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Speech characteristics after palatal closure in subjects with isolated clefts

– An exploration in Uganda –

Anke Luyten

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Faculty of Medicine and Health Sciences

Department of Speech, Language and Hearing Sciences

Speech characteristics after palatal closure in subjects with isolated clefts: an exploration in Uganda

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If you give a man a fish, he feeds his family for a day.
If you give him a fishing pole and teach him to fish,
he feeds his family for a lifetime.

Adapted from *Mrs. Dymond* by Anne Isabella Thackeray Ritchie (1885).

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Ghent, 13th of May 2014

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LIST OF PUBLICATIONS

This doctoral thesis is based on the following articles published or submitted in international peer-reviewed journals:

1. Luyten, A., D'haeseleer, E., Hodges, A., Galiwango, G., Budolfsen, T., Vermeersch, H., & Van Lierde, K. (2012). Normative nasalance data in Ugandan English-speaking children. *Folia Phoniatrica et Logopaedica*, 64, 131-136.

2. Luyten, A., D'haeseleer, E., Budolfsen, D., Hodges, A., Galiwango, G., Vermeersch, H., & Van Lierde, K. (2013). Parental satisfaction in Ugandan children with cleft lip and palate following synchronous lip and palatal repair. *Journal of Communication Disorders*, 46, 321-329.

3. Luyten, A., Bettens, K., D'haeseleer, E., De Ley, S., Hodges, A., Galiwango, G., Vermeersch, H., & Van Lierde, K. (2014). Impact of early synchronous lip and palatal repair on speech. *Folia Phoniatrica et Logopaedica*, DOI: 10.1159/000362501.

4. Luyten, A., Bettens, K., D'haeseleer, E., De Ley, S., Hodges, A., Galiwango, G., Bonte, K., Vermeersch, H., & Van Lierde, K. (2014). The impact of palatal repair before and after 6 months of age on speech characteristics. *International Journal of Pediatric Otorhinolaryngology*, 78, 787-798.

5. Luyten, A., Bettens, K., D'haeseleer, E., Dhondt, C., Hodges, A., Galiwango, G., Vermeersch, H., & Van Lierde, K. Impact of delayed one-stage palatal closure performed with the Sommerlad technique on speech in Ugandan youngsters and adults with cleft palate. *Submitted in International Journal of Language and Communication Disorders, March 2014.*

LIST OF ABBREVIATIONS

(C)PS	Palatoschisis met of zonder cheiloschisis
ANOVA	Analysis of Variance
asap	As soon as possible
BCLP	Bilateral cleft lip and palate
C(L)P	Cleft palate with or without cleft lip
CBM	Christian Blind Mission
CCG	Cleft comparison group
CEP	Cleft Evaluation Profile
CGPS	Cheilognatopalatoschisis
CL	Cleft lip
CL(P)	Cleft lip with or without cleft palate
CL/P	Cleft lip and/or palate
CLP	Cleft lip and palate
CoRSU	Comprehensive Rehabilitation Services in Uganda
CORU	Children's Orthopaedic Rehabilitation Unit
CP	Cleft palate
EG	Experimental group
EUROCAT	European Concerted Action on Congenital Anomalies and Twins
Ga	Group a
Gb	Group b
GDP	Gross Domestic Product
ICF	International Classification of Functioning, Disability and Health
ICF-CY	International Classification of Functioning, Disability and Health: Children and Youth Version

INAMI	Institut National d'Assurance Maladie-Invalidité
IPA	International Phonetic Alphabet
n.a.	Not available
NCG	Non-cleft control group
PAT-3	Photo Articulation Test – third edition
PI	Prediction interval
postop	Postoperatively
preop	Preoperatively
RIZIV	Rijksinstituut voor Ziekte- en Invaliditeitsverzekering
SD	Standard deviation
SE	Standard error
SLT	Speech-language therapist
SLTs	Speech-language therapists
SMCP	Submucous cleft palate
SNAP	Simplified Nasometric Assessment Procedures
Soft CP	Cleft of soft palate only
SWA	Satisfaction with Appearance Questionnaire
UCLP	Unilateral cleft lip and palate
UNICEF	United Nations International Children's Emergency Fund
VLIR-UOS	Vlaamse Interuniversitaire Raad voor Universitaire Ontwikkelings-samenwerking
VPI	Velopharyngeal insufficiency
WHO	World Health Organization

SUMMARY

Cleft lip and/or palate (CL/P) is a congenital craniofacial defect that arises on average in 1.7 per 1000 live births. This anomaly causes atypical facial appearance, hearing problems, malocclusions and speech disorders. Outcomes in terms of speech are influenced by timing of surgical cleft closure. In the Comprehensive Rehabilitation Services in Uganda (CoRSU) hospital, closure of the entire cleft during a single surgery prior to the age of 6 months is preferred in view of reducing default rates for second surgery and decreasing risks for malnutrition and death. However, some patients arrive the first time at the hospital in later childhood, youth or adulthood. No information about satisfaction and speech outcome was yet available for these Ugandan patients. Moreover, only few, if any studies systematically assessed satisfaction and speech following similar surgical timing protocols. Therefore, in view of searching for the optimal surgical treatment for patients with CL/P, the general aim of the current doctoral thesis was to verify satisfaction and speech in Ugandan patients with CL/P repaired in CoRSU by one experienced surgeon using the Sommerlad technique for palatal closure.

In view of clinics and further research, normative nasalance values were first obtained in Ugandan English-speaking males and females (age: 2;7 to 13;5 years) without craniofacial anomalies. No significant age and gender differences were observed.

Second, parental satisfaction was studied in Ugandan children with unilateral or bilateral cleft lip and palate (CLP) following synchronous lip and palatal closure. Overall high levels of satisfaction were noted for appearance of lip, nose and face, despite lower satisfaction levels for teeth appearance and speech. Comparison with an age- and gender-matched non-cleft control group revealed significant higher parental dissatisfaction for speech and appearance of teeth and nose in Ugandan patients with CLP.

Third, articulation and resonance characteristics of Ugandan patients with early synchronous closure of C(L)P (≤ 6 months) were assessed. Comparison with an age- and gender-matched non-cleft control group revealed various deviations from normal speech development. The Ugandan CP group showed significantly smaller consonant inventories as well as significantly more phonetic disorders, phonological processes and nasal emission/turbulence compared to the control group. In addition, Ugandan patients with C(L)P were compared to a Belgian CP group (matched for cleft type, age and gender) who underwent palatal repair after the age of 6 months. A Ugandan and Belgian age- and gender-matched non-cleft control group was included to control for language, culture and other environmental factors. Comparison of the Ugandan and Belgian CP group revealed at least similar articulation and resonance characteristics. No significant group differences were obtained for perceptual evaluation of resonance, mean nasalance values of oral speech samples, consonant inventories and most phonetic errors and phonological processes. However, the Belgian CP group showed significantly more distortions due to higher occurrence frequencies for (inter)dental articulation of apico-alveolar consonants.

Finally, articulation and resonance characteristics as well as patients' satisfaction with speech were verified in Ugandan patients following delayed one-stage soft and hard palatal closure (≥ 8 years). Comparison with an age- and gender-matched non-cleft control group revealed overall low satisfaction with speech and severely disordered articulation and resonance. The patient group showed significantly smaller consonant inventories, more phonetic and phonological disorders, more hypernasality and nasal emission/turbulence as well as higher mean nasalance values for oral and oronasal speech samples compared to the control group.

When findings of the current doctoral thesis are placed within a broader framework, early closure of the entire cleft during a single surgery seems to be an appropriate surgical timing protocol for resource-poor countries. However, prior to application in northern countries, more information on maxillofacial growth disturbances is required. Furthermore, when youngsters and adults present with untreated clefts, delayed cleft repair might be of value, although speech outcomes are poor.

SAMENVATTING

Schisis is een congenitale craniofaciale aandoening die voorkomt met een gemiddelde prevalentie van 1.7 per 1000 levendgeborenen. De afwijking gaat gepaard met een atypisch gelaat, gehoorproblemen, malocclusies en spraakstoornissen. De aanwezigheid van spraakstoornissen wordt beïnvloed door het tijdstip van chirurgische sluiting. In het Comprehensive Rehabilitation Services in Uganda (CoRSU) ziekenhuis gaat de voorkeur uit naar het gelijktijdige sluiten van de lip en het verhemelte tijdens één chirurgische ingreep vóór de leeftijd van 6 maanden. Zodoende wordt sluiting van de gehele schisis verzekerd en vermindert de kans op malnutritie en sterven. Desondanks melden sommige patiënten zich voor het eerst op latere leeftijd aan in het ziekenhuis. Informatie omtrent tevredenheid en spraak bij deze Oegandese schisispatiënten was tot nog toe niet beschikbaar. Bovendien onderzochten weinig tot geen studies systematisch de tevredenheid en spraak na gelijkaardige chirurgische benaderingen. Vertrekkende vanuit de zoektocht naar de optimale chirurgische behandeling van schisis, stelde dit doctoraat zich bijgevolg tot doel de tevredenheid en spraak te onderzoeken bij Oegandese schisispatiënten, bij wie de schisis gesloten werd in CoRSU door één ervaren chirurg die tijdens de palatoplastiek de Sommerlad techniek hanteerde.

Met het oog op klinische hulpverlening en verder onderzoek werden vooreerst normatieve nasometrische waarden opgesteld voor Oegandese Engelssprekende jongens en meisjes (leeftijd: 2;7 tot 13;5 jaar) zonder craniofaciale aandoeningen. Leeftijds- noch geslachtsverschillen werden geobserveerd.

Vervolgens werd de tevredenheid nagegaan bij ouders van Oegandese kinderen met unilaterale of bilaterale cheilognatopalatoschisis (CGPS) die een gelijktijdige chirurgische sluiting van lip en verhemelte ondergingen. Een hoge tevredenheid werd vastgesteld met betrekking tot het uitzicht van de lip, de neus en het gelaat. Voor het uitzicht van de tanden en de spraak werd een lagere tevredenheid verkregen. In vergelijking met een controlegroep zonder schisis (gematcht volgens leeftijd en geslacht) waren de ouders van Oegandese schisispatiënten significant vaker ontevreden met de spraak en het uitzicht van de tanden en neus.

Verder werden de articulatie- en resonantiekarakteristieken van Oegandese schisispatiënten bestudeerd na een vroege gelijktijdige sluiting van de (C)PS (≤ 6 maanden). In vergelijking met een controlegroep zonder schisis (gematcht volgens leeftijd en geslacht) werden verschillende afwijkingen van de normale spraakontwikkeling geconstateerd. De Oegandese schisispatiënten vertoonden significant kleinere consonantinventarissen en significant meer fonetische fouten, fonologische processen en nasale emissie/turbulentie in vergelijking met de controlegroep. Daaropvolgend werden Oegandese schisispatiënten vergeleken met een Belgische groep schisispatiënten (gematcht volgens type schisis, leeftijd en geslacht) bij wie palatoplastiek werd uitgevoerd na de leeftijd van 6 maanden. Een Oegandese en Belgische controlegroep zonder schisis (gematcht volgens leeftijd en geslacht) werden geïncludeerd om een mogelijke invloed uit te sluiten van verschillen in taal, cultuur en andere omgevingsfactoren. Articulatie- en resonantiekarakteristieken van Oegandese en Belgische schisispatiënten waren grotendeels vergelijkbaar. De groepen verschilden niet significant voor wat betreft perceptuele beoordeling van resonantie, gemiddelde nasometrische waarden voor orale spraakstalen, consonantinventarissen en de meeste fonetische fouten en fonologische processen. Belgische schisispatiënten vertoonden echter significant meer distorsies door significant hogere percentages van voorkomen voor (inter)dentale productie van apico-alveolaire consonanten.

Tot slot werden zowel articulatie- en resonantiekarakteristieken als tevredenheid met spraak onderzocht bij Oegandese schisispatiënten die een late gelijktijdige sluiting van het zachte en harde verhemelte (≥ 8 jaar) ondergingen. Vergelijking met een controlegroep zonder schisis (gematcht volgens leeftijd en geslacht) resulteerde in significant lagere tevredenheidsscores voor spraak en ernstig gestoorde articulatie en resonantie. In vergelijking met de controlegroep vertoonden de schisispatiënten significant kleinere consonantinventarissen, meer fonetische en fonologische stoornissen, meer hypernasaliteit en nasale emissie/turbulentie en hogere gemiddelde nasometrische waarden voor orale en oronasale spraakstalen.

Wanneer de bevindingen van dit doctoraat in een breder kader geplaatst worden, blijkt vroege sluiting van de gehele schisis tijdens één chirurgische ingreep een geschikte benadering voor derdewereldlanden. Alvorens het tijdstip van chirurgisch sluiting in

noordelijke landen kan worden aangepast, is echter meer informatie omtrent maxillofaciale groei vereist. Bovendien is gebleken dat jongeren en volwassenen met onbehandelde schisis baat kunnen hebben bij een late chirurgische sluiting, ondanks de slechte resultaten met betrekking tot spraak.

RÉSUMÉ

Une fente labiale et/ou fente palatine est une affection congénitale crâniofaciale qui se présente avec une prévalence moyenne de 1.7 sur 1000 naissances vivantes. La malformation cause une physionomie faciale atypique, des malocclusions et des troubles de l'audition et de la parole. Le moment précis de la réparation chirurgicale exerce un influence considérable sur la présence éventuelle de troubles de la parole. Dans l'hôpital Comprehensive Rehabilitation Services en Ouganda (CoRSU), la réparation simultanée de la lèvre et du palais dans une seule intervention chirurgicale avant l'âge de 6 mois est préférée afin d'assurer une fermeture entière de la fente et de réduire les risques de malnutrition et de mort. Néanmoins, certains patients arrivent à l'hôpital pour la première fois pendant leur enfance ou adolescence, ou à l'âge adulte. Jusqu'à présent, il n'existait pas d'information sur la satisfaction de ces patients ougandais, ni sur des éventuels troubles de parole. De plus, peu d'études, voire aucun, ont systématiquement évalué la satisfaction et la parole après des approches chirurgicales similaires. Par conséquent, le but général de ce doctorat était d'évaluer la satisfaction et la parole des patients ougandais souffrants d'une fente labio-palatine, dont la fente avait été réparée à CoRSU par un seul chirurgien compétent appliquant la technique de Sommerlad pendant la palatoplastie.

Afin de permettre de l'aide clinique et des recherches scientifiques plus approfondies, des valeurs normatives nasométriques ont été fixées pour des garçons et des filles ougandais anglophones (âgé de 2;7 à 13;5 ans) sans affections crâniofaciales. Des différences d'âge ou de sexe n'ont pas été observées.

En second lieu, la satisfaction des parents a été évaluée après la réparation simultanée d'une fente labio-palatine unilatérale ou bilatérale chez leurs enfants. Un taux de satisfaction élevé a pu être constaté en ce concerne l'aspect esthétique de la lèvre, du nez et du visage, à côté d'un taux plus bas pour l'esthétique des dents et pour la parole. En comparaison d'un groupe témoin sans fente labio-palatine (membres choisis en fonction de leur âge et leur sexe), les parents des patients ougandais étaient plus souvent insatisfait avec la parole et l'apparence des dents et du nez.

Les caractéristiques d'articulation et de résonance des patients ougandais ont ensuite été examinées après une réparation de la fente avant l'âge de 6 mois. Par rapport à un groupe témoin sans fente (membres choisis en fonction de leur âge et leur sexe), des différents anomalies ont été observés dans le développement normale de la parole. Les patients ougandais présentaient de façon significative des inventaires de consonnes plus petits, plus d'erreurs phonétiques, plus de procès phonologiques et plus d'émission/de turbulence nasale en comparaison du groupe témoin. Par la suite, les patients ougandais ont été comparés avec un groupe de patients belges (choisi en fonction du type de fente, de l'âge et du sexe) qui ont subi une palatoplastie après l'âge de 6 mois. Deux groupes témoin, un ougandais et un belge, ont été ajouté en vue d'exclure l'influence éventuelle de différences linguistiques, culturelles ou d'autres facteurs d'environnement. Les caractéristiques d'articulation et de résonance des patients ougandais et belges étaient en grande partie comparables. Les groupes ne présentaient pas de différences significatives en ce qui concerne l'évaluation auditive de la résonance, des valeurs moyennes nasométriques pour des échantillons orales de parole, des inventaires de consonnes et de la plupart des erreurs phonétiques et procès phonologiques. Cependant, les patients belges présentaient de façon significative plus de distorsions en raison de plus de production (inter)dentale de consonnes apico-alvéolaires.

Pour finir, les caractéristiques d'articulation et de résonance ainsi que la satisfaction vis-à-vis la parole ont été examinés auprès de patients ougandais qui avaient subi une réparation simultanée tardive (≥ 8 ans) du palais (dur et mou). La comparaison avec un groupe témoin sans fente a montré des taux de satisfaction considérablement moins élevés pour la parole, et une articulation et une résonance gravement déformée. Par comparaison avec le groupe témoin il est clair que les patients présentaient de façon significative des inventaires de consonnes plus petits, plus de troubles phonétiques et phonologiques, plus d'hypernasalité, plus d'émission/turbulence nasale et des valeurs nasométriques moyennes plus élevées pour des échantillons de parole orales et oronasales.

En plaçant les résultats de ce doctorat dans un cadre plus large, il paraît qu'une réparation tôt de la fente entière pendant une seule intervention chirurgicale est une stratégie chirurgicale appropriée pour les pays du tiers-monde. Mais avant de pouvoir adapter le moment précis de la réparation chirurgicale, il est indispensable d'avoir plus d'information sur la croissance maxillo-faciale. En outre, il est apparu que, malgré les mauvais résultats en ce qui concerne la parole, une réparation chirurgicale tardive peut être utile pour des adolescents et des jeunes avec une fente non soignée.

PART 1

General introduction

CHAPTER 1

CLEFT LIP AND/OR PALATE FROM VARIOUS POINTS OF VIEW

Cleft lip and/or palate (CL/P) is a frequently occurring congenital craniofacial anomaly with a largely unknown etiopathogenesis. This anomaly is associated with a variety of problems that determine the patient's disability. The current chapter will describe the potential etiology and prevalence of orofacial clefts. Moreover, CL/P will be placed within an holistic framework regarding disabilities and potential associated anomalies influencing the patient's quality of life will be discussed.

