

Global and communicative development skills in preschool children with cleft lip and palate

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ABSTRACT

Importance: Cleft lip and palate (CLP) is globally among the most common childhood malformations. This disorder impacts childhood development, including speech and language, and affects children worldwide.

Objective: To analyze child development skills (adaptive fine motor, gross motor, personal-social, and language) in preschool children with isolated CLP compared with children without this malformation.

Methods: The participants included an experimental group of 27 children with isolated CLP and a comparison group of 27 children without CLP aged between 48 and 59 months. The groups were evaluated using two instruments: the Denver Developmental Screening Test II (DDST-II) and the Avaliação do Desenvolvimento da Linguagem (ADL-Language Development Assessment). Data were analyzed by descriptive and inductive analyses, using the Student's *t*-test and the Mann-Whitney test, at a significance level of $P < 0.05$.

Results: All children in the comparison group performed within normal standards for their age range in the DDST-II and the ADL. The worst performance in the experimental group was observed in language skills, followed, in declining order, by adaptive fine motor, personal-social, and gross motor as measured by the DDST-II. Children with isolated CLP also performed poorly in receptive, expressive, and global language in the ADL. No statistically significant differences were observed in the experimental group's scores for the ADL and the DDST-II.

Interpretation: Developmental skill levels were below expectations for children of this age with isolated CLP.

KEYWORDS

Cleft lip and palate, Language, Child, Preschool

INTRODUCTION

Cleft lip and palate (CLP) is often related to communication disorders.¹ Speech impediments are related to the anatomical involvement of the structures of oral

production, velopharyngeal dysfunction, and dentofacial deformity, such as alterations in articulatory production and speech hypernasality.^{2,3}

Research suggests CLP is a risk factor for child development,

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with possible damage including that to social, emotional,⁴ motor, and cognitive skills.⁵ It affects quality of life and is associated with language disorders, even in cases of CLP without comorbidities or genetic syndromes.^{3,6-9}

The alterations in speech development of children with CLP are associated with damage to middle-ear functioning and is the primary cause of impaired cognitive development. This is because of insufficient stimulus or may be a consequence of speech disorders.¹⁰⁻¹²

Studies have demonstrated the significance of language disorders, separate from other risk factors, in other with CLP. The evidence suggests a delay in learning first words,¹³⁻¹⁵ a below-average achievement in receptive and expressive skills,^{9,13} and changes in vocabulary¹⁵⁻¹⁷ and syntax.¹⁸ However, other studies have indicated that language development is not different in children with or without CLP, with each population experiencing a delay in language acquisition.¹⁹

This study compared the child development skills (adaptive fine motor, gross motor, personal-social, and language) of preschool children with isolated CLP with those of children without malformation. We hypothesized that preschool children with CLP experience damage to their communicative and global performances. The characterization of these skills is necessary since research findings of language and development in children with clefts are controversial.

METHODS

Ethical approval

The Institutional Review Board approved this study at the university under report n.1.959.530 (CAAE: 63863917.9.0000.5417) and the hospital where the study was conducted under report n.1.994.369 (CAAE: 63863917.9.3001.5441). Data were collected in the Palatal Prosthesis sector. The written consent form was obtained from all participants.

Study population

The participants were divided into two groups: (1) the experimental group (EG), composed of 27 children with non-syndromic repaired complete unilateral CLP, assisted at the Hospital for Rehabilitation of Craniofacial Anomalies of the University of São Paulo (HRAC-USP), Bauru, São Paulo, Brazil, and (2) the comparison group (CG) comprising 27 children from the community who spontaneously responded to the request for participants.

The inclusion criteria for the CG were: 48 to 59 months old, of either sex, and no history of CLP or atypical development. Individuals in the EG had been submitted to primary palate repair, according to international protocols, before 2 years of age.²⁰ Individuals in the hospitalization

routine for surgical reasons or medical complications were excluded from the EG. Children with a history of middle ear disease or diagnosis of genetic syndromes or other malformations were excluded from both EG and CG.

Procedures

Both groups were analyzed using two different instruments, which were administered directly to the child and collected by a single evaluator.

