

**Critical Review:**  
**What are the effects of parent-mediated intervention for speech and language outcomes in children with cleft lip and palate?**

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This critical review examines the effects of parent-mediated intervention on speech and language outcomes in children with cleft lip and palate. Study designs include case-controls and between-group randomized clinical trials. Overall, there was found to be limited research available on the topic of interest. Further research studies are required before conclusive statements can be made regarding the efficacy of parent-mediated intervention on the speech and language outcomes in children with cleft lip and/or palate. Recommendations for future research and clinical practice are provided.

***Introduction***

Traditionally, intervention approaches for speech and language deficits in cleft lip and palate (CLP) have involved using modeling, stimulating environments and encouragement of spontaneous imitation of typical words and sounds (Edmonson & Reinhartsen, 1998). Within the traditional intervention model, a speech language pathologist, in addition to working with the child, endeavors to provide strategies to the parents that have been implemented in therapy. This traditional approach draws on a naturalistic intervention model in which everyday interactions are used to develop functional communication skills for a child (Scherer & Kaiser, 2007). While it would seem intuitive to use parents as interventionists within naturalistic models, therapy for children with CLP has remained primarily clinician-driven.

Research in other populations of children with speech and language deficits has shown that parent-mediated intervention (PMI) is a reliable service delivery model, which has positive speech and language outcomes. Girolametto, Pearce and Weitzman (1996, 1997) reported that parent-focused intervention is a feasible intervention model for late talking children with delays in expressive vocabulary and language abilities. As a result of the success of PMI in other non-CLP populations, researchers within the CLP field are now undertaking investigations to determine the efficacy of PMI models as a feasible alternative service delivery model (Scherer & Kaiser, 2007).

If PMI is shown to be an effective treatment approach for children with CLP, there is the potential for many clinical benefits. This approach has the potential to offer feasible alternatives to clinician-directed intervention in regions where CLP is common but where limited access to speech-language pathology services is

available (e.g. rural and third world countries). In urban centres where long waitlists are common, PMI could also be of significant benefit. Finally, PMI may help to reduce the financial burden on families given that in the traditional service delivery model children with CLP often require extended therapy (four to five years) (Edmonson & Reinhartsen, 1998). Before PMI can be employed as a feasible intervention model in children with CLP the effects of such a model on the speech and language development must be sufficiently determined through evidence-based research. The purpose of this critical review is to investigate the effects of PMI on speech and language outcomes in children with CLP.

***Objectives***

The primary objective of this paper is to provide a critical evaluation of existing literature on the effects of PMI for speech and language outcomes in children with CLP. A secondary objective is to offer evidence based recommendations for the use of this service delivery model in clinical practice as well as recommendations for future research.

***Methods***

Search Strategy

The computerized databases CINAHL, Pubmed, SCOPUS, Proquest Dissertations and Theses and Embase were used to find articles related to the topic of interest. Database searches were conducted using the following search strategies:

[(cleft lip) or (cleft Palate) and (intervention) or (program) and (speech) or (language)]

[(cleft lip) or (cleft palate) and (intervention) and (speech)]

[(cleft lip) or (cleft palate) and (intervention) or (program) or (treatment) and (speech) or (language) and (development)].

#### Selection Criteria

Studies selected for inclusion in this critical review were required to investigate speech and language developmental outcomes of children with cleft lip and/or palate whose parents had been involved in the therapy process via parent-training and parent-mediated intervention. With the exception of presence of a cleft lip and/or palate as well as parent involvement in speech therapy, no limitations were set on the demographics of the research participants.

#### Data Collection

Results of the literature search yielded 3 articles congruent with the aforementioned selection criteria. One article employed a case-control study, while two of the articles used a between group randomized clinical trial design.

### **Results**

#### **Studies using parent-mediated intervention**

Scherer, D'Antonio and McGahey (2008) carried out a case-control study to investigate the effectiveness of a parent-implemented, focused stimulation program on the speech characteristics of children with CLP below the age of three. They sought to answer two questions: Can parents be trained to deliver early intervention programs for children with CLP, and does a parent-implemented early intervention program result in positive changes in speech characteristics. A case group of 10 mother-unilateral CLP child pairs were matched to a control group of 10 mother-non-CLP child pairs. Inclusion criteria for participants with CLP consisted of: cleft lip with or without cleft palate in absence of a genetic syndrome; absence of significant medical or neurological impairments or preterm birth earlier than 36 wks gestation; and passing hearing screening. The case and control groups were matched for vocabulary production, age, gender and socioeconomic status, with vocabulary production being the primary matching variable. Pre and post-test measures consisted of standardized and informal procedures. The case group completed the *Sequenced Inventory of Communication Development, Revised (SICD-R)* and a 30-minute video and audiotaped language sample of mother-child interaction during a book-reading activity and free play. Transcribers who were blinded to the treatment phase generated transcripts. Analysis consisted of measures of the mother's use of speech and language and child measures consisted of four language measures and three speech measures. The non-CLP participants received no intervention so that intervention effects could be

compared to typical development over the study time period. Pre and post-intervention group differences for language and speech measures were compared for outcome measures.