Etiology of orofacial clefts

Formation of the lip and palate takes place between the 4th and 10th embryonic week *(Mossey et al., 2009)*. These embryonic processes can be disturbed by both genetic predisposition and environmental influences leading to development of orofacial clefts *(Dixon et al., 2011)*. Although the exact causal factors remain unknown, consensus exists about the multifactorial nature of the etiology *(Kohli & Kohli, 2012)*. Cleft lip with or without cleft palate (CL(P)) and cleft palate (CP) are recognized as a feature for respectively more than 200 and close to 400 genetic syndromes *(Wong & Hägg, 2004)*. Moreover, various genes and loci have yet been identified to be somehow related with the formation of cleft lip and/or palate (CL/P) *(Kohli & Kohli, 2012)*. Furthermore, larger recurrent risks are noted for siblings of patients with CL/P and higher concordance rates were observed in monozygotic compared with dizygotic twins *(Mossey et al., 2009)*. However, genetic causing factors insufficiently explain the presence of isolated orofacial clefts. In literature, a gene-environment interaction is suggested *(Romitti et al., 1999)*. Several environmental risk factors have yet been related with the occurrence of CL/P to some extent. Moderate increased risks for both CL(P) and CP are noted for maternal smoking *(Chung et al., 2000; Honein et al., 2007)* and alcohol consumption *(Molina-Solana et al., 2013; Shaw & Lammer, 1999)* during early pregnancy as well as for poor maternal nutrition *(Jia et al., 2011)*, a history of fever or a cold *(Molina-Solana et al., 2013; Wang et al., 2009)*, folium acid or zinc deficiency *(Jia et al., 2011; Molina-Solana et al., 2013)*, and maternal use of several medicinal drugs such as anticonvulsants *(Margulis et al., 2012; Werler et al., 2011)* or corticosteroids *(Kohli & Kohli, 2012; Park-Wyllie et al., 2000)* in the first trimester of pregnancy.

Prevalence of orofacial clefts

Orofacial clefts arise in 1.7 per 1000 live born babies, a frequency comparable with the prevalence of Down syndrome *(Mossey & Castilla, 2003)*. However, variation in prevalence is noted when variables such as cleft type, gender and ethnicity are taken into account. Based on 30 registers in 16 European countries, the European Concerted Action on Congenital Anomalies and Twins (EUROCAT) working group observed a birth prevalence of 0.62 per 1000 for CP between 1980 and 1996 *(Calzolari et al., 2004)*. A birth prevalence of 0.91 per 1000 was noted for CL(P) between 1980 and 2000 based on a network of 23 registers in 14 European countries *(Calzolari et al., 2007)*. Among these cases, 37% presented with cleft lip only (CL) and 63% showed cleft lip and palate (CLP). Overall, a predominance in females is observed for CP, whereas CL(P) most typically occurs in males *(Mossey et al., 2009)*. Male/female-ratios in large international studies varied from 0.83 *(Calzolari et al., 2004)* to 0.93 *(Mossey & Castilla, 2003)* for CP and from 1.70 *(Calzolari et al., 2007)* to 1.81 *(Mossey & Castilla, 2003)* for CL(P). Study of ethnical and geographical differences in prevalence of orofacial clefts revealed overall higher frequencies of CL/P in Asians. Lower frequencies are reported in Africans, with an intermediate prevalence for Caucasians *(Mossey & Castilla, 2003)*. The rates observed in migrant groups generally seem to resemble those of the originating area rather than the prevalence of the area they moved to *(Mossey et al., 2009)*. As described previously in this chapter, this racial and ethnical variation might be explained by a mixture of differences in environmental risk factors and genetic predisposition *(Croen et al., 1998)*. An overview of the birth prevalence of CL/P for various countries worldwide is provided in Figure 1.1.

Cleft lip and/or palate: a disability?

The extent to which patients with CL/P will feel disabled is not only determined by the cleft itself. According to the International Classification of Functioning, Disability and Health (ICF) presented by the World Health Organization (WHO),

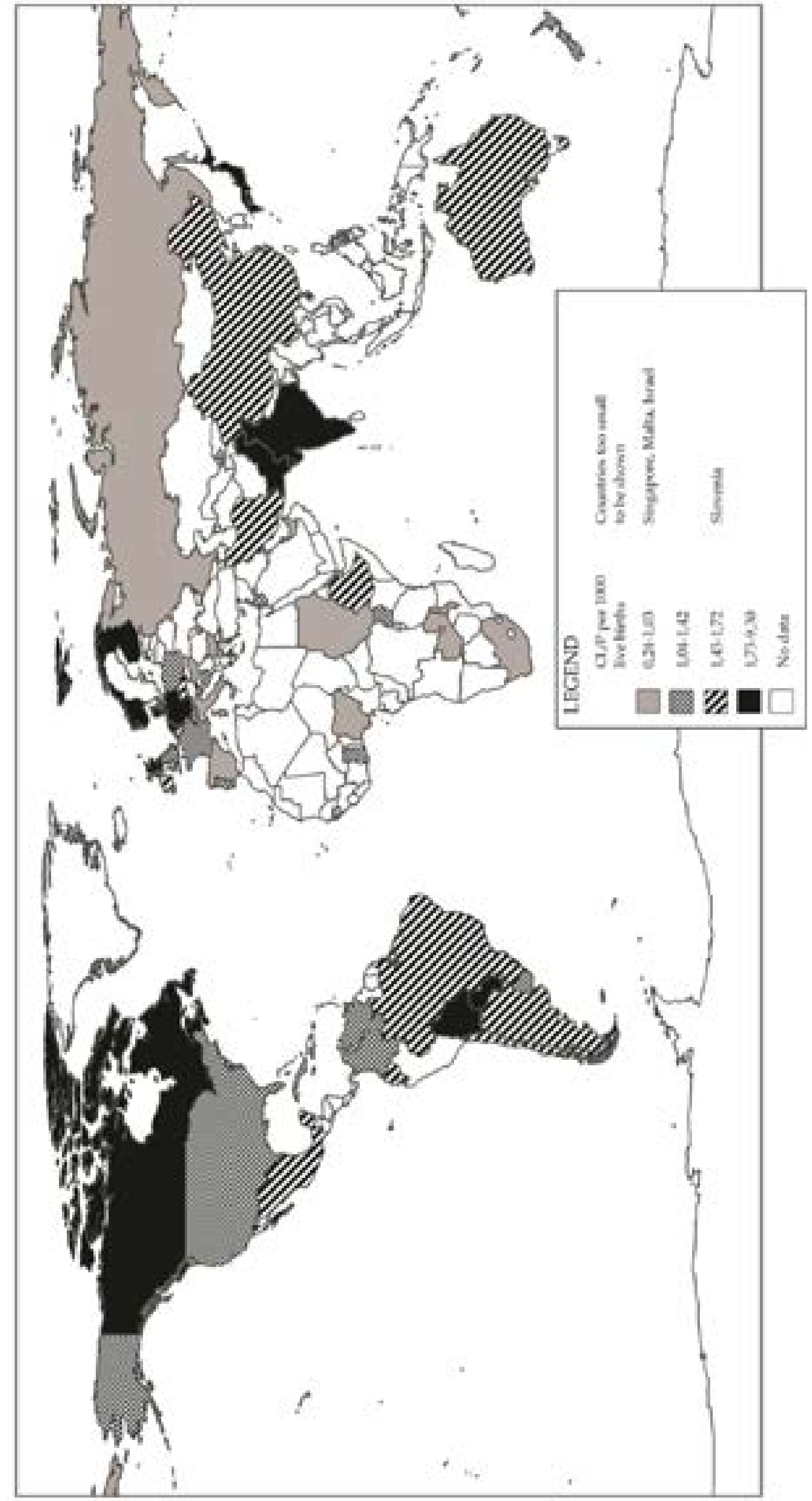

Figure 1.1 Birth prevalence of cleft lip and/or palate is presented for various countries worldwide. Classification is based on quartiles. The rates reported in following studies were included: *Tan (1988), Kozelj (1996), Murray et al. (1997), Msamati et al. (2000), Mossey & Castilla (2003), Al Omari & Al-Omari (2004), Elahi et al. (2004), Suleiman et al. (2005), Jamilian et al. (2007), Elliott et al. (2008) Dreise et al. (2011), Eshete et al. (2011), Agbenorku et al. (2013), Antoszewski & Fijałkowska (2013), Butali et al. (2013), Jahanbin et al. (2013), Kalanzi et al. (2013), Kalaskar et al. (2013).*

disabilities are defined as the outcome of a complex interaction between the patient's health condition and contextual factors *(World Health Organisation, 2002)*. As illustrated in Figure 1.2, this holistic framework does not just focus on the organic impairment of body structures (i.e. anatomical parts of the body) and functions (i.e. physiological and psychological functions of body systems). Implications on the patient's activities (i.e. execution of a task or action by an individual) and participation (i.e. involvement in a life situation) as well as the influence on the person itself (i.e. the special background of the individual) and his/her environment (i.e. the physical, social and attitudinal environment in which people live and conduct their lives) are also taken into account *(World Health Organisation, 2002)*. Consequently, interaction with attitudes of family, friends, neighbors, health professionals, teachers and strangers as well as social norms, practices and ideologies will partially influence the patient's disability *(Neumann & Romonath, 2012)*. Moreover, functional difficulties and aesthetical abnormalities potentially associated with CL/P might affect the extent of the patient's disability as they influence the patient's activities and participation (e.g. starting a conversation, forming relationships, education, employment). Therefore, observation of potential associated difficulties and abnormalities as well as of contextual factors is required to obtain the complete picture of the patient's disability.

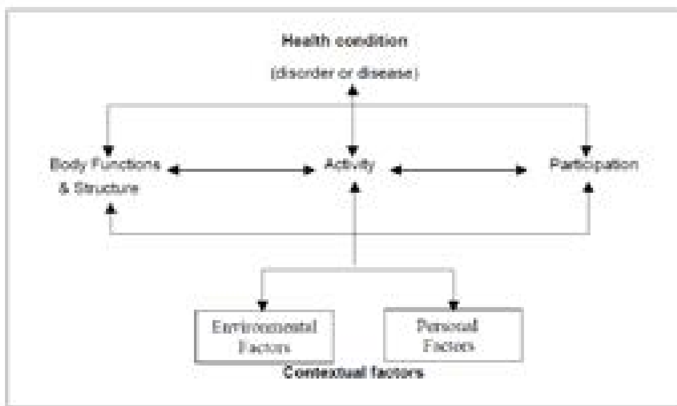

Figure 1.2 International Classification of Functioning, Disability and Health (ICF) as presented by the *World Health Organisation (2002).*

Implications of orofacial clefts

Babies born with an orofacial cleft will often have to contend with several problems and difficulties throughout their lives. In early infancy, feeding might be a first challenge to overcome, especially when an opening in the palate is involved. Severity of feeding problems seems to increase with the extent of the cleft and the presence of associated congenital anomalies *(Reid et al., 2007)*. Feeding difficulties particularly include the inability to suck and a disruption of the oral phase of swallowing *(Miller, 2011)*. Sucking disorders originate from failure to create negative intraoral pressure due to the coupling of the oral and nasal cavities *(Cubitt et al., 2012)*. Moreover, typically mentioned oral phase swallowing problems are increased feeding time lengths, nasal regurgitation and the consequential ulceration of the nasal mucosa, inadequate volume of oral intake and excessive air intake *(Bannister, 2001; Miller, 2011)*. In order to facilitate the feeding process, oral feeding facilitation techniques (e.g. adjusted positioning and liquid viscosity) as well as specialized feeding equipment (e.g. adjusted nipples and bottles) will often be required *(Miller, 2011)*.

Second, middle ear diseases such as otitis media with effusion and acute otitis media frequently occur in young children with CP and CLP up to 5-6 years due to failure of the Eustachian tube *(Goudy et al., 2006)*. Opening of the Eustachian tubes in order to equalize the middle ear pressure with the pressure of the environment as well as to drain middle ear fluid and secretions into the nasopharynx should be provided by contractions of the tensor veli palatini and levator veli palatini muscles, although the extent of contribution of the latter muscles is still uncertain *(Finkelstein et al., 1990; Ghadiali et al., 2010; Ishijma et al., 2002; Spauwen et al., 1991)*. In all young children, Eustachian tubes are oriented horizontally with the tensor veli palatini muscles laying in an unfavorable angle *(Willging & Kummer, 2008)*. Moreover, children with clefts of the palate show deviations in the anatomy of the Eustachian tube cartilages as well as abnormal insertion of the tensor veli palatini and levator veli palatini muscles, often leading to malfunctioning *(Lennox, 2001)*. The resulting accumulation of fluid in the aerated middle ear might cause a conductive hearing loss varying from 10 to 40 dB *(Lennox, 2001)*, what might result in speech and language difficulties *(Willging & Kummer, 2008)*.

A third frequently occurring consequence of particularly palatal clefts are speech difficulties. These speech problems, including predominantly abnormalities in articulation and resonance, are well-documented for patients with CL/P. An extensive overview of these disorders will be provided in Chapter 2.

In addition to the above-described functional disorders, facial aesthetics are usually affected in patients with orofacial clefts. Surgical closure of the CL will result in slight to severe scarification of the upper lip. Moreover, nasal deformities such as flattening of the alar rim and base at the cleft side may persist after primary correction of the cleft nose deformity *(Van Lierde et al., 2012)*. Furthermore, dental abnormalities are frequently reported in the cleft population *(Qureshi et al., 2012)*. These abnormalities consist in particular of developmental teeth agenesis, supernumerary teeth, displaced teeth and teeth malformations such as microdentia, fused teeth, enamel hypoplasia or taurodontism *(Al Jamal et al., 2010; Pegelow et al., 2012)*. The maxillary lateral incisors at the cleft side are the most vulnerable to be affected *(Baek & Kim, 2007)*.

Furthermore, dental (i.e. relationship of the teeth embedded in the jaws) and skeletal (i.e. relationship between the jaws) relationship are often altered in patients with orofacial clefts, resulting in malocclusions *(Peterson-Falzone et al., 2001)*. Crossbites of one or more teeth (i.e. maxillary elements are positioned lingually to the corresponding mandibular teeth) are the most commonly reported deviations from a normal dental arch form, presented in nearly all patients with clefts prior to orthodontic treatment (except for those with CL only) *(Peterson-Falzone et al., 2001; Semb & Shaw, 2001)*. Moreover, deviations of normal craniofacial growth, probably caused by a combination of intrinsic, iatrogenic and functional or adaptive factors *(Kreiborg et al., 2013)*, often result in hypoplasia of the maxilla and, consequently, in high incidence of class III malocclusion (i.e. mandibular first molar is positioned mesially relative to the maxillary first molar) *(Baek et al., 2002)*. These malocclusions may affect the production of specific phonemes such as apico-alveolar, labiodental and palatal consonants *(Bardach & Salyer, 1995; Campbell & Dock, 2008)*.

The afore-mentioned speech disorders, other functional anomalies and aesthetical abnormalities occurring in the CL/P population might induce the presence of psychosocial problems. Some studies found significantly more learning and especially behavioral

disorders as well as problems with self-concept, self-esteem, self-confidence, body image and satisfaction with facial appearance and speech in patients with CL/P compared to subjects without clefts *(Hunt et al., 2005)*. Moreover, *Wehby et al. (2012)* observed a significant association between low satisfaction with facial appearance and the risks for behavioral problems such as depression/anxiety, inattention/hyperactivity and somatic symptoms. In addition, according to *Ramstad et al. (1995)*, anxiety, depression and palpitations in subjects with CLP are twice as prevalent as in non-cleft control subjects and are strongly associated with appearance, dentition, speech and the desire for further treatment. Furthermore, the social life also seems to be influenced by the presence of a cleft. Literature revealed higher frequencies of school drop-out and unemployment, fewer friends and marriages as well as less participation in clubs and societies for patients with CL/P compared to non-cleft control subjects *(Hunt et al., 2005)*. The degree of psychosocial difficulties varies between patients. Although self-perception is the major factor influencing the level of psychological adjustment, peer interaction (e.g. teasing) as well as parental attitudes, expectations and degree of support substantially contribute to one's psychosocial functioning *(Sousa et al., 2009)*.

Due to this impact of orofacial clefts on physical, psychological and social health, the oral health-related quality of life in children and adults with CL/P seems to be diminished in comparison with non-cleft populations *(Foo et al., 2012; Ward et al., 2013)*. One important cause may be the presence of velopharyngeal insufficiency (VPI), which has been reported to be associated with significantly more negative quality of life scores compared to controls without VPI *(Barr et al., 2007)*. Nevertheless, increased satisfaction with treatment and treatment outcome seems to correlate with improved quality of life *(Munz et al., 2011)*. Therefore, in view of improving satisfaction with treatment and treatment outcome, a wide range of specialists (e.g. plastic and reconstructive surgeon, orthodontist, otorhinolaryngologist, speech-language therapist, maxillofacial surgeon, social worker, geneticist, dentist, psychologist, audiologist,…) needs to be involved, given the various interrelated problems associated with CL/P *(Hodgkinson et al., 2005)*. These specialists should not only make isolated evaluations within their own discipline. Interaction between disciplines is required as well *(Shprintzen, 1995)* in order to determine the appropriate timing and sequence of treatment, to optimize the functional, aesthetical and psychosocial outcome, and to improve as such the patient's quality of life *(Abarca et al., 2004)*.

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CHAPTER 2

SPEECH PRODUCTION IN PATIENTS WITH CLEFT LIP AND/OR PALATE

Activities and participation of patients with cleft lip and/or palate (CL/P) are influenced by the patients' communicative functioning and therefore by their speech intelligibility. Speech intelligibility can be defined by what the listener understands of the speakers' phonetic realizations *(Yorkston et al., 1996)*. It is "the product of a series of interactive processes such as phonation, articulation, resonance and prosody" with articulation being the strongest contributor *(De Bodt et al., 2002)*. Disorders of resonance, articulation and voice are frequently noted in this patient population. These disorders majorly result from oronasal fistulae and/or velopharyngeal insufficiency (i.e. an inconsistent and/or incomplete closure of the velopharyngeal valve caused by the anatomical defect and the associated functional loss *(Kummer, 2008b; Kummer, 2011b; Peterson-Falzone et al., 2006; Trost-Cardamone, 1989)*. The aim of the current chapter is to expound these specific resonance, articulation and voice abnormalities occurring in patients with palatal clefts.

Resonance disorders in patients with palatal clefts

Several types of resonance disorders arise in patients with cleft lip and palate or isolated cleft palate *(Harding & Grunwell, 1996; Henningsson et al., 2008; Kummer, 2008b; Kummer, 2011a; Peterson-Falzone et al., 2001; Peterson-Falzone et al., 2006)*. The resonance disorders frequently occurring in this patient population are described in the following paragraphs.

- Hypernasality is an extreme amount of acoustic energy resonating in the nasal cavities during the production of oral, voiced speech sounds *(Henningsson et al., 2008; Kummer, 2011a)*. Severity perception of this resonance disorder can be aggravated by a small mouth opening, high posterior tongue positioning, high respiratory effort as well as limited movement and/or increased tension of the articulators *(Peterson-Falzone et al., 2001)*.

