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ORIGINAL PAPER

Metacognitive strategies for enhancing language development in children with cleft palate

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Abstract
Background Children born with cleft palate frequently show compensatory articulation errors (CA), and they are also at risk for language delays. There is a need of studies on speech–language intervention in this patient group. The purpose of this paper is to study metacognitive strategies for enhancing language development in children with cleft palate.
Methods Twenty-six children with unilateral cleft lip and palate (UCLP) were studied and divided in two groups. The age of the patients ranged from 5 to 8 years. Language and articulation measurements for evaluating language development were made at the beginning and the end of the study. Both groups were treated with previously reported strategies routinely used for enhancing language. In addition, children from one group (active group) were exposed to metacognitive strategies which have been described as useful for enhancing expert thinking processes, such as think-aloud. For evaluating language development, all children were analyzed using the Situational-Discourse-Semantic Model.
Results The results indicate that children with UCLP and CA benefit from an intervention which also addresses specific aspects of language development. The patients included in the active group in which the metacognitive strategies were used showed a greater improvement as compared with the patients from the control group.
Conclusions Intervention in children with cleft palate and CA should address not only the articulation processes, but also specific aspects of language development. Metacognitive strategies could be an adequate option for enhancing language performance in this patient group.
Level of Evidence: Level III, therapeutic study

Keywords Cleft palate · Language · Speech · Therapy

Introduction

It is well known that children born with cleft palate are at risk for speech disorders. These disorders are usually referred as compensatory articulation (CA) and are secondary to velopharyngeal insufficiency (VPI), residual cleft, or fistula. CA has been considered a phonetic disorder as it may initially occur as a consequence of the cleft [1–3]. However, some authors define this impairment as a phonologic disorder stating that over time, these errors can become incorporated into the child's developing phonological system [4–6]. Since there are indications that the phonological system is integrated with other linguistic skills, some reports have suggested that the language of children with CA should be assessed and considered during intervention [6, 7].

In contrast with speech, at the present time the presence of language disorders in children with cleft palate has received relatively little attention in the literature. It is only recently that language has begun to be consistently considered as an important aspect in children with cleft palate. Hardin-Jones and Chapman [8] reviewed the literature regarding language and cognition in children with cleft palate. They found that there is a high incidence of learning and language disabilities in children with clefts and recommended that all linguistic areas should be

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considered during intervention. They stated that the lack of attention directed toward these areas is probably because the speech characteristics associated with VPI are the most salient problems encountered by clinicians treating these children and thus have taken priority in research for many years.

Rescorla and Dale [9] studied language in late talkers. They reported that the differences in language skills persisted until age 17, manifested as weaknesses in the ability to use high-order language for several functions including the following: narration, definition, explanation, description, and exposition. Thus, early problems with language may be long-lasting and should not be neglected.

In 2000 Pamplona, Ysunza et al. [10] studied the relationship between CA and the child's language system, including the ability to use semantic, syntactic, and discourse elements of language rules to express meaning. In this study, they found that children aged 3–8 years with CA differed in their overall development of language, and not just speech productions, from children with repaired cleft palates that do not show the CA speech patterns. These findings suggest that it would be necessary to provide children with CA with a more global treatment including higher levels of language such as abstract thinking. In contrast, Klinto et al. in 2015 [11] reported that no pattern was seen regarding a relationship between percent correct consonants and ability to retell a narrative in children with unilateral cleft lip and palate (UCLP). However, a larger proportion of children with UCLP than without displayed problems with retelling [11].

The whole language model described by Norris and Hoffman [12, 13] supports the statement that children with articulation deficits should be treated considering not only the peripheral aspects of speech but also higher levels of language organization. The Situational-Discourse-Semantic (SDS) Model was described as a valuable tool for conducting naturalistic observation and descriptive assessment of language development [12, 13]. The SDS Model provides a detailed description of three contexts (situational, discourse, and semantic) in 10 levels of cognitive and linguistic organization.

As mentioned herein, in the related scientific literature, linguistic development has not been considered as an important issue in patients with palatal clefts. Most of the emphasis has been aimed to enhance speech development and treat speech sound disorders. Thus, the use of strategies for enhancing language development in this population has been somehow overlooked. However, language and speech are deeply intertwined, and for a successful outcome, it is essential to address articulation as well as higher levels of linguistic organization. In recent years, several researchers have addressed language development in children with cleft palate at age 5–7 years and stated the need of further investigations in the area [8, 11, 14–17]. Thus, it seems necessary to study the use of strategies aimed to enhance linguistic development in this population.