Intervention consisted of training the mothers of participants with CLP to use a focused stimulation approach to facilitate the use of target words containing stop consonants. A multiple baseline design was used across two sets of target words over baseline, intervention and maintenance phases. Three baseline sessions took place in which mothers played with their children as they normally do. Intervention training followed during which parents received two to four 45-minute sessions until 80% accuracy in practice sessions were achieved. Training consisted of a description of the procedures, role-playing and parent-child practice of techniques with clinician coaching. The maintenance phase consisted of monitoring participants for generalization of target words.

The study had two primary questions. The first question was, can mothers be trained as reliable interventionists. The results of the study showed that yes, they can. Mothers were able to increase their use of target language facilitation strategies as measured by an increased number of words used ( $p = .046$ ); increased number of different words used ( $p \geq .000$ ) and increased use of expansions ( $p = .074$ ). These findings all showed large effect sizes and were therefore found to be statistically significant. The second question posed was does a PMI program result in positive changes in speech and language characteristics for children with cleft palate. The results showed that while participants with CLP made significant gains in speech and language measures ( $p = .002$  and  $p = .041$ ) with moderate to large effect sizes, they continued to have lower performance results than the non-CLP participants.

The proceedings by Scherer, D'Antonio and McGahey (2008) provide 2b level of evidence. This level of evidence uses some methods of scientific investigation, however, overall the methods employed are limited, providing only a moderate strength of evidence-based research. The rationale for the study was well presented and supported with appropriate research and an appropriate level of participant selection criteria was used to ensure generalization of the findings. Detailed information regarding pre and post-test measures were provided with the inclusion of a written intervention manual lending to the replicability of this study. The inclusion of a multiple baseline design allowed for a discussion of treatment effect. The results were analyzed appropriately, conducting both between and within group analyses and the results were well interpreted. Appropriate data was provided to ensure

reliability of the data, treatment fidelity and statistical analysis. The authors also provided a useful discussion of directions for future research in this area.

This case-control study was not without limitations and the authors clearly highlighted them. They discussed that no prospective comparison between participants with CLP who did or did not receive a PMI program was undertaken. Doing so would have increased the level of evidence of the study. Additionally, the authors believe that some parent or child participants may have had additional issues not considered in this study that decreased the effectiveness of this intervention approach. Consideration also needs to be given to the impact of a combined parent and clinician-implemented approach to treatment. It was noted that the study did not include a cost-benefit analysis. Finally, a small sample size was used which decreased the power of the study to detect potential differences and makes it more difficult to generalize the results to clinical practice.

Overall, given the strengths and limitations of the proceedings of Scherer et al. (2008) the results should be interpreted with caution when considering PMI as the primary treatment modality in clinical practice. The information presented in the study provides useful strategies to clinicians, which could be used to engage parents in the intervention process, however, without further evidence-based research with a stronger level of evidence the clinician would be prudent to use PMI with caution in their clinical practice.

### Studies using parent-involvement

Pamplona, Ysunza, Uriostegui (1996) conducted a between-group Randomized Clinical Trial (RCT) to investigate whether the active participation of parents in speech therapy enhances the linguistic performance of children with cleft palate. Twenty-one participants were found to meet the nine point inclusion criteria and were therefore included in the study. These participants were randomly divided into two groups. Group one (n=10) received whole language therapy while group two (n=11) received whole language therapy accompanied by their mothers. Age ranged from 3; 0 to 4; 8 for all 21 participants. Group one had a mean age of 3; 9 while group two had a mean age of 3; 7. All participants were found to demonstrate similar deficits on the *Batería de evaluación de la lengua Española (BELE)* linguistic development scales at baseline measures and all had below normal linguistic performance, for their age. Mother-child mode of interaction of group two was evaluated via video recording during a free play situation in order to consider aspects of the mothers' utterances such as semantic contingency and mode of interaction. Therapy was conducted for 1 hour, 3 times

a week for 8 months by a speech-language pathologist (SLP). Outcome measures included: linguistic performance, level of play and in group two mother's mode of interaction.