- Nasal emission includes the release of airflow through the nasal cavities, which usually accompanies or substitutes all or certain plosives and fricatives *(Henningsson et al., 2008; Kummer, 2011a; Peterson-Falzone et al., 2006)*. When this airflow is forced through an increased resistance in the nasal airway (intranasal or at the velopharyngeal valve), a turbulent 'snorting' noise will arise, which is called nasal turbulence *(Kummer et al., 2003; Kummer et al., 1992; McWilliams et al., 1990; Trost, 1981)*.
- Hyponasality refers to a reduced proportion of acoustic energy resonating in the nasal cavities during the production of especially nasal speech sounds *(Henningsson et al., 2008; Kummer, 2008b; Peterson-Falzone et al., 2006)*. In patients with palatal clefts, hyponasality is usually caused by an obstruction in the nasopharynx or at the entrance of the nasal cavity (e.g. an unwanted effect following speech improving surgery) *(Peterson-Falzone et al., 2001)*.
- Cul-de-sac resonance arises when acoustic energy resonates in a cavity with a dead end *(Peterson-Falzone et al., 2006)*. In patients with palatal clefts, this 'muffled speech' generally results from the combination of velopharyngeal insufficiency and an obstruction in the anterior/ventral part of the nasal cavity (e.g. deviated septum, stenotic nares) *(Kummer, 2008b)*. Moreover, this resonance disorder can be caused by a deep retraction of the tongue into the oral cavity *(Peterson-Falzone et al., 2001)*.
- Finally, mixed resonance is characterized by elements of at least two of the afore-mentioned resonance disorders (e.g. hyper/hyponasality) *(Kummer, 2011a)*.

Of all listed types of resonance disorders, hypernasality and nasal emission/turbulence are the most frequently observed in patients with palatal clefts.

Articulation disorders in patients with palatal clefts

Articulation includes the motor movement of articulators in order to obtain the correct distinctive features for the production of certain speech sounds *(D'Antonio & Scherer, 2008)*. When a child is unable to correctly produce specific age-expected speech sounds due to inappropriate motor movements, phonetic articulation errors occur *(Chapman, 1993)*. These errors might manifest as omissions, substitutions or distortions. Phonetic articulation

errors are frequently seen in patients with palatal clefts. Given that patients with palatal clefts often show difficulties to obtain sufficient positive intraoral air pressure due to the presence of velopharyngeal insufficiency and/or oronasal fistulae *(Kummer, 2011a)*, fricatives, affricatives (English language) and plosives are the most commonly affected speech sounds in this patient population *(Harding & Grunwell, 1996; Henningsson et al., 2008; Peterson-Falzone et al., 2001)*. Release of air through the nose during the production of these speech sound categories obligatorily leads to production of weak consonants and might even result in nasal consonant realizations *(Harding & Grunwell, 1996; Henningsson et al., 2008; Peterson-Falzone et al., 2006)*. Moreover, in order to produce sufficient intraoral air pressure, compensatory articulation regularly occurs *(Harding & Grunwell, 1998; Henningsson et al., 2008; Kummer, 2008b; Kummer, 2011a; Peterson-Falzone et al., 2006; Trost, 1981)*. During this phonetic articulation error, the place of articulation of native speech sounds is moved posteriorly, while the manner of articulation is maintained. Although constrictions are often performed inferiorly to the velopharyngeal valve (glottal or pharyngeal), backing of consonants to an oral place (velar or palatal) is also common. Several compensatory articulation errors can be distinguished based on the place and manner of articulation, (i.e. glottal stop [ʔ], glottal fricative [h], voiced/unvoiced pharyngeal stop [ʡ̬,ʡ], voiced/unvoiced pharyngeal fricative [ʕ,ʢ], voiced/unvoiced velar stop [g,κ], voiced/unvoiced velar fricative [ɣ,χ], voiced/unvoiced mid-dorsum palatal stop [ʒ,ɛ], voiced/unvoiced mid-dorsum palatal fricative [ʝ,ç]) *(Henningsson et al., 2008; Peterson-Falzone et al., 2006; Trost, 1981)*.

In addition to the frequent occurrence of phonetic articulation errors, patients with palatal clefts are at risk for development of linguistic phonological disorders *(Peterson-Falzone et al., 2001)*. Phonological disorders involve difficulties in the acquisition and use of meaningful contrasts between speech sounds resulting in age-inappropriate patterns of speech sound errors *(Chapman, 1993)*. These patterns can roughly be classified in syllable structure processes, substitution processes and assimilation processes *(Ingram, 1982)*. Young children with palatal clefts generally show similar phonological processes compared to typically developing children *(Peterson-Falzone et al., 2001)*. However, delays in phonological development are noted. These delays are usually caught up at the age of 4-5 years in case of successful palatal repair *(Chapman, 1993)*. However, in some children, early compensatory errors may become integrated in the developing phonological system resulting in sound preferences and persisting phonological processes *(Harding & Grunwell, 1996; Peterson-Falzone et al., 2001; Peterson-Falzone et al., 2006)*.

Voice disorders in patients with palatal clefts

Whether voice disorders occur more frequently in patients with CL/P compared to non-cleft populations is unclear. Some recently published studies found prevalence rates of voice disorders within the normal range (i.e. 6% to 34%) for patients with palatal clefts *(Hamming et al., 2009; Robinson, 2011)*. However, others conclude that voice disorders are considerably more common in this population compared to non-cleft subjects *(D'Antonio & Scherer, 2008; Peterson-Falzone et al., 2001; Van Lierde et al., 2004a)*.

Hoarseness, breathiness, low intensity and abnormal pitch are voice symptoms regularly noticed in patients with palatal clefts *(D'Antonio & Scherer, 2008)*. Hoarseness can be related to hyperadduction of the vocal folds secondary to compensatory articulation errors *(Kummer, 2008b)*. This hyperfunctional vocal use can subsequently cause organic voice disorders such as vocal fold thickening, vocal nodules and vocal fold inflammation *(D'Antonio & Scherer, 2008)*. Alternatively, hoarseness sometimes originates from congenital laryngeal anomalies related to syndromic clefts *(D'Antonio & Scherer, 2008; Kummer, 2008b)*. In contrast to hoarseness, the 'soft voice syndrome' including breathiness and reduced intensity, is a strategy to compensate for hypernasality and nasal emission rather than a secondary symptom, considering that lower subglottic pressure will minimize the presence of such resonance disorders *(Kummer, 2011a; Peterson-Falzone et al., 2001; Peterson-Falzone et al., 2006)*.

Factors influencing speech intelligibility in patients with palatal clefts

Patients with orofacial clefts form a very heterogeneous population. Variability in clinical expression (presence, type and severity) of speech disorders influencing the patients' overall speech intelligibility is huge. Some patients show normal resonance, articulation and voice, while others present with severe disorders affecting all aspects of speech intelligibility and resulting in unintelligible speech. According to literature, several factors may influence speech characteristics of patients with palatal clefts *(D'Antonio & Scherer, 2008; Kummer, 2008a; Mahoney et al., 2013; Peterson-Falzone et al., 2001; Peterson-Falzone et al., 2006; Sell & Grunwell, 2001)*.

These factors can be categorized in patient-related, treatment-related and environmental factors. An overview is provided in Table 2.1. Numerous studies compared speech of patients differing regarding one or more potential influencing factors. *Van Lierde et al. (2004b)*, for example, assessed speech characteristics of 2 groups of patients with different treatment-related characteristics. Fourteen patients had their soft palate repaired at 14 months on average by the Furlow technique; the hard palate was closed at a mean age of 8.10 years. Seventeen patients underwent a one-stage soft and hard palatal repair by the Wardill-Kilner procedure at 1.4 years on average. Both groups were treated by two different experienced surgeons. Statistical comparison revealed significantly more hypernasality and higher nasalance values for the Furlow-group, despite similar articulation and voice characteristics. Notwithstanding this large amount of studies assessing the influence of several variables on speech, it is still impossible to predict the speech characteristics that can be expected in a specific individual with an orofacial cleft. An unambiguous correlation between the contributing factors and speech outcome is not yet established. The final speech outcome will probably result from a complex interaction of several influencing factors. Consequently, when assessing the relationship between one predicting factor (e.g. age at palatal repair) and speech characteristics of a patient group with orofacial clefts, as many other potential influencing factors as possible should be controlled.

Table 2.1 Overview of the patient-related, treatment-related and environmental factors influencing the speech characteristics of patients with palatal clefts.

Patient-related factors	Treatment-related factors	Environmental factors
- Extent of the cleft - Palatal cleft width - Length of the soft palate - Velopharyngeal functioning - Middle ear diseases and resulting hearing status - Dental-occlusal status - Associated anomalies - Cognition - Chronological age - Motivation - …	- Efficacy of palatal repair (scarring and stiffness) - Age at primary palatal repair - Surgical technique - Surgeon's skill/experience - Remaining palatal fistulae - Secondary speech-improving surgery - Sensation in alveolar region despite scar tissue - Presence, timing, amount & efficacy of speech therapy - Treatment of middle ear diseases - Orthodontic correction of tooth position - …	- Socioeconomic status of the family - Linguistic status of the family - Parental acceptance - Parental stimulation - Parental motivation - …

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CHAPTER 3

TIMING OF PRIMARY CLEFT CLOSURE IN PATIENTS WITH CLEFT LIP AND/OR PALATE

The influence of age at primary cleft closure in patients with cleft lip and/or palate (CL/P) was subject of discussion during the previous decades *(Peterson-Falzone, 1996; Rohrich et al., 2000)*. Still, literature provides no consensus regarding the optimal timing of cleft closure. In the current chapter, the existing variety of surgical timing protocols will shortly be outlined. Moreover, a literature review will be provided regarding three types of surgical timing protocols: synchronous lip and palatal closure, soft and hard palatal closure prior to 6 months of age and primary palatal repair in youngsters and adults. This literature review will particularly focus on speech outcomes. Nevertheless, the importance of potential surgery-induced facial growth disturbances should not be underestimated. Although adults with untreated clefts seem to have potential for normal maxillary growth *(Mars & Houston, 1990)*, scar tissues and pressure forces resulting from lip and palatal repair performed in infancy or early childhood will particularly cause disturbances of the antero-posterior facial growth, what will lead to some extent of reduced maxillary length and maxillary retrusion, including smaller ANB and SNA angles *(Mars, 2008)*. Additionally, limitations in vertical (e.g. inclination of the maxillary plane relative to the anterior cranial base) and transverse (e.g. alveolar arch collapse) facial growth may occur as well *(De Mey et al., 2006; Rohrich et al., 2000)*.

In this chapter and in the entire doctoral thesis, univocal terminology will be used. Synchronous lip and palatal closure will refer to repair of the cleft lip, soft palate and hard palate during a single surgery in patients with cleft lip and palate (CLP). One-stage and two-stage palatal closure will indicate closure of the soft and hard palate during respectively one or two surgical intervention(s).

A multitude of surgical sequences and timing protocols

During the previous decades, surgeons and speech therapists were searching for the optimal timing to close the cleft lip and/or palate in order to obtain the best possible facial aesthetics and functions (i.e. feeding, hearing and speech) *(Peterson-Falzone, 1996; Rohrich et al., 2000)*. Unfortunately, it takes at least one or two decades to determine the effectiveness of surgical management in patients with CL/P considering that final speech and facial growth outcomes can merely be assessed in youngsters or even adults *(Berkowitz, 2013; Shaw et al., 2001)*. Moreover, research in this field is impeded by the large number of variables that are difficult to control such as physical variability in clefts, variation in the surgeon's skill and experience, lack of standardization of speech evaluations and the complexity of factors influencing the child's maturation, growth and development *(Peterson-Falzone, 1996)*. Consequently, no consensus yet exists concerning the optimal timing of surgical lip and palatal repair. As many different surgical sequences and timing protocols are still utilized worldwide as there are variations that can be applied *(Berkowitz, 2013)*. In Belgium, for example, various cleft teams use different timing protocols to close cleft lip and palate. The craniofacial team of the Ghent University Hospital opts for lip closure at the age of 3-4 months and one-stage repair of the soft and hard palate at 12 months on average. Furthermore, the craniofacial team of the University Hospital Saint-Luc in Brussels choose to perform neonatal lip closure followed by one-stage repair of the soft and hard palate at 3 months on average *(Castelein et al., 2006)*, while in the Queen Fabiola Children's University Hospital in Brussels, synchronous repair of the lip and (soft and hard) palate at a mean age of 3 months is preferred *(De Mey et al., 2009)*. However, the surgeons of the craniofacial teams in the Academic Hospital Monica in Antwerp *(Nadjmi et al., 2013a; Nadjmi et al., 2013b)*, the General Hospital Sint-Jan in Bruges and East-Limburg Hospital in Genk close the palate during a two-stage procedure (Antwerp: soft palate at 9 months and hard palate at 18 months; Bruges: soft palate at 12 months and hard palate at 48 months on average; Genk: soft palate at 9-15 months and hard palate at 48-60 months) after early lip repair. Moreover, in the Leuven University Hospital, the lip and hard palate are closed at a mean age of 3 months prior to soft palatal repair at about 1 year *(UZ Leuven, 2013)*. Similarly, the Eurocleft Project reported in 2001 that 201 centers in 30 European countries used 194 different surgical timing protocols and 17 sequences of operations to repair the lip, palate

and alveolus in patients with complete unilateral cleft lip and palate (UCLP) *(Shaw et al., 2001)*. The number of surgical interventions ranged from one to four whether or not combined with presurgical orthopaedics. The majority of the teams (135/201, 67%) closed the lip first during one or two surgeries followed by a one- or two-stage closure of the soft and hard palate.

Lip repair is performed by most craniofacial teams after 3 months of age *(Berkowitz, 2013)*. At this age, the risks for anesthesia are reduced and the lip and nose are sufficiently large to facilitate the detailed surgery *(Watson, 2001)*. However, some cleft teams opt for neonatal closure of the cleft lip *(Goodacre et al., 2004; Harris et al., 2010)*. Proponents associate neonatal lip repair with very good wound healing resulting in good aesthetics concerning lip scars and appearance of the nose as well as with lower needs for secondary lip correction *(Jiri et al., 2012)*. Moreover, feeding would be facilitated *(Jiri et al., 2012)* and less early mother-infant interaction difficulties would occur leading to improved cognitive outcomes at 18 months of age *(Murray et al., 2008)*. Conversely, opponents warn for immature cardiovascular and respiratory systems in neonates and the presence of associated congenital abnormalities that might still be undiscovered prior to surgery *(Watson, 2001)*.

The age to perform soft and hard palatal repair forms an even greater discussion in literature. Timing of this surgical closure depends on the preferences of the craniofacial team to emphasize (1) achievement of the best possible speech outcome, (2) maximal avoidance of inhibited facial growth disturbance or (3) both *(Berkowitz, 2013; Kaplan, 1981; Peterson-Falzone, 1996; Rohrich et al., 2000; Watson, 2001)*. Early closure of the soft and hard palate provides better chances for good speech abilities, although the risk for midfacial growth inhibition is larger *(Rohrich et al., 2000)*. This view is currently pursued by the craniofacial team of the Ghent University Hospital. Conversely, while accepting poorer speech results, some craniofacial teams do not provide palatal surgery until the maxillofacial growth is almost completed in order to minimize the facial growth disturbance *(Berkowitz, 2013)*. The middle path between both approaches consists of delayed hard palatal repair to avoid facial growth deformities as well as early soft palatal closure assuming that adequate speech is still achieved *(Nollet et al., 2005)*. All three approaches have supporters (e.g. *Rousseau et al. (2013), Paliobei et al. (2005), Reiser et al. (2013)*). However, within one approach, opinions vary on how early or how late surgery needs to be performed to enable the best possible speech or maxillofacial growth outcome as well as on the necessity to perform a one-stage or two-stage closure of the soft and hard palate.

Synchronous lip and palatal closure

Few craniofacial teams choose to close the cleft lip and (soft and hard) palate during a single operation. According to the Eurocleft Project, 10 out of 201 (5%) European teams included in the study performed a synchronous lip and palatal repair in patients with UCLP. Such synchronous lip and palatal repair can be applied in various ways *(Mueller et al., 2012)*. Similarly to other surgical approaches, differences can be noted in age at cleft closure, surgical technique and use of orthopaedic appliances.

Several authors provided a rationale for performing synchronous lip and palatal repair. First, this timing protocol seems to have positive psychosocial effects on the child and his/her parents. Multiple operations and hospitalizations are stressful for a family *(De Mey et al., 2009; Honigmann, 1996; Mueller et al., 2012; Savaci et al., 2005)* and might result in overprotective and overattentive parents as well as in anxious children *(Kaplan et al., 1974; Kaplan et al., 1980).* Support for this reasoning is provided by *Kramer et al. (2008)*, who reported that the number of surgeries correlated negatively with the child's psychological well-being. Second, the young children are spared from multiple anesthetics which reduces the potential medical risks *(Davies, 1966; De Mey et al., 2006)* as well as the risks for development of learning disabilities *(Wilder et al., 2009)*. Third, given that only one dissection is performed on an unscarred operating area, synchronous lip and palatal repair is associated with improved wound healing and lower incidences of fistulae and palate dehiscence *(Honigmann, 1996; Savaci et al., 2005)*. Finally, by reducing the number of surgeries and hospitalization periods, treatment costs and hospitalization expenses are minimized *(De Mey et al., 2009; Savaci et al., 2005)*.

Synchronous lip and palatal closure is considered to be a safe surgical procedure *(Hodges, 2010; Schwarz, 2006)*. Recently published studies on craniofacial morphology *(Corbo et al., 2005; De Mey et al., 2009; De Mey et al., 2006; Fudalej et al., 2010; Mueller et al., 2012; Savaci et al., 2005)*, dental arch relationship *(Corbo et al., 2005; Fudalej et al., 2009a; Fudalej et al., 2011)* and nasolabial aesthetics *(Fudalej et al., 2009b)* showed relatively favorable outcomes for this surgical protocol compared to results

following lip and palatal closure performed by multiple surgical interventions. Speech results after synchronous lip and palatal repair have however not attracted much attention from researchers. To our knowledge, 5 studies mentioned the impact of synchronous lip and palatal repair on speech *(Honigmann, 1996; Kaplan et al., 1974; Kaplan et al., 1980; Mueller et al., 2012; Schwarz, 2006)*. These studies broadly evaluated speech *(Honigmann, 1996; Kaplan et al., 1974; Kaplan et al., 1980)* or described only few aspects *(Mueller et al., 2012; Schwarz, 2006)*. Moreover, information about the speech assessment strategies, the chronological age at speech assessment and the composition of the cleft or non-cleft comparison groups is generally lacking. In addition, 2 studies systematically evaluated speech outcomes following synchronous lip and palatal repair *(De Mey et al., 2006; Hortis-Dzierzbicka et al., 2012)*. An overview of all studies is presented in Table 3.1. *De Mey et al. (2006)* could neither reveal significant differences for intelligibility at the age of 6 years, nor for nasality or articulation at 3 and 6 years of age between the experimental group and an age-matched CLP group who underwent soft palatal closure at 3 months on average and lip and hard palatal repair at a mean age of 6 months. However, literature provides some evidence that synchronous lip and palatal repair in early childhood overall leads to adequate speech production *(Honigmann, 1996; Hortis-Dzierzbicka et al., 2012; Kaplan et al., 1980; Mueller et al., 2012)*. *Honigmann (1996)* reported that one third (10/31, 33%) of the patients did not require speech therapy after synchronous lip and palatal repair before 12 months of age compared to all patients (108/108, 100%) who received early lip and soft palatal closure followed by later hard palatal repair. Furthermore, *Kaplan et al. (1980)* noted that 95% (20/21) of the patients showed excellent or satisfactory speech following synchronous closure of the CLP. Regarding resonance, *Mueller et al. (2012)* obtained no significant differences in mean nasalance values between the experimental group and the age- and gender-matched non-cleft control group. Moreover, *Hortis-Dzierzbicka et al. (2012)* described absence of hypernasality and nasal emission in respectively 89% (25/28) and 61% (17/28) of the patients after synchronous lip and palatal repair at 9 months on average. In the same study, compensatory articulation errors occurred in only 2 patients (2/28, 8%).

