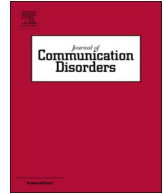




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## Delayed primary palatal closure in resource-poor countries: Speech results in Ugandan older children and young adults with cleft (lip and) palate

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### ABSTRACT

**Aims:** Unrepaired clefts still regularly occur in resource-poor countries as a result of limited health-care access. The purpose of the present study was to report resonance, airflow and articulation characteristics following delayed ( $\geq 8$  years) primary palatal closure.

**Methods:** Fifteen Ugandan participants with cleft (lip and) palate (CP  $\pm$  L) were included as well as 15 age- and gender-matched Ugandan subjects without clefts. Palatal closure was performed at a mean age of 15;10 years using the Sommerlad technique. Speech evaluations were carried out on a single occasion postoperatively (mean age: 18;10 years). Resonance and nasal airflow were perceptually evaluated and detailed phonetic and phonological assessments were carried out. Additionally, nasalance values were determined.

**Results:** Nasal emission occurred postoperatively in only 27% (4/15) of the patients, whereas resonance disorders and articulation errors were prevalent in 87% (13/15) of the patient group. Compared with the control group, a significantly higher prevalence of hypernasality and significantly higher nasalance values for all oral and oronasal speech samples were obtained in the CP  $\pm$  L group. Moreover, significantly smaller consonant inventories and significantly more phonetic and phonological disorders were observed.

**Conclusions:** Delayed palatal repair ( $\geq 8$  years) seems to be insufficient to eliminate nasal airflow errors, resonance abnormalities, and articulation disorders. In order to prevent patients' late presentation at specialized centers, the availability of high quality surgical cleft palate treatment should increase as well as people's awareness of the possibility and importance of early surgical intervention. Moreover, speech therapy following delayed palatal closure would be beneficial. Furthermore, a standardized and validated protocol for speech assessment in future studies is advocated.

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## 1. Introduction

Cleft lip and/or palate (CL/P) is a frequently occurring craniofacial anomaly, often associated with functional problems such as feeding, hearing and speech difficulties (Mossey, Little, Munger, Dixon, & Shaw, 2009). In most developed countries, children born with CL/P are counselled by a cleft and craniofacial team shortly after birth or even prenatally (Madahar, Murray, Orr, & Sandler, 2013). Multidisciplinary treatment adapted to the individual needs of the patient is then initiated. The cleft lip and palate are generally closed in early childhood during one to four operations (Shaw et al., 2001). As a result, older children and young adults with unrepaired clefts are rarely seen in developed regions.

In resource-poor countries unrepaired CL/P regularly occur in older children and young adults. According to studies conducted in Nigeria (Adeyemo et al., 2009) and Nepal (Schwarz & Khadka, 2004), reasons for late presentation include lack of knowledge of availability of cleft repair, large distance to health services, as well as insufficient financial resources for travel, accommodation, and medical expenses. Moreover, failure to complete palatoplasty after lip repair due to financial considerations and/or predominantly aesthetic concerns of parents seems to be widespread (Onah, Opara, Olaitan, & Ogonnaya, 2008). Furthermore, in these resource-poor countries clefts are often associated with demonization, witchcraft, the curse of God and ancestral punishment (Dagher & Ross, 2004). As a consequence, psychosocial problems, social isolation, school absence, unemployment and celibacy may be even more pronounced in these patients, in comparison to patients with repaired clefts in developed countries (Bradbury & Habel, 2008; Hunt, Burden, Hepper, Stevenson, & Johnston, 2006).

Several surgeons reported on the surgical treatment of older children and young adults with unrepaired clefts (Adekeye & Lavery, 1985; Holdsworth, 1954; Hoppenreijts, 1990; Koberg & Koblin, 1973; Law & Fulton, 1959; Ortiz-Monasterio et al., 1974; Ortiz-Monasterio, Serrano, Barrera, Rodriguez-Hoffman, & Vinageras, 1966; Schwarz, 2006). However, speech assessment after delayed cleft repair included mostly descriptive impressions by professionals who were not speech-language therapists (SLTs) (Sell & Grunwell, 1994), without mentioning whether speech was affected by resonance disorders, nasal airflow errors and/or articulation errors. Typically, these studies provided limited information regarding patients' cleft type, palatal closure technique, assessed speech samples and perceptual rating strategies.

