

Early Intervention for Speech Impairment in Children With Cleft Palate

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Objective: This study explored the effectiveness of a parent-implemented, focused stimulation program on the speech characteristics of children younger than 3 years with cleft lip and palate. The research questions included the following: (1) Can parents be trained to deliver an early intervention (EI) program for children with cleft palate? (2) Does a parent-implemented EI program result in positive changes in speech characteristics?

Participants: Ten mother-child pairs in which the child had cleft lip and palate (CLP) and 10 mother-child pairs in which the child did not have a cleft (NCLP). The children ranged in age from 14 to 36 months of age and were matched between the CLP and the NCLP groups for vocabulary size, age, and socioeconomic status.

Main Outcome Measures: Group differences (CLP and the NCLP) for preintervention and postintervention language and speech measures were compared.

Results: The results of this study showed that the mothers could be trained to deliver the intervention reliably. Furthermore, the results indicated that the intervention resulted in increased sound inventories, increased speech accuracy, and reduced use of glottal stops for the children with clefts.

Conclusions: While the intervention resulted in speech gains for the children with clefts, speech measures did not exceed those made by the children without clefts. The results of the study have implications for service delivery models where the services of speech-language pathologists are limited.

KEY WORDS: *cleft palate, early intervention*

A careful review of the literature shows a variety of communication impairments in young children with cleft palate. The literature suggests that prior to 3 years of age, toddlers with cleft palate demonstrate limited vocabulary, restricted sound inventories, and the emergence of compensatory articulation errors (Estrem and Broen, 1989; Scherer, 1999; Chapman et al., 2003; Salas-Provance et al., 2003). Presently, there are few data-based studies examining the efficacy of early intervention (EI) models for children with cleft palate younger than 3 years (Scherer, 1999; Pamplona et al., 1999, 2004). These studies demonstrate that language and phonological intervention programs are effective in facilitating both language and speech performance. These models have appeal for this population because of their strong validity for this age group and considerable data-based evidence regarding their efficacy with other groups of children with speech and language

disabilities (Girolametto et al., 1986, 1996, 1997; Kaiser, 1993; Kaiser et al., 1996, 1998; Kaiser and Hancock, 2003). Previous studies have suggested that EI programs for children with cleft palate may result in improvements in both vocabulary and, secondarily, speech production (Scherer, 1999; Pamplona et al., 2004).

Compensatory articulation errors are of particular concern in this population. These are abnormal speech sound patterns frequently heard in the speech of individuals with cleft palate (Golding-Kushner, 2001). Broadly defined, these are sounds that are made by shifting the place of articulation posteriorly in the vocal tract. A number of compensatory errors have been described (Trost, 1981; Peterson-Falzone et al., 2006). These error patterns often emerge in young children with clefts prior to 3 years of age. The studies of early vocal development have described a range of compensatory articulation patterns including glottal stops, velar and pharyngeal fricatives, posterior nasal fricative, and nasal substitutions (O'Gara and Logemann, 1988; O'Gara et al., 1994; Chapman et al., 2001; Salas-Provance et al., 2003; Morris and Ozanne, 2003). However, the most common and distinctive compensatory articulation error for young children is the glottal stop substitution (Kuehn and Moller, 2000; Peterson-Falzone et al., 2001). The glottal stop is made by stopping the airstream at the level of the vocal folds, thus approximating a stop consonant and preventing nasal

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air flow. Glottal stops are typically substituted for some or all of the stop consonant sounds and occasionally for fricatives as well. One hypothesis regarding the high occurrence of glottal stops in the speech of children with cleft palate is that parents may prefer words produced with compensatory stopping of air compared to words with correct place of articulation but with nasal air escape (Bradford and Culton, 1987). Parents may unintentionally reinforce these abnormal patterns early in the child's development, thus increasing the frequency of these aberrant compensatory errors.

Clinically, the glottal stop is especially problematic because this sound does not occur in most languages, and therefore its unusual nature draws attention to the speaker. Additionally, glottal stop substitutions are resistant to traditional forms of articulation therapy (Kuehn and Moller, 2000; Peterson-Falzone et al., 2001). Specific data on the occurrence of compensatory articulation errors in young children with cleft palate is lacking. Golding-Kushner (2001) estimated that at least 25% of preschool aged children with clefts undergo speech therapy for compensatory errors, with glottal stops being the most frequent form of compensatory error. Current approaches for treating glottal stop errors in young children involve intervening after these patterns are established (Golding-Kushner, 2001; Kuehn and Moller, 2000; Peterson-Falzone et al., 2006). Children with cleft palate are often enrolled in prolonged periods of speech therapy in attempts to eliminate these glottal stop errors. The problem is exacerbated by the fact that there are few clinicians experienced in effective techniques for reducing the unusual articulation errors associated with cleft palate. This lack of experienced clinicians frequently leads to ineffective interventions (Pannbacker et al., 1990).

Given the difficulties encountered in remediating glottal stop errors, it appears prudent to consider intervention models that may prevent the habituation of compensatory speech patterns in young children with clefts rather than waiting to treat them after they have developed and become established. Over the past two decades, EI programs have been used as an effective means for preventing or remediating early speech and language disabilities in many populations. There is a vast literature documenting the effectiveness of clinician-implemented EI programs for facilitating speech and language development. More recently, research has demonstrated significant gains in speech and language using parent-implemented EI programs (Hancock and Kaiser, 1998; Kaiser and Hancock, 2003; Scherer and Kaiser, in press). While there is solid scientific data to support the use of both clinician- and parent-implemented EI programs for many clinical populations, there are little data regarding the use of EI for children with cleft palate.

Health care and education budgets are shrinking, and services for children with special needs such as cleft palate are limited (Nagarajan et al., 2006). As services become less

available, adjustment in service delivery models may be needed to take advantage of parents' and the child's community in providing interventions (Koop, 1987). Therefore, the development and validation of a data-based, parent-implemented EI program for children with cleft palate has great appeal. Such a program would also be attractive not only for use in industrialized regions of the world but also for children with cleft palate in regions of the world where there are few or no speech-language pathologists (SLPs).

The purpose of the present study was twofold: (1) to determine whether parents can be trained to deliver an EI program for children with cleft palate and (2) to determine whether a parent-implemented EI program for children with cleft palate results in positive changes in speech characteristics, specifically, reduction or elimination in glottal stop compensatory articulations and increased vocabulary, speech sound accuracy, and sound inventory.

