The Cleft Palate-Craniofacial Journal 50(1) pp. e1–e17 January 2013

ORIGINAL ARTICLE

Speech and Language Therapy Interventions for Children With Cleft Palate: A Systematic Review

Alyson Bessell, M.Sc., C.Dip., Ph.D., Debbie Sell, L.R.C.S.L.T., F.R.C.S.L.T., Ph.D., Penny Whiting, B.A., M.Sc., Ph.D., Sue Roulstone, Med. Ph.D., F.R.C.S.L.T., Liz Albery, M.R.C.S.L.T., B.Sc., M.Phil., Martin Persson, M.Sc., M.P.H., Ph.D., Andrea Verhoeven, B.App.Sc., Margaret Burke, M.Sc., Andy R. Ness, D.P.H., Ph.D., F.F.P.H.M.

Objective: **(1) To examine the evidence for the effectiveness of differences in timing and type of speech and language therapy for children with cleft palate with or without a cleft lip and (2) to identify types of interventions assessed.**

Design: **Nine databases, including MEDLINE and EMBASE, were searched between inception and March 2011 to identify published articles relating to speech and language therapy for children with cleft palate with or without cleft lip. Studies that included at least 10 participants and reported outcome measures for speech and/or language measures were included. Studies where the experimental group had less than 90% of children with cleft palate with or without cleft lip were excluded. Two reviewers independently completed inclusion assessment, data extraction, and risk of bias assessment for all studies identified.**

Results: **A total of 17 papers were evaluated: six randomized control trials and 11 observational studies. Studies varied widely on risk of bias, intervention used, and outcome measures reported. None of the studies had a low risk of bias. In terms of intervention approaches, seven studies evaluated linguistic approaches and 10 evaluated motor approaches. Outcomes measures did not support either approach over the other, and based on data reported it was difficult to ascertain which approach is more effective for children with cleft palate with or without cleft lip.**

Conclusions: **The review found little evidence to support any specific intervention. Key uncertainties need to be identified and adequately powered, methodologically rigorous studies conducted to provide a secure evidence base for speech-language therapy practice in children with cleft palate with or without cleft lip.**

KEY WORDS: *cleft palate, speech, speech therapy, systematic review*

Dr. Bessell is Systematic Reviewer, School of Psychology, University of Plymouth, United Kingdom. Dr. Sell is Speech and Language Therapist, North Thames Regional Cleft Lip and Palate Service, Great Ormond Street Hospital for Children, London, United Kingdom. Dr. Whiting is Senior Research Fellow, School of Social and Community Medicine, University of Bristol, Bristol, United Kingdom. Dr. Roulstone, Faculty of Health and Life Sciences, University of the West of England. Ms. Albery is Speech and Language Therapist, South West Regional Cleft Lip and Palate Service, Frenchay Hospital, Bristol, United Kingdom. Dr. Persson is Project Manager, School of Oral and Dental Sciences, University of Bristol, Bristol, United Kingdom. Ms. Verhoeven is Senior Speech Pathologist, Cleft Palate/Craniofacial Team, The Royal Children's Hospital Melbourne, Australia. Ms. Burke is Information Specialist, School of Social and Community Medicine, University of Bristol, Bristol, United Kingdom. Dr. Ness is Professor of Epidemiology, School of Oral and Dental Sciences, University of Bristol, Bristol, United Kingdom.

This paper was presented at the Craniofacial Society of Great Britain and Northern Ireland Annual Conference in York, United Kingdom, April 2011.

Submitted August 2011; Accepted March 2012.

Address correspondence to: Dr. Alyson Bessell, University of Plymouth, School of Psychology, Portland Square, Drake Circus, Plymouth, United Kingdom, PL4 8AA. E-mail Alyson.bessell@plymouth.ac.uk.

DOI: 10.1597/11-202

It is estimated that cleft palate with or without lip (CP±L) occurs in approximately 1 in 700 live births, making CP±L one of the most common congenital malformations (Vieira, 2008). A cleft lip is usually repaired at 3 months of age and a cleft palate from 6 to 12 months (Vieira, 2008); although, in the past repairs were carried out later (sometimes up to 3 years of age). Cleft palate surgery is more controversial with considerable variation internationally in timing, various stages and sequence of the repair process, and the surgical technique used (Lohmander, 2011).

Speech is considered one of the primary outcome measures of palate repair. Although cleft palate repair is now usually carried out within the first year of life, children continue to present with speech difficulties. An audit of cleft services conducted in 1998 (Clinical Standards Advisory Group, 1998) found that around 29% of children with unilateral cleft lip and palate (UCLP) needed further surgery to improve elements of

speech, such as hypernasality (Sell et al., 2001). Hardin-Jones and Jones (2005), in their analysis of the speech of 212 preschool children with repaired cleft palates, found that 68% had received speech therapy and that despite advances in the overall management of these children, the majority still required speech therapy.

Within the field, outcome measures for "speech" have been highly controversial, with a wide range of approaches used including both objective and subjective measures. For example, objective measures have included acoustics and electropalatography (EPG). Perceptual measures, associated with judging the speaker's intelligibility, are part of an overall evaluation of speech. They usually include judging resonance, especially hypernasality and hyponasality, and judging nasal emission, which is a measure of consonant production.

Fortunately, as detailed below, there has been some progress in international consensus regarding perceptual outcome measures, with greater recognition of the complex process for ensuring reliable and valid perceptual outcomes (Sell, 2005; John et al., 2006; Lohmander et al., 2009). Language outcome measures have also been reported, usually based on formal standardized assessments and/or a judgment of language performance in terms of morphology, phonology, syntax, semantics, and pragmatics.

Within the field of speech and language intervention for children with CP±L, traditional articulation approaches that focus on modifying atypical production of speech sounds (Van Riper, 1978) have been widely used. With the development of the discipline of clinical linguistics, phonetic and phonological approaches started to be applied to the assessment and intervention of speech sound disorders in the 1970s (Grunwell, 1993). More recently psycholinguistic approaches have also been used with this population (Stackhouse and Wells, 1997). The psycholinguistic approach investigates the underlying speech processing skills, including both speech input and output, compared with existing stored lexical representations. Its relevance for speech and language therapy is that irrespective of the speech or medical diagnosis, the level of breakdown in the speech processing profile is identified and targeted in therapy (Stackhouse, 2011). This approach is usually used in combination with other approaches, especially phonetic and phonological. Some speech and language therapists have offered early intervention in the form of babble workshops for parents and their infants, which aim to modify deviant and restricted early sound development (Russell and Grunwell, 1993); whereas, the majority of other approaches take place from the age of 3 extending into the school years. Two other approaches are input modeling, which aims to stimulate speech output by increasing awareness of speech sounds (Harding and Bryan, 2002) and oral motor activities. Approaches vary greatly from therapist to therapist, and no uniform approach has been taken.

There have been no systematic reviews to summarize the current state of the evidence base for speech intervention in children with CP±L. The most comprehensive literature review was produced by Enderby and Emerson (1995), who concluded that research to investigate the effectiveness of interventions for speech impairment "was somewhat sparse" (p. 103) and was of dubious quality. In particular, they noted the lack of reliable and sensitive speech and language outcome measures, as well as problems with the heterogeneity of the populations under study. This current review aimed to identify the types of intervention that have been assessed. The review also aimed to assess the evidence for the effectiveness of current theoretical models of speech-language therapy (SLT) interventions, the most appropriate age of delivery, and the optimum intensity, duration, and delivery method for improving the speech of children with CP±L.

Methods

Identification of Studies

MEDLINE, EMBASE, ISI Web of Knowledge, CINAHL, PsycINFO, Cochrane Central Register of Controlled Trials (CENTRAL), Cochrane Database of Systematic Reviews, Database of Abstracts of Reviews of Effects (DARE), and the Health Technology Assessment Database were searched from inception to March 2011. The search terms included indexing and text words associated with cleft lip and/or palate and speech and language therapy (see Appendix A for the MEDLINE strategy). No language restrictions were applied. In addition, Web sites including National Institute of Clinical Excellence (NICE) and the meta-Register of Controlled Trials (mRCT) were searched, and the references of identified studies were screened.

Inclusion Criteria

Studies had to fulfill the following criteria to be included in the review.

Study Design

Studies included both randomized and non-randomized controlled trials (trials are studies that attempt to control for key elements of bias within the design). Observational comparative studies, either within or between groups, were also included to identify types of interventions assessed.

Patients

Children with CP±L were included in the review. Studies had to include at least 90% children with nonsyndromic CP±L or include syndromes with no known developmental delay (e.g., Stickler, hemifacial

microsomia). Children with isolated cleft lip only were excluded.

Intervention

Any SLT intervention was included within the review.

Comparison

No intervention control group or a different SLT intervention were used as comparators.

Outcome

Any speech and/or language outcome was included within the review. Studies reporting only psychosocial outcomes were excluded.

The results of the searches were screened independently by two reviewers (A.B. and P.W.). Articles considered potentially relevant were obtained and assessed for inclusion independently by two reviewers (A.B. and M.P.). All three reviewers were experienced researchers. Disagreements were resolved through consensus or referral to a fourth reviewer (D.S., an experienced clinician) when necessary and possible.