Table 3.1 Overview of the studies reporting speech results of patients with cleft lip and palate following synchronous lip and palatal repair.

Author	n	Cleft type	Age at CLP closure	Age at speech assessment	Comparison group(s)	Speech assessment	Speech results
Kaplan et al. (1974) (Vietnam)	13	UCLP BCLP	3-4m	n.a.	***Cleft***: 13 CP or CLP *CLP closure*: One-stage palatal repair at 10-12m *Matching*: n.a. ***Non-cleft***: n is n.a. *Matching*: n.a.	n.a.	Comparisons between experimental group (EG), cleft comparison group (CCG) and non-cleft control group (NCG): ***Babbling***: EG normal ↔ CCG delayed; ***First sounds (nasals, sibilants)***: EG normal ↔ CCG delayed; ***One-word response***: EG normal ↔ CCG delayed; ***Two- to three-word response***: EG delayed, but earlier than in CCG; ***Initial nasal sounds***: EG 40% adequate and 60% comparable with CCG.
Kaplan et al. (1980) and **Kaplan et al. (1982)** (Vietnam)	28	CLP	3-8m (mean 4m)	1-7y	none	n.a.	***Preverbal vocalization***: 71% (5/7) normal, 29% (2/7) dull mumbling; ***Nasality, phonation, intelligibility and fluency of speech***: 52% (11/21) excellent, 43% (9/21) satisfactory, 5% (1/21) poor speech because of delay and nasality.
Honigmann (1996) (Switzerland)	31	UCLP BCLP	3-12m (median 6m)	n.a.	***Cleft***: 108 (cleft type n.a.) *CLP closure*: Lip + soft palate, later hard palate *Matching*: n.a.	n.a.	***Speech therapy***: Experimental group: 33% (10/31) not required (no nasality, suprapalatal resonance or nasal grimace) ↔ cleft comparison group: 100% (108/108) required.
De Mey et al. (2006) (Belgium)	18	UCLP	2.8-3.1m (mean 3m)	3y and 6y	***Cleft***: 26 UCLP *CLP closure*: Soft palate: 2.8-3.2m (mean 3m) Lip + hard palate: 5.5-6.8m (mean 6m) *Matching*: Age	*Speech samples:* Spontaneous & standardized speech assessment in French or Flemish *Perceptual rating by one senior speech therapist:* - Intelligibility - Nasality - Articulation	***Intelligibility***: *At 3 years*: significant group differences (p<0.05). EG: 63% good, 37% mild deficiency, 0% major problem ↔ CCG: 52% good, 32% mild deficiency, 18% major problem. *At 6 years*: no significant group differences. 80% normal intelligibility; ***Nasality***: No significant group differences. *At 3 years*: 30% no nasality problems, 50% inconstant nasality, 20% constant nasality. *At 6 years*: 40% no nasality problems, <10% constant nasality; ***Articulation***: No significant group differences. *At 3 years*: 20% important disorders; *At 6 years*: 40% normal articulation; ***Velopharyngoplasty required***: EG: 6% (1/18) at 5.5y ↔ CCG: 12% (3/26) at mean age of 5y.
Schwarz (2006) (Nepal & Tibet)	4	CLP	10-25y (mean 15y)	10;9-25;10y (mean 16y)	none	*Speech samples:* n.a. *Perceptual evaluation:* Cleft Audit Protocol for Speech and Great Ormond Street Speech Assessment: - Intelligibility - Nasality	***Speech intelligibility***: 25% (1/4) poor, 50% (2/4) fair, 25% (1/4) good; ***Nasality***: 25% (1/4) moderate nasality, 50% (2/4) mild nasality, 25% (1/4) mixed hyper/hyponasality.

Hortis-Dzierzbicka et al. (2012) (Poland)	28	UCLP	6-13m (mean 9m)	9.1-10.5y (mean 10y)	none	*Speech samples:* - Syllables - Short balanced utterances - Counting to 10 *Perceptual rating of speech by one phoniatrician and one speech therapist:* - Hypernasality - Nasal emission - Articulation - Facial grimace	***Hypernasality***: 89% (25/28) absent, 7% (2/28) mild, 4% (1/28) severe; ***Nasal emission***: 61% (17/28) absent, 39% (11/28) present; ***Articulation***: *Compensatory articulations*: 93% (26/28) absent, 4% (1/28) oral backing, 4% (1/28) pharyngeal compensations; *Minor articulation disorders*: 43% (12/28) absent, 57% (16/28) present (particularly interdental and lateral realizations); ***Facial grimace***: 64% (18/28) absent, 29% (8/28) inconsistent or mild, 7% (2/28) severe; ***Overall speech***: 54% (15/28) entirely normal.
Mueller et al. (2012) (Switzerland)	15	UCLP	mean 7m	6-18y (mean 13y)	***Non-cleft***: n: n.a. *Matching:* Age and gender	*Speech samples:* 4 oral sentences *Objective assessment:* Nasometry II 6400	***Nasometry:*** No significant differences in mean nasalance values compared to the age- and gender-matched non-cleft group.

CLP: cleft lip and palate, UCLP: unilateral cleft lip and palate, BCLP: bilateral cleft lip and palate, n.a.: not available, EG: experimental group, CCG: cleft comparison group, NCG: non-cleft control group

By interpreting these results, one should bear in mind that differences are noted regarding age at CLP closure, surgical techniques, age at speech assessment and speech assessment strategies, what might have influenced the children's speech abilities. When, in addition, the several limitations of these studies are taken into account, it can be concluded that further research regarding speech results after synchronous lip and palatal repair is required.

Speech following primary palatoplasty prior to 6 months of age

As described earlier in the current chapter, it is generally believed that early closure of the cleft (soft and hard) palate provides better chances for adequate speech abilities. However, no consensus exists about how early primary palatoplasty must be completed to optimize the chances for normal speech. Several authors compared speech results after two different surgical timing protocols, what often resulted in improved speech outcomes for the earliest closure group. *Hardin-Jones & Jones (2005)*, for example, compared resonance and articulation characteristics of 192 children with various cleft types that were categorized in three groups based on age of palatal closure: ≤ 13 months (n=61), 13 to 17 months (n=66) or ≥ 18 months (n=65). Although no significant group differences were noted regarding the presence of compensatory articulation errors (i.e. backing to a glottal or pharyngeal place of articulation), the percentage of patients with moderate to severe hypernasality significantly increased with age at palatal repair. *Willadsen (2012)* compared early speech development of two groups of 17 children with UCLP following soft palatal closure at 4 months of age and hard palatal repair at the age of 12 or 36 months. Moreover, comparison was made to an age- and gender-matched non-cleft control group. Significantly better outcomes were noted for the early compared to the late hard palatal closure group regarding lexical and phonological development, vocalization and articulation.

When early speech development of infants with palatal clefts is compared to that of typically developing children, differences can be identified even as early as 6 months of age *(Peterson-Falzone et al., 2006)*. Between 6 and 10 months of age, typically developing children start to produce canonical babbles *(Chapman et al., 2001)*. During this period, babies generate alterations of consonants and vowels, what provides the opportunity to practice sounds and sound sequences in order to develop control and precision required for the production of words *(Stout et al., 2011)*. Babies with palatal clefts frequently show delayed onsets and lower frequencies of canonical babbling compared to typically developing children *(Chapman et al., 2001; Peterson-Falzone et al., 2006; Scherer et al., 2008; Stout et al., 2011)*. Furthermore, differences in composition of the consonant inventory are noted *(Chapman et al., 2001; Peterson-Falzone et al., 2006; Scherer et al., 2008; Stout et al., 2011)*. The consonant inventories of babies with palatal clefts are smaller and contain more consonants which require little or no intraoral air pressure (e.g. nasals, glides, liquids). Moreover, more glottal stops and bilabial consonants as well as fewer velar and alveolar consonants are observed *(Chapman et al., 2001; Peterson-Falzone et al., 2006; Scherer et al., 2008; Stout et al., 2011)*. Consonants that frequently occur in canonical babbling will also arise in the children's first words *(Peterson-Falzone et al., 2006)*. Consequently, the speech sound limitations observed in babies with palatal clefts during the first year of life will most likely affect the children's later speech and language performance *(Scherer et al., 2008)*.

Some authors hypothesized that these speech disorders occurring in later life can be avoided when the soft and hard palate are closed prior to 6 months of age *(Abdel-Aziz, 2013; Copeland, 1990; De Mey et al., 2006; Desai, 1983; Doucet et al., 2013; Grobbelaar et al., 1995; Kaplan, 1981)*. As such, the velopharyngeal sphincter is adequate at the start of the canonical babbling period. Consequently, the typical deviations observed in patients with palatal clefts during the prelinguistic speech development might fail to occur and cannot be integrated in the neuromuscular speech patterns, what might result in absence of compensatory speech disorders later in childhood *(Kaplan, 1981; Randall et al., 1983; Ysunza et al., 1998)*. Prior to application of this surgical timing protocol, maxillofacial growth should be considered as well. Although, to our knowledge, no long-term maxillofacial growth studies have yet been performed after palatal closure prior to 6 months of age, *Ysunza et al. (1998)* reported similar maxillofacial growth disturbances at the age of 4 years when palatal repair is performed at 6 or 12 months of age.

To our knowledge, 12 articles, 8 descriptive *(Abdel-Aziz, 2013; Barimo et al., 1987; Copeland, 1990; De Mey et al., 2006; Desai, 1983; Kaplan et al., 1982; Kaplan et al., 1980)* as well as 4 comparative studies *(Doucet et al., 2013; Grobbelaar et al., 1995; Kaplan et al., 1974; Ysunza et al., 1998)*, reported speech results of patients with palatal clefts following surgical closure of the cleft soft and hard palate prior to 6 months of age. An overview of these studies is provided in Table 3.2. Overall, relatively good speech outcomes were reported for patients treated with this timing protocol. Speech intelligibility

Table 3.2 Overview of the studies reporting speech results in patients with cleft lip and/or palate following (soft and hard) palatal closure prior to 6 months of age.

Author	n and cleft type	Palatal closure: age and surgical technique	Age at speech assessment	Control group	Speech assessment	Speech results
Kaplan et al. (1974) (Vietnam)	(a) 13 UCLP/ BCLP (b) 13 UCLP/ BCLP/ CP	*Age:* 3-4m *Age* : 10-12m *Surgical technique:* von Langenbeck	n.a. n.a.	yes, matched for age	n.a.	Comparisons between Group A (Ga), Group B (Gb) and control group: ***Babbling***: Ga normal vs. Gb delayed; ***First sounds (nasals, sibilants)***: Ga normal vs. Gb delayed; ***One-word response***: Ga normal vs. Gb delayed; ***Two- to three-word response***: Ga delayed, but earlier than in Gb; ***Initial nasal sounds***: Ga 40% adequate and 60% comparable with Gb.
Kaplan et al. (1980) and **Kaplan et al. (1982)** (Vietnam)	28 UCLP/ BCLP	*Age:* 3-8m (mean 4m) *Surgical technique:* von Langenbeck	1-7y	no	n.a.	***Preverbal vocalization***: 71% (5/7) normal, 29% (2/7) dull mumbling; ***Nasality, phonation, intelligibility and fluency of speech***: 52% (11/21) excellent, 43% (9/21) satisfactory, 5% (1/21) poor speech because of delay and nasality.
Desai (1983) (UK)	100 UC(L)P/ BC(L)P	*Age:* ≤ 16w *Surgical technique:* 95 Wardill-Kilner 5 von Langenbeck	n.a.	no	n.a.	***Hypernasality***: 0% (0/100); ***Pharyngoplasty***: 0% (0/100)
Randall et al. (1983) (USA)	17 (cleft type n.a.)	*Age:* 3-7m *Surgical technique:* n.a.	>2y	no	*Perceptual analysis by one speech therapist:* - Resonance - Articulation - Vocal quality	***Resonance***: 24% (4/17) hypernasality and 12% (2/17) hyponasality; ***Articulation***: 65% (11/17) normal; ***Vocal quality***: 65% (11/17) normal; ***Secondary pharyngoplasty***: required in 12% (2/17).

Barimo et al. (1987) (USA)	22 BCLP/ UCLP/ CP	*Age:* 3-8m (mean 6m) *Surgical technique:* Complete reconstruction of the velopharyngeal sling with interdigitation of the muscles and minimal dissection of the mucoperiosteum	1;10-8;11y	yes, matched for age	*Speech samples:* 1;10-2;6y: Phonological data collected from spontaneous utterances 3;1-8;11y: Templin-Darley Screening Test of Articulation 3;1-5;11y: Bzoch Error Pattern Screening Articulation Test	*Articulation*: 1;10-2;6y (n=5): Absence of nasal emission, glottal stops and pharyngeal fricatives. Age appropriate phoneme type and frequency. 3;1-8;11y (n=17): 0% failed the screening articulation test. 3;1-5;11y (n=8): 89% of consonants correct articulation, 8% substitution errors, 0% distortion errors, 2% omission errors. Absence of glottal stops and pharyngeal fricatives.
Copeland (1990) (UK)	53 UCLP 17 BCLP 14 CP 16 soft CP	*Age:* 9-25w (mean 16.4w) *Surgical technique:* 92 Wardill-Kilner 8 von Langenbeck	3.8-6.3y (mean 5.5y)	no	*Speech samples:* Edinburgh Articulation Test, conversational sample based on picture description, counting from 1 to 10 *Perceptual rating by one speech therapist:* - Intelligibility - Nasal resonance - Articulation	***Intelligibility:*** 87% (87/100) acceptable speech, 13% (13/100) unacceptable speech; ***Nasal resonance and nasal emission***: 81% (81/100) normal resonance and no nasal emission or minimal degrees of one of these, 93% (93/100) normal/minimal abnormal resonance, 7% (7/100) moderate/severe abnormal resonance, 91% (91/100) normal/minimal nasal emission, 9% (9/100) moderate/severe nasal emission; ***Articulation***: 57% (57/100) normal or some immature features, 6% (6/100) very immature phonology, 36% (36/100) atypical errors.

Grobbelaar et al. (1995) (South-Africa)	184 soft CP	*Age:* 2w – 18m (median 6.2m) *Surgical technique:* 25 Dorrance 41 Wardill 19 Perko 20 Furlow Z-plasty 79 von Langenbeck	4-24y (mean 9.6y)	no	*Perceptual consensus rating:* - Intelligibility - Resonance - Articulation Perceptual evaluation of velopharyngeal competence confirmed by consensus evaluation of videofluoroscopy and/or nasoendoscopy	***Speech*:** Palatal repair ≤ 6 months: 95% (73/77) normal* ↔ palatal repair > 6 months: 92% (98/107) normal* (*normal resonance, articulation and intelligibility); ***Velopharyngeal inadequacy (VPI)*:** Palatal repair ≤ 6 months: 5% (4/77) VPI ↔ Palatal repair > 6 months: 8% (9/107) VPI; Significant differences in speech and less velopharyngeal incompetence after palatal repair before vs. after 6 months of age (except for the Dorrance repair).
Ysunza et al. (1998) (Mexico)	(a) 41 UCLP (b) 35 UCLP	*Age:* 12m *Age:* 6m *Surgical technique:* Minimal incision palatopharyngo-plasty	4y 4y	yes, no info about matching available	*Clinical assessment by two speech therapists:* - Nasal resonance - Spanish articulation test Consensus diagnose based on video-nasopharyngoscopy and multiview videofluoroscopy	***Articulation scale***: Below normal limits for the patient's age in all cases. Significantly better in Group B vs. Group A; ***Velopharyngeal insufficiency*** *(VPI, p>0.05)*: Group A: 20% (8/41) VPI ↔ Group B: 17% (6/35) VPI; ***Compensatory articulation errors (CAE) in patients with VPI*** *(p<0.05)*: Group A: 62% (5/8) CAE ↔ Group B: 0% (0/6) CAE.

De Mey et al. (2006) (Belgium)	(a) 26 UCLP (b) 18 UCLP	*Age:* Soft palate: 2.8-3.2m (mean 3m) + hard palate: 5.5-6.8m (mean 6.2m) *Age:* Hard & soft palate: 2.8-3.1m (mean 3m) *Surgical technique:* Malek protocol (soft palate) and vomer flap (hard palate)	3y and 6y 3y and 6y	no	*Speech samples:* Spontaneous speech, standardized speech assessment in French or Flemish *Perceptual rating by one senior speech therapist:* - Intelligibility - Nasality - Articulation	***Intelligibility*****:** *At 3 years*: significant group differences (*p*=0.05). Group A: 52% good, 32% mild deficiency, 18% major problem ↔ Group B: 63% good, 37% mild deficiency, 0% major problem. *At 6 years*: no significant differences between groups. 80% normal intelligibility; ***Nasality*****:** No significant group differences. *At 3 years*: 30% no nasality problems, 50% inconstant nasality, 20% constant nasality. *At 6 years*: 40% no nasality problems, < 10% constant nasality; ***Articulation*****:** No significant differences between groups. *At 3 years*: 20% important disorders; *at 6 years*: 40% normal articulation; ***Velopharyngoplasty required:*** Group A: 12% (3/26) at mean age of 5 years ↔ Group B: 6% (1/18) at 5.5 years. Speech satisfactory in both groups in the majority of children. However, earlier intelligibility of speech in group B.
Abdel-Aziz (2013) (Egypt)	21 soft CP	*Age:* 3-6m (mean 4m3w) *Surgical technique:* Furlow Z-plasty	4y	no	*Speech samples:* Counting from 1 to 20, phonetically selected speech sample *Perceptual rating:* - Hypernasality - Nasal emission Assessment of velopharyngeal closure by nasopharyngoscopy	***Hypernasality***: 86% (18/21) absent, 14% (3/21) mild hypernasality; **Nasal emission**: 100% (21/21) absent; ***Velopharyngeal closure*** *(VPC):* 86% (18/21) competent VPC, 5% (1/21) borderline competent VPC, 10% (2/21) borderline incompetent VPC; ***Secondary pharyngoplasty***: none required

Doucet et al. (2013) (France)	(a) 20 UCLP (b) 20 UCLP	*Age:* Soft palate: 2.8-3.4m (mean 3.1m) + hard palate: 5.6-6.6m (mean 6.1m) *Surgical technique:* Sutured in midline without intravelar veloplasty (soft palate) + vomer flap (hard palate) *Age:* Soft palate: 5.7-6.3m (mean 6.0m) + hard palate: 16.9-18.7m (mean 17.8m) *Surgical technique:* Sommerlad (soft palate) + 2-layer hard palatal repair	3.0-3.6y (mean 3.3y) 2.9-3.7y (mean 3.3y)	no	*Speech samples:* n.a. *Perceptual rating by one senior speech therapist:* - Speech intelligibility - Hypernasality - Nasal emission - Nasal turbulence - Articulation Perceptual evaluation of velopharyngeal insufficiency confirmed by aerophonoscopy	***Speech intelligibility*****:** Significant group difference (*p*<0.05). Group A: 30% (6/20) adequate ↔ Group B: 75% (15/20) adequate; ***Hypernasality*****:** No significant group difference (*p*>0.05). Group A: 20% (4/20) absent, 35% (7/20) mild, 25% (5/20) moderate, 20% (4/20) severe; ***Nasal emission*****:** No significant group difference (*p*>0.05). Group A: 25% (5/20) absent, 35% (7/20) mild, 35% (7/20) moderate, 5% (1/20) severe; ***Nasal turbulence*****:** No significant group difference (*p*>0.05). Group A: 60% (12/20) absent, 40% (8/20) mild; ***Articulation*****:** Significantly more delays of ≥ 1y in group A (*p*<0.05). No significant group differences regarding consonant inventory, presence of bilabial consonants, velar substitution, compensatory articulations (i.e. glottal stops, pharyngeal fricatives, posterior nasal fricatives) (*p*>0.05); ***Velopharyngeal insufficiency*****:** Significant group difference (*p*<0.05). Group A: 55% (11/20) present ↔ group B: 15% (3/20) present.