METHODS

Participants

This study was approved by the East Tennessee State University Institutional Review Board, and signed consent was obtained from the children's parents or legal guardians. Twenty children and their mothers participated in the study over a 3-month period. Ten children with unilateral cleft lip and palate (CLP) were matched with 10 typically developing children without CLP (NCLP). The children with CLP were recruited from Children's Special Services, Regional Tennessee Department of Health, and Tennessee's Early Intervention Service. The children with NCLP were recruited from East Tennessee State University's Child Study Center and the community. Inclusionary criteria for the children in the CLP group were the following: (1) cleft lip with or without cleft palate in the absence of a genetic syndrome as assessed by a geneticist, (2) absence of significant medical or neurological impairments or preterm birth earlier than 36 weeks gestation as evidenced on the medical history form, and (3) passing a hearing screening consisting of pure-tone acuity and tympanometry measures. Hearing screenings were interpreted as a pass if the acuity measures were passed. In addition, the children in the NCLP group had no identified speech, hearing, or language impairments reported by the child's parent or preschool teacher.

The children were matched for vocabulary production, age, gender, and socioeconomic status (SES). The primary matching variable for all subjects was vocabulary production, while age, gender, and SES were considered secondary matching variables. The vocabulary production level was determined by the MacArthur Communication Development Inventory (CDI) (Fenson et al., 1993). The children with CLP were matched as closely as possible to the NCLP children. Children with CLP with a score of 50

TABLE 1 Age, Gender, Socioeconomic Status, and the Expressive Vocabulary Raw Score From the MacArthur Communicative Development Inventory (CDI; Fenson et al., 1993) for the Children With Cleft Palate (CLP) and Children Without Clefts (NCLP)

Child	Age (Mo)	Age of Palate Repair (Mo)	Gender	Socioeconomic Status	CDI Score
CLP					
1	30	12	F	2	59
2	24	11	M	4	11
3	28	11	M	2	1
4	35	12	F	2	222
5	35	10	M	1	324
6	18	12	M	1	9
7	21	12	F	2	52
8	27	11	M	2	10
9	31	12	F	4	36
10	25	12	F	2	123
NCLP					
1	21		F	1	89
2	15		F	2	44
3	17		M	1	10
4	30		F	1	321
5	29		M	1	483
6	22		F	1	40
7	14		F	1	58
8	15		M	1	42
9	18		M	1	49
10	21		F	3	144

words or fewer were matched to a child without a cleft palate with 50 words or less. Children whose parents reported more than 50 words were likewise matched with an NCLP child with more than 50 words. The difference between CDI scores for the CLP and NCLP groups was not significantly different ($t = 0.73$, $df = 18$, $p = .477$), indicating that the groups were similar in their vocabulary size as reported by their parents.

To verify the matching criteria, the participants were compared on a standard measure of language development. The Sequenced Inventory of Communicative Development (SICD-R) (Hedrick et al., 2002) scores for the children with CLP and children in the NCLP group were compared prior to the study. The two groups were not significantly different from each other for both receptive ($t = 0.53$, $df = 18$, $p = .73$) and expressive language ($t = 0.88$, $df = 18$, $p = .60$). This finding was expected because the children were matched on a language measure prior to the intervention.

Table 1 shows the age, age of palate repair, gender, SES, and expressive vocabulary score from the CDI for the children in the CLP and NCLP groups. The children in the NCLP group ranged in age from 14 to 30 months (mean, 20.2), whereas the children in the CLP group ranged in age from 18 to 35 months (mean, 27.4). A *t*-test comparison revealed that the CLP group was not significantly older than the NCLP group ($t = 1.33$, $df = 19$, $p = .19$). The mean age of palate repair was 11.5 months. The children in the CLP group were evenly split between boys and girls, while the children in the NCLP group had six girls and four boys. Socioeconomic status was established from a case history form completed by the parent that included

information on the parent's education background and type of employment. Socioeconomic status was determined by placing the family in one of five socioeconomic categories based on the method described by Eilers et al. (1993). Eight of the families of the children with clefts were in the mid to high categories (1 to 3), and the remaining two families were in the mid to low categories (4 to 8). All 10 of the families of children in the NCLP group were placed in mid to high categories. A chi-square comparison of the SES categories indicated no significant difference in the distribution of SES for children in the CLP and NCLP groups (chi-square = 6.29, $df = 3$, $p = .09$).

Procedures

Pre-Post Test Measures

The children participated in standardized and informal testing procedures at the initiation of the study and following intervention (or for the NCLP group, 3 months after pretest). The NCLP group did not receive an intervention in order to compare the effects of the intervention to the typical course of speech and language development during the intervening period. These sessions took place in the child's home, preschool, or the East Tennessee State University Speech and Hearing Clinic. The test measures for both testing sessions included the following procedures: (1) administration of the SICD-R (Hedrick, et al., 2002), which provided a receptive and expressive language age score, and (2) collection of a 30-minute videotaped and audiotaped language sample involving mother-child interaction during a book-reading activity and free play with toys. The toys used to elicit both preintervention and postintervention language samples were controlled for initial and final consonants and were placed together in play themes including bathing and feeding baby, play mat with transportation toys, barn with animals, zoo animals, and books. The toys and books used to elicit target words in the language sample are presented in Appendix A.

A transcription of each language sample was generated from the videotapes and audiotapes using the Systematic Analysis of Language Transcripts (SALT) (Miller and Chapman, 2004). The transcribers were blinded to the children's treatment phase. This analysis contained measures from the mothers' use of speech and language. The maternal measures included (1) number of total words used, (2) number of different words, (3) the mean length of utterance (MLU), (4) the percentage of responsive labels, (5) the percentage of commands/requests, and (6) the percentage of expansions. (Expansions are defined as repeating a lexical item and generally maintaining the topic while shifting the central meaning or changing the referential context, such as "Child: Get that! Adult: You want that over there"; Fey et al., 1999).

The children's measures included (1) the total number of words, (2) the number of different words, (3) the MLU, and

(4) the percentage of responses. The child speech measures were computed using the PROPH+ component within the Computerized Profiling System (Long and Fey, 1993). Consonant distortions were counted as correct for both common and uncommon clinical distortions (Shriberg, 1993). Common distortions included such productions as lateralized production of /s/, while uncommon distortions included nasal emission on consonants. The rationale for not counting these common distortions in the Percent Consonants Correct-Revised (PCC-R) calculation is that they are developmentally appropriate for young children. Analyses were completed on the responses to elicited target words in the sample from Appendix A and the middle 50 words in the language sample. The 50-word criterion was to equate the preintervention and postintervention speech samples for the children. Three speech measures were determined from the analysis: (1) the percentage of glottal stops, (2) the number of true consonants in the phonetic inventory (i.e., true consonants exclude glide or glottal consonants), and (3) the PCC-R (Shriberg et al., 1997).

