Occurrence of consonant production errors in liquid phonemes in children with operated cleft lip and palate

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ABSTRACT

Information about the prevalence of consonant production errors, including compensatory articulations (CA), in individuals with cleft lip and palate (CLP) who speak Brazilian Portuguese is limited, particularly regarding liquid sounds. The literature primarily reports the occurrence of CA for plosive and fricative sounds, since occurrence of CAs in sounds that require higher amounts of oral air pressure is expected. While the use of CA during liquid sound production is not expected, clinical experience suggests that individuals with CLP present with inadequate backing, elevation, and anteriorization of the tongue as well as tongue clicks during production of /r/ and /l/. Objectives: Describe the occurrence of consonant error productions during liquid sounds for children with CLP; compare the occurrence between children operated with the Furlow and von Langenbeck techniques for palatoplasty; and compare the occurrence between children operated between 9-12 months and 15-18 months of age at primary palatoplasty. Material and Methods: A sample of 397 children (237 males and 160 females) with operated unilateral CLP was studied. In this group, 163 underwent palatoplasty with the Furlow procedure and 234 with the modified von Langenbeck procedure. Age at palatoplasty was between 9 and 12 months for 189 children and between 15 and 18 months for 208 children. Data about production of /l/, /r/, /R/, /λ/ and consonant clusters /l/ and /r/ were obtained from speech pathology records. Speech pathologists registered the speech findings after an auditory-perceptual evaluation of the participants at the sixth year of age. Results: The use of middorsum palatal place (MDP) of production was identified for 2% of the sample. Tongue anteriorization of the /l/ production was observed for 55% of the children. No significant difference was found related to surgical technique, but children operated earlier developed the use of the consonant cluster /r/ sooner than children operated later (p=0.040). Conclusion: We found a low occurrence of use of cleft related CA during attempts of production of liquid phonemes, and the variable age at primary palatoplasty significantly interfered with the acquisition of consonant cluster /r/.

Key words: Articulation disorders. Cleft lip and palate. Liquid phonemes. Speech
INTRODUCTION

Cleft lip and palate (CLP) is a common congenital anomaly, occurring at a frequency between 1:500 and 1:700 births\(^{17}\). According to Pegoraro-Krook, et al.\(^{15}\) (2004), when cleft affects only the lip and premaxilla (CL), usually there is no severe speech disorders. In CL, when speech disorders are present, intelligibility is affected by sound distortions resulting from dento-occlusal conditions or by lip retraction due to surgical scars. When cleft involves the palate, either as an isolated cleft palate (CP) or cleft lip and palate (CLP), oral communication can be severely affected by speech and hearing disorders, with implications for nutrition and psychosocial and educational development as well as facial aesthetics. The communication disorders associated with CP may be due to several factors: (1) unoperated cleft, (2) velopharyngeal dysfunction (VPD) resulting from the failure of the primary surgery to establish a functional velopharyngeal mechanism, (3) dento-occlusal conditions, and (4) hearing loss due to associated chronic otitis media, among other factors.

Surgical repair of the palate aims to correct the cleft with minimum compromise to facial growth while establishing anatomic and functional conditions for the velopharyngeal mechanism to function adequately, preventing the development of VPD and associated speech disorder\(^{12,21}\). Surgical treatment and its outcome, however, can severely impact the growth of the middle third of the face and the development of speech and hearing\(^{12,15}\). Authors suggest that the technique used for primary palatoplasty and the age of the child at palatoplasty have a relationship with the occurrence of speech and hearing disorders in children with CLP or VPD\(^{13-18}\). That is, according to the literature, some children are at higher risk of developing speech disorders due to the type of surgical correction they received for CLP.