Data Extraction and Analysis

Descriptive Information

Two reviewers (A.B. and A.V. [an experienced clinician]) independently extracted the following descriptive data from the included studies: author, publication year, country of study, study design, population characteristics, setting, inclusion/exclusion criteria, intervention details, outcome measures, and results data.

Types of Intervention Assessed

Observational studies were also included for analysis of the interventions. The authors used a qualitative approach to synthesize data across studies (Dixon-Woods et al., 2005) and focused on three main areas: theoretical or therapeutic perspective, method of delivery (setting, person delivering the intervention), and timing of the intervention (age of intervention, intensity, and frequency of the intervention).

Meta-Analysis of Trials Only

Outcome Measures

Primary outcome measures of speech and language were extracted, followed by secondary outcomes of psychosocial outcomes (e.g., confidence, quality of life, social integration).

Effect Sizes

Standard mean differences (SMDs) and/or effect sizes together with 95% confidence intervals (CIs) were extracted for continuous outcomes, and odds ratios (ORs) together with 95% CIs were extracted for dichotomous outcomes. These figures were taken directly from the papers or calculated based upon raw data provided within the papers where necessary. Effect sizes and CIs were plotted using forest plots. This approach was used because the studies were too heterogeneous, both clinically and statistically, to permit statistical pooling and so a narrative synthesis was conducted.

Disagreement was resolved through discussion and consultation with the fourth reviewer (D.S.). Study authors were contacted for additional information where necessary and possible.

Assessment of Risk of Bias

Two reviewers (A.B. and A.V.) independently assessed studies using the Cochrane Risk of Bias tool (Higgins and Green, 2011) to rate each of the following five components as high, low, or unclear risk of bias: (1) method of sequence generation (how the randomization sequence was generated, e.g., random number table), (2) method of allocation concealment (how the randomization was concealed from researchers/clinicians), (3) method of blinding of the outcome assessor (how the study ensured the outcome assessor did not know the condition to which the participant had been allocated), (4) selective reporting of outcome data (not reporting all outcomes in the results that are mentioned in the method or failing to use standard outcome measures within a particular field of research), and (5) completeness of outcome data (whether all attritions were accounted for and whether an intention to treat analysis was performed). In the case of SLT, it is not possible for the treating clinicians or participants to be blind to their treatment allocation; therefore, adequate blinding was considered to have taken place if both data analysis and outcome assessment were blinded. Selective reporting of outcome data was considered to have taken place when papers reported only outcomes that had a favorable result or if the outcome measures assessed did not include standard measures that experts in the area would expect to have been reported.

Results

Types of Intervention Assessed

The searches identified 1305 references, of which 17 were included in the review (Fig. 1). The total number of

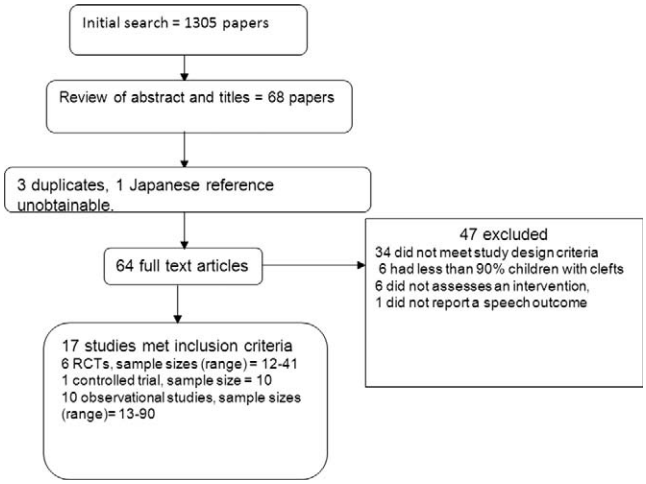

FIGURE 1. Flowchart of review search results and numbers of included papers.

participants across the 17 studies meeting inclusion criteria was 654, with a range of 10 to 90 participants per study. Six randomized controlled trials (RCTs) (n = 150 participants) and 1 controlled trial (n = 20 participants) were included in the meta-analysis. Another 10 observational studies were included for the evaluation of interventions: six cohort studies, three before-and-after studies, and one with an unclear design. Studies were conducted in Mexico (seven studies all from the same research center [n = 278 unique participants]), United States (four studies), China (four studies), U.K. (one study) and Denmark (one study). Four studies were conducted prior to 1990, six were conducted between 1990 and 2000, and the remaining seven studies took place after 2000. Table 1 provides an overview of the included studies with more detailed descriptions available in Appendix B.

Meta-Analysis

Assessment of Risk of Bias

The assessment of risk of bias was based upon the combined judgments of the five separate components (see Table 2). All six of the RCTs were judged as having unclear risk of bias due to unexplained sequence generation, unclear allocation concealment, inadequate blinding of outcome assessment, and the presence of missing outcome data (see Table 2). Only one study reported adequate sequence generation (Pamplona and Ysunza, 2000), none reported adequate allocation concealment, and four studies had adequate blinding of outcome assessment. All of the studies reported complete outcome data, and there was no evidence of selective outcome reporting (Table 2).

Outcome Measures (Effects on Speech)

All seven trials (six RCTs and one controlled trial) that are included in the meta-analysis reported data on speech. The exact outcome measures varied across studies with three dichotomizing the outcome into the proportion of children with adequate or inadequate speech, persistent or modified compensatory articulation disorder (CAD), and positive change or no change in speech (Fig. 2). Two further studies used ordinal data scales to report levels of speech ability and levels of CAD. Both these scales had four levels that were reorganized into two categories for analysis as binary data. Speech ability was grouped as low or high and levels of CAD as low (none or mild) and high (moderate or severe). All other outcomes were continuous and included scales to assess intelligibility, acceptability of speech, and correction of CAD, as well as validated scales for linguistic level, all based on free speech samples (Fig. 3).

Two studies (Ysunza et al., 1997; Gibbon et al., 2001) suggested a beneficial effect of computer-generated speech assessment (EPG or nasoendoscopy) combined with visual feedback versus no feedback.

One suggested that EPG compared with conventional therapy not involving visual feedback increased the proportion of children showing an improvement in *s* (OR, 10.7, 95% CI, 0.40 to 287.8) and *t* sounds (OR, 3.3, 95% CI, 0.2 to 54.5). However, the study was very small (n = 12) and CIs were wide (Gibbon et al., 2001). The other study provided evidence for a reduction in the severity of CAD (OR, 96.3, 95% CI, 3.4 to 2715.3) for nasoendoscopy combined with feedback compared with nasoendoscopy alone (Ysunza et al., 1997) (see Fig. 2 for a summary of these studies).Two studies by Pamplona and Ysunza (2000) and Pamploma et al. (1996) suggested a greater improvement in linguistic level with parental interaction compared with no parental interaction (SMD, 1.5, 95% CI, 0.84 to 2.24; SMD, 0.89, 95% CI, 0.00 to 1.79). Two other studies showed an improvement in time to correct CAD for whole language (SMD, –0.36, 95% CI, –1.08 to 0.36) and motor approach (SMD, –1.96, 95% CI, –2.85 to –1.08) compared with a purely phonological approach (Pamplona et al., 1999; Pamplona et al., 2004). The controlled study (Scherer et al., 2008) showed no effect of focused stimulation compared to intervention on receptive language (see Fig. 3 for a summary of these studies).

Outcome Measures (Psychosocial Outcomes)

Although none of the other studies assessed psychosocial outcomes, two studies reported on level of play (Pamplona et al., 1996; Pamplona and Ysunza, 2000). One study (Pamplona and Ysunza, 2000) suggested that

parental interaction improved play compared with no interaction (SMD, 0.44, 95% CI, –0.43 to 1.31); whereas, the second study by the same author (Pamplona et al., 1996) suggested that interaction reduced play (SMD, –0.43, 95% CI, –1.05 to 0.19). Therefore, the findings are inconclusive regarding the effects of speech and language therapy interventions on play. The results are shown in Figure 4.

Evaluation of Interventions

Descriptive Information: Therapeutic Approach

The therapeutic approaches used in the studies fall broadly into two groups: motor and articulation versus linguistic approaches (see Fig. 5). Figure 5 shows the differences among these approaches and the kinds of techniques incorporated within them. This section summarizes the different approaches taken to highlight the types of interventions currently in use with children with CP±L. One study compared more than one approach (Pamplona et al., 1999): the traditional motor approach compared with the phonological approach (see "Multiple Approaches" section).

Motor Approaches

Seven studies focused on motor movements specifically in the context of speech, often referred to as the traditional articulation approach or phonetic approach (Van Demark, 1974; Van Demark and Hardin, 1986; Ma et al., 1990; Chen et al., 1996; Pamplona et al., 1999; Yang et al., 2003; Hardin-Jones and Chapman, 2008). In this approach a single phoneme at a time is worked upon in therapy. Once elicited, often a challenge in this type of etiology, it is practiced within consonant-vowel–type nonsense syllables, before moving onto establishing the sounds in words and sentences, and finally working on carryover in connected speech. Throughout the intervention process, auditory awareness is emphasized to the participant. These techniques follow an articulatory approach (see Fig. 5).