Intervention

Only the mothers of children in the CLP group were trained to use a focused stimulation approach to facilitate the use of target words that contained stop consonants. A multiple baseline design across two sets of target words was conducted over three phases: baseline, intervention, and maintenance. Baseline consisted of the mothers playing with their children for 20 minutes using a generic set of toys and books that contained target words. The mothers were instructed to play as they typically did with their child. Following three baseline sessions for both target word sets, the mothers of the children with CLP received training in the intervention. The training consisted of a description of the focused stimulation procedure (with the written handbook included in Appendix B), role-play of scripted examples of the procedure, demonstration of the technique by the clinician with the child, and parent-child practice of the technique with coaching from the clinician following the parent-training procedures in Kaiser and Hancock (2003). A flow chart of the procedure is provided in Appendix C. The parents of the children with CLP were trained to model target words at least three times during the play intervention. Once the parents were able to perform this consistently, they were trained to respond to their child's turn through expansions if their child produced the word or repetition of the model (corrective model) if the child did not respond to the first model. The parents received two to four 45-minute training sessions until they achieved 80% accuracy in the practice sessions with their child. Training on the first set of target words began and continued until the child produced all the target words spontaneously at least once in the session. Once the first set of words reached the criterion, the intervention began on the second set of target words until they also reached the

criterion. Generalization of the target words was monitored during treatment and in a maintenance phase following treatment for each set of words. During this same period, the parents and children in the NCLP group underwent the baseline and generalization procedures but did not receive the training or intervention procedures.

Reliability

Twenty percent of both the SALT (Miller and Chapman, 2004) and phonetic transcripts were randomly selected by the first author. Five-minute segments at the midpoint of the language sample for each child in the study were edited, without participant identifiers, onto a compact disk (CD) by a graduate student unfamiliar with the project. The first author selected numbered clips without viewing the CD. These clips were retranscribed by a second transcriber familiar with SALT transcription and phonetic transcription of speech disorders in children. Interrater and intrarater language transcript reliability yielded a weighted Kappa of .66, indicating good agreement (Altman, 1991). The calculations for the PCC-R were calculated by a second transcriber, which yielded a weighted Kappa of .68, indicating good agreement. Interrater and intrarater phonetic transcription reliability yielded a weighted Kappa of .43, indicating a moderate agreement (Altman, 1991). In addition, 20% of the mothers' utterances were recoded as responsive labels, expansions, and command/requests. The percentage agreement between the two transcribers for coding the mothers' utterances yielded a weighted Kappa of .61, indicating good agreement.

Treatment Fidelity

To determine how well the mothers used the focused stimulation procedure during the intervention, a flow sheet of procedural components was used to monitor use of the procedures (Appendix A). The procedural accuracy of the mothers was assessed following their model of target words during the treatment sessions. Two sessions during the treatment phase were assessed with the flow sheet by a clinician not associated with the study, and a weighted Kappa was computed for parental compliance with the procedure. The comparison yielded a weighted Kappa of .62, indicating that all the mothers used the intended focused stimulation procedures for both sampled sessions during the treatment.

Analysis

For continuous data, the mean and standard deviation were used to summarize values for each study group. Means were compared with the *t* test (group 1 versus group 2 comparisons) or the paired *t* test (pre versus post comparisons within a group). A probability level of .05 or smaller was used to indicate statistical significance. The

TABLE 2 Study Group Comparisons for Mothers' SALT (Miller and Chapman, 2004) Measures†

Measurement	Group	Pre		Post		p Level
		Mean	SD	Mean	SD	Pre Versus Post
Number of words	1	108.5	51.2	155.3	73.9	.046*
	2	1483	530	1349	485	.224
p level	1 versus 2		.000*		.000*	
Number of different words	1	45.5	24.2	92.6	37.8	.000*
	2	244.3	50.4	252.1	49.9	.221
p level	1 versus 2		.000*		.000*	
MLU	1	3.43	0.70	3.81	0.85	.226
	2	4.27	0.58	4.45	0.27	.304
p level	1 versus 2		.009*		.046*	
% Responsive labels	1	8.04	7.47	10.14	8.08	.514
	2	17.57	12.46	2.25	3.02	.002*
p level	1 versus 2		.057		.015*	
% Expansions	1	5.96	7.78	14.00	9.75	.027*
	2	1.48	1.59	3.05	2.11	.003*
p level	1 versus 2		.108		.007*	
% Command/requests	1	33.76	17.00	23.31	14.50	.074
	2	37.29	6.53	36.80	9.87	.839
p level	1 versus 2		.553		.028*	

† The values for pretest, posttest, and change (differences from pretest to posttest) are the group mean and then the standard deviation. The number of subjects in each group is 10. Group 1 = children with cleft lip and palate; group 2 = noncleft children. MLU = mean length of utterance. SALT = systematic analysis of language transcripts (Miller & Chapman, 2004).

* Statistical significance, $p < .05$.

effect size of significant findings was determined using Cohen's d . The effect size is a name given to a family of indices that measure the magnitude of a treatment effect. Unlike significance tests, these indices are independent of sample size. A small effect size is represented by a d of 0.25, 0.50 a moderate effect size, and 1.00 a large effect size (Meline and Schmitt, 1997).

RESULTS

Can Parents Be Trained to Deliver an EI Program for Children Younger Than 3 Years?

Results demonstrated that the mothers who participated in this study could be trained successfully to deliver a focused stimulation EI program. Pretreatment and posttreatment measures show that the mothers of children with CLP increased their use of specific targeted language facilitation strategies. Table 2 shows that the mothers who received training increased the total number of words they used with their child ($p = .046$, $d = 0.73$), increased the number of different words used ($p \geq .000$, effect size $d = 1.48$), and increased the use of expansions ($p = .027$, $d = 0.91$). These significant findings showed large effect sizes. The mothers who received training also decreased the use of command/requests, but this change did not reach statistical significance ($p = .074$). Other measures shown in Table 2 did not show a significant change over the 3-month period of the study.

Inspection of the data comparing the mothers of children in the CLP group with the mothers of children in the NCLP group shows that, 3 months later, the mothers in the latter group were using a greater number of total words, greater number of different words, and higher MLU, suggesting that these mothers were using a greater complexity of

language when addressing their children. However, a comparison of premeasures and postmeasures shows that the mothers of children in the NCLP group (who did not receive training) did not show significant changes in any of these variables except an increase in percentage expansions ($p = .003$, $d = 0.84$) and a decrease in the use of responsive labels ($p = .002$, $d = 1.68$). When group differences between the mothers was examined posttreatment, the mothers of the children with CLP showed a greater use of responsive labels ($p = .015$, $d = 1.29$) and expansions ($p = .007$, $d = 1.55$) and fewer commands/requests ($p = .028$, $d = 1.08$). All these differences between the groups had large effect sizes.

Does a Parent-Implemented EI Program for Children With Cleft Palate Younger Than 3 Years Result in Positive Changes in Speech and Language Characteristics?