Nevertheless, a few studies systematically reported evaluations of speech following delayed palatal repair by at least one SLT (Murthy, Sendhilmathan, & Hussain, 2010; Sell & Grunwell, 1990; Whitehill, Stokes, & Yonnie, 1996; Zhao et al., 2012). An overview of these studies is provided in Table 1. As seen in Table 1, backing to a velar, pharyngeal or glottal place of articulation was found to be the most common phonetic error (Sell & Grunwell, 1990; Whitehill et al., 1996). In addition, many patients still showed hypernasality and nasal emission after delayed palatal closure (Murthy et al., 2010; Sell & Grunwell, 1990; Zhao et al., 2012).

Conservative palatal repair (i.e. Veau or von Langenbeck technique) was performed in all patients reported by Sell and Grunwell (1990), whereas Murthy et al. (2010) and Zhao et al. (2012) applied the Sommerlad technique in respectively 69% (91/131) and 52% (44/84) of the patients. A substantial portion of the patients described by Zhao et al. (2012) presented without hypernasality and/or nasal emission after delayed palatal repair, but no significant differences in the incidence of a secondary operation were noted after different surgical techniques. In contrast, Murthy et al. (2010) reported both hypernasality and nasal emission in almost all patients, but a better improvement in hypernasality was reported following radical muscle dissection as described by Sommerlad (2003). Although recommended (Lohmander et al., 2009; Sell, 2005), neither study included a detailed articulation analysis or objective assessment of resonance. Furthermore, the presence or absence of other potential resonance disorders such as hyponasality and cul-de-sac resonance was not mentioned. Finally, a risk of researcher bias is present in the study of Zhao et al. (2012), considering that only one SLT evaluated the patients' speech once. Given these study limitations and the variation in speech results, further research on speech outcome following delayed palatal repair is indicated.

In the past few decades, an increasing number of plastic and maxillofacial surgeons as well as cleft and craniofacial teams have visited resource-poor countries to treat patients with clefts and train local health care providers (Garfein, Hom, & Mulliken, 2008). In Uganda, Christian Blind Mission and Smile Train funded in 2005 the establishment of a plastic and reconstructive unit, which moved in 2009 to the Comprehensive Rehabilitation Services in Uganda (CoRSU) hospital in Kisubi. Despite these efforts, late presentation of patients with an unrepaired CL/P at an older age still occurs regularly in Uganda.

In Uganda 41 different languages of particularly the Bantu and Nilotic language families are currently spoken (Lewis, Simons, & Fenning, 2014). However, English remained one of both official languages of this country at the end of the colonial period in 1962 and is still deeply rooted in media, education and administration (Mpuga, 2003). Nevertheless, the most widely spoken language in the southern part of the country is Luganda (Lewis et al., 2014). Some notable differences exist between Luganda and English regarding the sound system and phonotactics. An overview of all consonants included in the sound system of both languages is provided in Table 2. As seen in Table 2, English and Luganda have 18 similar consonants. In the Ugandan English dialect, the English fricatives and affricates (i.e./θ/,/ð/,/ʃ/,/ʒ/,/tʃ/, and/dʒ/) that do not occur in the native language, are often produced incorrectly. Unlike English, Luganda is a tonal language, which contains single and geminate consonants. Consonants do not occur at the end of a syllable and syllable initial nasals are often followed by plosives or fricatives. The liquids/l/and/r/are allophones: their use depends on the preceding vowel. Consequently, devoicing of final voiced consonants, addition of/k/or/g/after/ŋ/and liquid gliding are often noticed in the Ugandan English dialect.

The main purpose of the current study was to report the resonance, nasal airflow (nasal emission and nasal turbulence) and articulation characteristics of Ugandan English-speaking older children and young adults with non-syndromic cleft palate with or without cleft lip (CP ± L) following palatal closure after the age of 8 years. To our knowledge, no research has yet been performed regarding speech development in Ugandan English-speaking children. Therefore, the age of 8 years is chosen given that phonetic and phonological development in typically developing native American-English-speaking children is considered by some to be completed

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