Throughout the late 1970s and 1980s, research on metacognition focused on understanding the relationship between metacognitive skills and elementary students' classroom performance. Flavell's [18] research on children's memory development underscored the importance of metacognition in influencing behavior. Markman [19] linked comprehension to constructive processing, and she believed it was absent in young children. Two categories of metacognition can be identified: knowledge about cognition and regulation of cognition. Metacognition researchers highlighted the importance of supporting students' planning, monitoring, and self-regulating strategies through interventions to enhance learning. Within the past 20 years, educators have adopted metacognitive strategies in instructional design for students of all ages [20].

The purpose of this paper is to study whether the use of strategies originally designed for improving expert-thinking processes, such as think-aloud, can be effective for speech and language intervention in children with cleft palate.

Material and methods

Participants and assessment procedure

This study was carried out at the Hospital Gea González in Mexico City. The Bioethics Committee and Internal Review Board of the Hospital approved the protocol, and the study was performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki's and its later amendments. Before the inclusion of each child into the study group, the parents or legal guardians were carefully explained about the procedures and the methodology of the protocol. All parents of the children included in the study group, agreed to participate, and gave their informed consent prior to the inclusion in the study.

Sample size was calculated at an Alfa of 95 % confidence interval and a beta power of 80 % for a comparative study of two groups. The distribution of the severity of CA across children treated at the Cleft Palate Clinic of the Hospital Gea Gonzalez during the last 2 years was considered for defining the sample size. The aim was to detect a difference of at least 20 % between language development scores as measured by the SDS Model. According to these calculations, a minimum of 13 children classified in each group should be included in the study.

Children with UCLP and CA were studied in the contexts of storytelling in order to assess their language abilities.

To qualify for the study group, children had to meet the following criteria:

(a) Unilateral, complete cleft lip and palate (UCLP) [21]
(b) No known neurological or genetic syndromes

(c) No identified severe language disorders according to the SDS Model evaluation practiced in our clinic routinely and reported previously [10]
(d) Palatal repair of the UCLP performed according to the surgical routine of the cleft palate clinic. This routine includes the following: surgical repair of the lip and primary palate between 1 and 3 months and surgical repair of the secondary palate between 5 and 8 months with a minimal incision palatopharyngoplasty [22]
(e) VPI after palatal repair demonstrated by perceptual assessment, videonasopharyngoscopy and multi-view videofluoroscopy
(f) CA in association with VPI had to be demonstrated during a complete Speech and Language Pathology evaluation.
(g) Absence of postoperative fistulae
(h) Chronological age between 5 and 8 years of age at the time of selection for the study group
(i) Normal hearing demonstrated by conventional pure-tone audiometry

According to the sample size calculation, the first 26 children who met the inclusion criteria were studied. The age of the children ranged from 5 to 7 years and 11 months. The median age was 6 and 4 months. All children received a complete clinical evaluation of speech, language, and voice. The assessment also included a linguistic analysis during storytelling/retelling. Special attention was focused on the linguistic organization with the SDS Model [12, 13]. For this purpose, children were videotaped interacting with a trained speech-language pathologist (SLP) during storytelling for 30 min. A Go-Pro Hero4, 4K camera was used for video capture; the device included 12.0 MP digital still resolution; Protune; Auto Low Light mode; SuperView; QuikCapture; Wi-Fi; Bluetooth. All recordings were performed in a sound proof room. A 20-min segment was selected where a high level of verbal interaction occurred.

Transcription and analysis of transcriptions

All SLP's participating in this study had been performing transcriptions of cleft palate children for at least 3 years. For assessing the reliability of the language evaluation in the SDS Model, a blind procedure was utilized, whereby all analyses were independently conducted by two trained SLP's (first and second authors of this paper).

The 20 min of interaction were transcribed verbatim by one of the SLP's for analyzing the semantic level of ideas expressed, discourse organization, and the level of situational displacement. The transcribed samples were then randomly assigned to the other SLP who performed an independent analysis. Whenever there was a disagreement, each case was discussed until a consensus was reached.

Each sentence was coded for the level of meaning (semantics) expressed using the criteria established in the SDS Model [12, 13]. The children had to score in the middle levels of the model in order to be able to work with the selected strategies for this study. For that reason, children that scored at the lowest levels were not considered as candidates. In the SDS-model, the level of representation present in the activity is specified in the situational context.