Table 3 shows the SICD-R scores for the children in the CLP group and children in the NCLP group at initial assessment and 3 months later. The children in the CLP and NCLP groups did show significant progress in their language development between pretreatment and posttreatment and moderate to large effect sizes for both SICD-R–receptive (CLP: $p = .002$, $d = 0.88$; NCLP: $p = .005$, $d = 0.91$) and SICD-R–expressive language measures (CLP: $p = .041$, $d = 0.79$; NCLP: $p = .006$, $d = 1.03$). However, the two groups were not significantly different from each other at pretreatment (receptive language pretreatment: $t = 0.53$, $df = 18$, $p = .73$; receptive language posttreatment: $t = 0.98$, $df = 18$, $p = .39$) or posttreatment (expressive language pretreatment: $t = 0.88$, $df = 18$, $p = .60$; expressive language posttreatment: $t = .29$, $df = 18$, $p = .80$) for both receptive and expressive language.

TABLE 3 Study Group Comparison for Receptive and Expressive Language Ages for the Sequenced Inventory of Communicative Development-Revised (SICD-R)†

Measurement	Group	Pre		Post		p Level
		Mean	SD	Mean	SD	Pre Versus Post
SICD-RA	1	23.0	7.50	29.20	6.55	.002*
	2	22.0	4.71	26.80	5.67	.005*
p level	1 versus 2		.73		.39	
SICD-EA	1	21.20	4.24	27.90	11.08	.041*
	2	20.0	5.66	26.80	7.32	.006*
p level	1 versus 2		.60		.80	

† The values for pretest and posttest are the group mean and then the standard deviation. The number of subjects in each group is 10. Group 1 = children with cleft lip and palate; group 2 = noncleft children; SICD-RA = receptive language age score (in months) on the SICD; SICD-EA = expressive language score (in months) on the SICD-R.

* Statistical significance at $p < .05$.

Table 4 shows the language performance of the children in the CLP group prior to and following the parent-implemented intervention, compared with the children in the NCLP group matched for pretreatment language age. Results show that following the intervention, the children with CLP demonstrated significant increases in the number of words ($p = .012, d = 0.94$), number of different words ($p = .000, d = 1.04$), and MLU ($p = .007, d = 0.94$). Effect sizes were calculated to determine the clinical significance of these findings using Cohen's d . The effect sizes were large for all three measures, indicating that the magnitude of the group differences was large. During the same time interval, the children in the NCLP group whose mothers had not received training also showed a significant increase in language measures, including the total number of words ($p = .002, d = 0.36$), number of different number of words ($p = .000, d = 0.46$), and MLU ($p = .021, d = 0.25$). However, the effect sizes for these three measures were small for the NCLP group. As expected, the statistical comparison of the two groups showed that they did not differ from one another on measures of language performance prior to the intervention. (Recall that the children were purposefully matched on language performance prior to the study, so there should not be a difference between the groups on this variable.) Following the intervention, both groups showed significant improvement in number of words, number of different words, and MLU, and the groups were not significantly different from each other.

Figures 1 through 3 show the children's performance on the speech measures collected during the preintervention and postintervention from the responses to the standard target words (Appendix A) and the middle 50 utterances of the spontaneous language samples. Figure 1 shows the number of true consonants prior to and following the intervention period for the children in the CLP and the NCLP groups. The children in the CLP group used significantly fewer true consonants than the children in the NCLP group both prior to treatment ($t = 8.09, df = 18, p < .0001, d = 3.81$) and following treatment ($t = 7.63, df = 18, p < .0001, d = 3.59$). Both groups demonstrated a significant increase in number of true consonants (CLP: $t = 3.37, df = 9, p = .008$; NCLP: $t = 3.61, df = 9, p = .005$). These significant findings showed large effect sizes (CLP, $d = 2.24$, and NCLP, $d = 2.40$).

Figure 2 shows the performance of individual subjects in the intervention study for the number of true consonants prior to and following the intervention compared with the mean performance of the NCLP group. Inspection of Figure 2 shows that 8 of the 10 children in the intervention study demonstrated an improvement in the number of true consonants in their inventories. It appears that the greatest change occurred for children 30 months and older.

Figure 3 shows the percentage consonants correct (PCC-R) in the children's speech. Prior to the intervention, the CLP group had a significantly lower PCC-R than the children in the NCLP group ($t = 3.09, df = 18, p = .006$,

TABLE 4 Comparison of Language Performance for the CLP and NCLP Children Prior to and Following the Intervention Period†

Measurement	Group	Pre		Post		p Level
		Mean	SD	Mean	SD	Pre Versus Post
Number of words	1	44.9	66.7	162.0	163.8	.012*
	2	292	344	418	350	.002*
p level	1 versus 2		.053		.058	
Number of different words	1	19.6	28.9	56.8	41.1	.000*
	2	64.8	65.6	97.4	74.7	.000*
p level	1 versus 2		.069		.156	
MLU	1	0.92	0.49	1.54	0.79	.007*
	2	1.41	0.98	1.65	0.93	.021*
p level	1 versus 2		.182		.777	

† The values for pretest, posttest, and change (difference from pretest to posttest) are the group mean and then the standard deviation. The number of subjects in each group is 10. Group 1 = children with cleft palate; group 2 = noncleft children; MLU = mean length of utterance.

* Statistical significance at $p < .05$.

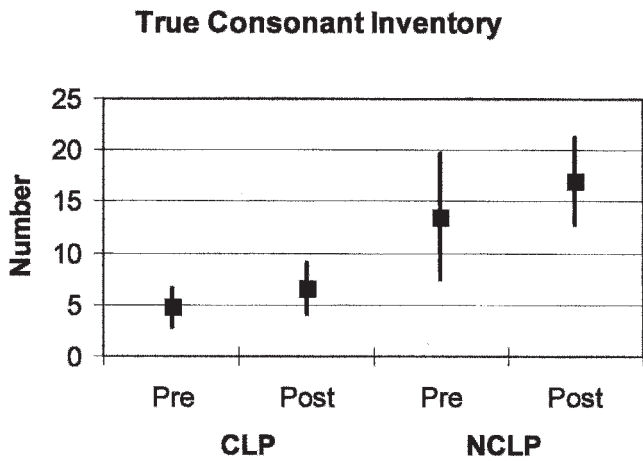


FIGURE 1 Mean and standard deviation for pretreatment and posttreatment true consonant inventory for the children with and without cleft lip and palate.

$d = 1.45$). Following the intervention period, there was a significant difference between the groups, with the CLP group showing poorer PCC-R than the NCLP group ($t = 3.92, df = 18, p = .001, d = 1.84$). The CLP group demonstrated a significant improvement in PCC-R following the intervention ($t = 3.76, df = 9, p = .004, d = 2.5$), while the NCLP children showed no change in PCC-R over the same time interval ($t = 0.59, df = 9, p = .58$).