*Denver Developmental Screening Test II (DDST-II)*²¹

As described by Frankenburg et al,²¹ screening has been used worldwide and standardized in more than a dozen countries (Brazilian Portuguese included). Health professionals apply it to children from birth to 6 years of age. It consists of 125 items, distributed in four areas of development: Personal-social (PS), Language (LGG), Adaptive fine motor (AFM) and Gross motor (GM), measuring the risks of delaying neuropsychomotor development. Its application consists of direct skills testing, behavioral observation, and information reported by caregivers.

The results were interpreted according to the DDST-II guideline criteria (pass or fail in the screening) and analyzed to assess whether each developmental skill the child displayed was in line with expected skill levels for their age

*Avaliação do Desenvolvimento da Linguagem (ADL-Language Development Assessment)*²²

This scale was standardized for Brazilian Portuguese speaking children by Menezes,²² who identified changes in the learning and development of oral language and the different etiologies of specific language changes in children aged 1 to 6 years and 11 months. It was used to evaluate receptive and expressive oral language (in tasks related to language domains: phonology, morphology, syntax, semantics, and pragmatics). The sample age range tasks were grouped in concepts such as quantity, quality (adjectives), spatial and temporal relationship, and sequence. Compensatory articulations (CA) were not considered, as suggested in the instrument guidelines.²²

For our analysis, 1 point was assigned to each correct response and 0 to each error or missing response. The results were classified according to the standard score: 115–85 corresponded to the normal standard; 84–77 to mild disorder; 76–70 to moderate disorder; and equal to or below 69 to severe disorder.

Data analysis

The categorical variables were described in number and percentages, and the continuous variables were expressed as mean \pm standard deviation. Group comparisons used a significance level of $P < 0.05$. The Fisher test was used to assess categorical variables, and the Mann-Whitney test

was used for continuous variables. The Student’s *t*-test was used for comparing means and the Spearman test for data correlations.

RESULTS

This study included a convenience sample of 54 children who were regularly registered at a specialized referral hospital. The EG group comprised 15 boys (56%) and 12 girls (44%), with a mean age of 53.0 ± 3.5 months. The CG group comprised 9 boys (33%) and 18 girls (67%), with a mean age of 53.0 ± 4.0 months and matched socioeconomic and educational levels.

All children in the CG performed within normal standards for their age range according to the DDST-II, taking into account the pass-fail criterion (the child shows no “delay” and, some children presented only one error, which for the test means “attention to development”). In the EG, 17 children (63%) failed and were classified as having delayed development; 10 children (37%) passed, highlighting the difference in general performances between the EG and CG ($P = 0.0001$). In the EG, language represented the most disorders (48%, 13 children), followed by adaptive fine motor skills (29%, 8 children), personal-social skills (25%, 7 children), and gross motor skills (11%, 3 children). Performance was lower for all four domains in the EG. The mean performance of the EG in all skills measured by the DDST-II was below that expected for the age range (Table 1), with a statistically significant difference between groups in language, adaptive fine motor, and personal-social skills.

The CG children’s performance in the ADL was 100% normal. In the EG, however, 15% of the children performed below expectations, with the difference appearing between the means of groups (EG 104.26 ± 15.08 vs. CG 114.56 ± 5.21 , $P = 0.001$). Table 2 shows the outcomes of the ADL test. Again, the EG’s performance was lower for receptive language, expressive language, and global score compared with the CG.

No statistically significant differences were observed in the EG’s scores between the DDST-II and the ADL (Table 3). In addition, 13 of the 17 children who failed the DDST-II were classified within the normal standards according to the ADL, suggesting that the DDST-II instrument was more sensitive to detect changes in the study group than the ADL.