Two studies focused on the more general motor movements that are used when sucking and maintaining oral pressure but not in the context of speech (see Fig. 5). The approach focuses on sucking and blowing and on general lip and tongue movements not associated with speech motor movements (Regan and Versaci, 1977; Ma et al., 2003).

Two other studies that took a primarily motor approach included those that focus on the use of visual feedback through the use of EPG (Gibbon et al., 2001) or nasopharyngoscopy (Ysunza et al., 1997) (see Fig. 5). These had their roots in traditional articulatory approaches yet not only provided the usual auditory feedback but also additional visual feedback.

Linguistic Approaches

Seven studies (Van Demark and Hardin, 1986; Pamplona et al., 1996; Pamplona and Ysunza, 2000; Pamplona et al., 2004; Pamplona et al., 2005; Scherer et al., 2008; Pamplona et al., 2009) used linguistic approaches that included phonological approaches (where children are introduced to multiple sounds or sets of sounds working toward the child's understanding and production of the rule-based phonological sound system), focused stimulation (where the child is presented with vocabulary targeted at particular sounds), or whole-language approaches (where the emphasis is on creating successful communication experiences for the children) (see Fig. 5 and Table 1).

Multiple Approaches

Only one study contrasted the different approaches (Pamplona et al., 1999). This study compared the traditional articulation approach with the phonological linguistic approach. This study, although an RCT, had a small sample size (n = 29) and was rated as having unclear risk of bias due to unexplained sequence generation and allocation concealment (Table 2). The remaining studies compared different types of linguistic approaches with one another, compared one type of intervention with no intervention, or compared parental interaction with no interaction.

One study tested the phonological approach against the whole-language approach (Pamplona et al., 2004). Again, this study was a small (n = 30) RCT but was found to have a low risk of bias. All the other studies were case series. Additionally, none of these studies could be compared because each used different outcome measures.

Finally, two additional studies looked at the inclusion of the additional effect of visual feedback (EPG and nasopharyngoscopy) when used alongside a motor or phonetic approach (Ysunza et al., 1997; Gibbon et al., 2001). These two RCTs were small and had an unclear risk of bias (n = 29).

Descriptive Information: Delivery of Intervention (Setting and Facilitator)

The settings of the interventions varied. Whereas most were conducted in the clinic setting, four studies were carried out in the home and three studies assessed the effectiveness of a therapy provided at a summer camp (see Table 1). The three studies looking at summer camps (Van Demark and Hardin, 1986; Pamplona et al., 2005; Pamplona et al., 2009) did not find a difference between this setting and a clinic setting. However, Pamplona et al. (2005) reported that the costs of running an intensive summer camp course were

TABLE 1 Characteristics of Included Studies

Study	*N*	*Location*	*Cleft Type*	*Age*	*Study Design*	*Intervention*	*Comparator Intervention*
Pamplona et al., 1999	29	Mexico	UCLP*	3–7 y	RCT	Motor: Trad	Linguistic: phono
Gibbon et al., 2001	12 (8 male*)	U.K.	UCLP (7), BCLP* (2), SCP (3)	5–18 y	RCT	Motor: EPG	Conventional therapy with no visual feedback
Ysunza et al., 1997	17	Mexico	UCLP	11–13 y	RCT	Motor: nasoendoscopy + feedback	Motor: nasoendoscopy, no feedback
Pamplona et al., 2004	30	Mexico	UCLP	3–7 y	RCT	Linguistic: WL*	Linguistic: phono
Pamplona et al., 1996	21	Mexico	UCLP	3–5 y	RCT	Linguistic: WL with parental interaction	Linguistic: WL without parental interaction
Pamplona and Ysunza, 2000	41	Mexico	UCLP	3–4 y	RCT	Linguistic: FS parental interaction	Linguistic: FS no parental interaction
Scherer et al., 2008	10 (plus 10 children without CLP as control; 9 male)	USA	CLP, non CLP	1–3 y	CT	Linguistic: FS*	Linguistic: Phono WL at summer camp
Hardin-Jones and Chapman, 2008	30 (plus 10 children without clefts, 33 male)	USA	BCLP (6), UCLP (20), hard and soft palate (3), SCP (1)	2–3 y	Cohort	Motor: Trad	No intervention
Yang et al., 2003	16 (8 male)	China	CP with obturator plate	5–29 y	Cohort	Motor: Trad	No intervention
Ma et al., 1990	30 (12 male)	China	CP	4–6 y	Cohort	Motor: Trad	No intervention
Van Demark, 1974	67	Denmark	CP	5–7 y	Cohort	Motor: Trad	No intervention
Pamplona et al., 2009	50	Mexico	CP	Mean 42 ± 11 mo	Cohort	Linguistic: WL at summer camp	Linguistic: WL in clinic
Pamplona et al., 2005	90	Mexico	UCLP (45), CP (45 control)	3–10 y	Cohort	Linguistic: Phono WL at summer camp	Linguistic: Phono WL in clinic
Chen et al., 1996	33 (15 male)	China	CP	4–16 y	Before and after	Motor: Trad	N/A
Regan and Versaci, 1977	45	USA	CP±L	18 mo–7 y	Before and after	Motor: nonspeech	N/A
Van Demark and Hardin, 1986	13 (9 male)	USA	CP±L	6–12 y	Before and after	Linguistic: Phono*	N/A
Ma et al., 2003	67 (42 male)	China	CP	4–11 y	Unclear (abstract only)	Motor: nonspeech focus	No intervention

* Not all studies reported gender. Figures are provided where reported. UCLP = unilateral cleft lip and palate, BCLP = bilateral cleft lip and palate, WL = whole language, FS = focused stimulation, phono = phonological.

TABLE 1 Extended

Setting	*Facilitator*	*Intensity*	*Duration*	*Results*	*Follow-Up*
Clinic	Therapist	Not stated	Not stated	Standard mean difference = 1.936	Until compensatory articulation had been corrected (6–22 mo)
Clinic	Therapist	Not stated	Not stated	"s" sound: odds ratio = 10.714 "t" sound: odds ratio = 3.333	None
Clinic	Therapist	Not stated	Not stated	Odds ratio = 96.333	12 wk
Clinic	Therapist	Not stated	Not stated	Standard mean difference = 0.361	4–27 mo
Clinic	Therapist	104 h, hourly sessions	3 times a wk for 8 mo	Standard mean difference = 0.895 Levels of play (secondary outcome): standard mean difference = −0.433	8 mo
Clinic	Therapist	3 h	4-monthly in small groups	Standard mean difference = 1.539 Levels of play (secondary outcome): standard mean difference = 0.441	12 mo
Home	Parents	Not stated	Not stated	Expressive language: standard mean difference = 0.117 Receptive language: standard mean difference = 0.392	None
Home	Therapist	Not stated	Not stated	Auditory comprecation: intervention mean = 105.4, comparator mean = 107.22	27 mo of age
Clinic	Therapist	Not stated	Not stated	Formant frequency of *A*: intervention mean = 2391, comparator mean = 2391 Formant frequency of *E*: intervention mean = 2146, comparator mean = 2466 Formant frequency of *I*: intervention mean = 3277, comparator mean = 2970 Formant frequency of *U*: intervention mean = 2873, comparator mean = 2890	6 mo
Clinic	Therapist	8–13 h	Once a wk for 6 mo	Correct lip/tongue movements: intervention mean = 12, comparator mean = 12 Correct blowing: intervention mean = 16, comparator mean = 16 Correct mandibular movements: intervention mean = 18, comparator mean = 18	6 mo
Clinic	Therapist	Not stated	Not stated	Intervention mean = 35.11, comparator mean = 49.62	2 y
Summer camp/ clinic	Therapist	Standard therapy: 58.5 h, 2 × 45 min/wk Summer camp: 60 h, 4 h/d	Standard therapy: 9 mo Summer camp: 5 d/wk for 3 wk	Situational speech: odds ratio = 0.603 Discourse (contextual) speech: odds ratio = 1.446; Semantic speech: odds ratio = 1.556	9 mo
Summer camp/ clinic	Therapist	Standard therapy: 69 h, 2 h/wk Summer camp: 60 h, 4 h/d	Standard therapy: 8 mo Summer camp: 5 d/wk for 3 wk	Odds ratio = 3.143	Up to 12 mo
Clinic	Therapist	15–124 h, daily for 0.5–1 h	1–2 times a wk	Mean before therapy = 38.43, mean after therapy = 89.54	None
Home	Parents	Not stated	Not stated	23 out of 27 had adequate speech	None
Summer camp	Therapist	26 h, 1 h/d	26 d	Articulation defectiveness: mean before therapy = 3.88, mean after therapy = 2.54 Nasality: mean before therapy = 1.94, mean after therapy = 1.31	9 mo
Home	Parents	5–10 h	20–40 d		12 mo

TABLE 2 Risk of Bias in RCTs*

Study	*Study Design*	*Sequence Generation*	*Allocation Concealment*	*Method of Blinding of Outcome Assessor*	*Completeness of Outcome Data*	*Reporting of Outcome Data*
Pamplona et al., 2004	RCT	Low	Unclear	Low	Low	Low
Gibbon et al., 2001	RCT	Unclear	Unclear	Unclear	Low	Low
Pamplona and Yzunsa, 2000	RCT	Unclear	Unclear	Low	Low	Low
Pamplona et al., 1999	RCT	Unclear	Unclear	Low	Low	Low
Ysunza et al., 1997	RCT	Unclear	Unclear	High	Low	Low
Pamplona et al., 1996	RCT	Unclear	Unclear	Low	Low	Low
Scherer et al., 2008	CT	High	High	Low	Low	Low

* Low = low risk of bias; High = high risk of bias; Unclear = information in the paper not sufficient to assess risk of bias; CT = controlled trials.