Figure 4 shows the performance of the individual subjects with CLP in the intervention study group for PCC-R prior to and following the intervention compared with the mean performance of the NCLP group. Inspection of the data shows that all but one child demonstrated increases in PCC-R following the intervention. The trajectory of change for the children with CLP in the present study over the period of the intervention indicates rapid increases in their speech sound accuracy relative to the performance of the children in the NCLP group.

Figure 5 shows the percentage of glottal stops in the children’s utterances calculated during the speech sample. The children in the NCLP group had no glottal stops prior

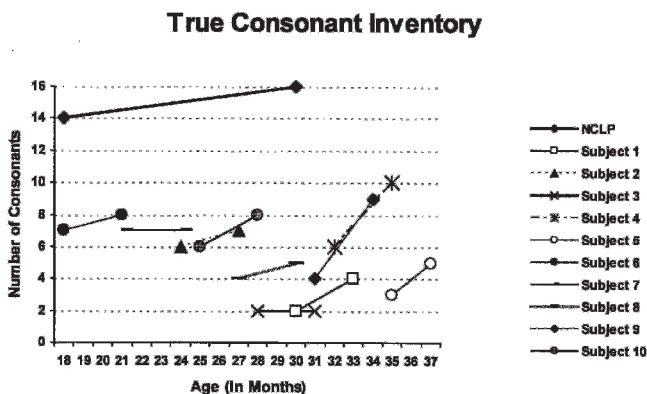


FIGURE 2 Individual pretreatment and posttreatment true consonant inventories for the children with cleft lip and palate and the true consonant group mean for the children without cleft lip and palate.

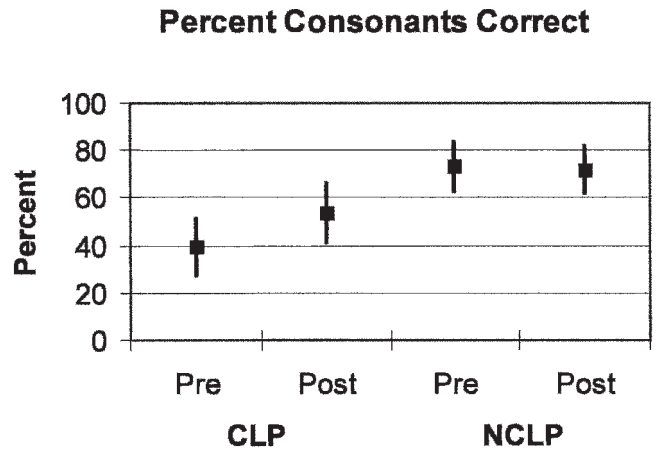


FIGURE 3 Mean and standard deviation for pretreatment and posttreatment Percent Consonants Correct–Revised for the children with and without cleft lip and palate.

to or following the intervention. The children in the CLP group demonstrated a high use of glottal stops prior to the intervention and a significant reduction in the percentage of glottal stops following the intervention ($t = 2.61, df = 9, p = .02, d = 1.74$). The effect size for this result was large, suggesting that the magnitude of the reduction in glottal stop use was large.

Figure 6 displays the percentage of glottal stops in the speech sample for each child in the study group prior to and following the intervention compared with the mean performance of the NCLP group. Nine of the 10 children with CLP in the treatment study used glottal stops. All nine of these children showed a decrease in the percentage of glottal stops used following intervention.

DISCUSSION

There are two main models for the delivery of EI programs: one is clinician delivered, and the other is delivery by parents under the supervision of a speech

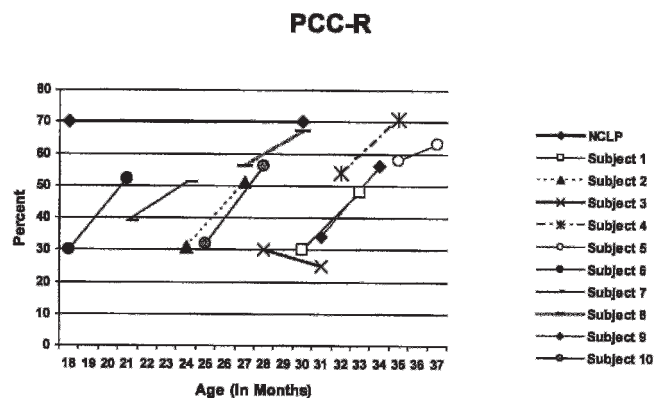


FIGURE 4 Individual pretreatment and posttreatment Percent Consonants Correct–Revised (PCC-R) scores for the children with cleft lip and palate and the PCC-R group mean for the children without cleft lip and palate.

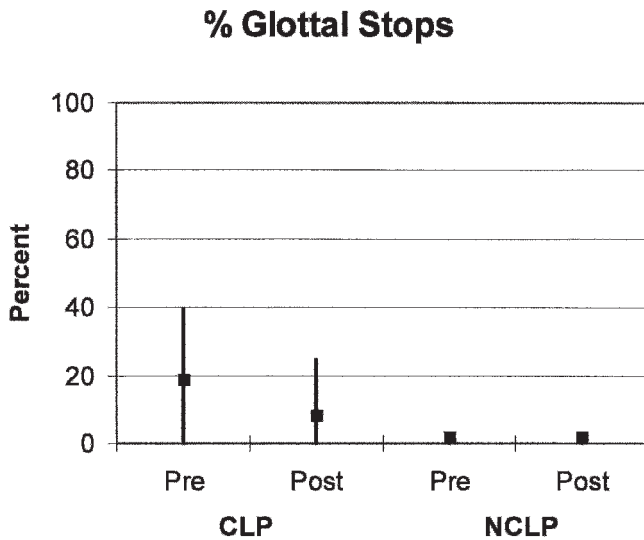


FIGURE 5 Mean and standard deviation for pretreatment and posttreatment percentage of glottal stops for the children with and without cleft lip and palate.

clinician. The present study was designed (1) to determine if parents could be trained to deliver an EI program for children younger than 3 years with cleft palate and (2) to determine if a parent-implemented EI program resulted in positive changes in speech and language characteristics.

Can Parents Be Trained?

The results of the study demonstrated that mothers of children with CLP can be trained to deliver a focused stimulation, parent-implemented EI program. The training delivered in this study resulted in changes in the mothers' use of communication strategies to promote vocabulary use and speech production. These mothers of children with CLP, who received training, demonstrated an increased use of modeling of target words, increased expansions of their children's utterances, and decreased use of commands. The training effects observed in this study are similar to those reported in the literature for the parents of children with other clinical impairments such as developmental delay, phonological impairment, and expressive language impairment (Girolametto et al., 1986, 1996, 1997). The results of this study also show that the mothers of children with CLP were able to learn the methods in a relatively short time interval with limited clinician instruction and were further able to maintain accurate use of the strategies over the 3-month course of the study.