The production of speech requires coordinated movements of the lips, tongue, jaw, and palate, together with the activity of the vocal folds and respiratory muscles\(^{3,11,20}\). The expiratory airflow is modified in the oral cavity, allowing the establishment of two classes of sounds: vowels and consonants. Vowels are produced with a relatively open vocal tract and no significant constriction to the airstream. Production of consonants involves complete or partial obstruction of air in the oral cavity\(^{1,20}\). More specifically, the production of consonants requires contact between articulators at a specific area in the vocal tract. The place where the airstream is manipulated (place of production) has an impact on the type of sound that is generated. The sounds /p/, /b/ and /m/, for example, are produced with contact between the upper and lower lips, named bilabial sounds due to their place of production. The /f/ and /v/ are produced with approximation of the upper teeth and lower lip, named labiodental sounds. Sounds made with the tip or blade of the tongue are named coronal when the tongue articulates with the alveolar ridge (such as /s/, /z/, /l/ and /r/), and are named linguodental (such as /t/, /d/ and /n/), when the tip of the tongue articulates with the teeth. Production of the velar sounds such as /k/, /g/ and /R/ involves contact or approximation of the back of the tongue to the back of the hard palate, while production of palatal sounds /ʃ/, /ʒ/, /ŋ/ and /ʌ/ involves contact of the middle part of the tongue and hard palate\(^{3,20}\).

The consonants are also classified according to the manner of articulation, which refers to the type of constriction resulting from the articulatory contact. Thus, plosives sounds involve complete obstruction of the airstream followed by a sudden release that is heard as a plosion. Plosive sounds include /p/, /b/, /k/, /g/ and /R/. Nasal sounds are the only ones produced with acoustic energy being displaced simultaneously to oral and nasal cavities, including /m/, /n/ and /ŋ/. Nasally produced consonants are those involving a close approximation of the articulators, leading to an audible friction of the airstream, such as /l/, /ɾ/, /ʃ/, /ʒ/ and /ʒ/. Affricate sounds involve both an obstruction followed by a constricted release of the airstream, such as /tʃ/ and /dʒ/. Nasal sounds are the only ones produced with acoustic energy being displaced simultaneously to oral and nasal cavities, including /m/, /n/ and /ŋ/. Finally, a phoneme is called liquid when it is produced with a much wider passage of air compared to the narrow constriction observed for the fricatives. Liquid sounds include /l/, /ɾ/, /ʃ/, /ʒ/ and /ʒ/. Affricate sounds involve both an obstruction followed by a constricted release of the airstream, such as /tʃ/ and /dʒ/.

The word literature suggests that individuals with CP and/or VPD often have difficulties producing consonants that require a high amount of intraoral air pressure, as plosives, fricatives, and affricates\(^{2,17,23}\). According to Warren\(^{24}\) (2004), the inability to generate and/or maintain adequate levels of intraoral pressure for production of plosion or friction may lead to the development of atypical places of production, called compensatory articulations (CAs). As described by Trost-Cardamone\(^{23}\) (2004), when a CA is used, the manner is usually preserved, while the place of production is atypically posteriorized. Atypical place of production used for CA may involve the use of the pharynx (such as in the pharyngeal fricatives, posterior nasal fricatives, or pharyngeal plosives), the use of the glottis or larynx (such as in the glottal stops or laryngeal fricatives), or even oral but backed places of production (such as velar
The use of middorsum palatal stop (MDPS) is described in literature for the plosives /t/, /d/, /k/ and /g/. The presence of VPD, or a palatal fistula, can explain some adjustment in tongue placement during speech, leading to compensations like the MDPS, for example. When a crossbite is present, for example, the jaw may be protruded beyond the maxilla and the tongue and may be inadequately anteriorized or even crowded backward, depending on how the tongue adjusts to the remaining oral space. In some cases, when the tongue protrudes beyond the limits of the upper jaw, the use of the tongue dorsum (instead of the tip) in contact with the middle of the palate is used in substitution for liquid sounds, a production called middorsum palatal stop (MDPS). That is, when the place and manner of articulation the cause of the compensatory adjustment, both MDPS and MDP can be observed.