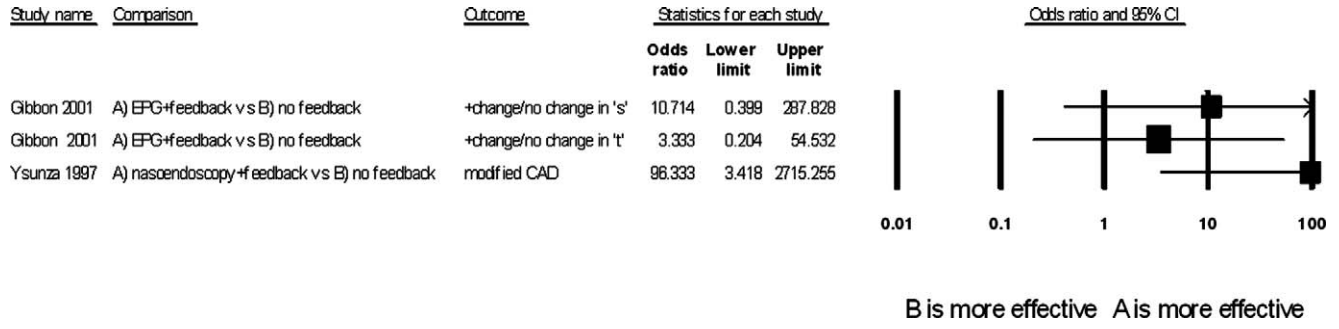

FIGURE 2. Odds ratios for binary primary outcomes.

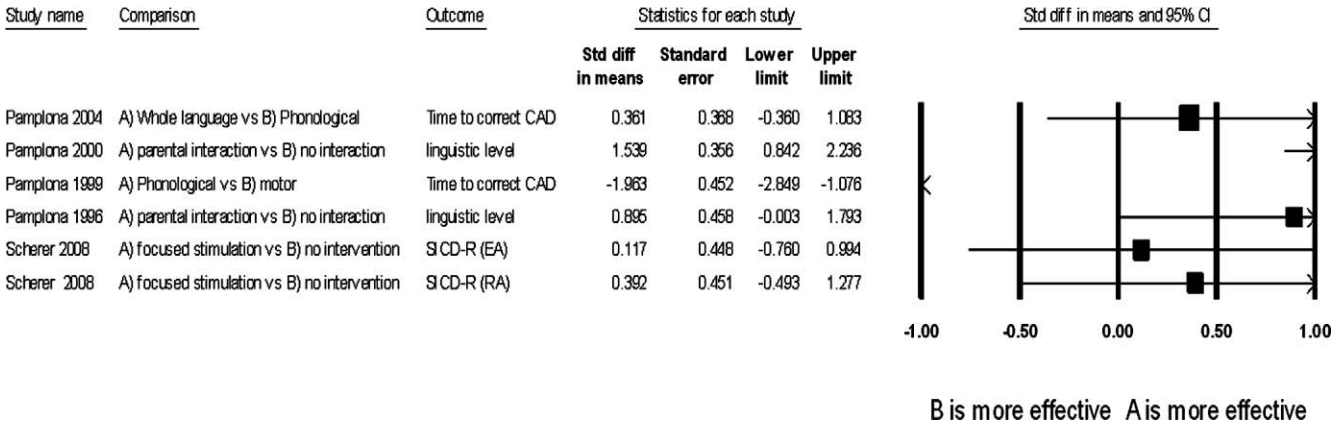

FIGURE 3. Standard mean differences for continuous primary outcomes.

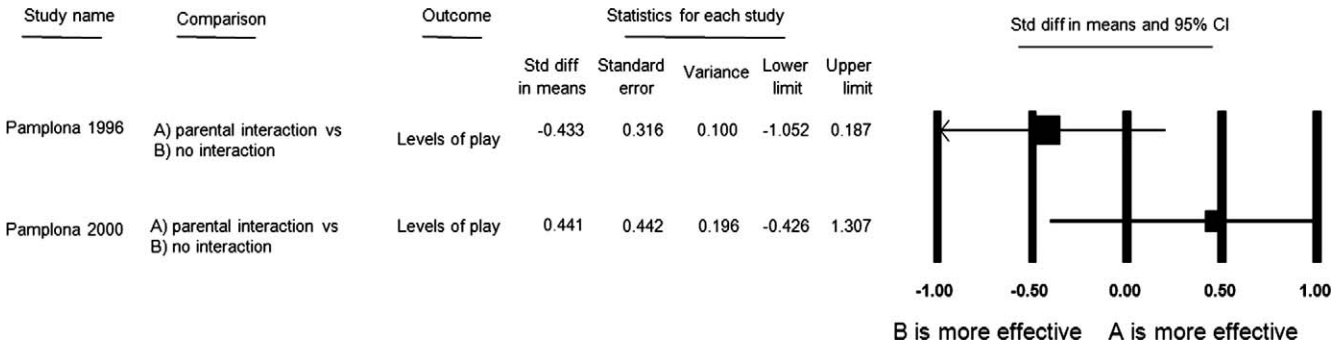

FIGURE 4. Standard mean differences for continuous secondary outcomes.

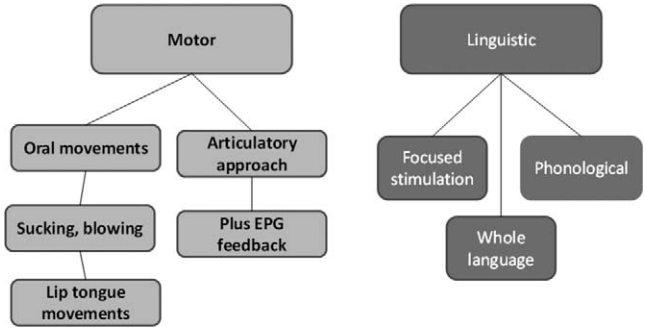

FIGURE 5. Speech and language therapy approaches to cleft lip and palate.

considerably cheaper than the same therapy administered in the standard way within clinics (US $100 and US $412 US, respectively; see paper for details).

Interventions were delivered by parents or speech and language professionals. Those interventions using parents produced general advice for them with interventions designed to be used regularly at home, particularly focusing on language. These contrasted sharply with therapy targeted at consonant errors that require speech and language professionals, trained parents, speech and language assistants, or teaching assistants (see Table 1).

Descriptive Information: Timing of Intervention (Duration, Intensity, and Age of Delivery)

Only eight studies reported the duration of the intervention. Of these, five studies were delivered over 20 to 60 days. One intervention ran for 6 months, one for 8 months, one for 9 months, and a remaining intervention was conducted for 1 year (see Table 1 for details).

The intensity of the interventions also varied from three times a year, weekly, twice weekly, three times a week, or five times a week to daily (Table 1).

The age of therapeutic intervention varied across the studies. Overall, interventions took place between 12 months and 29 years, often with large age ranges included within the same study. For example, the study by Yang et al. (2003) included 49 participants aged 5 to 29 years (see Table 1).

Discussion

Summary

This review identified 17 studies of various SLT interventions in children with CP±L. Of these studies only seven were trials (six RCTs and one controlled trial) and contributed to the meta-analysis of effectiveness.

Effectiveness of Interventions

Most studies suggested beneficial effects of the interventions on speech and language outcomes, but effect sizes varied and confidence intervals were wide, making it difficult to draw overall conclusions based on the current published evidence in studies of children with CP±L. Only two studies assessed psychosocial outcomes, and these reported conflicting results despite comparing the same interventions and being conducted by the same authors. Differences between studies meant there was no consistent evidence to support specific settings for interventions. The review could not determine the optimal duration (number of sessions) or intensity (regularity of sessions) of therapy or who was best placed to facilitate such interventions (e.g., parents, teaching assistants, or speech therapists or pathologists).

Assessment of Risk of Bias

All had small sample sizes and had an unclear risk of bias. It was noted that five of the seven trials included in the analysis were published by a single author and completed at the same site. This raises concern about generalizability of the data.

Evaluation of Intervention Types

To evaluate the types of interventions used, an additional 10 small observational studies were included, all of which had methodological weaknesses. A variety of SLT techniques were reported in the studies. In most cases, the descriptions of the therapeutic approaches were limited. The studies did not provide sufficient evidence to support any particular theoretical approach or provide enough information about the optimum length, setting, or age of intervention.

Intervention Effectiveness: Current Thinking and Practice

There is some literature that provides further evidence from relevant noncleft populations. McCauley et al. (2009) undertook a systematic review in non–cleft-related speech impairment on the effect of nonspeech oral motor exercises on speech physiology, speech production, and intelligibility. They concluded that there is insufficient evidence to support or refute their use to produce effects on speech. A Cochrane review of EPG also concluded that there was no evidence on which to recommend the technique for children with clefts (Lee et al., 2009).