UCLP: unilateral cleft lip and palate, BCLP: bilateral cleft lip and palate, C(L)P: cleft palate with or without cleft lip, CP: cleft of hard and soft palate, soft CP: cleft of soft palate only, n.a.: not available was indicated to be 'acceptable' in 87% (87/100) of 5.5-year-old patients *(Copeland, 1990)* and 'normal' in 80% of 6-year-old patients *(De Mey et al., 2006)*. However, only 30% (6/20) of the 3.3-year-old patients described by *Doucet et al. (2013)* showed 'adequate' speech intelligibility. Hypernasality was reported to be absent in all patients reported by *Desai (1983)* as well as in respectively 76% (13/17), 86% (18/21) and 93% (93/100) of the children included in the studies of *Randall et al. (1983)*, *Abdel-Aziz (2013)* and *Copeland (1990)*. In contrast, only 30/40% of the 3/6-year-old children described by *De Mey et al. (2006)* showed 'no nasality problems'. Furthermore, *Doucet et al. (2013)* noted mild to severe hypernasality in 80% (16/20) of the early closure group. Nasal emission occurred in none of the patients described by *Barimo et al. (1987)* and *Abdel-Aziz (2013)*, while *Copeland (1990)* and *Doucet et al. (2013)* observed nasal emission in 9% and 75% of the patients, respectively. The amount of patients having normal articulation following palatal closure prior to the age of 6 months ranged in literature from 40% *(De Mey et al., 2006)*, over 57% *(Copeland, 1990)* to 65% *(Randall et al., 1983)*. Although *Barimo et al. (1987)* noted absence of glottal stops and pharyngeal fricatives in all children aged 1;10-2;6 years (5/5, 100%), *Doucet et al. (2013)* observed compensatory articulation (i.e. glottal stop, pharyngeal fricative or posterior nasal fricative) in 20% (4/20) as well as velar substitution in 25% (5/20) of the included patients.

In comparison with patients whose palatal clefts were closed after the age of 6 months, children with palatal closure prior to the age of 6 months majorly showed improved speech results. The clinical impressions reported by *Kaplan et al. (1974)* included earlier onset of babbling, uttering of first sounds, one-word response and two- to three-word response following early (3-4m) compared to later (10-12m) palatal closure. Furthermore, *Grobbelaar et al. (1995)* observed significantly more normal resonance, articulation and intelligibility as well as significantly less velopharyngeal insufficiency in patients with palatal cleft repair before compared to after 6 months of age. In addition, *Ysunza et al. (1998)* noted significantly better articulation scales after palatal repair at 6 compared to 12 months of age, although they were below the normal limits for all patients. However, *Doucet et al. (2013)* reported better early speech outcomes after early soft and delayed hard palatal repair compared to early two-stage soft and hard palatal closure. The delayed cleft palate (CP) closure group contained significantly more patients with adequate speech intelligibility. Moreover, significantly less velopharyngeal insufficiency and articulation delays of at least one year

were noted for this group. The authors attributed these differences to variation in surgical technique, considering that intravelar veloplasty as described by *Sommerlad (2003)* was applied in the delayed CP closure group, but not in children with early palatal repair. Nevertheless, no significant group differences were observed for different types of resonance disorders and compensatory articulation. Drawing conclusions about speech following palatal repair before 6 months of age based on the afore-mentioned studies is hampered given the variation in several influencing factors such as cleft type, surgical palatal closure technique, age at speech assessment and type of speech evaluation. Moreover, information regarding cleft type *(Randall et al., 1983)*, surgical technique *(Randall et al., 1983)*, age at speech assessment *(Desai, 1983; Kaplan et al., 1974; Randall et al., 1983)*, characteristics of the included control group *(Barimo et al., 1987; Kaplan et al., 1974; Ysunza et al., 1998)*, the assessed speech sample *(Desai, 1983; Doucet et al., 2013; Grobbelaar et al., 1995; Kaplan et al., 1982; Kaplan et al., 1974; Kaplan et al., 1980; Randall et al., 1983; Ysunza et al., 1998)* and the speech evaluation strategies *(Barimo et al., 1987; Desai, 1983; Kaplan et al., 1982; Kaplan et al., 1974; Kaplan et al., 1980; Randall et al., 1983; Ysunza et al., 1998)* is often lacking or described insufficiently in view of replication. Taken into account the study variations and limitations as well as the restricted number of publications concerning speech following palatal repair prior to 6 months of age, it can be concluded that additional research in this field seems to be required.

Speech following primary palatoplasty in youngsters and adults

In literature, it is generally accepted that delayed palatal closure results in maximal avoidance of facial growth inhibition, despite decrease of speech abilities *(Berkowitz, 2013; Kaplan, 1981; Peterson-Falzone, 1996; Rohrich et al., 2000; Watson, 2001)*. Several authors reported maxillofacial growth to be normal or increased in patients with primary cleft closure during adolescence or adulthood *(Hoppenreijs, 1990; Lambadusuriya et al., 1988; Law & Fulton, 1959; Murthy, 2009; Ortiz-Monasterio et al., 1974; Ortiz-Monasterio et al., 1966; Will, 2000)*. Regarding speech, patients with unrepaired palatal clefts are generally unable to close the velopharyngeal sphincter and to built-up sufficient intraoral air pressure required for plosives and fricatives. As a consequence, air and acoustic vibrations obligatorily flow through the nose leading to nasal emission and hypernasality *(Harding & Grunwell, 1998; Kummer, 2008)*. In order to establish the necessary meaningful phoneme distinctions between individual consonant targets, compensatory errors most often arise *(Harding & Grunwell, 1998)*. In youngsters and adults with unrepaired clefts, these compensatory errors are well-integrated in the neuromuscular speech patterns considering that they are used for such a long time *(Ortiz-Monasterio et al., 1974)*. After primary palatoplasty, compensatory speech disorders are hypothesized to remain. However, the obligatory speech disorders might decrease or disappear due to the anatomical restoration.

Few studies documented in detail speech outcome following primary palatoplasty in youngsters or adults. The first reports were largely anecdotal and reported impressions of non-speech-language therapists *(Sell, 2008)*. Some studies contained small sample sizes *(Adekeye & Lavery, 1985; Holdsworth, 1954; Hoppenreijs, 1990; Schwarz, 2006)*, poorly documented the included patients regarding cleft type, age at cleft closure, age at speech assessment or surgical closure techniques *(Adekeye & Lavery, 1985; Holdsworth, 1954; Hoppenreijs, 1990; Koberg & Koblin, 1973; Law & Fulton, 1959; Ortiz-Monasterio et al., 1974; Ortiz-Monasterio et al., 1966)*, and/or broadly evaluated speech without mentioning the type of speech sample or inter/intrarater reliability scores *(Adekeye & Lavery, 1985; Holdsworth, 1954; Hoppenreijs, 1990; Koberg & Koblin, 1973; Ortiz-Monasterio et al., 1974; Ortiz-Monasterio et al., 1966)* what frequently resulted in conflicting findings. To our knowledge, only 4 studies systematically described the speech characteristics of patients following palatal repair in youngsters or adults *(Murthy et al., 2010; Sell & Grunwell, 1990; Whitehill et al., 1996; Zhao et al., 2012)*. *Sell & Grunwell (1990)* evaluated resonance (hypernasality) and articulation characteristics of 18 Sri Lankan adolescents (mean age 14 years) with various cleft types on 3 occasions: preoperatively, 8 months postoperatively and 12 months postoperatively after a period of speech therapy. Severe to moderate hypernasality occurred in all patients preoperatively which improved in only 4 patients (4/18, 22%) with soft CP after palatal repair. Preoperatively as well as 8 months postoperatively, articulation was characterized by an overall glottal and pharyngeal place of articulation and by a high frequency of nasal substitutions and approximations. *Whitehill et al. (1996)* described the use of electropalatography to treat the speech disorders of a Cantonese-speaking 18-year-old Chinese women with bilateral cleft lip and palate (BCLP) following palatal closure at the age of 13 years. Prior to speech therapy, speech was characterized by moderate hypernasality (score 4/6), nasal emission and a moderate articulation disorder. Deviations in place of articulation included velar placement of alveolar plosives, fricatives and affricatives as well as bilabial production of /f/ and /v/. The manner of articulation was characterized by nasal plosion of initial plosives /p,t,k/. *Murthy et al. (2010)* compared preoperative and postoperative speech

characteristics of 131 Indian Tamil-speaking patients with UCLP or BCLP who received surgical palatal repair after the age of 10 years. Postoperatively, speech intelligibility was rated as 'normal' to 'occasional repetition of words required' in 55% (72/131) of the patients. Hypernasality was observed in all patients ranging from mild (84/131, 64%) to moderate or severe (47/131, 36%). Moreover, the majority of the patients showed mild (102/131, 78%) to moderate (21/131, 16%) nasal emission as well as mild (58/131, 44%) to moderate (63/131, 48%) articulation errors. In comparison with the preoperative condition, all speech parameters improved significantly. Finally, *Zhao et al. (2012)* reported the speech results of 224 Chinese patients with various cleft types following palatal repair at 2-5 (n=140) and 5-24 (n=84) years of age. In the latest CP closure group, absence of hypernasality and nasal emission was noted in 36% (30/84) and 37% (31/84), respectively. However, the majority of this group showed mild (30/84, 36%) to moderate (19/84, 23%) hypernasality as well as mild (27/84, 32%) to moderate (23/84, 27%) nasal emission. For both speech characteristics, significant differences were obtained compared to the earlier CP closure group.

From this literature review, it can be carefully concluded that palatal repair in youngsters and adults generally seems to result in maintenance of severely disordered speech (speech intelligibility, resonance, articulation), although small improvements might occur compared to the patients' preoperative speech abilities. However, the number of detailed described studies that systematically evaluate speech outcome in this patient population is limited. Moreover, variations occur regarding patient-related characteristics (e.g. cleft type), treatment-related factors (e.g. surgical technique, age at palatal repair) and study design (e.g. speech samples, rating scales). As a consequence, further expansion of this research field seems to be indicated.

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CHAPTER 4

CLEFT LIP AND/OR PALATE IN RESOURCE-POOR COUNTRIES

Impairments such as cleft lip and/or palate (CL/P) occur in all societies. Perception towards this impairment and the way of categorizing and treating affected individuals is however influenced by culture and context *(Wickenden, 2008)*. As a consequence, understanding of existing traditional beliefs is important in view of rehabilitation and medical care *(Louw et al., 2006)*. Given that the studies of the current doctoral thesis were generally conducted in a resource-poor country, i.e. the Republic of Uganda, the existing traditional cultural beliefs about etiology of CL/P in resource-poor countries will be discussed in the current chapter. Furthermore, the importance of well-considered modern health care for CL/P in these countries is outlined and information will be provided on treatment facilities of this congenital anomaly in the Republic of Uganda.

Cultural beliefs on cleft lip and/or palate in resource-poor countries

In resource-poor countries, a multitude of unique cultures and societies exists. Each has his own view on clefts which is influenced by local religion and traditional beliefs. Notwithstanding the diversity, certain commonalities can be found regarding beliefs on etiology of clefts in interviews with Indian *(Weatherley-White et al., 2005)*, South-African *(Louw et al., 2006; Patel & Ross, 2003)*, Nigerian *(Olasoji et al., 2007)*, Sri Lankan *(Bradbury & Habel, 2008)*, Venezuelan *(Bradbury & Habel, 2008)*, Ugandan *(Bradbury & Habel, 2008)* and Egyptian *(el-Shazly et al., 2010)* patients and/or parents of patients with CL/P as well as in South-African traditional healers *(Dagher & Ross, 2004)*. The common thread is one of blame and guilt. The subjects in these studies generally attributed the origin of clefts to (1) the will of God, (2) a curse of Satan, evil spirits or witches, and/or (3) ancestors' punishment for sins or insufficient sacrifices. Moreover, somewhat bizarre explanations are frequently mentioned including wrong doings by mothers during pregnancy such as laughing with a cleft patient, going out or using a sharp object during an eclipse, and speaking evil words. Others give rather medically based incorrect explanations such as infection or obstruction of the womb, consequence of a previous miscarriage, prematurity, and the nurse cutting the patient's lip, while cutting the umbilical cord. Only few subjects acknowledged the influence of genetics and/or environmental factors such as drugs and medicines as a potential cause for clefts.

These cultural beliefs about causation will influence the way societies in resource-poor countries handle patients with clefts. Many patients are stigmatized and deprived of normal social interaction *(Bradbury & Habel, 2008; el-Shazly et al., 2010; Weatherley-White et al., 2005)*. For example, the majority of the Ugandan parents reported by *Bradbury & Habel (2008)* mentioned during interviews negative responses from the community, varying from mocking to outright rejection. Moreover, in various cultures, prospects for education, marriage and employment seems to be poor, particularly in patients with unrepaired clefts *(Bradbury & Habel, 2008; el-Shazly et al., 2010; Weatherley-White et al., 2005)*. However, parents and patients often believe that the opportunities for education, marriage and employment will increase after surgical cleft closure. In some communities, females with CL/P seem to have increased risks for experiencing social isolation and disadvantages in education and marital prospects compared to males with similar anomalies *(Bradbury & Habel, 2008; Weatherley-White et al., 2005; Wickenden, 2008)*. Furthermore, Indian *(Weatherley-White et al., 2005)* and Ugandan *(Bradbury & Habel, 2008)* parents of children with clefts are generally more concerned about the cleft lip (CL) compared to the cleft palate (CP), considering that a disfigured appearance has a greater impact on the patients' social life. The role of the CP in later speech performance remains however generally unacknowledged.

In addition to the influence on overall social attitudes, cultural beliefs about the etiology of clefts will also directly or indirectly reflect the methods for patients and parents of patients with CL/P to seek help *(Louw et al., 2006; Olasoji et al., 2007)*. In resource-poor countries, two types of treatment approaches are represented for CL/P, including traditional healers and modern health care. A traditional healer can be described as "someone who is recognized by the community in which he/she lives as competent to provide health care by using vegetable, animal or mineral substances and certain other methods based on the social, cultural and religious backgrounds as well as the prevailing knowledge, attitudes and beliefs regarding physical, mental and social well-being and the causation of disease and disability in the

community" *(Dagher & Ross, 2004)*. According to the WHO, more than 80% of the population in resource-poor African countries visits traditional healers for receiving health care with or without seeking for modern treatment or surgery *(Bodeker et al., 2005)*. Traditional healers seem to form an integral part of a family's approach to health and illness, while modern medicine is often considered to be of secondary importance *(Louw et al., 2006)*. As a consequence, patients with CL/P and their families will often ask traditional healers for help.

Modern health care in resource-poor countries

In many resource-poor countries, specialized health care does not exists *(Aziz et al., 2009)*. Therefore, cleft missions have been carried out since several decades by visiting teams from the North such as Europe and North-America *(Aziz et al., 2009; Donkor et al., 2007; Hodges & Hodges, 2000; Pham & Tollesfon, 2007)*. Despite general consensus on the necessity of interdisciplinary treatment for patients with orofacial clefts *(Shprintzen, 1995)*, these visiting teams consist predominantly of plastic and maxillofacial surgeons and assisting medical staff (e.g. anesthetists, nurses), while other important specialists such as orthodontics, speech-language therapists (SLTs) and otorhinolaryngologists are less frequently involved *(Akinmoladun & Obimakinde, 2009)*. A wide range of preparation, organization and sustainability is noticed in these cleft care missions *(Garfein et al., 2008)*. Numerous teams travel to resource-poor countries for short periods treating as many patients as possible. Therefore, patient selection based on eligibility and priority for surgery is often required *(Aziz et al., 2009; Pham & Tollesfon, 2007; Weatherley-White et al., 2005)*. However, the missions' goal increasingly seems to shift from treating the largest number of patients to training and supporting medical professionals and organizations *(Garfein et al., 2008)*. Particularly large organizations such as Operation Smile *(www.operationsmile.org)* and Smile Train *(www.smiletrain.org)* emphasize the importance of teaching local surgeons advanced techniques in order to decrease dependence on visiting teams and to create long-term self-sufficient care *(Garfein et al., 2008)*. Moreover, improvement of local infrastructure as well as high quality treatment by qualified surgeons without providing untried or experimental surgery are considered to be essential *(Aziz et al., 2009; Spauwen, 2013)*. Similar purposes were stated during the VLIR-UOS project (ZEIN2009EL28) of the Ghent University with the mission of "Creation of a reference center for congenital facial cleft and benign jaw tumors in Uganda" (promotor: prof. dr. Hubert Vermeersch, copromotor: prof. dr. Kristiane Van

Lierde), in which this doctoral thesis was incorporated. During this project, quality of care, empowerment, training and sustainability were considered to be of paramount importance.

In comparison with northern countries, the surgical timing protocol in resource-poor countries is increasingly changed due to the large number of failures to complete all surgical procedures *(Lehman, 2008)*. Default rates for palatoplasty following successful lip repair in young patients with cleft lip and palate (CLP) seem to vary from 44% (20/45) *(Onah et al., 2008)* to 90% (9/10) *(Hodges, 2010)*. Explanations might be sought in financial considerations *(Hodges, 2010)* and the afore-mentioned cultural importance of normal aesthetics *(Bradbury & Habel, 2008; Weatherley-White et al., 2005)*. Consequently, closure of the entire cleft during a single surgery, including synchronous lip and palatal closure for patients with CLP and one-stage soft and hard palatal closure for patients with CP, is advised for all healthy children from resource-poor countries who have limited access to secondary surgery *(Hodges & Hodges, 2000; Lehman, 2008; Onah et al., 2008)*. When it seems impossible for any reason to perform synchronous closure of the CLP, one-stage palatal repair should be carried out first, followed by closure of the CL after 3 to 6 months *(Lehman, 2008)*. As such, parents will return to complete lip closure considering the social implications of CL. In case the second surgery would be delayed, the functional outcome would not be compromised.