The authors found that there was no significant difference between either group for level of play post-intervention ( $p > 0.05$ ) using a Fisher exact test. Significant linguistic gains were made in both groups, however, participants who received therapy with their mothers scored significantly higher ( $p < 0.05$ ). Pamplona, Ysunza and Uriostegui (1996) concluded that the results of the study support the statement that linguistic development in the cleft palate child is strongly related to adult-child mode of interaction, which provides evidence for the inclusion of parents as active participants in therapy sessions.

This study used a well-defined set of inclusion criteria and ensured groups were both randomized and equally balanced by number of participants, age and language abilities. The study provided level 1 research evidence. This label is given to studies that have the highest level of scientific evidence and which use rigorous methods when investigating patient outcomes. While the authors ensured a high level of evidence, nevertheless several limitations existed in this study. The most notable limitation was that no post-treatment follow-up was conducted, thus durability of the treatment effect could not be reported. Additionally, there was limited analysis and statistical data provided and no inclusion or discussion of reliability measures used. Furthermore, in discussing the results the authors acknowledged that improvements seen could have been due to the increased number of adult models to which the children were exposed when more than one adult participant was present and not necessarily due to the inclusion of the child's parent. Thus, the proceedings by Pamplona et al. (1996) provide moderate to high strength of support towards the increased linguistic performance of children with cleft palate as a result of parental participation in speech therapy.

Pamplona and Ysunza (2000) carried out a follow-up study to Pamplona et al. (1996) in which they continued to investigate whether including mothers as active participants in speech therapy would improve language development of children with cleft palate and additional language delays. This study included 41 CLP participants with moderate language delay. The participants were assessed prior to entrance into the study and those with roughly the same language level were randomly assigned to either the control (n=20) or experimental (n=21) group. The authors chose to use a double-blind procedure with analyses conducted by 2 Speech-Language Pathologists (SLP). This resulted in a

95% agreement in classification of child behaviours pre-intervention and a 94% agreement post-intervention. Intervention consisted of three one-hour sessions of speech and language therapy per week carried out by a SLP using symbolic play activities, during a 12-month period. The design of the control and experimental treatment groups was based on the study by Pamplona et al. (1996). Both groups were similar in age with a range from 3; 0 to 4; 8. Group similarities included age, play and language ability. The mean age of the control group was 3; 8 compared to 3; 7 for the experimental group. All participants had moderate language impairments. Participants were evaluated via video recording during a free play activity with a SLP. Recordings were taken pre and post intervention. The recordings were transcribed and an analysis of play skills was done using the *Westby's Play Scale*. Language performance was assessed using an adaptation of *Bloom and Lahey's Communicative Categories*. Gain scores for language and play were calculated by subtracting the pre-intervention rating from the post-intervention rating.

Results showed no significant difference when levels of play were compared using a Fisher's exact test ( $p > 0.05$ ), indicating that the number of children who made gains in type of play skills was similar between groups. However, there was found to be a significant difference ( $p < 0.05$ ) when examining gains in language performance. A Fisher's exact test showed that overall, the experimental group made primarily two and three level gains while the control group had more limited one and two level gains. The study concluded that participants accompanied by their mothers to therapy sessions had significantly better language skills than participants with no mother participation.

This study provides level 1 research evidence giving it the highest level of scientific evidence. Appropriate statistics and standardized measures were used in this study. Strengths of this experiment include equally balanced control and experimental groups. As well, improvements were made from Pamplona, Ysunza and Uriostegui (1996), including a larger sample size and a 12, rather than 8, month intervention period. This strengthens the power of these findings. Furthermore, strict inclusion criteria were used in this follow-up study, which permitted the authors to clearly highlight their outcome measures. The proceedings of Pamplona and Ysunza (2000) provided sufficient detail so that the methodology and measures were well understood and could be easily replicated.

Limitations of this study include that pertinent demographic data, including participant gender, number of participants with primary or secondary clefts,

socioeconomic status of participants, parental education levels and limited analysis and statistical data were not included. Follow-up information regarding treatment effect was also not included in this study, thus long-term benefits of the intervention approach used are unknown. This impedes the use of these results in clinical practice.

This between-group RCT study provides a moderate to high level of evidence toward parental involvement in speech and language therapy. The level of evidence would have been further strengthened had treatment effect data been included. Overall, Pamplona and Ysunza (2000) lend support to the active participation of parents in speech therapy sessions.