Many of the recent phonological therapy approaches that have been published within the speech impairment literature have not been tested with this group (Bowen, 2009). Cleft speech is conceptualized as an articulation disorder with phonological consequences (Grundy and Harding, 1995). Therefore, future studies should aim to evaluate the evidence to support the use of speech-related motor techniques, such as the traditional articulation approaches in comparison with or in addition to linguistic techniques such as phonetic and phonological approaches. In addition, focused stimulation or whole-language approaches should be evaluated further in preschool children. Other specific intervention models that require further testing include multisensory input modeling (Harding and Bryan, 2002), which provides children with multiple exemplars of target behavior to raise conscious awareness of how sounds are made. As yet there is only clinical-level evidence for the success of this intervention strategy.

Speech Outcomes

Within cleft SLT interventions there is wide variation in the nature of the outcome measures used (see Appendix B), in part because the aims of the studies are also hugely variable, from those aiming to change speech sound production, improve velopharyngeal closure, or improve language skills. It is noteworthy that those measuring speech do not always meet the current standards for reporting outcomes. For example, Sell and Grunwell (2001) discuss the problem of simplistic and inadequate speech reporting systems where speech is reported using 4-point gross classification scales, as found in several studies in this review. Sell (2005) and Lohmander and Olson (2004) have each described current standards for reporting outcomes in speech studies, and most of the studies reviewed fall below these. When measuring speech outcome, current thinking is that perceptual speech assessment (human listener) rather than instrumental assessment has the greatest face validity (Kuehn and Moller, 2000). It is also important to evaluate different speech parameters (Lohmander and Olsson, 2004; John et al., 2006; Henningsson et al., 2008), such as that of hypernasality/hyponasality, nasal airflow errors, and consonant production, as separate entities. There is, however, considerable controversy on the use of intelligibility as an outcome measure (Witzel, 1991; Whitehill, 2002; Henningsson et al., 2008) due to it being affected by speaker variables, listener variables, external factors such as context, the message content, and background noise, and the complexity of its measurement.

In early intervention studies measures of language as well as speech are appropriate because there is some evidence that language-based interventions improve speech production (Scherer et al., 2008). Furthermore, it is important to interpret speech outcomes within the framework of development and particularly language skills in the early years, given the known risk for delayed expressive language development in children born with CP±L and the impact this may have on phonetic and phonological development.

Additional Outcomes

Historically SLT has focused on outcome studies almost exclusively at the level of the impairment, for example, the disorder as a result of pathology. The World Health Organization (1980, 2001) has advocated broader outcomes describing the impact of the impairment on an individual's functional performance (activity) and his or her social consequences (participation). Therefore, there is a need to measure the impact of the speech impairment on a person's social functioning (McCormack et al., 2009). In this review studies that measured only psychosocial factors were excluded because speech is generally viewed as the primary outcome for SLT intervention, but secondary outcome assessment of psychosocial functioning is important.

Alongside the clinical assessment of effectiveness, it is also important for clinicians to consider the costs of interventions and the effect on social functioning (McCormack et al., 2009). For example, summer camps or intensive residential courses may be a more cost-effective method of delivery that may prove beneficial from a psychosocial perspective and help increase motivation to improve.

Study Design

Systematic reviews base the evidence for intervention models on a hierarchy of methodological designs (Centre for Reviews and Dissemination, 2001). The criterion standard for evidence of effectiveness is taken from high-quality RCTs with large sample sizes. Although RCTs are the criterion standard, they are not always possible. In these instances observational studies can provide useful evidence, but they should be conducted to the highest methodological standard with sample sizes based on calculations that allow the detection of realistic and clinically important effects.

Some have argued that systematic reviews of SLT interventions are problematic due to the limited research base within the field (Pring, 2004). Although it is true that there are few well-designed, adequately powered RCTs, there are observational studies that may provide evidence to guide current practice and future research (Pring, 2004). In these cases it is still possible for these observational studies to be reviewed in a systematic way; though meta-analyses may not be possible,

narrative syntheses of intervention models within systematic reviews provide both researchers and clinicians with a firmer basis on which to design future interventions. It is still important, however, for future studies to adopt the RCT methodology in order to reduce the risk of bias in study designs.

Limitations of the Review

The review examined SLT interventions of varying types. This makes it difficult to compare effect sizes or to pool them for the purposes of meta-analysis. Studies had small sample sizes, and no study findings were replicated either in the same or in other centers.

For the purposes of this review all studies with a sample size of fewer than 10 were excluded. This was due to the high risk of bias present in these studies and the absence of a control group. Pring (2004) stated that case studies are useful to assess whether a potential treatment has an effect that justifies further investigation in an RCT. Case studies can also give an indication as to why a treatment effect has been found (Pring, 2004). These case studies need closer inspection by researchers to highlight key approaches that could be assessed in future studies. But it is important that such studies are replicated with larger participant numbers in order to lend some support to the techniques.

It is unclear whether it is possible to generalize results across different languages, countries, and cultures. The possible difficulties associated with generalizing the findings from different countries must be considered in relation to this review. Seven of the studies were conducted in Mexico and another four were conducted in China where time of surgery and resources may be very different from those of countries such as the U.K. and the United States. In the developing world context, Sell et al. (2011) summarized the challenges of speech outcome studies, described different models of speech therapy delivery, and highlighted attitude, cultural, and language issues for both service providers and users. The differences in resources between developing and developed countries may make comparisons difficult. Most obvious is the lack of timely surgery and speech and language therapy professionals in many parts of the developing world. Sell et al. (2001) described many differences and considerations such that caution should be exercised in applying interventions in one context to patient groups in another totally different context. With regard to languages, not only do the consonant sound systems and the phonotactic structures differ, and as a result, their susceptibility to the cleft condition, but also there are languages that are tonal in nature. The impact of the latter in speech therapy intervention studies in cleft palate is unknown. Notwithstanding, it is generally considered that the characteristics of cleft speech are essentially universal (Henningsson et al., 2008).

Implications for Practice

This review was unable to provide any evidence to support any specific model of SLT intervention in relation to theoretical perspective, duration, age, setting, intensity, and delivery.

Implications for Research

Future researchers within the field of SLT intervention need to consider addressing the key uncertainties within the literature and addressing these using adequately designed studies that are well reported. In order for reviews to be most useful in the future it is important that papers of SLT interventions include detailed information about the intervention and its underlying therapeutic approach. It is also necessary to describe the intervention setting, who provided it, and the frequency and intensity of the intervention in sufficient details to allow replication (Marshall et al., 2010). Where possible, future studies should take the form of RCTs and use core outcome measures as standard.

The review has highlighted the need to compare motor-based phonetic approaches with linguistic approaches in future studies.

Conclusion

We found a lack of evidence to support any one existing intervention. The review was unable to identify the optimum setting, duration, intensity, age, or facilitation method of SLT interventions. The studies included in this review provide some suggestions for future models of intervention. However, further research is required to identify the elements of SLT interventions that may be effective, including their impact on other aspects of children's communication and psychosocial well-being.

Acknowledgments. Thank you to Alla Sorokin, Chihiro Sugiyama, and Joanna Wyrwoll for providing support with translations, and thank you to Anne Harding-Bell for providing information about the content of babble workshops for babies with clefts.

References

Bowen C. *Children's Speech Sound Disorders*. Oxford: Wiley-Blackwell; 2009.

Centre for Reviews and Dissemination. *Undertaking Systematic Reviews of Research on Effectiveness, CRD Report No. 4*. 2nd ed. (2001). Available at http://www.york.ac.uk/inst/crd/pdf/Systematic_Reviews.pdf. Accessed May 30, 2012.

Chen R, Wang G, Sun Y, Ma L, Li X. A new speech training method for patients following cleft palate repair. *Chin J Stomatol*. 1996;31:220–223.

Clinical Standards Advisory Group. *Cleft Lip and/or Palate. Report of a CSAG Committee*. London: Department of Health; 1998.

Dixon-Woods M, Agarwal S, Jones D, Young B, Sutton A. Synthesising qualitative and quantitative evidence: a review of possible methods. *J Health Serv Res Policy*. 2005;10:45–53.

Enderby P, Emerson J. *Does Speech and Language Therapy Work? A Review of the Literature*. London: Whurr Publishers; 1995.

Gibbon F, Hardcastle WJ, Crampin L, Reynolds B, Razzell R, Wilson J. Visual feedback therapy using electropalatography (EPG) for articulation disorders associated with cleft palate. *Asia-Pac J Speech, Language and Hearing*. 2001;6:53–58.

Grundy K, Harding A. Developmental speech disorders. In: Grundy K, ed. *Linguistics in Clinical Practice*. London: Whurr Publishers; 1995:329–358.

Grunwell P. *Analysing Cleft Palate Speech*. London: Whurr Publishers; 1993.

Hardin-Jones M, Chapman KL. The impact of early intervention on speech and lexical development for toddlers with cleft palate: a retrospective look at outcome. *Lang Speech Hear Serv*. 2008;39:89–96.