Thanks to international organizations and visiting teams an increasingly number of surgical lip and palatal closures is performed in accordance with this adapted surgical timing protocol in young children living in resource-poor countries. However, the number of unrepaired adult cases is still significantly higher compared to northern countries *(Akinmoladun & Obimakinde, 2009)*. The majority of these untreated patients is impoverished and lives in rural areas *(Aziz et al., 2009)*. As a consequence, many parents are not aware that this anomaly can be surgically treated *(Adeyemo et al., 2009; Aziz et al., 2009; Hodges et al., 2009; Schwarz & Khadka, 2004)*. Moreover, access to modern health care is often limited for rural families given its concentration in urban areas and the associated long travelling time to these centers *(Adeyemo et al., 2009; Schwarz & Khadka, 2004; Sommerlad, 2008)*. Furthermore, parents' beliefs about the etiology of clefts might affect their willingness to bring their children for surgery *(Adeyemo et al., 2009; Akinmoladun & Obimakinde, 2009; Aziz et al., 2009; Sommerlad, 2008)*. In addition, the influence of financial resources may not be underestimated. Most parents are unable to fund the

required investigations and surgical treatment of their affected child and cannot pay the travelling costs to the urban center and the subsistence costs during the stay in the hospital *(Akinmoladun & Obimakinde, 2009; Donkor et al., 2007; Hodges et al., 2009)*. Finally, other possible explanations for the presence of unrepaired clefts in adults are late recognition of the deformity in case of isolated CP *(Onah et al., 2008)* and the patient's fear of death from surgery *(Adeyemo et al., 2009)*.

In order to optimize modern health care for patients with clefts in resource-poor countries and to reach as many patients as possible, the above-mentioned reasons for delaying cleft repair should be tackled. Greater collaboration between modern medical practitioners and traditional healers in CLP management *(Olasoji et al., 2007)* as well as awareness campaigns *(Wilson & Hodges, 2012)* might counter the influence of sociocultural beliefs and the unawareness about the possibility of cleft repair. Moreover, financial and medical help as well as training programs provided by international organizations such as Smile Train and Operation Smile should be encouraged to handle the financial constrictions, to improve the local infrastructure, and to increase the number of well-skilled specialists.

Cleft lip and/or palate in the Republic of Uganda

The Republic of Uganda is a poor country located in East-Central Africa, bordered by South-Sudan, Kenya, Tanzania, Rwanda and the Democratic Republic of Congo. This tropical state covers an area of 241559 sq km *(State house office, 2013)*. According to data reported by *UNICEF (2003b)* for 2011, about 34.5 million people live in this country with 16% living in urban areas. An estimated 19 million (55%) is younger than 18 years and 6 million children (19%) have an age under 5. Life expectancy at birth amounts 54 years. Seventy percent of the population aged 15 years or older are lettered. Thirty-eight percent lives below the international poverty line of $1.25 per day. The majority of the Ugandans is Roman Catholic (42%), Protestant (42%) or Muslim (12%) *(Central Intelligence Agencey, 2013)*. The official languages of Uganda are (particularly) English and (secondly) Swahili *(Uganda Law Reform Commission, 2006)*. However, about 41 individual languages are currently spoken in this country, including Luganda, the most widely spoken language in the southern part of the country *(Lewis et al., 2013)*.

The Republic of Uganda has a poor health care infrastructure. No obligatory medical insurance system exists. Moreover, the number of health care workers in Uganda is very limited. Table 4.1 presents the number of physicians, nurses and midwifes, pharmacists, dentists and SLTs employed in Uganda as well as the density per 10000 citizens. A comparison is made with Belgium, a wealthy country located in Europe. As seen in Table 4.1, the density of physicians per 10000 people is a fiftieth of what it is in Belgium. This difference is even larger for the density of paramedical specializations such as dentists (1/83) and SLTs (1/917). During the last few years, the number of SLTs increased significantly. According to the survey conducted by *Fagan & Jacobs (2009)*, 2 SLTs were employed in Uganda. The only Speech-Language Therapy department was located at Mulago National Referral Hospital in the capital city, Kampala *(Robinson et al., 2003)*. However, in 2008, a bachelor program for Speech-Language Therapy was established at the Makarere University, the only training program for SLTs in East and Central Africa. To date, 19 graduates finished this training program, of which 15 are currently working in Uganda. Considering that the number of long-term SLT expats increased to 4 in 2013, 19 SLTs are currently employed in this country.

Table 4.1 Comparison of the number of health care workers between Uganda *(World Health Organisation, 2006)* and Belgium *(RIZIV-INAMI, 2010)*. The density per 10000 people is provided as well. For calculations of density in Belgium, the population size in 2004 and 2013 was retrieved from the *Belgian Federal Government (2013)*.

	Uganda		**Belgium**	
	n	**Density (per 10000)**	**n**	**Density (per 10000)**
Physicians (in 2004)	2209	0.8	41734	40.1
Nurses and midwifes (in 2004)	19325	7.3	67295	64.7
Pharmacists (in 2004)	688	0.3	11618	11.2
Dentists (in 2004)	363	0.1	8660	8.3
Speech-language therapists (in 2013)	19	0.006	6089	5.5

CL/P occurs in Uganda between 0.73 *(Dreise et al., 2011)* and 1.34 *(Kalanzi et al., 2013)* per 1000 live births. Despite the low birth prevalence, CL/P is a commonly occurring congenital anomaly in this country. Given that the annual number of births for Uganda is estimated at 1.5 million *(UNICEF, 2003b)*, 1128 to 2070 children are born with CL/P each year.

Surgical closure of clefts can be performed in just a few Ugandan hospitals. The number of plastic surgeons working in Uganda is limited and until recently, no recognized training program for plastic and reconstructive surgery was yet available in East Africa *(Hodges et al., 2009)*. As a consequence, 80% of the CL/P surgeries in south-western Uganda are performed by plastic surgery teams funded by organizations such as Smile Train and Interplast *(Walker et al., 2010)*.

In 2005, a specialist plastic and reconstructive unit was established at the Children's Orthopaedic Rehabilitation Unit (CORU) of Mengo hospital in Kampala *(Hodges et al., 2009)*. This unit moved in 2009 to the Comprehensive Rehabilitation Services in Uganda (CoRSU) hospital *(www.corsu.or.ug)* in Kisubi (Wakiso district) a private non-profit and non-government organization established by Christian Blind Mission (CBM, *www.cbm.org*) and Smile Train and funded by international and local donors. In this well-equipped hospital, surgical treatment is provided for patients with CL/P, burn contractures, complications of soft tissue infections or tumors and orthopaedic anomalies. The plastic surgeries are carried out by two trained permanent plastic and reconstructive surgeons and a handful of visiting specialized surgeons. One permanent plastic and reconstructive surgeon treats all patients with CL/P. Since the beginning, more than thousand CL/P closures have been performed for free thanks to financial support of Smile Train. These patients came from all over the country and even across the borders. Some of them came of their own accord, others were traced by social workers or community-based rehabilitation staff. A few times a year, outreach services are provided by CoRSU's plastic and reconstructive surgeon to assess and surgically treat patients in remote areas. Moreover, in January 2012, a postgraduate training program for plastic and reconstructive surgery was started at CoRSU. This program is based in and approved by Mbarara University in Uganda and directed by CoRSU's senior plastic and reconstructive surgeon, dr. A. Hodges. Currently, 2 plastic surgery students are in training in this hospital.

Treatment of babies with CL/P at CoRSU consists of surgical cleft closure, preceded by nutritional support when necessary and sometimes followed by speech-language therapy. Nutritional support is often required in Ugandan babies with palatal clefts. As mentioned in Chapter 1, babies with palatal clefts regularly show feeding difficulties *(Cubitt et al., 2012;*

Miller, 2011). Most Ugandan parents can however not afford formula preparations and specialized feeding supports *(Hodges et al., 2009)*. As a consequence, these babies are frequently malnourished at time of arrival. In CoRSU, malnourished children who require surgery are provided nutritional support by a nutritionist. Vitamin supplementation is applied and mothers are learned how to feed their child with a spoon and a cup. Subsequently, closure of the cleft is performed as soon the baby reaches a target weight of 3 kg, irrespective of age *(Cubitt et al., 2012)*.

Clefts of the lip and/or palate are generally closed during a single operation, considering the reasoning about high default rates for palatoplasty after successful lip repair *(Hodges, 2010; Lehman, 2008; Onah et al., 2008)*. Internationally accepted surgical techniques are applied (e.g. Millerd [unilateral CL] and modified Mulliken [bilateral CL] technique for lip repair, Sommerlad technique for palatal closure). In order to prevent further malnourishment, this synchronous lip and palatal repair is often performed before the age of 6 months. Surgical intervention at such a young age can be carried out safely in CoRSU thanks to the presence of well-trained anesthetists and the necessary specialized anesthetic equipment. However, not all patients present at such a young age.

After surgical cleft closure, patients with palatal clefts are generally referred to the Speech-Language Therapy department of CoRSU, established in 2010 in cooperation with the VLIR-UOS project of Ghent University. Advice is provided to parents of young children with CL/P regarding stimulation of the child's speech-language development. In case of seriously disordered speech, speech therapy is offered. Since October 2012, an SLT graduated from the Makarere University is employed in CoRSU for one day per week. This man provides speech-language therapy to patients with communication disorders due to CL/P or other developmental or acquired causes. Of course, this ambulant treatment is only feasible for patients living not too far from the hospital. Therefore, patients are sometimes referred to Mulago National Referral hospital in Kampala for speech therapy in case this is closer to their home and the parents can afford the treatment.

In conclusion, CoRSU is a rather modern hospital with a good quality infrastructure and well-trained medical health care providers, located in a resource-poor country, Uganda. However, when compared to craniofacial teams operating in northern countries such as

the cleft team of the Ghent University Hospital in Belgium, several differences can be noted in patient population, health care system, team composition, surgical approach and available equipment. Table 4.2 provides an overview of differences and similarities between both teams, hospitals and countries. Although high standards are already achieved in this resource-poor context (particularly for plastic surgery and anesthetics), additional financial, clinical and research investments are required in order to reach an increasing number of East African patients with CL/P and to provide a more extensive interdisciplinary treatment, including speech-language therapy, hearing assessment and management as well as dental and orthodontic treatment.

Table 4.2 Comparison of patient population, health care system, team composition, surgical approach and available equipment for assessment of speech, hearing and cognition between the cleft and craniofacial teams of CoRSU in Uganda, a resource-poor country, and the Ghent University Hospital in Belgium, a wealthy country.

	CoRSU/Uganda	**Ghent University Hospital/Belgium**
	PATIENT POPULATION	
Urbanized population[a]	16%	97%
Life expectancy at birth[a]	54y	80y
Birth prevalence of CL/P[b]	0.73-1.34 per 1000	1.54-1.58 per 1000
Babies born with CL/P each year[a]	1128-2070	189-194
Beliefs about medical etiology	Unrecognized	Recognized
Consequences of cleft: education	School absence	No influence
Consequences of cleft: job	Decreased prospects	No influence
Consequences of cleft: marriage	Decreased prospects	Limited influence
Desired treatment	Traditional (often) + modern approach	Modern approach
	HEALTH CARE SYSTEM	
Public spending for health as % of GDP[a]	2%	7%
Obligatory medical insurance	Unavailable	Available
Number of health care providers	Insufficient	Sufficient
Number of cleft teams in the country	1	At least 7
Access to hospital	Limited	Good
Travelling time to hospital	Often several hours/days	Maximum ± 2h
	TEAM COMPOSITION	
Plastic and reconstructive surgeon	Available	Available
Anesthetist	Available	Available
Nurse	Available	Available

Orthodontist	Unavailable	Available
Otorhinolaryngologist	Unavailable	Available
Speech-language therapist	Available	Available
Maxillofacial surgeon	"Unavailable"	Available
Social worker	Available	Available
Geneticist	Unavailable	Available
Dentist	Unavailable	Available
Psychologist	Unavailable	Available in hospital
Audiologist	Unavailable	Available in hospital
SURGICAL APPROACH		
Timing cleft repair in case of CLP	Synchronous lip and palatal repair asap (depending on weight)	- Lip repair at 3-4m - One-stage palatal repair at ±12m
Timing lip repair in case of CL	Asap	3-4m
Timing palatal repair in case of CP	One-stage repair asap (depending on weight)	One-stage repair at ±12m
Technique for palatal repair	Sommerlad	Sommerlad
Technique for lip repair (UCLP)	Millard	Millard
Technique for lip repair (BCLP)	Modified Mulliken	Modified Mulliken
Major morbidity and mortality	Limited/non-existent	Limited/non-existent
Long-term follow-up	Patients often not return	Until ± 18y
Outcome studies for speech[c]	Unavailable	Available
AVAILABLE EQUIPMENT FOR SPEECH, HEARING AND COGNITIVE ASSESSMENTS		
Nasoendoscope	Unavailable	Available
Videofluoroscope	Unavailable	Available
Nasometer	Unavailable	Available
Normative nasalance values	Unavailable	Available
Articulation (picture-naming) test	Unavailable	Available
Video camera	Unavailable	Available
Audiometer	Unavailable	Available
Tympanometer	Unavailable	Available
Intelligence test	Unavailable	Available

Abbreviations
GDP: Gross Domestic Product, CLP: cleft lip and palate, UCLP: unilateral cleft lip and palate, BCLP: bilateral cleft lip and palate, CL: cleft lip, CP: cleft palate, asap: as soon as possible
References
[a]Uganda: *UNICEF (2003b)*, Belgium: *UNICEF (2003a)*
[b]Uganda: *Dreise et al. (2011)* and *Kalanzi et al. (2013)*, Belgium: *Mossey & Castilla (2003)*
[c]Belgium: *Van Lierde et al. (2002)*, *Van Lierde et al. (2003)*, *Van Lierde et al. (2003); Van Lierde et al. (2004)*

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CHAPTER 5

RESEARCH GOALS

Between 2005 and 2009, Ugandan patients with cleft lip and/or palate (CL/P) were surgically treated at the Children’s Orthopaedic Rehabilitation Unit (CORU) of Mengo hospital in Kampala. In 2009, the unit for plastic and reconstructive surgery moved to the Comprehensive Rehabilitation Services in Uganda (CoRSU) hospital in Kisubi. All patients presenting in these units have been surgically treated by one experienced plastic and reconstructive surgeon who used advanced internationally accepted surgical techniques and a surgical timing protocol adapted to the requirements for resource-poor areas. To date, two studies with peri- and early postoperative results *(Hodges, 2010; Hodges et al., 2009)* as well as one study about the nutritional status of the presenting babies *(Cubitt et al., 2012)* were already carried out for this population. However, neither speech outcomes, nor parental satisfaction have yet been verified. Nevertheless, good speech intelligibility and satisfaction outcomes are required to reduce the influence of the congenital anomaly on patients’ activities and participation in view of optimizing the patients’ quality of life *(World Health Organisation, 2002).* While searching for the optimal surgical treatment in patients with CL/P in terms of good speech intelligibility, the main purpose of the current doctoral thesis was to assess satisfaction and speech outcome of Ugandan patients with surgically closed CL/P treated by one experienced surgeon using the Sommerlad technique for palatal repair. In order to obtain this research aim, specific research questions were formulated.

Nasometry can be used to objectively assess the presence of resonance disorders in patients with CL/P by comparing their mean nasalance values with those of persons without clefts. However, differences in normative mean nasalance values have been reported in literature across languages *(Putnam Rochet et al., 1998)*. Moreover, some studies noted a significant influence of gender and/or age *(Van Lierde et al., 2003)*. Prior to this doctoral thesis, no normative nasalance values were available for Uganda. Establishment of such normative values was however necessary for clinical purposes as well as for further research on resonance outcome in the Ugandan cleft population. Consequently, the first purpose of this doctoral thesis was to establish normative nasalance values in typically developing Ugandan English-speaking children and to determine the influence of gender and age. This topic is handled in Chapter 6.

According to the *World Health Organisation (2007)*, the extent to which a patient with CL/P feels to be disabled depends among others on the interaction with environmental and personal factors. Considering that the parents' view substantially influences the children's attitude towards their own appearance and speech *(Bull & Rumsey, 1988)*, a relevant manner to determine treatment success is parental satisfaction with facial appearance and speech. Hence, our second aim was to verify parental satisfaction with facial appearance and speech in Ugandan patients with unilateral or bilateral cleft lip and palate following a synchronous lip and palatal repair. The results were compared with judgments by parents of non-cleft control children. Moreover, the impact of possible influencing variables was assessed. This study is described in Chapter 7.

As mentioned in Chapter 3, timing of palatal repair influences speech characteristics in children with palatal clefts *(Hardin-Jones & Jones, 2005; Willadsen, 2012)*. Significant differences were even noted between canonical babbles of babies with unrepaired palatal clefts and those of typically developing children *(Peterson-Falzone et al., 2006)*. Although some authors reported better speech outcomes for early (≤ 6 months) compared to later (> 6 months) palatal closure *(Grobbelaar et al., 1995; Kaplan et al., 1974; Ysunza et al., 1998)*, methodological weaknesses hamper interpretation and generalization of these results and necessitate further research. Therefore, the third goal of this doctoral thesis included description of articulation and resonance characteristics of Ugandan children with palatal clefts following palatal closure prior to the age of 6 months using the Sommerlad technique, in comparison with age- and gender-matched non-cleft children. Moreover, we aimed to compare this Ugandan patient group with a Belgian cleft palate group (matched for cleft type, age and gender) who underwent later palatal repair by means of the same surgical technique. These topics are presented in Chapter 8 and 9.

Finally, the number of studies systematically describing speech after delayed palatal closure is limited. Although overall severely disordered speech has been noted, variations in variables related to patients, surgery and speech assessment makes it difficult to draw

conclusions. Consequently, the final objective was to describe resonance and articulation characteristics in Ugandan youngsters and adults with palatal clefts following palatal closure after 8 years of age. This study is described in Chapter 10.

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PART 2

Publications

CHAPTER 6

NORMATIVE NASALANCE DATA IN UGANDAN ENGLISH-SPEAKING CHILDREN

Based on:

Luyten, A., D'haeseleer, E., Hodges, A., Galiwango, G., Budolfsen, T., Vermeersch, H., & Van Lierde, K. (2012). Normative nasalance data in Ugandan English-speaking children. *Folia Phoniatrica et Logopaedica*, 64, 131-136.

Abstract

Aims: The aim of this study was to obtain normative nasalance values for typically developing Ugandan English-speaking children as a reference point for clinical practice and further research.