While MDPS is used in substitution for plosive sounds and the plosion characteristic is present, MDP is used in substitution for liquid sound that does not involve high pressure, so plosion is absent. Articles describing the use of MDP place of articulation for liquid sounds were not found. Nonetheless, our clinical experience suggests that patients with CLP can make articulatory adjustments involving inadequate backing, elevation, protrusion, or even clicks of the tongue, particularly during production of /r/ and /l/. Besides backing of liquid oral targets, developmental articulatory errors (phonological in nature) and significant delays in speech sound development are also reported in the literature for children with CLP. Additionally, consonant cluster reductions and simplifications of liquid phonemes were described for children with cleft who have normal expressive language. Information on prevalence of consonant production errors in Brazilian Portuguese speakers, however, was not found. Consonant clusters /r/ and /l/ in contact with the middle of the hard palate (instead of the alveolar region) can be observed.

MATERIAL AND METHODS

This retrospective study was approved by the Ethics Committee of the Hospital for Rehabilitation of Craniofacial Anomalies at the University of São Paulo (HRAC/USP). The population of interest for this study consisted of 460 children of both genders who presented with unilateral complete operated cleft lip and palate not associated with syndrome or other anomalies that could impair speech, language, and hearing. A retrospective review of the healthcare records of the referred children was done with special attention for the evaluation conducted by the speech-language pathologist (SLP) at the sixth year of life. At the sixth year of life, Brazilian children were expected to have completed phonological acquisition of speech sounds and to be compliant to speech assessment. Evaluation of consonant production was performed by six SLPs during repetitions of words and phrases, and these data was obtained from the healthcare records of the participants. The SLPs’ rated both, manner and place of production of liquid consonants produced by children speaking the Brazilian Portuguese language. The protocol for auditory-perceptual ratings included observations of productions of consonants /l/, /r/, /l/, /r/ and consonant clusters /r/ and /l/. An adaptation of Henningsson, et al. (2008) recommendations for reporting consonant production errors was used for this study. These authors suggested categorizing consonant production errors into the following rating categories: a) within normal limits, b) abnormal backing of oral targets to post uvular places, c) abnormal backing of oral
targets but place remains oral, d) nasal fricatives, e) nasal consonant for oral pressure consonants, f) nasalized voiced oral pressure consonants, g) weak oral pressures, h) other oral misarticulations (including errors related to dentofacial and oral structure deviations), i) developmental errors. While Henningsson, et al. (2008) reported mid dorsum palatal stops and mid dorsum palatal fricatives/affricates as “abnormal backing of oral targets but place remains oral”, in this study this parameter was also used to report abnormal pre-uvular backing of liquid targets (middorsum palatal liquids). Furthermore, this study included observation of tongue anteriorization (beyond incisors) and sound distortions. Instead of an overall developmental error category, this study reported delay in acquisition of liquids and simple substitutions during liquid targets.

Data were recorded in an Excel table with frequency of occurrence of consonant production errors during liquid sounds calculated in percentages. Mann-Whitney test was used to verify whether the difference in occurrence of sound production errors was significant (p<0.050) for the groups distributed according to palatoplasty technique and according to age at palatoplasty.

RESULTS

According to the protocol followed at the research site the primary palatoplasty for the participants included in this study was conducted either with the modified von Langenbeck technique (VL) or the Furlow technique (FW). Age at primary palatoplasty varied between 9-12 months (early closure) and 15-18 months (late closure). Out of the 460 children of interest for this study, only 397 (66%) had speech evaluation completed at the sixth year of life. Therefore, data reported in this study was retrieved from 397 speech evaluations obtained from 237 males (60%) and 160 females (40%). A total of 163 children underwent palatoplasty with the FW procedure (41%) and 234 with the VL procedure (59%).

While 189 children (48%) received early primary palatoplasty (9-12 months), 208 children (52%) received late primary palatoplasty (15-18 months). No secondary surgeries had been performed by the time the speech evaluations reported in this study were conducted (sixth year of life).

As indicated in Table 1, liquid sound productions within normal limits were observed for most of the children at the sixth year of age (average=71%). Consonant production errors observed included: (a) mid dorsum palatal liquids (2%); (b) tongue anteriorization (14%); sound distortions (2%); delay in acquisition of liquids (4%); and simple substitutions (6%).

