Speech outcome in unilateral complete cleft lip and palate patients: a descriptive study

ABSTRACT

**Aim** In this study, resonance and articulation disorders were examined in a group of patients surgically treated for cleft lip and palate, considering family social background, and children’s ability of self monitoring their speech output while speaking.

**Materials and methods** Fifty children (32 males and 18 females) mean age 6.5±1.6 years, affected by non-syndromic complete unilateral cleft of the lip and palate underwent the same surgical protocol. The speech level was evaluated using the Accordi’s speech assessment protocol that focuses on intelligibility, nasality, nasal air escape, pharyngeal friction, and glottal stop. Pearson product-moment correlation analysis was used to detect significant associations between analysed parameters.

**Results** A total of 16% (8 children) of the sample had severe to moderate degree of nasality and nasal air escape, presence of pharyngeal friction and glottal stop, which obviously compromise speech intelligibility. Ten children (10%) showed a barely acceptable phonological outcome: nasality and nasal air escape were mild to moderate, but the intelligibility remained poor. Thirty-two children (64%) had normal speech. Statistical analysis revealed a significant correlation between the severity of nasal resonance and nasal air escape (p ≤ 0.05). No statistical significant correlation was found between the final intelligibility and the patient social background, neither between the final intelligibility nor the age of the patients.

**Conclusion** The differences in speech outcome could be explained with a specific, subjective, and inborn ability, different for each child, in self-monitoring their speech output.

**Keywords** unilateral cleft lip and palate; speech disorders; intelligibility; speech assessment; speech outcome.

Introduction

Velopharyngeal incompetence (VPI) is a speech disorder due to incomplete closure of the velopharyngeal sphincter during swallowing, blowing, whistling, and word articulation; it is common to observe VPI in case of congenital cleft lip and/or palate. Orofacial cleft is a congenital malformation characterised by an incomplete development of the structures which separate nasal from oral cavities (i.e. lip, alveolus, hard and soft palate) [Rullo et al., 2006]. It can result in various morphological and functional disorders [Schuster et al., 2006], and one of the most severe manifestations of this malformation is the unilateral complete cleft of the lip and palate (UCLP). A UCLP can establish both aesthetical facial defects [Rullo et al., 2006] and speech and voice disorders; frequently in these patients it is possible to perceive consonant misarticulation and velopharyngeal incompetence (VPI), due to an incomplete closure of the oro-pharyngeal ring during the articulatory movements in speech [Pulliken et al., 2001; Akinbami, 2007].

The surgical approach to this malformation has the main goals of functional rebuilding of the oro-pharyngeal walls, in order to allow an adequate speech development [Timmons et al., 2001], and functional and aesthetic rehabilitation of the whole face [Rullo et al., 2006]. Unfortunately speech disorders can endure even after an adequate surgical restoring intervention [Schuster et al., 2006]; this is why speech rehabilitation in cleft children represents a fundamental step in the treatment strategies. Nowadays there is not a universally accepted method to evaluate this cleft characteristics [Galliot et al., 2007; Schuster et al., 2006; Timmons et al., 2001], but it is accepted that the language development assessment, in a cleft patient, is essentially clinical and somewhat subjective [Timmons et al., 2001].

The purpose of this study was to examine the phono-articulatory defects of a group of consecutive surgically treated UCLP patients, in relation to the children’s ability to monitor their own speech production, by means of word repetition and language, using a complete assessment method of phonetic and phonological examination.
Patients and methods

Patients
Fifty consecutive patients (32 males and 18 females) aged between 5 to 8 years (mean age 6.5±1.6), affected by a non-syndromic unilateral cleft of the lip and palate (UCLP), were selected for this study. All patients were operated by the same surgeon at the Second University of Naples, Dental Clinic. None of the selected patients showed any kind of other pathologies correlated to the cleft, no remaining palatal fistula, neither a significant degree of hearing loss, as indicated by the audiometric examinations, exhibited by the patients at the time of our evaluation.

Treatment protocol
All patients underwent three primary surgical steps in order to correct the congenital malformation. The functional reconstruction of the lip and nose was carried out at about 6 months of life in accordance with the Delaire functional chelirhinoplasty technique [Markus and Delaire, 1993]. Subsequently, functional closure of the soft palate was performed between 12 to 14 months of age, in accordance with the Delaire technique for the functional reconstruction of the palatal muscular layers; finally the hard palatoplasty, between 18 to 36 months of age, completed the primary surgical procedures [Markus et al., 1993].