Methods: Sixty-nine typically developing Ugandan children (35 males and 34 females, 2;7-13;5 years of age) participated in the study. Nasalance scores were obtained with the Nasometer while children repeated 4 sustained sounds, 14 repeated syllables, 15 sentences (12 oral, 3 nasal) and 2 texts ('Rainbow Passage' and 'Zoo Passage'). Data were analyzed for gender and age dependence.

Results: No significant effects of age and gender on nasalance values were obtained; hence, normative values for the overall group were reported. The average nasalance scores for Ugandan English-speaking children were 17% and 64% for the oral and nasal sentences and 33% and 14% for the oronasal and oral text, respectively.

Conclusion: The normative values are important as a reference point to assess the impact of several surgical procedures and several surgical timing strategies on speech in Uganda.

Key Words

Resonance • Nasalance • Nasometry • Norms • Uganda

Introduction

To assess and study nasalization and disorders of nasalization, head and neck surgeons, otorhinolaryngologists and speech-language therapists commonly rely on perceptual auditory evaluations of speech and objective assessment techniques *(Schneider & Shprintzen, 1980)*. Perceptual evaluation, the most frequently used method to assess nasality disorders, is generally based on perceptual judgments of connected speech in combination with uncomplicated subjective tests like the Gutzmann test *(Gutzmann, 1913)* and the Bzoch tests *(Bzoch, 1989)*. Due to the influence of secondary characteristics such as articulation and speech rate on perceptual judgments of nasality, a general consensus exists in literature on the necessity of objective tests in addition to subjective assessment techniques for the assessment of nasality (e.g. endoscopy, videofluoroscopy, ultrasound and radiography). A frequently used instrument for objective assessment of nasality is the Nasometer, introduced in 1986 by *Kay Elemetrics Corporation (2010)*. This instrument, developed by *Fletcher & Bishop (1973)*, uses two microphones on either side of a horizontal sound separator plate that rests on the upper lip. The microphones assess the relative amount of oral and nasal acoustic energy in a subject's speech. The signal from each microphone is individually filtered and digitized by custom electronic modules and, subsequently, the sound is processed by a computer. The resultant signal is a ratio of 'the nasal acoustic energy' to 'the nasal-plus-oral acoustic energy'. This ratio is multiplied by 100 and expressed as a 'nasalance score' (%) *(Kay Elemetrics Corporation, 2010)*. The Nasometer provides useful information about the speech of patients with velopharyngeal disorders *(Dalston et al., 1991a)* and upper airway impairment *(Dalston et al., 1991b; Dalston et al., 1991c; Williams et al., 1990)*. Several investigations have confirmed that Nasometer results correlate positively to perceptual judgments *(Dalston et al., 1993; Dalston & Warren, 1986; Dalston et al., 1991a; Fletcher, 1976; Hardin et al., 1992; Van Lierde et al., 2002)* and the reproducibility of nasometric measures has been proven in several studies *(Litzaw & Dalston, 1992; Mayo et al., 1996; Seaver et al., 1991; van Doorn & Purcell, 1998; Van Lierde et al., 2003)*.

A review of the literature reveals that numerous investigations have established normative values of nasalance scores for several languages. Table 6.1 summarizes the mean nasalance scores for standard nasometric tests in different languages. Various studies suggest that nasalance scores depend on language and regional dialect *(Leeper et al., 1992; Nichols, 1999; Putnam*

Table 6.1 Mean nasalance scores in percent for oronasal, oral and nasal sentences or texts as revealed by various studies.

Author	Language	Gender M/F	Age years	Oronasal	Oral (0% nasals)	Nasal	Nasometer model
Seaver et al. (1991)	Canadian English	56/92	16-63	29.0 (11.5%)	13.0	23.0/24.0 (35%)	6200
Kavanagh et al. (1994)	Canadian English	16/36	18-33	37.1 (11.5%)	13.4	65.4 (35%)	6200
Putnam Rochet et al. (1998)	Canadian English	149/166	9-85	31.0-35.4 (11.5%)	9.3-13.7	59.5-63.7 (35%)	6200
Litzaw & Dalston (1992)	American English	15/15	mean 26	36.0 (11.5%)	19.0	63.0 (35%)	6200
Mayo et al. (1996)	American English	40/40	mean 23	-	17.6	59.3 (35%)	6200
van Doorn & Purcell (1998)	Australian English	122/123	4-9	-	13.1	59.6 (35%)	6200
Sweeney et al. (2004)	Irish-English	34/36	4-13	26 (11%)	14.0-16.0	51.0 (55%)	6200-3
Haapanen (1991)	Finnish	5/37	3-54	-	12.5-13.6	69.4 (57%)	6200-2
van de Weijer & Slis (1991)	Dutch, The Netherlands	20/20	7-30	32.0 (11.7%)	11.8	52.3 (54.9%)	not indicated
Van Lierde et al. (2001)	Dutch, Flanders	28/30	19-27	31.5/36.1 (11.7%)	10.9	54.2/57.4 (57%)	6200
Van Lierde et al. (2003)	Dutch, Flanders	18/15	7-13	31.9 (11.7%)	11.3	51.6 (57%)	6200
Putnam Rochet et al. (1998)	Canadian French	60/93	9-85	24.0-30.1 (13.8%)	8.8-14.5	33.4-40.3 (28%)	6200
Anderson (1996)	Puerto Rican Spanish	40 females	21-43	36.0 (18%)	22.0	62.1 (33.3-60%)	6200
Nichols (1999)	Mexican Spanish	73/79	6-40	-	17.0	55.3 (20%)	6200-2
Tachimura et al. (2000)	Japanese	50/50	19-35	-	9.1	-	6200
Mishima et al. (2008)	Japanese	31/37	mean 24	-	10.3/15.6	-	6400
Müller et al. (2000)	Saxon German	23/28	18-74	41-33-37	13.0	67.2 (50%)	6200
Prathanee et al. (2003)	Thai	188	6-13	35.6 (16%)	14.3	51.0 (48%)	not indicated
Hirschberg et al. (2006)	Hungarian	75	5-25	31.7-39.5 (25%)	11.0-13.4	50.6-56.0 (33%)	6400
Brunnegård & van Doorn (2009)	Swedish	92/128	4-11	29.5 (12.9%)	15.7-12.7	56.5 (26.8%)	6400

The proportion of nasal consonants in the oronasal and nasal samples is indicated in parentheses.

Rochet et al., 1998; Seaver et al., 1991; van de Weijer & Slis, 1991; Van Lierde et al., 2001), gender *(Hutchinson et al., 1978; Leeper et al., 1992; Mishima et al., 2008; Nichols, 1999; Prathanee et al., 2003; Seaver et al., 1991; Van Lierde et al., 2001)*, age *(Brunnegård & van Doorn, 2009; Haapanen, 1991; Hirschberg et al., 2006; Hutchinson et al., 1978; Leeper et al., 1992; Nichols, 1999; Prathanee et al., 2003; Putnam Rochet et al., 1998; Trindade et al., 1997; Van Lierde et al., 2003)* and the Nasometer model *(Awan et al., 2011; Watterson et al., 2005)*. However, others did not reveal significant differences regarding age *(Kay Elemetrics Corporation, 2010; Litzaw & Dalston, 1992; Van Lierde et al., 2003)*, gender *(Brunnegård & van Doorn, 2009; Hirschberg et al., 2006; Kavanagh et al., 1994; Litzaw & Dalston, 1992; Mayo et al., 1996; Sweeney et al., 2004; Tachimura et al., 2000; van de Weijer & Slis, 1991; van Doorn & Purcell, 1998)* or dialect *(Brunnegård & van Doorn, 2009; Kavanagh et al., 1994; Mishima et al., 2008)*. Studies that observed significant gender differences revealed higher nasalance scores in female subjects compared to males, with the exception of the investigation of *Prathanee et al. (2003)* in typically developing Thai children. With regard to age, significant differences were mainly reported in studies that compared children with adults, but rarely in studies comparing groups of children *(Brunnegård & van Doorn, 2009; van Doorn & Purcell, 1998)*. The general consensus in literature regarding differences in nasalance scores due to differences in language and dialect implies development of normative data for each language and dialect to be necessary. To our knowledge, no normative nasalance values have been published for Ugandan English-speaking children. However, the existence of normative data would be advantageous to Ugandan cleft clinics in the assessment of the nasal resonance in patients with cleft palate and/or the investigation of the impact of different surgical procedures or surgical timing strategies.

From 1897 until 1962, the United Kingdom ruled Uganda *(Fage, 1982)*. English, the language of the colonizing power in that period, was introduced into government and public life and became deeply rooted in administration, media and education *(Mpuga, 2003)*. As a consequence, English remained the official language when Uganda gained independence from Britain in 1962. However, the native languages of the Ugandan population influenced the English language concerning pronunciation *(Bobda, 2001)*, semantics and grammar. In Uganda, approximately 40 different local languages are currently spoken *(Lewis et al., 2013)*. In particular, Luganda and other Bantu languages, Swahili and Nilotic languages are widely spoken *(Mpuga, 2003)*.

Currently, medical services for treatment of cleft lip and palate in Uganda are very limited. A different treatment plan is applied in the Comprehensive Rehabilitation Services in Uganda (CoRSU) hospital in Kisubi, Uganda, compared to northern countries. Instead of performing a two-stage surgery with early lip repair at the age of 6 weeks and surgical palatal closure at the age of 11 months, a synchronous closure of the lip and palate is performed before the age of 6 months. Prior to the investigation of resonance disorders in children with early repair of the cleft palate, normative data need to be obtained. As a consequence, the main purpose of the present study is to establish normative nasalance values in typically developing Ugandan English-speaking children. With such normative values, resonance disorders in Ugandan children with congenital cleft palate can be objectively assessed and monitored and the effect of different surgical techniques and surgical timing strategies can be examined. An additional objective of the present study is to determine whether gender and age differences influence the mean nasalance scores in Ugandan English-speaking children.

Participants and methods

Participants

Initially, 77 children were recruited for this study. All participants lived in an orphan home in Entebbe, a city in the southeast of Uganda. The children attended All Nations, a primary and secondary school for middle-class students. All participants spoke English as their second language. Prior to undertaking the study, the participants were assessed by a medical doctor. Children with craniofacial anomalies, pathology of the oral, nasal and velopharyngeal structures, hearing problems and/or a temporary nasal obstruction on the day of testing were excluded from the study. Furthermore, a consensus evaluation was performed to exclude children with perceptual resonance disorders and articulation problems (K.V.L., E.D.). Audio-recorded speech samples were collected using the MacKay-Kummer Simplified Nasometric Assessment Procedures (SNAP) test *(Kummer, 2005; MacKay & Kummer, 1994)*, in which children were asked to repeat sounds, syllables and sentences. Listener judgments indicated 3 children with slight hypernasality, 1 subject with slight hyponasality and 4 children with articulation disorders. Those children were excluded from this study. Eventually, 69 participants, 35 males and 34 females, were selected for this study. The children ranged in age from 2;7 to 13;5 years (mean age 6;8 years).

Methods

The research protocol was approved by the Ethical Committee of the Ghent University Hospital, Belgium (EC2011/269). The investigation was performed at the orphan home in Entebbe by two speech-language therapists (K.V.L., E.D.). Testing was completed in a quiet room. The Nasometer (model II 6450), manufactured by *Kay Elemetrics Corporation (2010)*, was used to obtain nasalance values. Prior to data collection, the Nasometer was calibrated according to the manufacturer's instructions *(Kay Elemetrics Corporation, 2010)*. Placement of the headgear and necessary adjustments were also conducted according to the manufacturer's specifications. The English stimuli used to obtain the normative nasalance values were derived from the MacKay-Kummer Simplified Nasometric Assessment Procedures (SNAP) test *(Kummer, 2005; MacKay & Kummer, 1994)*. This test was developed in 1994 and renormed in 2005. The children were asked to repeat 3 different types of speech samples. The first consisted of the sustained oral vowels /a/, /i/ and /u/ and the sustained nasal consonant /m/, produced for 2 seconds at a comfortable pitch and loudness. Secondly, the children were asked to repeat 14 CV-syllable sequences, containing the vowels /a/ or /i/ combined with a plosive (/p,t,k/), fricative (/s,ʃ/) or nasal (/m,n/) consonant. The third speech sample included 15 simple sentences, divided into 5 groups. In each sentence group, attention was focused on various consonants: bilabials, alveolars, velars, sibilants and nasals. Four sentence groups contained only oral sounds and the nasal sentence group contained 37% (18/49) nasal sounds. Each sentence was repeated twice, while the child was cued by a picture. For children aged 6 years and over, the 'Rainbow Passage', an oronasal text with 9.7% (31/318) nasal sounds *(Fairbanks, 1960)*, and the 'Zoo Passage', a text containing only oral sounds *(Fletcher, 1972)*, were also performed. The passages were elicited via phrase repetition.

Results

Statistical analyses were completed using SPSS software (version 16.0). Normality of each of the 25 dependent variables was tested using the Kolmogorov-Smirnov and Shapiro-Wilk test. The distribution of many of these variables deviated from normal and in such instances,

non-parametric equivalent tests were used in the analysis of the data. To determine whether age and nasalance values were related, Spearman rank-correlation coefficients were calculated for each of the 25 dependent variables; none of these correlations was significant. The scores of female and male children on each of the dependent variables were then compared using Independent Sample t-tests or Mann-Whitney U tests (depending on the data distribution) to assess whether gender had a significant effect on nasalance. Given the large number of statistical comparisons completed, a p-value of 0.05 was used as a cutoff for significance; Bonferroni post hoc analyses were carried out to ascertain whether the age and gender differences were statistically significant. The statistical analysis indicated that gender did not affect nasalance values for any of the 25 dependent variables. Given that neither age nor gender had significant effects on nasalance values, normative data is reported in Table 6.2 for the group as a whole (i.e. collapsed across age and gender). The sum mean nasalance scores for the oral and nasal sentences was 17% and 64%, respectively. Regarding the oronasal 'Rainbow Passage' and the oral 'Zoo Passage', mean nasalance scores of 33% en 14% were observed.

Discussion

In Uganda, at CoRSU in Kisubi, clefts of the lip and palate are generally repaired during a synchronous repair before the age of 6 months. No investigations have been performed yet regarding the impact of this early palatal repair on resonance. In order to assess speech outcome, normative data in Ugandan English-speaking children are required. As a consequence, the purpose of the present investigation was to obtain normative nasalance values in Ugandan English-speaking children.

The obtained normative group mean nasalance scores appear slightly higher in comparison to mean nasalance scores reported for other languages and other English variants. Although this might be due to sample variation, these differences support the suggestion that nasalance scores are influenced by languages and regional dialects. Comparison of the normative values for Ugandan English with the normative nasalance scores of other languages and dialects is an area for further research. According to *Mishima et al. (2008)*, the establishment of normative nasalance values for each language is very helpful in cleft palate

Table 6.2 Mean nasalance scores in percent ± standard errors (SE), SD and 95% prediction intervals (95% PI) of the sustained sounds, the syllable repetitions, the sentences focusing on different consonants and the oronasal and oral texts are shown for the entire group of Ugandan English-speaking children.

	n[a]	Nasalance score ± SE	SD	95% PI
/a/	65	11.7 ± 1.1	8.55	0-29.0
/i/	65	24.1 ± 1.4	11.05	1.8-46.4
/u/	65	17.4 ± 1.4	11.14	0-39.7
/m/	65	90.5 ± 0.7	5.97	78.4-100
/papapa/	56	9.0 ± 0.5	3.68	1.5-16.5
/pipipi/	56	15.7 ± 1.0	7.15	1.1-30.2
/tatata/	56	9.1 ± 0.6	4.19	0.6-17.6
/tititi/	56	19.3 ± 1.0	7.58	3.9-34.7
/kakaka/	56	8.0 ± 0.5	3.57	0.7-15.3
/kikiki/	56	20.0 ± 1.2	8.64	2.6-37.4
/sasasa/	56	8.0 ± 0.6	4.62	0-17.3
/sisisi/	56	18.5 ± 1.2	8.58	1.1-35.9
/ʃaʃaʃa/	56	8.0 ± 0.6	4.45	0-17.6
/ʃiʃiʃi/	56	18.0 ± 1.3	9.80	0-37.8
/mamama/	56	66.5 ± 0.8	6.14	54.2-78.8
/mimimi/	56	81.0 ± 0.7	5.18	71.0-92.0
/nanana/	56	68.0 ± 1.0	7.32	53.2-82.8
/ninini/	56	81.0 ± 0.6	4.49	71.9-90.1
Bilabials	59	16.1 ± 1.0	7.38	7.02-25.2
Alveolars	59	15.3 ± 1.0	7.35	0.4-30.2
Velars	59	19.1 ± 1.1	8.69	1.5-36.7
Sibilants	59	18.4 ± 1.2	9.02	23.9-36.6
Nasals	59	63.8 ± 1.5	11.28	41.0-86.6
Oronasal text	43	33.4 ± 1.1	7.25	18.5-48.3
Oral text	44	14.4 ± 1.0	6.44	1.3-27.5

[a]Number of subjects that completed the test.

clinics for diagnosis and follow-up of cleft palate speech. The general consensus in literature is that objective assessment of resonance, generally performed with the Nasometer, needs to be supported by perceptual judgments in the assessment of resonance disorders. The Nasometer should not be used as a sole instrument. As a consequence, establishment of normative values for nasalance in the current study is necessary to assess resonance disorders in Ugandan children with cleft palate and to perform follow-up evaluations after surgical treatment and/or speech-language therapy.

Two aspects need to be considered in the present study. At first, the assessment was performed in English, the children's second language. English is still one of the official languages in Uganda and is used in educational settings. Although differences exist between English and Luganda regarding phonetic and phonological structure, it was impossible to perform the assessment in Luganda or other local languages/dialects, like Swahili, Nyankole and Lusoga because the investigators did not master these languages and dialects. Furthermore, perceptual judgment of spontaneous speech could not be performed because children refused to tell English stories in their spontaneous speech, but they were very cooperative to repeat sounds, syllables, sentences and texts. As a consequence, listener judgments of repeated sounds, syllables and sentences, derived from the MacKay-Kummer SNAP test, were used to exclude children with abnormal nasal resonance.

No clinically significant gender and age differences were obtained in the nasometric tests. Our findings regarding gender differences in nasalance scores correspond with most of the published international data *(Brunnegård & van Doorn, 2009; Hirschberg et al., 2006; Kavanagh et al., 1994; Litzaw & Dalston, 1992; Mayo et al., 1996; Sweeney et al., 2004; Tachimura et al., 2000; van Doorn & Purcell, 1998; Van Lierde et al., 2001)* and correspond with the suggestions of *Brunnegård & van Doorn (2009)*, who stated that investigations concerning the establishment of normative nasalance values in Germanic languages (e.g. Dutch, Spanish, English variants and Swedish) regularly could not reveal significant gender differences in contrast to languages deriving from other families (e.g. Thai, Japanese). In literature, significant age-related differences for adults were observed in several studies *(Haapanen, 1991; Hirschberg et al., 2006; Hutchinson et al., 1978; Leeper et al., 1992; Nichols, 1999; Putnam Rochet et al., 1998; Trindade et al., 1997; Van Lierde et al., 2003)*. Our findings support the investigations of *Seaver et al. (1991)*, *van de Weijer & Slis (1991)* and *van Doorn & Purcell (1998)*, who could not reveal significant differences in age groups.

