligibility of single words (Children's Speech Intelligibility Measure) and connected speech to three Group and individual results presented. Following treatment 15/16 children were more intelligible to familiar and/or unfamiliar listeners. Six weeks and one week before therapy, one week and six weeks after therapy completion. No treatment integrity checks; longer term effects of intervention were not evaluated. |</p>
<table>
<thead>
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<td>Puyuelo 2005</td>
<td>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 &quot;absence of articulated speech&quot;. Hearing and language comprehension within normal limits.</td>
</tr>
<tr>
<td></td>
<td>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 phrase length/syllables per breath.</td>
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<tr>
<td></td>
<td>Two blocks of treatment. Each block comprised 11 months of twice weekly therapy, each session lasting 30 minutes.</td>
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<tr>
<td></td>
<td>Impairment scores on Spanish adaptation of Robertson Dysarthria Profile (Robertson 1982). Spectrographic analysis of a repeated sentence.</td>
</tr>
<tr>
<td></td>
<td>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.</td>
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- 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.

- Interventions 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 phrase length/syllables per breath.

- Familiar and three unfamiliar listeners. Listeners blind to time of recording for intelligibility measures. Questionnaire on the acceptability of therapy, using Likert scales.

- 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.

- Before intervention, between first and second interventions, after intervention two. Exact timing of measures not specified.

- No control group; long duration of treatment; no control of maturational effects; no blinding of assessor.
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<td>Ray 2001</td>
<td>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.</td>
<td>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.</td>
<td>Four-point rating scale of function of lips, jaw and tongue, by one orthodontist and two speech language pathologists. Percentage errors then converted to five-point scale.</td>
<td>Group difference in pre and post therapy scores for lip and tongue position and for percentage phonemes correct.</td>
<td>No blinding of assessors; no maturational or experimental control; no follow-up.</td>
</tr>
<tr>
<td>Robson 2009</td>
<td>Same as Pennington et al 2009</td>
<td>See Pennington et al 2009</td>
<td>Perceptual measures: 16 therapists rated severity of voice impairment from recordings blind to time of recording using validated four point scale.</td>
<td>Slight reduction in fundamental frequency, intensity and jitter of children’s voices. Slight increase in speaking time between pauses.</td>
<td>See Pennington 2009 No long term follow-up.</td>
</tr>
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</table>
Acoustic measures: HNR, RMS amplitude, shimmer, APQ, jitter, RAP, PPD, PEP, F0 mean, rate with pauses, rate without pauses, time with pauses, time without pauses.
**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; Nessell 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 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; Hedge 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 (Weissner 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 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.
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/
20. ((singl$ or doubl$ or trebl$ or tripl$) adj (mask$ or blind$s)).tw.
21. placebo$.
22. placebos.tw.
23. random$.
24. research design/
25. (latin adj square).tw.
26. or/17-25
27. 26 not 15
28. 27 not 16
29. comparative study/
30. exp evaluation studies/
31. follow-up studies/
32. prospective studies/
33. (control$ 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 Gehoer. (The current titles are given for journals experiencing name changes since 1980.)


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; articula-
tion; 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
to be calculated using the Kappa statistic. Individual criteria
were to be rated according to the Cochrane Handbook for Sys-
tematic 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 alloca-
tion 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

Adequate blinding of outcome assessors; researchers must be blin-
d to the type of intervention received by participants. 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) Adequate blinding of outcome assessors.
(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 continu-
ous 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 investi-
gated 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 nar-
rative 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 robust-
ness of review findings by investigating the impact of study qual-
ity: effects of randomisation; inadequate concealment; blinding
of outcomes; 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 remain-
ing 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.

ACKNOWLEDGEMENTS

We thank Cerebra for funding the salary of Sheila Robson and the UK National Institute for Health Research for supporting Lindsay Pennington’s salary during this review. This report is independent research arising from a Career Development Fellowship supported by the National Institute for Health Research. The views expressed in this publication are those of the author(s) and not necessarily...
those of the NHS, the National Institute for Health Research or the Department of Health

REFERENCES

References to studies excluded from this review

Fischer-Brandies 1987 (published data only)

Fox 2005 (published and unpublished data)

Fox 2008 (published and unpublished data)

Hartley 2003 (published data only)

Marchant 2008 (published and unpublished data)

Pennington 2006 (published and unpublished data)

Pennington 2008 (published and unpublished data)

Pennington 2009 (published and unpublished data)

Puyuelo 2005 (published data only)

Ray 2001 (published data only)

Robson 2009 (published data only)

Additional references

Bax 2006

Cahill 2002

Chapman 2001

Cornwell 2003

Dickinson 2007

Hayden 1994

Higgins 2003

Hodge 1999

ICAN 2007

Kennes 2002
Lepage 1998

Love 1992

Morgan 2008

Netsell 2001

Odding 2006

Pennington 1999

Petheram 2001

Richter 2005

Robinson 2002

Schmidt 2005

Strand 1995

van Mourik 1996

Weismer 2006

Yorkston 1996

Yorkston 1999

* Indicates the major publication for the study
**CHARACTERISTICS OF STUDIES**

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

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<td>Fox 2008</td>
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<td>Marchant 2008</td>
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<tr>
<td>Puyuelo 2005</td>
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<tr>
<td>Ray 2001</td>
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<td>Robson 2009</td>
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DATA AND ANALYSES

This review has no analyses.

WHAT’S NEW

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

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HISTORY

Protocol first published: Issue 1, 2008
Review first published: Issue 4, 2009

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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.

SOURCES OF SUPPORT

Internal sources

- New Source of support, Not specified.
External sources

- Cerebra (salary support for Sheila Robson), UK.
- National Institute of Health Research, UK.
  Salary support to Lindsay Pennington