If the child could interpret and talk about the pictures, this was scored as level 4=symbolic representation. If the child could talk about something that happened before but it is not in the actual time anymore such as talking about what he did during the weekend, a level 6 = Descontextualized egocentric was assigned. When the child used language to describe an event from the perspective of an observer, this was scored as level 7 = descontextualized-descentered.

The Semantic context was scored for the level of meaning expressed during storytelling.

The samples were evaluated for the highest level of meaning expressed by the child. In the SDS Model, the first 2 levels are nonverbal responses, and these were not scored. The lowest score achieved by the children in this study, level 4, description, was assigned if the child talked about actions, as in "Go to sleep now," or "The boy is taking a bath." Level 5=Attribution, was assigned if the child described characteristics or emotions, such as "My car is rolling fast" or "The mother is sad." Level 6=Interpretation, was scored if the child made a prediction or mentioned causality or similar insight, as in "I think it is going to crash" or "He is going to sleep because he is very tired". The highest score assigned, level 7, was given if the child made an inference that required combining personal experience or prior knowledge with the action, as in "He is going to be in trouble because he is hiding instead of going upstairs to eat dinner." The child was assigned the semantic level equivalent to the highest produced that occurred with high frequency (i.e., more than five occurrences) during storytelling.

The discourse was scored according to the highest level of organization shown. Levels 1 and 2 (a single discrete action with no continued interest) were not seen by any of the subjects. Level 3 was assigned to organization in list form, as in "There is a mommy, and there is a dog, and he is taking a bath and she is hiding", with no temporal connections.

Level 4 was assigned if temporal connections were made between events, as in "He is taking a bath and now he will dry off. Then the boy brushed his teeth." Level 5 was assigned when causality was established between actions, as in "Dad cooked dinner while mom did the laundry. Dad called everyone to dinner, but the boy did not come because he was hiding. Dad became very angry." Levels 4 and 5 of discourse required temporal or causal links across actions or events, and therefore required the child to play or tell a sequence of at least three

related actions. The child was assigned the discourse level equivalent to the highest produced during storytelling.

The coding resulted in one number assigned for situation, one for discourse, and one for semantics for each child. The first coding was done by one of three randomly assigned examiners. A second researcher also coded the samples for all three measures. Inter-coder agreement calculated with Cohen's Kappa was 0.95.

Intervention

The children with UCLP were randomly divided into two groups. Children assigned to the active group were matched by gender with patients included in the control group. The age range of the children from both groups was kept as similar as possible. The linguistic levels of all children were evaluated with the SDS Model as described previously [10, 12, 13].

Both groups received 20 sessions of speech therapy aimed to stimulate language and correct compensatory articulation according to the principles of the Whole Language Model [12, 13].

All encounters were 45-min sessions, and they were provided twice per week on weekdays. The total period of intervention was 10 weeks.

Intervention was aimed to reinforce correct speech sounds while enhancing cognitive-linguistic organization. For language, the object of our study, the treatment goals were set depending on the SDS Model levels of language of each group. Within these main principles of intervention, strategies described for language intervention such as modeling, expansions, and/or cloze procedure were applied in both groups. In addition, only in the case of the active group, the SLP used metacognitive strategies described for reading comprehension and for developing expert-thinking processes in addition to the strategies mentioned before [24]. Reading and storytelling were the main context for intervention. In both groups, strategies for language and articulation were used before, during, and after the storybook reading. The children were invited to reflect and discuss about the content of the book. One of the most common metacognitive strategies is "think-aloud" [23]. During "think-aloud," the clinician verbalizes thoughts aloud while reading or talking, thus modeling the process of comprehension and what children do before, during, and after reading a selection for using expert thinking processes. The idea is to teach explicitly the use of the strategies. Oczkus [23] developed a practical way to teach and think about the strategies using symbols for each one for helping children to understand and remember them. We used this methodology to enhance learning and using the strategies.

The following metacognitive strategies were explicitly taught to children included in the active through "think-aloud":

Make connections When we hear/read, we naturally make connections to our own experiences and knowledge. This knowledge helps us relate to the characters or setting and that improves our understanding of the topic helping us to build meaning (example, "This reminds me of_____ because_____.").

Predicting Skill readers/language users use their connections to help them predict what will happen next. They also use what they know about the text structure for predicting (example, "I think _____will happen next because_____".).

Inferring It has been stated that readers that infer are the best readers [25]. Norris and Hoffman [12, 13] mention that inferring requires combining personal experience or prior knowledge with the content of the narration. Inferring includes thinking in information not explicit in the context or text. An example could be "I can say that_____ and it makes me think _____".