### **Discussion**

This systematic review revealed that there is currently limited research in the area of PMI for children with CLP, given that only three articles met the inclusion criteria of this review. Furthermore, the few articles available together lack strong evidence-based research which causes some reservation about application of the results to clinical practice. A significant weakness of the literature presented in this critical review is that only one article used PMI as the primary treatment modality. An additional variable that limits the ability to conduct research of any kind with high levels of evidence in the CLP population is the low occurrence of CLP in the general population, contributing to small sample sizes in the studies. This factor is further compounded by the heterogeneity of the population, given that children can present with either cleft lip and/or palate with or without the presence of a genetic syndrome and therefore will have varying degrees of speech and language deficits. The influence of these variables makes conducting research on PMI in the CLP population difficult. Thus, it is difficult to draw conclusive findings with regards to the effects of PMI on speech and language outcomes of children with CLP.

As stated by Scherer and Kaiser (2007), the literature independently presents research limitations. However, they state that, as a whole the literature suggests naturalistic language intervention mediated by parents can perhaps be an effective early intervention approach as results suggest it does facilitate speech and language development. Thus the need for further investigation seems warranted. Further support for this treatment approach comes from Pamplona et al. (2001) who suggest that encouraging communication development in CLP children with speech and language difficulties should encompass a child's natural partnerships rather than limiting it to direct clinical therapy, since children's learning and development is supported by their interpersonal contacts. This was gleaned from

research, which shows that, when provided with instructions, mothers of children with CLP were able to change their communicative styles and mode of interaction. As a result they suggest that parents ought to be active members of therapy (Pamplona et al. 2001).

Work done by Brothers (2002) supports the statement that CLP children show gains from PMI as her investigation found that this method resulted in participants developing increased expressive language and sound accuracy in addition to changes in vocabulary use and decreased glottal stop productions. While this study had a small sample (n=4) and has not been peer-reviewed, it does provide suggestive results from which future studies could be developed.

Finally, a related issue, which suggests benefits of PMI concentrates on the importance of a parent-focused approach given that parental support is central to the inclusion of therapy goals in daily life (Hardin & Jones, 2007). Without this their research suggests that progress is likely to be less efficient and more protracted.

#### *Future research considerations*

It is recommended that further research be conducted in order to clarify and confirm the effects of PMI on the speech and language development of children with CLP. In order to generate evidence based research with strong levels of evidence it is recommended that future research consider the following:

- a. Future research should employ study designs that offer a stronger level of evidence with larger sample sizes and PMI as the primary treatment approach, such that results can be more confidently and appropriately applied to clinical practice.
- b. Future research should include an examination of long-term maintenance of treatment effects, in order to support the efficacy of the intervention approach.
- c. Future research studies should control for variables such as type of deficit (e.g. primary or secondary cleft), given the heterogeneity of this population, and should also consider participants involvement in pre-study speech and language therapy.
- d. A cost-benefit analysis of PMI should be undertaken, such that conclusions can be drawn regarding the efficacy of the treatment approach and efficiency of resource allocation.

- e. Future research should investigate the strengths and weaknesses of naturalistic models, such as focus stimulation and enhanced milieu training, when used in PMI for children with CLP.
- f. Future research must determine appropriate candidates for PMI within the CLP population taking into consideration potentially influential variables (e.g., socioeconomic, parent educational levels, community resources, etc.), in order to determine efficacy and effectiveness of this service delivery model.
- g. The use of confederates, such as early childhood educators and other professionals should be considered as possible alternative interventionists.

#### *Clinical Implications*

Overall, there is currently limited evidence-based research to support the use of a PMI model as the primary treatment modality for children with CLP. However, given that the findings were suggestive of success in these studies it would appear that PMI in CLP may be a worthwhile intervention approach for clinicians to consider, as the research presented in this critical review showed that some gains were possible. While PMI for children with CLP would not be recommended as the primary intervention approach at the present time, it bears on the clinician to consider using the principles of the model presented in this clinical review within the clinical setting, given the supportive anecdotal findings as well as the proven success of PMI in other populations with speech and language delays. Given the inherent constraints of conducting studies with this population the research does appear to be persuasive enough to consider using this approach for the possible short-term gains as revealed in the studies presented, with clinician-directed support. Therefore, it is recommended that clinicians cautiously consider the research presented when working with children with CLP and use their intuitive clinical judgment when choosing to use PMI in their clinical practice.

Children with CLP present with a unique set of speech and language difficulties not seen in other populations and which often require extensive therapy. Any support and intervention approaches available to clinicians should be used to provide services to these children. Parent's as interventions is one such approach. Parent involvement as active participants in their child's therapy can and should be encouraged in order to achieve maximal benefits from therapy. In doing so clinicians can help create naturalistic environments which support the development of skills taught in the

clinical setting in order to facilitate gains in a child's speech and language development.

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