Speech assessment
The articulation level was evaluated by a speech therapist, by means of a previously reported approach [Rullo et al., 2009].
- Phono-articulatory level with phonetic balance.
- Semantic-lexical output level by means of repetition of a list of phonetically well-balanced words (configuration Consonant-Vowel-Consonant-Vowel, and cluster of consonant in series CCCV).
- Morpho-syntactic structure by means of guided conversation and pictures, in order to stimulate spontaneous answers.
- Facial mimic muscular movements, blowing and breathing ability.

For the assessment of speech problems we used a slightly modified version of the Accordi’s speech assessment protocol [Accordi et al., 2001; Accordi and Agnelli, 2006] (Table 1). This is a complete and reproducible method to evaluate the main cleft-type characteristics, and consonant production; moreover this protocol highlights the fundamental key areas of intelligibility, nasality (hypernasal or hyponasal resonance), nasal air escape, pharyngeal friction (active articulation of fricatives and affricates), and glottal stop [Henningsson et al., 2008; Jhon et al., 2006]. These parameters were evaluated, during clinical examination on a four-point scale, ranging from 0 (severe degree of alteration) to 3 (absence of any defect) (Table 1). The final score, from 0 to 15, represents the sum of the single assessment for each evaluated parameter. On the basis of these results the patients were divided into three phonetic classes. Furthermore, each child was attributed to a lower, middle or upper social class, on the basis of the results of the social interview, in particular on the basis of the parental educational level, analyzed by a counselor, during an interview (Table 2).

Statistical analysis
A Pearson product-moment correlation analysis was performed to calculate the correlation coefficients between the judged categories (Sigma stat software). A p value ≤ 0.05 was considered to be statistical significant.

<table>
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<tr>
<th>Nasal resonance</th>
<th>Nasal air escape</th>
<th>Pharyngeal friction</th>
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<td>Normality</td>
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TABLE 1 Scores of the Accordi’s speech assessment protocol.

Keys
- Nasal resonance: Normality: absence of nasal resonance; Level I: nasal resonance mild and occasional, without relevant phonological disorders; Level II: moderate nasal resonance, with relevant phonological disorders; Level III: severe and consistent nasal resonance, with reduction of intelligibility.
- Nasal air escape: Normality: absence of nasal air escape; Level I: mild and occasional nasal air escape only in few consonants; Level II: moderate nasal air escape in all the consonants; Level III: severe and consistent nasal air escape in all the consonants.
- Pharyngeal friction: Normality: absence of pharyngeal friction; Level I: mild and occasional pharyngeal friction only in fricative sounds; Level II: moderate pharyngeal friction, with systematic substitution of fricatives consonants; Level III: severe and consistent pharyngeal friction, with systematic substitution of fricatives and affricates sounds.
- Glottal stop: Normality: absence of glottal stop; Level I: mild and occasional glottal stop in few consonants; Level II: moderate glottal stop with systematic substitution of velar consonants; Level III: severe and consistent glottal stop in the majority of consonants.
- Intelligibility: Normality: absence of phonological disorders; Level I: mild and occasional phonological alteration, without compromising the intelligibility; Level II: moderate phonological disorders with mild to moderate compromising of intelligibility; Level III: severe, consistent and systematic compromising of intelligibility.
Results

The percentage and the distribution of the analysed phonetic defects are reported in Table 3.

According to the final score obtained after the speech evaluation, we divided the children in three different phonetic categories (Table 4).

- Class A (final score 0–5): 8 children (16%), in the selected sample, had a negative result, with a severe to moderate degree of nasality and nasal air escape in all consonants (level III-I); the presence of active pharyngeal friction with a systematic substitution of fricatives and affricates of severe degree (level III) was also identified.
- Class B (final score 6–10): 10 children (20%) showed an almost acceptable phonological outline; nasality and nasal airflow were mild to moderate (level II-I); no pharyngeal friction, neither glottal stop were evident, but nasal flare was showed by the majority of the children of this group. Despite this, the final intelligibility of these patients resulted severely affected by the VPI.
- Class C (final score 11–15): 32 children (64%) showed a perfectly normal speech, without any phonological alterations, excellent blowing ability and normal breathing mechanism.

Statistical analysis of the whole sample showed a significant correlation between the severity of nasal resonance and nasal air escape (p ≤ 0.05), and both, nasal resonance and nasal airflow, positively correlate with the presence of grimace (p ≤ 0.05); moreover there was a significant correlation between nasal resonance and the final intelligibility (p ≤ 0.05). No statistically significant correlation was found between the final intelligibility and the patient social context of life, neither between the final intelligibility and the age of the patients. Moreover the distribution of the patients in the three phonetic classes seems not to be influenced by age.