Questioning Question deepens comprehension. For comprehending, skill listeners/readers make questions, before, during, and after the reading. These ensure the validity of future thoughts and guides comprehension.

Summarizing Oczkus [23] state that this skill includes a host of challenging tasks, including recalling important events or details, ordering points, and using synonyms or selected vocabulary. Summarizing supports language learning process to restore information addressing the most important ideas and

Table 1 Comparison of situational context levels (SDS Model) at the onset of the intervention

Patient	Active-group 2	Control-group 1
1	4	4
2	4	4
3	4	4
4	4	4
5	4	4
6	4	4
7	4	3
8	4	3
9	7	5
10	6	5
11	7	5
12	6	6
13	6	6

Table 2 Comparison of discourse context levels (SDS Model) at the onset of the intervention

Patient	Active-group 2	Control-group 1
1	5	3
2	3	4
3	4	4
4	4	4
5	4	4
6	4	4
7	5	4
8	4	4
9	6	5
10	5	5
11	5	6
12	5	6
13	6	3

relationships between them (example, "First_____ Then _____ at the end_____".).

Evaluating Good language users/readers get to be judges (author, character, or text). Evaluating involves determining importance and encourages reflection (example, "I agree-disagree because_____".).

Statistical analyses

To determine if the groups were equivalent, Student's *t* tests were run for the variable of chronological age. At the end of the intervention, a Wilcoxon signed-rank test was performed to compare the levels of progress of both groups according to the three contexts of the SDS Model [12, 13].

Table 3 Comparison of semantic context levels (SDS Model) at the onset of the intervention

Patient	Active-group 2	Control-group 1
1	4	4
2	4	4
3	4	4
4	4	4
5	4	4
6	4	4
7	4	4
8	4	4
9	6	6
10	6	6
11	6	6
12	6	5
13	6	5

Table 4 Comparison of situational context levels (SDS Model) after the intervention

Patient	Active group 2	Control group 1
1	7	5
2	6	5
3	7	5
4	7	6
5	7	4
6	7	5
7	6	5
8	7	4
9	8	6
10	8	7
11	8	7
12	8	7
13	8	6

A Mann Whitney *U* test was used in order to compare the distribution of language levels between groups at the onset and at the end of the intervention period.

For all statistical analyses, $p<0.05$ (two-tailed) was considered to indicate significant differences.

Results

A student's *t* test demonstrated a non-significant difference between ages in both groups.

At the onset of the follow-up period, the distribution of language levels, as assessed by the SDS Model was similar in both groups of patients. All children were present with at least one level delay.

Table 5 Comparison of discourse context levels (SDS Model) after the intervention

Patient	Active-group 2	Control-group 1
1	6	5
2	6	6
3	6	5
4	6	6
5	6	5
6	6	5
7	6	6
8	6	4
9	7	6
10	8	6
11	7	8
12	6	6
13	8	5

Table 6 Comparison of semantic context levels (SDS Model) after the intervention

Patient	Active-group 2	Control-group 1
1	7	7
2	7	7
3	7	5
4	7	6
5	7	5
6	7	7
7	7	6
8	7	5
9	8	7
10	8	7
11	8	7
12	8	5
13	8	5

In Tables 1, 2, 3, 4, 5, and 6 results of children of both groups at onset and after the intervention are displayed.

There were non-significant differences in the distribution of language levels between groups at the onset of the intervention period ($P>0.05$) (Tables 1, 2, and 3).

There was a significant improvement ($P<0.05$) in the levels of linguistic development after the intervention in both groups, involving the three contexts of the SDS Model. In other words, all children showed significant improvement after the intervention, regardless of the different methods and strategies used for each group.

The levels of linguistic development in children with cleft palate after the intervention showed significant differences ($P<0.05$) between groups in the three contexts of the SDS Model (Table 7); children of the active group showed greater improvements as compared to the control group.

Discussion

The research was conducted with children with UCLP present with residual VPI and CA. Both groups were homogeneous in age and linguistic levels at the onset of the study; hence, they were comparable. The purpose of this paper was to study if metacognitive strategies described to enhance reading comprehension could have an effect in language development. It was hypothesized that children with UCLP with speech disorders could benefit and improve their linguistic performance with the use of these strategies because they enhance comprehension and the use of higher language processing. The results of this study seem to support this hypothesis.