DISCUSSION

Owing to frequent speech disorders in children with CLP caused by velopharyngeal dysfunction, few studies focus on the performance of linguistic skills in this population. The focus of this study is on the possible hearing and stimulus risks¹⁰⁻¹² that may affect receptive and expressive language, which can worsen speech disorders caused by the anatomical and functional conditions in these children.^{2,3}

TABLE 1 Comparison of skills assessed by the DDST-II between the experimental group and comparison group

Items	Experimental group	Comparison group	Mean difference (95% CI)	P
PS	50.33 ± 7.15	53.78 ± 4.01	-3.45 (-6.61, -0.28)	0.03
AFM	50.89 ± 5.28	53.78 ± 4.01	-2.89 (-5.45, -0.33)	0.03
LGG	50.89 ± 3.89	53.78 ± 4.01	-2.89 (-5.03, -0.75)	0.01
GM	52.19 ± 4.13	53.78 ± 4.01	-1.59 (-3.82, 0.63)	0.16

DDST-II, Denver Developmental Screening Test II; PS, personal-social; LGG, language; AFM, adaptive fine motor; GM, gross motor; CI, confidence interval.

TABLE 2 Comparison of skills assessed by ADL between the experimental group and the comparison group

Items	Experimental group	Comparison group	Mean difference (95% CI)	P
Receptive	101.33 ± 13.26	109.41 ± 4.81	-8.08 (-13.52, -2.63)	0.004
Expressive	106.44 ± 15.93	116.44 ± 6.85	-10.00 (-16.70, -3.30)	0.004
Global	104.26 ± 15.08	114.56 ± 5.21	-10.30 (-5.03, -0.75)	0.001

ADL, Language Development Assessment; CI, confidence interval.

TABLE 3 Comparison of performance of the experimental group in ADL and DDST-II

Related test	Category/Classification	DDST-II		P
		Pass	Fail	
ADL classification	Normality	10	13	0.33
	Mild disorder	0	1	
	Moderate disorder	0	2	
	Severe disorder	0	1	

ADL, Language Development Assessment; DDST-II, Denver Developmental Screening Test II.

The present study’s hypothesis was supported: children with CLP aged 48 to 59 months (4 years of age) evidenced damage in their communicative and global performances compared with children without CLP. These findings are consistent with research that suggests CLP’s presence is a factor that causes a delay in overall child development,^{4,5} including speech and language disorders.^{3,9,13-18}

Our sample of children with CLP showed delayed development: 63% failed in the DDST-II, performed poorly in the skills analyzed (personal-social: $P = 0.03$; adaptive fine motor: $P = 0.03$; language: $P = 0.01$), and displayed a significant language deficit. These findings are consistent with prior research, which reports that the failure rate of 5-year-old children with CLP in the DDST-II is indicative of poor language performance.²³ Another study using the Kent Infant Developmental Scale and the Minnesota Child Development Inventory instruments on 36-month-old children also indicated poor performance in

language and adaptive fine motor skills.²⁴

In the ADL, which assessed receptive and expressive language and the overall score, 15% of the EG showed more alterations compared with the CG. This corroborates studies that have reported damage to receptive and expressive language skills in this population.^{9,25}

A comparison of the children's performances revealed a more significant incidence of language disorders (48%) when evaluated by the DDST-II, compared with the ADL (15%). The complexity of language requires the application of various tools to investigate all expressive and receptive language levels, to ensure that the combination of these results can provide the diagnostic conclusion for language as early as possible.^{10,26}

Although the findings are evidence of a possible language disorder in children with CLP, some studies have reported similar findings in children with or without CLP.²⁰ This demonstrates the importance of conducting more research on children with CLP. The aim should be to establish language evaluation protocols and to take into account all linguistic aspects that will contribute to the rehabilitation of these children.

There are still a few studies that used the DDST-II, which is an instrument for developmental screening in children.²³⁻²⁵ For the present study, this instrument met our goal of identifying children at risk. However, future studies should consider using other global development instruments to compare global aspects and specific language development to better understand children with CLP.

Parents and caregivers of children with CLP are often concerned about the child's communication skills, but have difficulty expressing their concerns about speech/language aspects.²⁷ It is therefore important to raise awareness of the wider effects of CLP.²⁸ Early interventions are performed by speech therapists trained to identify and treat speech and language disorders. A multidisciplinary approach is important because the age at which children receive primary palate surgical repair (approximately 12 months) is associated with better speech outcomes.^{20,29,30} This can positively influence language development.

In conclusion, child development skills were found to be lower than expected in children with CLP, with a significant deficit in receptive and expressive language.

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CONFLICT OF INTEREST

None.

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