Hardin-Jones MA, Jones DL. Speech production of preschoolers with cleft palate. *Cleft Palate Craniofac J*. 2005;42:7–13.

Harding A, Bryan A. *The Use of Multisensory Input-Modelling to Stimulate Speech Output Processing. A Therapy Demonstration Video for Speech And Language Therapists for the Treatment of Speech Disorders*. [DVD]. Cambridge, U.K.: Cleft Lip and Palate Network; 2002.

Henningsson G, Kuehn DP, Sell D, Sweeney T, Trost-Cardamone JT, Whitehill TL. Universal parameters for reporting speech outcomes in individuals with cleft palate. *Cleft Palate Craniofac J*. 2008;45:1–15.

Higgins JPT, Green S. *Cochrane Handbook for Systematic Reviews of Interventions*. Oxford: The Cochrane Collaboration; 2011.

John AK, Sell D, Harding-Bell A, Sweeney T, Williams A. CAPS-A—a validated and reliable measure for auditing cleft speech. *Cleft Palate Craniofac J*. 2006;43:272–288.

Kuehn D, Moller KT. Speech and language issues in the cleft palate population: the state of the art. *Cleft Palate Craniofac J*. 2000;37:348–348.

Lee ASY, Law J, Gibbon FE. Electropalatography for articulation disorders associated with cleft palate. *Cochrane Database Syst Rev*. 2009;(3):CD006854. doi:10.1002/14651858.CD006854.pub2.

Lohmander A. Surgical intervention and speech outcomes in cleft lip and palate. In: Howard S, Lohmander A, eds. *Cleft Palate Speech Assessment and Intervention*. Oxford: Wiley-Blackwell; 2011:55–82.

Lohmander A, Willadsen E, Persson C, Henningsson G, Bowden M, Hutters B. Methodology for speech assessment in the scancleft project—an international randomised clinical trial on palatal surgery: experiences from a pilot study. *Cleft Palate Craniofac J*. 2009;46:347–362.

Lohmander-Agerskov A, Olson M. Methodology for perceptual assessment of speech in patients with cleft palate: a critical review of the literature. *Cleft Palate Craniofac J*. 2004;41:64–70.

Ma J, Tian Y, He Y. Voice training to palatoschisis children. *Chin J Clin Rehabil*. 2003;7:164.

Ma L, Wang G, Zhang L, Bai X. A preliminary report of speech training following the cleft palate surgery and its effect. *Chin J Stomatol*. 1990;25:86–89.

Marshall J, Goldbart J, Pickstone C, Roulstone S. Application of systematic reviews in speech-and-language therapy. *Int J Lang Commun Disord*. 2010;46:261–272.

McCauley RJ, Strand E, Lof GL, Schooling T, Frymark T. Evidence-based systematic review: effects of nonspeech oral motor exercises on speech. *Am J Speech Lang Pathol*. 2000;18:243–260.

McCormack J, McLeod S, McAllister L, Harrison L. A systematic review of the association between childhood speech impairment and participation across the lifespan. *Int J Speech Lang Pathol*. 2009;11:155–170.

Pamplona MC, Ysunza A. Active participation of mothers during speech therapy improved language development of children with cleft palate. *Scand J Plast Reconstr Surg Hand Surg*. 2000;34:231–236.

Pamplona MC, Ysunza A, Espinosa J. A comparative trial of two modalities of speech intervention for compensatory articulation in cleft palate children, phonologic approach versus articulatory approach. *Int J Pediatr Otorhinolaryngol*. 1999;49:21–26.

Pamplona MC, Ysunza A, Patino C, Ramirez E, Drucker M, Mazon JJ. Speech summer camp for treating articulation disorders in cleft palate patients. *Int J Pediatr Otorhinolaryngol*. 2005;69:351–359.

Pamplona MC, Ysunza A, Perez G, Vergara S. Summer school speech therapy for children with cleft palate and language disorder. *Gaceta Medica De Mexico*. 2009;145:475–479.

Pamplona MC, Ysunza A, Ramirez P. Naturalistic intervention in cleft palate children. *Int J Pediatr Otorhinolaryngol*. 2004;68:75–81.

Pamplona MC, Ysunza A, Uriostegui C. Linguistic interaction: the active role of parents in speech therapy for cleft palate patients. *Int J Pediatr Otorhinolaryngol*. 1996;37:17–27.

Pring T. Ask a silly question: two decades of troublesome trials. *Int J Lang Commun Disord*. 2004;39:285–302.

Regan JB, Versaci A. A home program for improving voice and speech quality of infants with repaired cleft palate. *R I Med J*. 1977;60(8):384–385. 409.

Russell J, Grunwell P. Speech development in children with cleft lip and palate. In: Grunwell P, ed. *Analysing Cleft Palate Speech*. London: Whurr Publishing; 1993:19–47.

Scherer NJ, D'Antonio LL, McGahey H. Early intervention for speech impairment in children with cleft palate. *Cleft Palate Craniofac J*. 2008;45:18–31.

Sell D. A review of issues in perceptual speech analysis in cleft palate and related disorders. *Int J Lang Commun Disord*. 2005;42:103–121.

Sell D, Grunwell P. Speech assessment and therapy. In ACH Watson, D Sell, P Grunwell (eds). *Management of Cleft Lip and Palate*. London: Whurr Publishers; 2001:258–285.

Sell D, Grunwell P. Speech assessment and therapy. In: Watson ACH, Sell D, Grunwell P, eds. *Management of Cleft Lip and Palate*. London: Whurr Publishers; 2001:258–285.

Sell D. A review of issues in perceptual speech analysis in cleft palate and related disorders. *Int J Lang Comm Dis*. 2005;42:103–121.

Sell D, Grunwell P, Mildinhall S, Murphy T, Cornish TA, Bearn D, Shaw WC, Murray JJ, Williams AC, Sandy JR. Cleft lip and palate care in the United Kingdom—the Clinical Standards Advisory Group (CSAG) Study. Part 3: speech outcomes. *Cleft Palate Craniofac J*. 2001;38:30–37.

Sell D, Nagarajan R, Wickenden M. Cleft Palate Speech in the Majority World: Models of Intervention and Speech Outcomes in Diverse Cultural and Language Contexts. In: Howard S, Lohmander A, eds. *Cleft Palate Speech Assessment and Intervention*. Oxford: Wiley-Blackwell; 2011:105–125.

Stackhouse J. Psycholinguistic assessment and intervention. In: Howard S, Lohmander A, eds. *Cleft Palate Speech Assessment and Intervention*. Oxford: Wiley-Blackwell; 2011:239–255.

Stackhouse J, Wells B. *Children's Speech and Literacy Difficulties: Psycholinguistic Framework*. London: Whurr Publishers; 1997.

Van Demark DR. Some results of speech therapy for children with cleft palate. *Cleft Palate-Craniofac J*. 1974;11:41–49.

Van Demark DR, Hardin MA. Effectiveness of intensive articulation therapy for children with cleft palate. *Cleft Palate Craniofac J*. 1986;23:215–224.

Van Riper C. *Speech Correction: Principles and Methods*. 6th ed. Englewood Cliffs, N.J: Prentice-Hall; 1978.

Vieira AR. Unravelling human cleft lip and palate research. *J Dent R*. 2008; 87:119–125.

West S, King V, Carey TS, Lohr KN, McKoy N, Sutton SF, Lux L. Systems to rate the strength of scientific evidence. Evidence report/ technology assessments: 47; 2002.

Whitehill TL. Assessing intelligibility in speakers with cleft palate: a critical review of the literature. *Cleft Palate Craniofac J*. 2002;39: 50–58.

Whitehill TL, Goetzee C, Hodge, M. Intelligibility. In: Lohmander A, Howard S, eds. *Cleft Palate Speech: Assessment, Analysis and Intervention*. In press. London: Wiley Publishers.

Witzel MA. Speech evaluation and treatment. *Oral and Maxillofacial Surgery Clinics of North America*. 1991;3:501–516.

World Health Organisation. *International classification of function and disability*. Geneva: WHO; 2001.

World Health Organisation. *International classification of impairments, disabilities and handicaps (ICIDH)*. Geneva: WHO; 1980.

Yang X, Li N, Bu L, Cai X, Tong Q. The analysis of formant characteristics of vowels in the speech of patients with cleft palate. *W China J Stomatology*. 2003;21:451–454.

Ysunza A, Pamplona M, Femat T, Mayer I, Garcia-Velasco M. Videonasopharyngoscopy as an instrument for visual biofeedback during speech in cleft palate patients. *Int J Pediatr Otorhin*. 1997;41: 291–298.