When looking only into the records which reported production errors for liquid consonants, we observed that tongue anteriorization during /l/ was the most common finding (55%), followed by tongue anteriorization during /r/ and /λ/ (13%). When used, mid dorsum palatal place of articulation for liquid sounds was verified during /l/ (5%), /r/ (3%), /λ/ (1%) and consonant clusters (1%).

Percentage occurrence of production errors during liquid sounds was recalculated after distributing the samples according to palatoplasty technique (Table 2) and according to age at primary palatoplasty (Table 3). The differences observed between the groups distributed according to the surgical technique for palatoplasty (FW and VL) were no greater than 5% and were not statistically significant (p>0.050). The differences observed between groups distributed according to the age at primary palatoplasty (early and late) were statistically significant only for consonant cluster /r/ (p=0.04). In this case, the group that underwent palatoplasty early (between 9 and 12 months of age), showed a significantly lower occurrence of delay in the acquisition of the sound than the group that underwent palatoplasty later (between 15 and 18 months of age).

### Table 1- Distribution of percentage occurrence of consonant production errors during liquid targets

| Sound Types | Adequate | Mid dorsum palatal | Tongue anteriorization | Distortion | Late acquisition | Substitution |
|-------------|----------|---------------------|------------------------|------------|-----------------|-------------|
| /l/         | 33%      | 5%                  | 55%                    | 1%         | 0%              | 6%          |
| /r/         | 68%      | 3%                  | 13%                    | 5%         | 1%              | 10%         |
| /R/         | 91%      | 0%                  | 0%                     | 1%         | 1%              | 7%          |
| /λ/         | 78%      | 1%                  | 13%                    | 1%         | 0%              | 7%          |
| Cluster /r/| 82%      | 1%                  | 1%                     | 2%         | 11%             | 3%          |
| Cluster /l/| 74%      | 1%                  | 2%                     | 2%         | 11%             | 10%         |
| Average     | 71%      | 2%                  | 14%                    | 2%         | 4%              | 6%          |
Table 2- Distribution of percentage occurrence of consonant production errors during liquid targets for data distributed according to surgical technique (Furlow versus von Langenbeck) for palatal repair

|                  | /l/ | /r/ | /R/ | /l/ | Cluster /l/ | Cluster /r/ | Average |
|------------------|-----|-----|-----|-----|-------------|-------------|---------|
| No alteration    |     |     |     |     |             |             |         |
| Fw               | 32% | 71% | 92% | 78% | 84%         | 72%         | 72%     |
| VL               | 34% | 66% | 91% | 78% | 81%         | 74%         | 71%     |
| Difference       | 2%  | 5%  | 1%  | 0%  | 3%          | 2%          | 2%      |
| Mid dorsum       |     |     |     |     |             |             |         |
| Fw               | 5%  | 3%  | 0%  | 1%  | 1%          | 1%          | 2%      |
| VL               | 0%  | 3%  | 0%  | 1%  | 0%          | 0%          | 1%      |
| Difference       | 5%  | 3%  | 0%  | 0%  | 1%          | 1%          | 2%      |
| Palatal          |     |     |     |     |             |             |         |
| Fw               | 5%  | 3%  | 0%  | 0%  | 1%          | 1%          | 2%      |
| VL               | 0%  | 3%  | 0%  | 1%  | 0%          | 0%          | 1%      |
| Difference       | 5%  | 3%  | 0%  | 0%  | 1%          | 1%          | 2%      |
| Tongue anteriorization | 58% | 12% | 0%  | 0%  | 0%          | 0%          | 0%      |
| Fw               | 54% | 14% | 0%  | 15% | 0%          | 2%          | 15%     |
| VL               | 4%  | 2%  | 0%  | 4%  | 2%          | 0%          | 2%      |
| Difference       | 4%  | 2%  | 0%  | 4%  | 2%          | 0%          | 2%      |
| Distortion       |     |     |     |     |             |             |         |
| Fw               | 0%  | 4%  | 0%  | 1%  | 2%          | 2%          | 2%      |
| VL               | 1%  | 5%  | 1%  | 1%  | 2%          | 2%          | 2%      |
| Difference       | 1%  | 1%  | 1%  | 0%  | 0%          | 0%          | 1%      |
| Late acquisition |     |     |     |     |             |             |         |
| Fw               | 0%  | 1%  | 1%  | 0%  | 9%          | 9%          | 3%      |
| VL               | 1%  | 1%  | 1%  | 0%  | 12%         | 12%         | 5%      |
| Difference       | 1%  | 1%  | 1%  | 0%  | 3%          | 3%          | 1%      |
| Substitution     |     |     |     |     |             |             |         |
| Fw               | 5%  | 9%  | 7%  | 6%  | 4%          | 8%          | 7%      |
| VL               | 6%  | 11% | 7%  | 8%  | 3%          | 12%         | 8%      |
| Difference       | 1%  | 2%  | 0%  | 2%  | 1%          | 4%          | 2%      |