The mothers of the children with CLP were not significantly different from the mothers of the NCLP children prior to the intervention on three critical language facilitation measures, including percentage of responsive labels, percentage expansions, and percentage use of commands and requests. However, following the intervention, the two groups were significantly different, with the mothers of the children with CLP, who received the

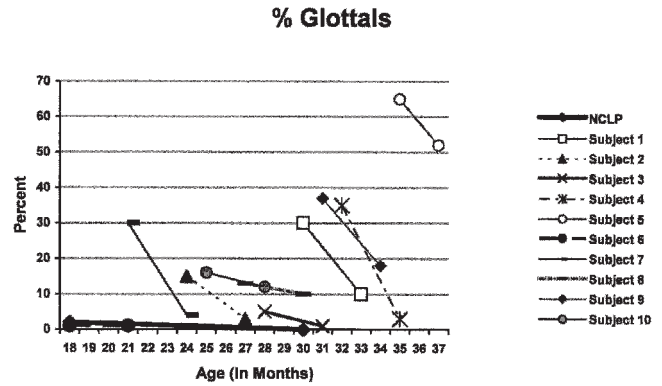


FIGURE 6 Individual pretreatment and posttreatment percentage glottal stop use for the children with cleft lip and palate and the percentage glottal stop group mean for the children without cleft lip and palate.

intervention, showing a higher percentage of responsive labels and expansions and lower percentage of commands/requests than the mothers of the NCLP children. These data suggest that the mothers of children with CLP were more responsive to their children's communication level than the mothers of noncleft children, who used vocabulary diversity and sentence complexity far in advance of their children's language level.

Does Parent-Implemented EI Result in Changes in the Speech and Language of Children With Cleft Palate?

The parent-implemented EI program used in this study was efficient in that it resulted in gains in both speech and language. Evidence of this can be seen in the growth of vocabulary, consonant inventory, and PCC-R. The association between vocabulary gains and speech production gains in young children with clefts was demonstrated previously in a clinician-implemented EI study (Scherer, 1999). The present study showed similar findings, demonstrating gains in speech production skills following a vocabulary-based EI program implemented by parents.

While the children with CLP, who received the intervention, made significant gains during the intervention they remained significantly impaired when compared to the performance of the children without CLP on the measures of true consonant inventory and speech accuracy following the intervention. This finding suggests that the children with CLP made clinically significant improvements in speech and language measures over the period of this study even though they did not achieve performance commensurate with the NCLP children. While the goal of EI is normalization of speech and language performance, the gains made by the children with CLP can be viewed with optimism given the very short duration of the treatment period in this study.

Given the results of this study, what is the likely mechanism responsible for the changes observed in the speech of the children with clefts? One theory of

phonological learning suggests that children change their productions in response to repeated practice (Stoel-Gammon, 1992). As the children attempt the target words in response to their mother's models, they can compare and potentially modify their production relative to the adult model. Subsequently, the parent provides corrective feedback following their child's production, thus completing the feedback loop for the child. Training the parent to increase the frequency of target word models provided the opportunity for the child's phonological practice.

Several observations regarding the data warrant further discussion. First, many of the children with CLP in this study showed use of glottal stop productions early in their speech and language acquisition. In response to specific facilitation of phonological practice provided by the parents, the children replaced their glottal productions with oral stop consonants. This pattern suggests that oral stop acquisition for children with CLP may display characteristics of phonological learning apart from velopharyngeal status. Second, the data in this study suggest that with respect to acquisition of new consonants, the most phonologic change occurred for the children 30 months and older. However, change in PCC-R was approximately the same for children of different ages. These data suggest that children with CLP were improving their speech accuracy during the intervention regardless of their age. But the greatest gains in new consonant acquisition occurred after 30 months of age. Although the number of participants in this study was small, this finding may have implications for the timing and nature of goals selected for early speech and language treatment. Future intervention studies should address this issue.

Comparison to the Developmental Course of Children With Clefts

The literature regarding the early speech production skills of children with CLP consistently demonstrates that children with cleft lip and palate manifest reduced consonant inventories, reduced speech accuracy, and the use of compensatory articulations (Scherer et al., 1999; Chapman et al., 2001, 2003). The results of this study showed clear improvement in all of the monitored aspects of speech production, including increased true consonant inventory, increased speech accuracy as evidenced by improved PCC-R, and reduced use of a prominent and particularly deviant compensatory articulation pattern—the use of glottal stops. All of these gains are likely to result in an increase in the number of sounds in the child's repertoire that are then available for use in word production, thus increasing vocabulary production. The gains in speech accuracy and reduction in glottal stops translate into improved speech intelligibility.

The reduction of glottal stop use as a result of the parent intervention is of particular theoretical interest and clinical importance. There are few studies in the literature that

address the efficacy of traditional therapy techniques for eliminating glottal stops once they are established. However, it is generally agreed that the elimination of these sound patterns should be addressed early (Peterson-Falzone, et al., 2001, 2006). There are no methods described and tested for preventing the occurrence of compensatory articulation errors nor studies that address the elimination of these patterns in children younger than 3 years when these sounds are present but have not yet been fully habituated.

Therefore, perhaps the most significant finding of this study was the reduction of glottal stops in young children with CLP as a result of the parent-implemented intervention. This study clearly provides data showing that glottal stop production can be reduced in children younger than 3 years without traditional therapy techniques that require the child's focused attention on place and manner of articulation using drill-and-practice methods. Inspection of Figure 6 shows that 9 of the 10 children with CLP in this study demonstrated the use of glottal stops. All nine of these children demonstrated a reduction in use of this compensatory articulation pattern. In a recent study, the percentage of glottal stop use was assessed longitudinally in a group of children with CLP aged 12 to 30 months (Scherer, 2001). The incidence of glottal stops (in terms of the number of children producing them) rose from 8% at 18 months of age to 13% at 30 months of age, indicating a gradual rise in percentage glottal stop use, while seven of the nine children in the intervention study showed a reduction in glottal stop use. Even the oldest child in the present intervention study who had the highest percentage of glottal stops showed a marked improvement from 64% to 52% following the 3-month intervention. These results demonstrate the benefit of addressing these error patterns prior to stabilization of the errors into the child's phonologic sound system and motoric patterns.

The literature on phonological development suggests that young children learn speech through repeated practice with sounds and monitoring of their practiced productions (Stoel-Gammon, 1992; Vihman, 1992). The focused stimulation intervention program in this study specifically targeted parent stimulation that facilitated the children's sound practice in words and provided corrective feedback following their word attempts. It is likely that this process, then, facilitated elimination of the glottal stop productions for those children with the capacity for complete velopharyngeal closure. The children in the present study were too young to complete accurate assessments of velopharyngeal function. However, anecdotally, it was our clinical judgment that all but two of the children (subjects five and six) demonstrated evidence of velopharyngeal competence.