Discussion

Speech anomalies in patients with clefts are multifactorial; anatomical and functional defects associated with UCLP and its surgical rehabilitation are, probably, the main causes of speech problems, often related to hypoplasia and hypomobility of the levator and tensor veli palatini muscles and their abnormal course and insertion into the palate [Timmons et al., 2001; Merrik et al., 2007]. Likewise, it seems logical that, if cleft muscles are not repositioned or are inaccurately relocated, only a minority of patients will achieve completely normal speech [Marsch et al., 1989], with the development of a VPI condition. The velo-pharyngeal canal is normally opened in case of nasal phonemes articulation (/m/, /n/, /ŋ/); indeed, alterations of vocal tone due to nasal air escape during phonation (hyper-nasality), are developed in case of VPI condition. This kind of air loss is responsible for the misarticulation of oral, occlusive and fricative consonants. Therefore, if patients are not able to control oral and nasal air emission, they will develop compensatory mechanisms, such as glottal stop and pharyngeal friction. This is why functional palatoplasty pays particular attention to the restoration of anatomical continuity of the musculature with the potential for better function of the tensor veli palatini [Markus et al., 1993; Merrik et al., 2007], in order to improve speech and hearing functions. The social context in which the children live must also be considered for the development of cognitive and intellectual capabilities of the toddlers; poor social circumstances could increase the risk of speech problems [Pulliken et al., 2001; Hunt et al., 2006; D’Mello and Kumar, 2007]. Nevertheless, analysing the whole sample, we did not find a significant correlation
between the final intelligibility and the familial social context of patients. This remark leads us to speculate that each child has specific inborn capabilities in self-controlling the spontaneous language, independently from parental education and logopaedic rehabilitation. Obviously it would be correct to consider as inborn capabilities of each child during speech evaluation, but, unfortunately, it remains an inviable datum.

Analyzing the sample with regard to each phonetic class, we detected a low grade of correlation between final intelligibility and social status (\(p < 0.06\)), only in the phonetic class C. This result could probably confirm that the “family factor” could positively influence speech development, especially if the patient already has inborn capabilities to control air emission from nasal and oral cavities. The children belonging to the phonological Class A showed poor linguistic and phonetic development, despite the majority of them had attended speech therapy for a long period; moreover the social background does not seem to influence the linguistic level of this group. Naturally they showed insufficient inborn phonological self-controlling ability, and they usually had a dialectal linguistic code, with a very fast speech. As previously reported, in fact, accent, and speed of speech could interfere with a correct consonant production and language development [Accordi et al., 2001]. Concerning phonetic Class B, results are debatable because, notwithstanding final scores higher than those obtained in Class A, the final intelligibility remains quite poor (level II–III); it is clear that the intelligibility is not enough to describe, alone, the whole speech characteristics of the patients, but it is the only parameters that could provide an overall assessment of the speech of a child [Timmons et al., 2001]. For this reason we can state that the results obtained in phonetic class B are comparable with the ones of phonetic Class A, except for the assessment of glottal stop and pharyngeal friction which appear better in class B. Similarly, the Class B patients, independently from the social background, had attended speech therapy without satisfactory results, most likely because of strongly consolidated speech defects. As for phonetic Class C, it should be mentioned that the majority of the patients enrolled in this study belonged to this class; they appear to speak well, without any kind of consolidated phonetic and phonological defects, both in rote speech and spontaneous language. Moreover, most of these patients had never undergone logopaedic treatment. Only few of them underwent a brief speech therapy cycle, aimed just to correct vocal resonance. Obviously a particular favourable familial and social context, could further facilitate verbal cognitive abilities and intellectual development [Merket et al., 2007]; notwithstanding, the correct speech development is a multifatorial process. Finally, the real role of external factors, like family or social circumstances, rather than anatomical and functional factors, should be further investigated. How much the inborn and individual capabilities of each patient could interfere with language development [Ballini et al., 2012; Paolantonio et al., 2009; Perillo et al., 2011]? Are there, perhaps, anatomical and congenital conditions, like breathing aerodynamics during speech, palatal and tongue muscular strength, to be considered, during speech evaluation?

**Conclusion**

This study tested the speech outcome in a group of schoolchildren affected by unilateral cleft lip and palate. The results suggest that in UCLP patients the phonetic and phonological development is clearly determined by the surgical strategies; but, despite the same surgical technique applied by the same surgeon, the results are different in terms of speech abilities.

**References**