Fw: Samples of patients who received palatoplasty with Furlow technique
VL: Samples of patients who received palatoplasty with von Langenbeck technique
Difference: difference in % between groups Fw and VL

Table 3- Distribution of percentage occurrence of consonant production errors during liquid targets for data distributed according to age at primary palatoplasty (early versus late)

|                  | /l/ | /r/ | /R/ | /l/ | Cluster /l/ | Cluster /r/ | Average |
|------------------|-----|-----|-----|-----|-------------|-------------|---------|
| No alteration    |     |     |     |     |             |             |         |
| 9-12             | 33% | 70% | 92% | 78% | 86%         | 75%         | 72%     |
| 15-18            | 35% | 67% | 91% | 78% | 79%         | 74%         | 71%     |
| Difference       | 2%  | 3%  | 1%  | 0%  | 7%          | 1%          | 2%      |
| Mid dorsum palatal |    |     |     |     |             |             |         |
| 9-12             | 7%  | 4%  | 0%  | 1%  | 1%          | 1%          | 2%      |
| 15-18            | 3%  | 2%  | 0%  | 1%  | 0%          | 0%          | 1%      |
| Difference       | 4%  | 2%  | 0%  | 0%  | 1%          | 1%          | 1%      |
| Tongue anteriorization |    |     |     |     |             |             |         |
| 9-12             | 56% | 13% | 0%  | 12% | 1%          | 2%          | 14%     |
| 15-18            | 54% | 13% | 0%  | 13% | 1%          | 2%          | 14%     |
| Difference       | 2%  | 0%  | 0%  | 1%  | 0%          | 0%          | 1%      |
| Distortion       |     |     |     |     |             |             |         |
| 9-12             | 0%  | 5%  | 1%  | 3%  | 3%          | 3%          | 3%      |
| 15-18            | 1%  | 5%  | 0%  | 0%  | 0%          | 1%          | 1%      |
| Difference       | 1%  | 0%  | 1%  | 3%  | 2%          | 2%          | 2%      |
| Late acquisition |     |     |     |     |             |             |         |
| 9-12             | 0%  | 0%  | 2%  | 0%  | 7%          | 8%          | 3%      |
| 15-18            | 0%  | 1%  | 1%  | 0%  | 14%         | 14%         | 5%      |
| Difference       | 1%  | 0%  | 0%  | 0%  | 7%*         | 6%          | 2%      |
| Substitution     |     |     |     |     |             |             |         |
| 9-12             | 4%  | 8%  | 5%  | 6%  | 2%          | 8%          | 6%      |
| 15-18            | 7%  | 12% | 8%  | 8%  | 5%          | 9%          | 8%      |
| Difference       | 3%  | 4%  | 3%  | 2%  | 3%          | 1%          | 3%      |

9-12: Samples of patients who received palatoplasty between 9 and 12 months
15-18: Samples of patients who received palatoplasty between 15 and 18 months
Difference: difference in % between groups 9-12 and 15-18
*statistically significant difference (p=0.04)
DISCUSSION

In this study we observed a low occurrence of production errors during liquid sounds for children with CLP (28%). Midfacial growth deficiency and dento-occlusal abnormalities, especially crossbites (common in children with CLP), may explain articulation of tongue dorsum and a more posteriorized area of the hard palate. Other possible structural deviations that may contribute to this finding include anteriorized tongue frenulum and dentition stage of children at sixth year of age (primary and mixed dentition). As reported in the literature, midfacial growth deficiency and dento-occlusal abnormalities are suggested by some authors to be the reason for adjustment of the tongue to a reduced oral space.