Implications for Service Delivery Models

An important implication of the results of this intervention study pertains to considerations regarding

service-delivery models for speech-language therapy. As health care and education budgets shrink, there are limits placed on the number of therapy sessions allowed and mandates for increasing the numbers of children on a clinician caseload. It is essential that we look to new service-delivery models. In the United States and many industrialized countries, the number of sessions allowed or the number of clients who must be cared for prevents the kinds of services often necessary to treat the communication impairments of many children with CLP. Therefore, it is essential that we provide evidence to support intervention programs that might lessen the amount of time required by the SLP. Parent-implemented EI programs address this need (Gibbard et al., 2004). The results of this study clearly demonstrate that an EI program supervised by an SLP but provided by a parent on a daily basis can lessen the communication handicap of young children with CLP.

In addition to the benefits just described, the results of this study regarding parent-implemented EI have important implications for regions of the world where there are few or no SLPs. This type of program is not meant to supplant the services of an SLP and would be best supervised by a fully qualified SLP. However, in many locations of the world, in fact, in most of the world, there are few or no SLPs. In these regions, it is likely that a parent-implemented therapy program such as the one described in this study could be modified for use by allied health workers, rural health workers, or community based rehabilitation workers. For example, D'Antonio and Nagarajan (2003) have suggested that there are a variety of solutions for providing SLP services to children with CLP in locations in the world where there are no SLPs. They suggested that the solutions that are most likely to work in the immediate future are those that require little funding and take advantage of existing resources and build on newer models of rehabilitation. These authors (Nagarajan, et al., 2006) further suggested that a principal means for increasing speech services for children with CLP in most of the world is to train village health workers, school teachers, special educators, parents, and successful older individuals in the community with CLP to provide some of the basic speech intervention that is needed. However, prior to the data provided in this study, there has been little evidence-based information to suggest methods that might be useful for developing a non-clinician-implemented program for use by individuals in regions where there are no SLPs. Perhaps most importantly, the method described in this study demonstrates a reduction of communication deficits in very young children before they have become habituated and thus require intervention by more highly trained individuals. Therefore, one of the major contributions of this study is the evidence it provides to support the benefits of a non-clinician-implemented EI program. The intervention program used in this study can now be evaluated with an expanded set of intervention providers to determine whether similar beneficial outcomes in

language and speech in young children with cleft lip and palate can be obtained in regions where few or no SLPs are available. In addition, a larger cohort study comparing the impact of the intervention provided by parents or other health workers is warranted. A follow-up study should address the impact of a longer intervention period on the normalization of speech and language performance of children with CLP and long-term maintenance of parent use of facilitation strategies during the intervention.

Limitations

While the use of EI did show improvements in several aspects of the children's speech and language, there are some limitations that should be addressed. First, this study did not provide a prospective comparison between a group of children with CLP who received a parent-implemented intervention program and a group of children with CLP who did not receive intervention over the same time interval. This study required comparison to an NCLP group at the same language development level to discern whether the impact of the treatment moved the children with CLP to the normal developmental trajectory for speech and language performance.

Similarly, the present study suggests that some parents or children may have additional issues that reduce the effectiveness of intervention. For example, there was one child in the treatment group who had the most severe impairment and demonstrated the least gains following the intervention. This child appeared to have additional deficit areas that became apparent during the study. The presence of a global developmental delay may reduce the observed pace of progress. In addition, further investigation is needed to determine the impact of a combined approach to treatment, which might include a parent-implemented and clinician-implemented intervention. Finally, future research should consider a cost analysis comparing the financial costs and therapeutic benefits between a parent-and-clinician-implemented intervention in this population.

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APPENDIX A

LIST OF WORDS TARGETED IN THE LANGUAGE SAMPLE

<i>Sound</i>	<i>Initial Word Position</i>	<i>Final Word Position</i>
m	money	mom
n	night-night	moon
ŋ	—	ring
p	puppy	cup
b	baby	cub
t	tiger	boat
d	duck	bird
k	car	truck
g	goat	egg
f	feet	off
v	vine	move
s	soap	house
z	zoo	shoes
th	—	bath
sh	sheep	wash
ch	cheese	peach
dg	juice	fudge
w	wagon	—
j	yoyo	—
l	lion	ball
r	ring	roar

Books: *The Very Hungry Caterpillar*, by Eric Carle (1970); *Brown Bear, Brown Bear, What Do You See?*, by Bill Martin Jr. and Eric Carle (1970); *Goodnight Moon*, by Margaret Wise Brown (1947).

APPENDIX B

WRITTEN INTERVENTION MANUAL

Manual for Parents of Young Children With Cleft Lip and Palate

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Early Speech and Language Development in Children With Cleft Lip and Palate

Young children with cleft lip and palate approach speech and language development at a disadvantage. They have not had the opportunity to practice sounds with a typical oral mechanism until their palates are repaired. There have been several studies of the speech of children with cleft lip and palate before and after their palates are repaired. These studies show that children with cleft lip and palate make fewer sounds before their palates are repaired and they continue to use fewer sounds long after their palates are repaired. Young children with cleft lip and palate have trouble saying sounds that represent one class of sounds called stop consonants. Stop consonants include the sounds p, b, t, d, k or c, and g. The stop consonants are made by using the lips and tongue to build up air pressure in the mouth and release it. These sounds are among the earliest sounds acquired by young children and are contained in many words used by toddlers. Think of the first words of young children such as daddy, dog, baby, ball, go, pop, and cup. They all include stop consonants. These sounds are

difficult to produce for young children with cleft lip and palate because they require that the palate close off the nose completely. Recent studies show that not only do children with cleft lip and palate use fewer sounds but also they start talking later and have smaller vocabularies. The purpose of this program is to train you to increase the vocabulary of your child by using words that have been specially selected to teach sounds that may be difficult for your child with a repaired cleft.

Glottal Stop Sounds

Some children with cleft lip and palate use another sound to replace stop consonants called the glottal stop. Instead of using a stop consonant, the child may substitute a glottal stop. The glottal stop is a sound made with the vocal cords in the throat rather than the lips or tongue. When a child uses a glottal stop instead of a stop consonant, it is an indication that the child does not know how to use their new palate or that their new palate may not be working adequately. When children are in the early stages of learning to talk, we do not know if they haven't learned to use their palates after surgery or if the palate itself cannot close off the nose. One approach to reduce the use of glottal stops is to teach the child to use stop consonants instead of glottal stops. The program that you will be using will hopefully help your child to learn to use stop consonants instead of glottal stops.