Appendix A Search Strategy for MEDLINE (on OVID)

1 Cleft Palate/ (14,334)
2 Cleft Lip/ (9934)
3 hare lip$.tw. (90)
4 harelip$.tw. (327)
5 Palatoschisis.tw. (81)
6 cleft lip$.tw. (7164)
7 cleft palate$.tw. (7454)
8 orofacial$ cleft$.tw. (443)
9 facial cleft$.tw. (742)
10 oral cleft$.tw. (387)
11 craniofacial cleft$.tw. (79)
12 Velopharyngeal Insufficiency/ (1156)
13 Velopharyngeal Insufficiency.tw. (448)
14 or/1–13 (19806)
15 Speech Therapy/ (4380)
16 (speech adj3 therap$).tw. (2416)
17 (speech adj3 clinic$).tw. (468)
18 (speech adj3 intervention$).tw. (154)
19 (speech adj3 train$).tw. (373)
20 (speech adj3 rehabilitat$).tw. (468)
21 "Early Intervention (Education)"/ (1046)
22 (early adj3 educat$).tw. (1287)
23 (early adj3 interven$).tw. (11,627)
24 or/15–23 (20,059)
25 exp Speech Production Measurement/ (4196)
26 (speech adj3 assess$).tw. (847)
27 (speech adj3 screen$).tw. (84)
28 (speech adj3 measur$).tw. (1188)
29 (speech adj3 outcome$).tw. (403)
30 (speech adj3 evaluat$).tw. (881)
31 (articulation adj3 test$).tw. (167)
32 or/25–31 (6812)
33 24 or 32 (26,029)
34 14 and 33 (1080)
35 limit 34 to ("all child (0 to 18 years)" or "young adult (19 to 24 years)") (816)
36 (Child$ or infant$ or baby or babies or boy$1 or girl$1 or pre-school$ or preschool$ or toddler$ or young people or young adult$).tw. (1,048,686)
37 34 and 36 (386)
38 35 or 37 (832)

Appendix B Detailed Study Characteristics

Study	*Methods*	*Participants*	*Intervention*	*Outcome*	*Results*
Pamplona et al., 2004	Mexico. Single-center RCT.	30 children with UCLP postprimary surgical correction with compensatory articulation disorder (CAD) associated with velopharyngeal incompetence. Exclusions: hearing loss, neurological deficits, language disabilities, postsurgical fistula.	Intervention A: Phonological approach. Modification of groups of sounds, treatment goals based on child's active phonological rules, establishment and maintenance of phonological contrasts. Small group including speech pathologist, 2–3 children and mothers. 1-h sessions, twice weekly until CAD treated. Intervention B: Whole-language approach involving play and storybooks, and sounds associated with CAD treated indirectly—reinforcement of correct sounds and enhancement of cognitive linguistic organization. Small group including speech pathologist, 2–3 children and mothers. 1-h sessions, twice weekly until CAD eliminated.	Total time taken to correct CAD in sessions assessed by 2 independent examiners based on a free speech sample.	Standard mean difference = 0.361
Gibbon et al., 2001	Conducted in U.K. Single-center RCT.	12 participants: 7 children with UCLP, 2 with bilateral cleft lip and palate (BCLP) and 3 with clefts of the soft palate (SCP) following surgical correction, with compensatory articulation disorder, and a history of speech therapy pre-EPG, aged 5–18 y, 8 male.	Intervention A: Electropalatography (EPG) (4 sessions, followed by non-EPG therapy (another 4 sessions). Individual sessions lasting 30–45 min. Targeted anterior placement for /t/ and/or /s/ and groove formation for /s/. Administered by specialist cleft center speech and language therapists with training in EPG. Same therapist for EPG and non-EPG for each participant. Intervention B: 4 non-EPG sessions followed by 4 sessions of EPG. Individual sessions lasting 30–45 min. Targeted anterior placement for /t/ and/or /s/ and groove formation for /s/. Administered by specialist cleft center speech and language therapists with training in EPG. Same therapist for EPG and non-EPG for each participant.	EPG and acoustic data taken during read-aloud word list using natural conversational rate. 3 time points: pretherapy, post-fourth session, and posttherapy. 10 words analyzed: 5 containing /t/ and 5 containing /s/. COG score and qualitative observations by EPG experts.	"s" sound: odds ratio = 10.714 "t" sound: odds ratio = 3.333
Pamplona and Ysunza, 2000	Conducted in Mexico. Single-center RCT.	41 children with UCLP post–primary surgery aged 3–4.8 y (mean = 3 y 8 mo). Exclusions: congenital abnormalities, developmental disabilities, hearing loss, language delay, neurological deficits, history of velopharyngeal incompetence after surgical correction, history of postoperative fistula.	Intervention A: Mothers involved in therapy. Small working groups of 2 children, speech pathologist, and mothers. Symbolic play activities: bath time, mealtime, bedtime scenarios, and nonfamiliar events. 1 h, 3 times weekly for 12 mo. Materials: dolls, doll accessories, cars and accessories. Mean age 3.7 y (3–4.5 y). Intervention B: No parental involvement. Small working groups of 2 children and speech pathologist. Symbolic play activities: bath time, mealtime, bedtime scenarios, and nonfamiliar events. 1-hour, 3 times weekly for 12 mo. Materials: dolls, doll accessories, cars and accessories. Mean age, 3.8 y (3–4.8 y).	Linguistic level (levels 1–5): prelinguistic, 1-word utterances, semantic combination, simple sentences, and complex. Assessed by 2 independent listeners watching free play videotape for 40 min at preintervention and postintervention. Level of play (1–3): relational, symbolic, or imaginative assessed by independent assessors by videotape for 40 min.	Standard mean difference = 1.539 Levels of play (secondary outcome): standard mean difference = 0.441
Pamplona et al., 1999	Conducted in Mexico. Single-center RCT.	29 children with repaired UCLP aged 3–7 y. Exclusions: hearing difficulties, language impairment, or neurological disorders.	Comparison of two therapies to correct compensatory articulation. Therapy A: Therapy according to traditional standard errors in articulation treated on a phoneme-by-phoneme basis. 1-h sessions twice a week. Administered by speech pathologist. Group sessions of 2–3 children. Therapy B: Therapy according to phonological approach—treatment goals set depending on phonological rules that are active in child's system, focused on modification of groups of sounds and emphasis on establishment of previously neutralized contrasts. 1-h sessions twice a week. Administered by same speech pathologist as Therapy A. Group sessions of 2–3 children.	Total time of speech therapy from onset of therapy until the complete normalization of articulation as assessed in a phonological analysis from a free speech sample (videotape). Assessed by two examiners with 8 y of experience in the procedure, both involved in the study.	Standard mean difference = 1.936
Ysunza et al., 1997	Conducted in Mexico. Single-center RCT.	17 children with UCLP following surgical closure with compensatory articulation disorder associated with velopharyngeal incompetence and negative lateral pharyngeal wall displacement during speech. Exclusions: syndromes known to cause developmental delay, hearing difficulties, postoperative fistula.	Intervention A: Speech therapy 3 times a week for 60 min delivered by a speech pathologist aimed at correcting compensatory articulation. No visual feedback. Intervention B: Speech therapy 3 times a week for 60 min aimed at correcting compensatory articulation plus visual biofeedback sessions twice a week for 25 min each using videonasopharyngoscopy delivered by a speech pathologist. Therapy involved modifying negative movements of the lateral pharyngeal walls using phoneme samples.	Modification or persistence of negative movements of the lateral pharyngeal walls at 12 wk as rated by an examiner inspecting the image from videonasopharyngoscopy.	Odds ratio = 96.333

Appendix B Continued

Study	*Methods*	*Participants*	*Intervention*	*Outcome*	*Results*
Pamplona et al., 1996	Conducted in Mexico. Single-center RCT.	21 nonsyndromic children with UCLP post–surgical correction aged 3–4.8 y with velopharyngeal competence. Exclusions: Children with syndromes, congenital abnormalities, postoperative fistula, hearing difficulties, neurological deficits.	Intervention A: Nonparental involvement. Whole-language therapy. Recreation of everyday significant events. Groups of 2 children and 1 speech pathologist. Mean age 3 y 9 mo. 1 h 3 times weekly for 8 mo. Intervention B: Parental involvement. Whole-language therapy. Recreation of everyday significant events. Groups of 2 children, mothers, and 1 speech pathologist, mean age, 3 y 7 mo. 1 h 3 times weekly for 8 mo.	Linguistic level (levels 1–5): Prelinguistic, 1-word utterances, semantic combination, simple sentences and complex. Assessed by 2 independent listeners watching free play videotape for 40 min at preintervention and postintervention. Level of play (1–3): Relational, symbolic, or imaginative assessed by independent assessors by videotape for 40 min. Parental interaction: Modified in Intervention B at postintervention versus no modification.	Standard mean difference = 0.895 Levels of play (secondary outcome): standard mean difference = −0.433
Pamplona et al., 2009	Conducted in Mexico. Cohort study.	50 patients with cleft palate. Mean age, 42 mo.	Intervention A: Whole-language therapy delivered at summer camp for 3 wk Monday–Friday for 4 h a day. Intervention B: Whole-language therapy delivered using standard speech therapy in a clinic setting for 9 mo twice a week for 45 min.	Comparison of initial and final levels of development using situational, discourse, semantic levels.	Situational speech: odds ratio = 0.603 Discourse (contextual) speech: odds ratio = 1.446 Semantic speech: odds ratio = 1.556
Hardin-Jones and Chapman, 2008	Conducted in USA. Cohort study.	Comparison of a total of 30 children with cleft lip and/or palate not referred for therapy, those referred for therapy and received it, those not receiving therapy, and an additional 10 children without clefts aged 27 mo. 6 had BCLP, 20 UCLP, 3 hard and soft palate cleft, 1 SCP. Exclusions: cleft lip only, syndromes known to cause developmental delay.	Intervention A: Therapy received between 9 and 21 mo (mean, 16 mo). Delivered weekly for 60–75 min at home, or twice weekly for 30 min at home. Administered by speech and language pathologists with 1–19 y of experience (mean, 10 y). Stimulation of consonant sounds using modeling and tactile placement cues and encouraging vocabulary development through play with additional oral movements. Comparator A: Patients who had not been referred for treatment. Comparator B: No therapy but were referred. Comparator C: Children without cleft (normal speech development).	Preschool language scale at baseline. 17 and 27 mo of age: assessments using audio and video recordings while interacting with primary caregivers in home assessed using International Phonetic Alphabet (IPA).	Auditory comprehension: Intervention mean = 108.1, comparator mean = 106.44 Expressive communication: Intervention mean = 105.4, comparator mean = 107.22
Scherer et al., 2008	Conducted in USA. Controlled study.	10 children with CLP and 10 children without CLP, aged 14–35 mo. 11 girls, 9 boys. Exclusions: genetic syndromes, neurological impairment, hearing difficulties, preterm birth earlier than 36 wk of gestation.	Therapy delivered only to children with CLP. Children without clefts were used as a comparison group demonstrating normal speech and language development. Mothers trained to use focused stimulation approach, consisting of description of technique, role play of scripted procedure examples, demonstration by clinician and parent-child practice with clinician coaching. Modeling of target words at least 3 times during play session and then use of expansions if child did not respond correctly. 2–4 training sessions of 45 min until 80% accuracy with technique achieved.	Sequenced Inventory of Communicative Development Inventories–Revised (expressive & receptive language). Transcripts—assessment of language sample by blinded raters. Systematic analysis of language transcripts: Number of total words, number of different words, mean length of utterance, percentage of responses. Speech: Percentage of glottal stops, number of true consonants, Percent Consonants Correct-Revised.	Expressive language: standard mean difference = 0.117 Receptive language: standard mean difference = 0.392