Particularly, the use of MDPS has been related to presence of VPD and/or palatal fistula. Following these authors explanation for occurrence of MDPS, one can also suggest that middorsum palatal liquids would result from speech developing at the presence of open structure. After experiencing vocalizations and babbling with inadequate anatomical structure, posteriorized tongue posture would become habituated, persisting even after the primary palatoplasty or correction of palatal fistula and VPD. The use of atypical place of production, such as pharynx or larynx, as place of articulation was not found for liquids sounds in the present study. This finding agrees with previous statements that CAs, such as glottal stops and pharyngeal fricatives, are used for sounds that require higher amounts of intraoral air pressure.

Significant difference in the occurrence of atypical place of production was not found when data was distributed according to surgical technique (Furlow and von Langenbeck) or according to age at primary palatoplasty (early and late). Establishing a direct relationship between occurrences of atypical place of articulation during liquids and surgical technique or age at palatoplasty, however, requires controlling variables such as the presence of VPD, fistula, and nasal resistance. Future prospective studies addressing these variables, therefore, are recommended.

Correct production of speech sounds depends on speech stimulation, as well as the child’s auditory and cognitive development. Because children with CLP are often hospitalized and commonly present hearing loss secondary to persistent middle ear fluid and otitis media, they are at higher risk of presenting consonant production errors. Hardin-Jones and Jones (2005) suggest that even with the advances observed in management of CLP, the majority of preschool children with cleft (62%) still show significant delays in speech sound development. According to Morris and Ozanne (2003), consonant cluster reductions and simplifications of liquid sounds are present in more than 50% of children with cleft who have normal expressive language. Trost-Cardamone (2004) also reports that around 50% of children with CLP treated in the United States need speech therapy during pre-school years. In our study, late acquisition (4%), distortions (2%), and sound substitution (6%) for liquids together represented only 11% of the errors. These findings suggest that speech therapy, which is usually offered to children with CLP, could have influenced speech development and these findings. Missing data on delivery of speech therapy was another limitation of this study which should be addressed in future investigations.

Liquid sounds in the Brazilian Portuguese language are usually completely acquired by the end of the 6th year of life (2004). Significant differences due to timing of palatal surgery, therefore, were not expected, since the children were operated between 9-12 months or 15-18 months. Studies have suggested that the earlier the surgery establishes adequate oral structure and velopharyngeal functioning, the greater the chances for children with CLP to develop adequate speech. In the present study, children operated earlier (9 and 12 months) developed the use of the consonant cluster /r/ sooner than children operated on later (15-18 months). While consonant clusters are one of the last phonological acquisitions their production depends on more complex articulatory adjustments, especially when it involves the liquid /r/. It appears that articulatory adjustments that are more refined, such as those involving the production for the cluster /r/, would be previously established in more favorable morphological conditions.

Finally, the overall functioning of the speech system is determined by structural and functional aspects of speech structures. Some changes in the articulatory production can be related to anatomic conditions such as deep palate, anterior open bite, crossbite, thickness of the alveolar ridge, degree of teeth protrusion or maxillary retrusion, tongue frenulum, and rotation and spaces between teeth. Other changes may be related to the presence and late maintenance of oral habits (such as use of pacifiers or teeth grinding) or hearing loss (middle ear pathologies). In this retrospective study, variables like dental conditions, oral habits, and functioning of the middle ear were not controlled, justifying further prospective investigations.

CONCLUSION

The present study revealed a low occurrence of consonant production errors during liquid
sound production in children with surgically corrected unilateral CLP. With the exception of tongue anteriorization during /l/ productions (55%) all other errors had very low occurrence. While no significant differences were observed during liquid production due to surgical technique, statistically significant difference was found for age of acquisition of consonant cluster /r/ between children operated early and those operated late.

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