Intervention followed whole language model principles [10, 12, 13], and the main context for intervention was storybook reading in addition to the strategies for enhancing language regularly used in our center; the active group was introduced with metacognitive strategies (activating prior knowledge, anticipate, clarify, question, making connections, summarize and evaluate), designed for enhancing reading comprehension [23]. Expert readers use these strategies [24, 25], and they may be used as well by any language user for comprehending or organizing a message. The results of this study support this statement since children with cleft palate who learned to use these strategies improved their language levels to a higher degree than the children who received traditional intervention. They were able to improve their level of abstraction according to SDS Model [12, 13]. The use of the strategies facilitated the use of inferences, evaluations, connections, and helped children to analyze the story and other real situations in more complex ways. They were more able to incorporate details in their narrations and took different perspectives when analyzing the plot of the story. This could indicate that metacognitive strategies may be useful for enhancing language development and this could be reflected in both, written and oral language. An interesting finding is that when the metacognitive strategies were used with children from the active group, they used emissions with a higher level of complexity and abstraction.

It has been described that children with cleft palate are at risk for language disabilities in addition to speech disorders [8]. There is a growing body of literature regarding language disabilities [9]. Studies have shown that these children in comparison with traditional developing children differed

Table 7 Statistical analyses

Pre-intervention			Post-intervention		
SDS levels	Active-group 2	Control-group 1	*p*	Active-group 2	Control-group 1
Situational	4.92 (1.26)	4.38 (0.96)	0.947	7.23 (0.73)	5.54 (1.05) 0.001
Discourse	4.62 (0.87)	4.31 (0.95)	0.953	6.46 (0.78)	5.62 (0.96) 0.011
Semantic	4.77 (1.01)	4.62 (0.87)	0.265	7.38 (0.51)	6.08 (0.95) 0.001

S-D-S mean (and standard deviation) levels of the three contexts (situational, discourse, semantic) in cleft palate patients included in the experimental and control group are displayed. The levels at the onset and at the end of the intervention were compared. Intervention between groups compared with Mann–Whitney *U* test

significantly and substantially in their receptive and expressive language skills. Rescorla [9] who studied language in late talkers, indicated that these differences persisted until age 17, suggesting that it is not a temporary delay even when at first it was assumed that late talkers were able to catch up to normative expectations in vocabulary and grammar by the time they entered kindergarten, but they continued to show weaknesses relative to traditionally developing peers in the ability to use complex, higher-order language for narration, definition, explanation, description, exposition, and reading [9–15]. This was also found by Scarborough and Dobrich's [26]. For this reason, it is important to address the language disorder in children with cleft palate, and the metacognitive strategies used in this study seems to be an option for enhancing language development in these population.

Moreover, Hoff [27] reported that children from families with low social and economic status have lower language skills in a variety of linguistic domains including vocabulary, grammar, narrative and phonological development, and speed of processing. For instance, the proportion of children living in poverty whose language development reflects influence of socioeconomic status is likely to be greater than 22 %. In our center, many children are from low socioeconomic status, and this implies a higher risk for language problems.

The strict inclusion criteria used for recruiting children for this study resulted in a reduced number of patients who were finally included in the study groups for this paper. The reduced number of children assessed herein does not allow drawing definite conclusions. Also, there may be other explanations for the increased advance in the group of children in which the metacognitive strategies were used. Further studies on speech and language intervention for children with cleft palate are necessary.

Conclusion

Treating only the compensatory articulation errors of children with cleft palate does not seem enough to promote speech and language development. Also, higher levels of language including abstract thought should be considered. In these contexts, phonologic information is integrated with all linguistic areas within communicative purposes and situations. Thus, the production of speech sounds is not the only goal. Rather, the goal is to become effective communicators and expert language users. Higher levels of thinking and language in children with cleft palate may also impact other domains on their life as school performance and social relations.

Intervention in children with cleft palate with CA should address not only the articulation processes but also specific aspects of language development. Metacognitive strategies could be an adequate option for enhancing language performance in this population.

Conflict of interest Maria Del Carmen Pamplona, Silvia Carolina Solis, Pablo Antonio Ysunza, and Santiago Morales declare that they have no conflict of interest.

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Ethical standards This project was approved by the Internal Review Board of Hospital Gea Gonzalez in Mexico City. All procedures for this study were approved by the appropriate ethics committee and have therefore been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments. As mentioned in the manuscript, all persons gave their informed consent prior to their inclusion in the study. Moreover, details that might disclose the identity of the subjects under study were omitted.

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