Appendix B Continued

Study	*Methods*	*Participants*	*Intervention*	*Outcome*	*Results*
Pamplona et al., 2005	Conducted in Mexico. Cohort study.	45 children with UCLP following primary surgical closure, with CAD in association with velopharyngeal incompetence aged 3–10 y. Control: 45 children with cleft palate following primary surgical closure with CAD in association with velopharyngeal incompetence following secondary speech surgery aged 3–10 y.	Intervention A: Summer camp for 3 wk, 4 h a day Monday–Friday. Mothers present. Maximizing articulation in naturalistic setting and within a linguistic context. Using phonological principles and whole-language model. Intervention B: Speech therapy to correct CAD 2 times weekly for 1 h for 12 mo using phonological principles and whole-language model. Small groups with mothers, 2–3 children and speech pathologist.	Level of CAD—No (Normal), Mild, Moderate or severe at pre and post as rated by examiners using videotaped 1-h play and storytelling sample.	Odds ratio = 3.143
Ma et al., 2003	Conducted in China. Abstract only. Unclear study design.	67 children with cleft palate aged 4–11 y, 42 boys, 25 girls. No specific exclusion criteria stated.	Voice training program. Reinforcement for velopharyngeal closure ability (1 mo postoperation), instruction to say "oh" loudly 30 times twice daily for 1 mo. Reinforcement of competence (2 mo postoperation). Instruction to blow balloon 40–60 times once or twice daily and play harmonica 1–2 times a day. Lip posture and movements: grin, pout, blow, say /p/, nibble underlip with upper, huff, and say silent /f/ for 10–15 min 2–3 times daily.	Song Ruyao evaluation of vocalization and speaking condition (4 grades): Excellent = no different to normal speech, fine = some differences in speaking but understandable, mistake = abnormalities to a great extent, bad = cannot be understood.	
Yang et al., 2003	Conducted in China. Cohort study.	Post–speech therapy group: 16 patients with cleft palate with pharyngeal obturator, aged 5–28 years (mean, 17.2 y), 8 males, 8 females who received speech therapy after this surgery. Pre–speech therapy group: 33 individuals with cleft palate (18 males and 15 females) aged 5–29 (mean, 15.8 y), with pharyngeal obturator, had participated in speech training classes from August 1999 to August 2000 who did not receive speech therapy after surgery. Control: healthy students aged 5–15 y. Exclusions: Received speech therapy post–cleft palate closure, oral or nasal fistulas, hearing difficulties or learning difficulties, severe bite deformity, local accent.	Intervention A: standard speech therapy care, described as speech training, wearing a velopharyngeal obstructor to assist pronunciation and homework practice. Comparator B: Healthy control group who received no intervention.	First three formant frequencies (F1, F2, and F3) assessed on vowel sounds *a*, *e*, *i*, and *u*, using computer-based recording instrument, preintervention and postintervention up to 6 mo following speech therapy.	Formant frequency of *A*: intervention mean = 2391, comparator mean = 2391 Formant frequency of *E*: intervention mean = 2146, comparator mean = 2466 Formant frequency of *I*: intervention mean = 3277, comparator mean = 2970 Formant frequency of *U*: intervention mean = 2873, comparator mean = 2890
Chen et al., 1996	Conducted in China. Before-and-after study.	33 children with CP between 1 mo and 5 y post–cleft palate repair and functional speech disorder aged 4–16 y (mean, 6.9 y), 15 males. No specific exclusion criteria.	Intensive speech therapy (1–2 times per d, 0.5–1 h a session for 1–2 mo). Lip movement training, tongue movement training, elicitation of correct consonants, and training of syllable combination production using Pinyin method, induction method, classification method, and games.	Speech intelligibility score at preintervention and postintervention and speech intelligibility for consonants before and after therapy. Classifications: category I: abnormal articulatory place, category II: abnormal manner of articulation, category III: both abnormal articulatory place and manner.	Mean before therapy = 38.43, mean after therapy = 89.54

Appendix B Continued

Study	*Methods*	*Participants*	*Intervention*	*Outcome*	*Results*
Ma et al., 1990	Conducted in China. Cohort study.	30 patients with cleft palate from 4–6 y with submucous cleft palate, incomplete and complete UCLP and BCLP, all having received von Langenbeck repair; 12 male, 18 female. No specific exclusion criteria.	Intervention A: postoperative speech training for 6 months, once a week for 20–30 min. Involving lip and tongue movements and blowing exercises, practicing pronouncing single vowels and double vowels, correct mandibular movement exercises and phonetic alphabet exercises. Comparator B: No therapy.	Intelligibility on experimental syllable diagram and on alphabet at pretherapy, 3 months, and posttherapy, and lateral head X-ray (stationary position, pronouncing "ee").	Correct lip/tongue movements: intervention mean = 12, comparator mean = 12 Correct blowing: intervention mean = 16, comparator mean = 16 Correct mandibular movements: intervention mean = 18, comparator mean = 18
Van Demark and Hardin, 1986	Conducted in USA. Before-and-after study.	13 children with cleft palate with or without cleft lip aged 6.8–12 y (mean, 8.7 y), 9 boys, 4 girls. All had velopharyngeal competence or marginal competence, with articulation errors judged by the first author to have potential to improve. Tolerant to 6 wk away from home as assessed by a psychologist. No specific exclusion criteria.	Summer camp delivery of systematic multiple sound approach to articulation. 1-h sessions per day for 26 d. Total of 104 h. Half individual sessions, half with another child. Administered by speech pathologists with experience in cleft, except for 3 children who received it from a graduate speech pathologist.	Rated on scales: articulation defectiveness scale of 1–7, severity of nasality scale 1–7, velopharyngeal competence scale 1–3. Used speech sample of conversational speech, Iowa Cleft Palate Articulation Test, responses on phonetically balanced sentences.	Articulation defectiveness: mean before therapy = 3.88 mean after therapy = 2.54 Nasality: mean before therapy = 1.94, mean after therapy = 1.31
Regan and Versaci, 1977	Conducted in USA. Cohort study.	45 consecutive children with CL±P post surgical correction. Mean age 19.8 mo at start of intervention. Mean age 6.9 y at completion of study.	Parents seen for half hour to explain and instruct on straw sucking. Advised to provide all liquids through a straw (a cup called "sip-it" was used). Parents responsible for enforcement of the procedure. Evaluation twice yearly for approximately 4 y.	4-point rating scale used by a professional listener to estimate voice and speech acceptability based on a substantiated speech sample. Listening by 2 skilled speech clinicians and 1 layperson all not involved in study. Parental judgments also gathered.	23 out of 27 had adequate speech
Van Demark, 1974	Conducted in Denmark. Cohort study.	67 children with cleft palate who had not received speech therapy. No specific exclusion criteria.	Intervention A: Received intervention between two speech assessment points. Comparison A: Had not received intervention between two speech assessment points.	Percentage correct on both the 44 and 70-item Iowa/Danish Pressure Articulation test as rated by a Danish speaking speech pathologist and the investigator (95% agreement). Type of errors made: oral distortion, substitution including glottal and pharyngeal and omissions.	Intervention mean = 35.11, comparator mean = 49.62