The Training Program

Your child has undergone a recent speech and language assessment to determine his or her level of development. Additionally, you completed a questionnaire about your child's speech and language use at home. From these measures, we will determine a list of words that include sounds that will help your child learn words with stop consonants.

The backpack is provided with several books, toys that accompany one of the books, and toys to engage play surrounding a cooking theme. The backpack includes all the basic materials that you will need for the training and are yours to keep. However, during the intervention, please keep all the materials in the backpack. The toys were selected to represent a large number of words that contain stop consonant sounds that will be the target of the intervention. Additionally, the toys represent a range of activities that toddlers enjoy. Your child may not play with all the toys initially but will engage with all of them as you begin to include them in your play.

Techniques for Using All Toys

Barney book. For the first two sessions, we ask that you look at this book with your child as you typically would.

Don't attempt to use the focused stimulation or other speech and language techniques with this book. We want to see how your child responds to the book so that we may judge his or her progress when you begin using the speech and language stimulation techniques.

The *Hungry Caterpillar book and toys*. Starting the third day of the program, look at the book together with your child. It is not necessary to read the book word for word. Rather, talk about the story and the items in the pictures. If your child prefers to look at the pictures and skip around the book, let them. Some young children have an interest in looking at books, while others prefer playing with toys. The backpack provides both a book and toys so that you may use them according to your child's interest. You will be given a list of words to use during play and book reading with your child. Use these words as many times as possible while looking at the book. Use the toys that go with the book to permit your child to act out the story either during or after looking at the book.

The additional food and dishes are provided to facilitate pretend play surrounding a cooking and eating theme. The toys from the book may be used in play as well. We recommend that you include some of your child's own toys into the play. Of course, those that begin with stop consonants are preferred, but any favorite toy may be included in the play.

Focused stimulation is a procedure parents can use to increase their children's vocabulary use. The principle behind the procedure is that the more you use a word while playing with your child, the more likely your child will use that word. So we will be giving you a list of words to use while you play with the toys provided in your child's backpack. You are encouraged to use some of your child's own toys to supplement the toys in the backpack. During the program, it is best to keep all the toys together so that they are readily available during the times that you spend with your child on the program. It is recommended that you plan 10 to 20 minutes each day to work on the program. In order for your child to get the most benefit from the program, it is recommended that you work with your child at least five times a week. A tape recorder and tapes have been provided for you to record your sessions with your child. I will make arrangements to collect the tapes from you.

Emphasis of First Sounds in Words

There is an adaptation of the focused stimulation in which you emphasize the first sound in the word. It is thought that this procedure helps your child to pay attention to these words, in particular, the first sounds in the words. This procedure requires that you emphasize the first sound of the word by adding an "h" after the sound. For example, "pat" becomes "phhhat" or "ball" becomes "bhall." Or make the first sound in the word a little louder

than the other sounds. Use these procedures with the word list given to you.

Setting up the Learning Environment

It is important that your child be able to see your face so that they may see how your mouth moves when you say words and so that you can see their facial expressions, level of interest, and what they are paying attention to. Sit so that you can see into your child's eyes. Get down on your child's level. Sit on the floor. Let your child sit in a chair, if they wish. It is common when reading a book to sit next to your child or have your child on your lap. We want your child to see your face as much as possible. So try sitting in front of a mirror with your child on your lap, or hold the book in your lap facing your child.

You will be given a list of words that have been selected based on the sounds contained in them and their usefulness to enhance your child's speech development. The sounds that will be selected are within the stop consonant class of sounds that are often problematic for young children with cleft lip and palate.

We want you to use these words as many times as you can while looking at the *Hungry Caterpillar* book and playing with the toys in the backpack. Please view the videotape provided in the backpack for examples of how to use the words selected for your child in the play activity.

Let Your Child Know You Are Listening

In addition to the focused stimulation procedure, there are some additional procedures that we know improve young children's speech and language development by promoting sustained engagement in conversations.

REPEAT. One of the best ways for your child to know that you are listening is to repeat words, sounds, and actions that they use. If your child says a word or a part of a word, repeat it. They may try to say the word again after you repeat it. For example, your child may say "ma" while picking up the mother figure. You say "Mommy," and your child may try to say "Mommy." Even if your child does not try to say the word again, you have told them that you have heard what they said. Please view the videotape for examples of this procedure.

INTERPRET. Another way to show your child that you are listening is to interpret what they say. Interpreting may mean guessing at what your child is trying to say and putting it into words. When you are pretty sure about your child's meaning, put it in a sentence. For example, if your child says "ba" when you put them in the bath, you say, "Yes, you are taking a bath." You have let your child hear the word pronounced in the adult form and in an appropriate sentence. However, sometimes it is difficult to know the meaning of your child's word attempts. In these cases, it may be helpful to repeat the child's word with a question in your voice. This encourages your child to

repeat their word attempt and perhaps be better understood. Please view the videotape for examples of this procedure.

COMMENT. Talk about what your child is doing even if they are not talking. This shows them that you are interested in what they are doing and models words for things they are focused on in their environment. This is also a good opportunity to work in words from the focused stimulation exercise. Please view the videotape for examples of this procedure.

MODELING. If your child is not saying anything, it helps to describe what they are doing by using simple, short sentences. This technique can also be used to emphasize certain words through focused stimulation.

EXPANSION. When your child says a word or short sentence, add to it when you take your turn. So if your child says “Dog,” you can expand what your child said to “The dog is running.” Add more information about what the child is talking about. In this way, you will be showing your child how to use words to describe what they are experiencing.

Common Problems Encountered by Parents

What if my child does not talk much at the beginning of the program? It takes some children a period of time to adjust

to the new play with toys and books. Continue to use the techniques in the program, particularly modeling and focused stimulation. Avoid excessive use of questions. The program does not require that your child use words immediately.

How long should I work with my child each day? Let your child guide you. At least 10 to 15 minutes is suggested, but some children will play for longer. We want this activity to be enjoyable for your child, but they may need encouragement from you to continue the play.

What do I do if my child does not want to end the play? This time spent with your child has a powerful appeal to young children, and they may want to extend it beyond what is manageable. Try to include the clean up as a regular part of the play. Have your child say “Bye” to each toy as you place them in the backpack.

What if I hear air coming out of the nose on the words? While this is a problem that requires further assessment for some children with repaired cleft lip and palate, young children may go through a period of learning how to use their palate. We will monitor your child’s nasal air emission during this program as they learn new words. If necessary, we will refer you and your child for further follow-up.

What do I do if I have questions during the program? We will be visiting you periodically, but you may call Dr. Nancy Scherer (423) 439-5254 at any time.

**APPENDIX C
FOCUSED STIMULATION FLOW SHEET**