This study collected normative nasalance values in normal Ugandan English-speaking children. The normative data obtained for Ugandan English-speaking children will provide important reference information for clinicians who assess nasality disorders in cleft palate clinics in Uganda and will be important to investigate the impact of palatal closure before 6 months of age on speech (resonance).

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CHAPTER 7

PARENTAL SATISFACTION IN UGANDAN CHILDREN WITH CLEFT LIP AND PALATE FOLLOWING SYNCHRONOUS LIP AND PALATAL REPAIR

Based on:

Luyten, A., D'haeseleer, E., Budolfsen, D., Hodges, A., Galiwango, G., Vermeersch, H., & Van Lierde, K. (2013). Parental satisfaction in Ugandan children with cleft lip and palate following synchronous lip and palatal repair. *Journal of Communication Disorders*, 46, 321-329.

Abstract

Aims: The purpose of the present case control study was to assess parental satisfaction with speech and facial appearance in Ugandan children with complete unilateral or bilateral cleft lip and palate (CLP), who underwent a synchronous lip and palatal closure. The results are compared with an age- and gender-matched control group.

Methods: The experimental group consisted of the parents or guardians of 44 Ugandan patients (21 males, 23 females) with complete unilateral or bilateral CLP (mean age 3;1 years). The control group included the foster mothers of 44 orphan children matched by age and gender (mean age 3;7 years). A survey based on the Cleft Evaluation Profile was used to assess the perceived satisfaction for individual features related to cleft care.

Results: Overall high levels of satisfaction were observed in the experimental group for all features (range 56%-100%). No significant differences could be established regarding age, gender, age at lip and palatal closure, cleft type or maternal vs. paternal judgments. In participants who were dissatisfied with the appearance of the lip, the time period between the cleft closure and the survey was significantly larger compared to satisfied participants. Furthermore, significantly lower levels of satisfaction were observed in the cleft group for speech and the appearance of the teeth and the nose compared to the control group.

Conclusion: Satisfaction appears to be rather mixed. Parents were most satisfied with

hearing and aspects of the face, in particular with the lip and the nose. Moderate satisfaction was observed regarding speech and appearance of the teeth.

Learning outcomes

As a result of reading this manuscript, the reader will be able to explain the attitudes of parents toward the surgical repair of their children's cleft lip and palate. As a result of reading this manuscript, the reader will be able to identify differences in parental attitudes toward synchronous lip and palatal repair.

Key Words

Cleft • Satisfaction • Speech • Facial appearance • One-stage palatoplasty • Rural countries

Introduction

Research and clinical practice in patients with cleft lip and/or palate (CL/P) often focuses on differences in body structures and functions *(Neumann & Romonath, 2012)*. However, according to the International Classification of Functioning, Disability and Health: Children and Youth Version (ICF-CY), developed in 2007 by the World Health Organization (WHO), other aspects, including activities and participation (e.g. eating, drinking, speaking), environmental factors (e.g. individual attitudes, family and friends) and personal factors (e.g. coping style, psychological assets) should not be overlooked *(World Health Organisation, 2007)*. Analysis of these personal factors necessitates assessment of how the aesthetic aspects of children with CLP are handled *(Neumann & Romonath, 2012)*. Therefore, the most relevant aspect to determine treatment success is satisfaction with facial appearance and speech as judged by both patients and parents *(Sinko et al., 2005)*. Satisfaction with appearance as well as ability to verbally communicate highly influences psychological well-being *(Turner et al., 1997)*. Persons with an aesthetic and/or speech disorder might therefore be more at risk for developing psychological problems such as social withdrawal *(Berger & Dalton, 2011; Bernstein & Kapp, 1981)*, depressions and anxiety disorders *(Feragen et al., 2009)*. However, as reported by *Hunt et al. (2005)*, the majority of children and adults with repaired cleft lip and palate (CLP) do not appear to experience major psychosocial problems.

In the past two decades, numerous studies have been conducted on patient and parental satisfaction with aesthetics and speech *(Broder et al., 1992; Noor & Musa, 2007; Oosterkamp et al., 2007; Van Lierde et al., 2012; Williams et al., 2001)*. In a literature review, *Hunt et al. (2005)* reported that children and adults with repaired CL/P are generally positive about their body, facial appearance and speech. However, several authors mentioned that some cleft features are considered less than satisfactory to a sizable group of patients. Regarding facial appearance after closure of the cleft, patients are the most dissatisfied with the nose, the upper lip, the teeth and the facial profile *(Marcusson, 2001; Noar, 1991; Noor & Musa, 2007; Oosterkamp et al., 2007; Semb et al., 2005; Van Lierde et al., 2012)*. Furthermore, patients report persistent dissatisfaction with speech *(Noor & Musa, 2007)*, hearing *(Van Lierde et al., 2012)* and nasal breathing *(Oosterkamp et al., 2007; Van Lierde et al., 2012)*. *Oosterkamp et al. (2007)* hypothesized that patient and parental expectations are an important factor influencing satisfaction. Therefore, if the treatment outcome proves in line with the patients' and parental expectations, a maximum satisfaction could be reached. Consequently, the authors stressed the importance of providing honest and realistic information about the possible outcome of surgery to create more realistic expectations and reduce dissatisfaction with treatment outcome.

The effect of race (i.e. people with certain hereditary characteristics) and/or ethnicity (i.e. people sharing a common and distinctive racial, national, religious, linguistic or cultural heritage) on satisfaction with the facial appearance and speech in these patients has rarely been reported in literature. To our knowledge, only two studies have been conducted in non-white populations. *Noor & Musa (2007)* and *Reekie (2011)* both determined satisfaction with CLP repair in patients of Asian ethnicity. *Noor & Musa (2007)* assessed satisfaction regarding cleft-related features in 60 Asian teenager-parent pairs in Malaysia, using the Cleft Evaluation Profile (CEP). Seventy-seven percent (46/60) of the patients and 75% (45/60) of the parents were satisfied with the overall clinical outcome. For both groups, the lowest level of satisfaction was observed regarding the appearance of the teeth. *Reekie (2011)* used the Satisfaction with Appearance Questionnaire (SWA) in order to compare 15 Asian and 95 Caucasian adults with clefts in the UK. Compared to the Caucasian group, Asians showed lower mean scores for all the SWA-components, except for profile and hearing. However, for cleft-related features, the only differences reaching statistical significance were satisfaction with the nose and speech. No controlled satisfaction study has yet been performed in an East African population. However, studying parental satisfaction on facial appearance and speech in East African children with CLP would be interesting, considering that disabled people in East Africa have limited opportunities for accessing education, health, housing and employment due to stigma and negative attitudes in the community *(International Labour Organization, 2009)*. Disabilities are often associated with evil spirits, curses and punishment for ancestral wrong doings *(Dagher & Ross, 2004)*, resulting in abuse and discrimination by strangers, neighbors and even family members and in denial of basic provisions such as food, clothing and shelter *(Human Rights Watch, 2010)*.

In Uganda, life expectancy at birth in 2010 was estimated at 54 years with an under-5 year mortality rate of 99 per 1000 children *(UNICEF, 2003)*. The main causes of death among children under 5 years of age are prenatal conditions, neonatal disease and malaria. For the

population of approximately 35 million people, fewer than 5000 doctors are active in the country *(Wasswa, 2012)*. Traditional healers are still strongly integrated into Ugandan Society. According to *Abbo (2011)*, at least one traditional healer lives in each village and the *World Health Organisation (2002)* estimates that more than 80% of the population attend traditional healers for health reasons, particularly in rural areas. Regarding CLP, the incidence in Uganda is 0.73 per 1000 live births, indicating that approximately 1100 babies are born with a cleft each year *(Dreise et al., 2011)*. *Walker et al. (2010)* reported that 80% of the CLP operations executed between 1 August 2007 and 31 July 2008 were performed by externally funded surgeons. Since 2009, children and adults with CL/P can be operated for free in the Comprehensive Rehabilitation Services in Uganda (CoRSU) hospital in Kisubi, a non-profit and non-governmental organization supported by Christian Blind Mission (CBM) and Smile train that focuses on children with physical impairment. In CoRSU, one experienced British plastic and reconstructive surgeon (A.H.) performs surgery in patients with CL/P. Moreover, local plastic surgeons are trained in order to stimulate maintenance and sustainability of cleft care. In 2010, a Speech-Language Therapy department was established in the hospital in order to follow-up speech and language development in children with cleft (lip and) palate and to diagnose speech and language disorders in children with e.g. developmental delay or hearing impairment. A graduate of the local Speech-Language Therapy program is being trained to provide services to the children with CLP and their families. Except for the Speech-Language Therapy department of Mulago Hospital in Kampala, this is currently the only department for Speech-Language Therapy in Uganda. According to *Fagan & Jacobs (2009)*, only two speech-language therapists (SLTs) were working in Uganda in 2009. However, since the establishment of a bachelor education at the Makarere University, the number of SLTs has increased to 19 in 2012.

Babies with unrepaired clefts of the palate generally show a significant weight loss in the first few months due to the inability to adequately breastfeed *(Miller, 2011)*. Much has been written about feeding adjuncts and milk substitutes *(Jones, 1988; Pandya & Boorman, 2001)*. However, there are only limited resources available in Uganda. Consequently these children are at great risk for malnutrition *(Cubitt et al., 2012)*. In order to improve nutrition and to prevent babies from failure to thrive and death, babies with CLP receive in CoRSU synchronous surgical repair once they reach a target weight of 3 kg, irrespective of age *(Cubitt et al., 2012)*.

As a consequence, the CLP closure is generally performed before the age of 6 months and, thus, before the onset of babbling. Previous experience pointed out that patients with CLP who have their lip repaired often do not return for palate repair because the importance of the palate in speech is not appreciated and given that the lip stigma has been removed for society *(Hodges & Hodges, 2000; Onah et al., 2008)*. Moreover, economic pressure on families makes it extremely difficult for them to travel long distances many times for surgery and follow-up *(Hodges, 2010)*. A synchronous repair of the CLP before the age of one year is rarely performed in cleft centers in Europe and the USA *(Fudalej et al., 2008)*. The influence of a synchronous lip and palatal closure on speech and facial appearance of patients with clefts might differ from the impact of a two-stage lip and palatal closure, especially when performed before the age of six months. Moreover, the effect on parental satisfaction after synchronous repair of the CLP has never been described in literature. Therefore, the main purpose of the current study is to determine parental satisfaction with treatment outcome in Ugandan children with complete unilateral (UCLP) and bilateral (BCLP) cleft lip and palate, closed during a synchronous lip and palatal repair. Moreover, the influence of age, gender, age at lip and palatal closure, time period between cleft closure and survey, cleft type and maternal vs. paternal judgments on satisfaction ratings is assessed. An additional aim of the study is to compare the results with an age- and gender-matched control group. Based on the results of previous reports, a significantly greater satisfaction with appearance and function is hypothesized in parents of children without a cleft compared to parents of children with CLP. Moreover, knowledge of satisfaction with treatment outcome allows detection of persisting problems or disorders and may result in changing timing and/or technique of the surgical procedures. In summary, the following questions were explored in the present study:

1. Are parents of Ugandan children with UCLP or BCLP satisfied with the child's facial appearance and speech after a synchronous repair of the lip and palate?
2. Which aspects influence parental satisfaction regarding facial appearance and speech (chronological age, gender, age at lip and palatal closure, time period between cleft closure and the survey, cleft type and/or maternal vs. paternal judgments)?
3. Does the parental satisfaction regarding facial appearance and speech in children with CLP differ from parental ratings of a control group?

Methods

The research protocol was approved by the Ethical Committee of the Ghent University Hospital, Belgium (EC2011/269).

Design

In order to control for possible influences of age, gender, race and ethnicity, a case control design was chosen, including comparison of parental satisfaction in Ugandan children with CLP and Ugandan orphan children. *Rumsey et al. (2004)* observed differences in appearance-related concerns between white and black adults with visible disfigurements. Furthermore, *Gilbert (1994)* noted racial and ethnical differences in satisfaction with appearance of teeth in South-African adults aged 20-64 years. Consequently, comparison with a Ugandan control group is recommended in order to fit the results in with the black race and the Ugandan ethnicity, as reference data for black East African patients with CLP are not yet available. Although the control children would be likely to be viewed more positively on appearance and speech compared to children with CLP, parental satisfaction regarding some cleft-related features might not differ due to the success of surgery.

Participants and sampling

All the parents or guardians of children with CLP who visited the Speech-Language Therapy department of the CoRSU hospital for the first time between September 2010 and January 2012 were asked to participate in the present study, of which 39 parents (26 mothers and 13 fathers) and 5 guardians (2 aunts and 3 grandmothers) agreed. These participants were the main caregivers of the children and descended from all strata of society. The procedure was first explained verbally to all participants in their mother tongue by a translator and, subsequently, the participants were asked to sign an English informed consent form. In case of illiteracy, the participants made a thumb-print.

All patients with CLP were treated by one experienced surgeon (A.H.). A synchronous lip and palatal closure was carried out. To perform lip closure, a modified Millard repair (UCLP) and a modified Mulliken technique (BCLP) were used and for palatal repair, the Sommerlad method with push back of the soft palate musculature was the standard technique. Follow-up was done by the surgeon (A.H.) in the perioperative period and on a regular basis in the first postoperative year. Furthermore, patients were referred to the Speech-Language Therapy department for speech evaluation and/or advice. Patients with speech problems from 2;6 years are assessed on repeated occasions.

The foster mothers of 44 orphan children were included in the age- and gender-matched control group. A Mann-Whitney U test showed no significant differences in chronological age between children with CLP and control children (p=0.132). All orphan children live in an orphan home in Entebbe, located at 4 km from CoRSU. In the orphan home, daily life is similar to a common well-to-do Ugandan family living in rural areas: in each of the ten houses, one local Ugandan foster mother cares for about ten children. A description of the children with CLP and the control children is presented in Table 7.1.

Table 7.1 Overview of the children with CLP and the orphan children, judged by the experimental group and the control group, respectively.

	Experimental group	Control group
n	44	44
Gender	21 males, 23 females	21 males, 23 females
Chronological age: mean (range)	3;1 years (1;0-7;8 years)	3;7 years (0;11-7;8 years)
Cleft type	32 complete UCLP 12 complete BCLP	-
Age cleft closure: mean (range)	1;0 years (0;1-7;6 years)	-
- ***Closure ≤ 6 months: n***	28	-
- ***Closure > 6 months: n***	16	-
Duration between surgery and survey: mean (range)	2;1 years (0;2-5;6 years)	-
Exclusion criteria	- Cleft type: CL, CP, submucous cleft (n=28) - Unrepaired cleft (n=8) - Syndromes (n=2) - Not the main caregiver (n=2)	- Speech and/or language pathologies (n=11) - Neurological anomalies (n=1)
CL: cleft lip, CP: cleft palate		

Materials and procedures

The survey, used to assess satisfaction with cleft-related features, was based on the Cleft Evaluation Profile (CEP) *(Turner et al., 1997)*. This assessment originates from the Royal College of Surgeons Cleft Lip and Palate Audit Group and is applied in several satisfaction studies regarding clefts *(Noor & Musa, 2007; Turner et al., 1997; Van Lierde et al., 2012)*. All items of the CEP are

related to features that play a major role in assessing function and facial appearance among patients with CLP *(Noor & Musa, 2007)*. In previous investigations, parents and/or patients were asked to rate their satisfaction for each item of the CEP on a 7-point Likert scale, ranging from very satisfactory to very unsatisfactory. However, many caregivers in the present study were not familiar with the use of gradation with a 7-point scale. Therefore, the parents and guardians of the patients with CLP and the foster mothers of the control children were asked to judge if they were satisfied or dissatisfied with the following items: speech, hearing and appearance of the teeth, the lip, the nose and the face. These six standardized questions were verbally asked in English, one of the official languages of Uganda, by an SLT of the Ghent University speech-language team and were translated to the participants' mother language by a local translator if the participants' comprehension of English was insufficient. Given that in the local Bantu languages separate words exist for 'language' and 'speak', the translation will probably not have influenced the participants' interpretation of the concept 'speech'. The order of the questions was similar for all participants. The responses to the interview which lasted 5 minutes were computerized without delay. For the experimental group, the interviews occurred in an office of CoRSU; the foster mothers were interviewed in the orphan home.

Statistical analyses

The SPSS software (version 19) was used for statistical analyses of the data. The parental judgments regarding the appearance of the teeth and speech were excluded from the analyses for children younger than 20 months and 18 months of age, respectively. At the age of 20 months, the incisors and canines are present in most typically developing children. Furthermore, the vocabulary generally increases exponentially from the age of 18 months.

To assess the influence of chronological age and time period between primary surgery and survey, the normality of both variables was assessed. Subsequently, a non-parametric Mann-Whitney U test and a parametric Independent Samples t-test were performed for these variables, respectively. For statistical analysis regarding age at cleft closure, children with a lip and palatal repair before the age of 6 months were divided from the children with a lip and palatal closure above the age of 6 months. A Kruskal-Wallis rank sum test indicated no significant age differences between children with cleft closure before and after the age of 6 months (p=0.075). Differences in satisfaction ratings related to gender, cleft type (UCLP vs. BCLP), age at cleft closure, judge (mother vs. father) or groups (control vs. experimental group) were separately analyzed using chi-square tests. All significance levels were set at α=0.05. Bonferroni corrections were applied when necessary.

Results

Results for the CEP regarding the experimental group and the control group are provided in Table 7.2.

Table 7.2 Results for the Cleft Evaluation Profile regarding the experimental group and the control group.

	Experimental group		Control group		p
	% satisfied (n)	% dissatisfied (n)	% satisfied (n)	% dissatisfied (n)	
Speech	56% (19/34)	44% (15/34)	100% (39/39)	0% (0/39)	*<0.001*
Hearing	98% (43/44)	2% (1/44)	100% (44/44)	0% (0/44)	0.315
Teeth	56% (18/32)	44% (14/32)	100% (38/38)	0% (0/38)	*<0.001*
Lip	93% (39/42)	7% (3/42)	100% (44/44)	0% (0/44)	0.071
Nose	86% (38/44)	14% (6/44)	100% (44/44)	0% (0/44)	*0.011*
Face	100% (44/44)	0% (0/44)	100% (44/44)	0% (0/44)	/

Speech

Fifty-six percent (19/34) of the parents and guardians of children with CLP were satisfied with speech, whereas 44% (15/34) were dissatisfied. No significant differences were established regarding age (p=0.271), gender (p=0.224), age at lip and palatal closure (p=0.459), time period between cleft closure and survey (p=0.435), cleft type (p=0.560) or maternal vs. paternal judgment (p=0.514). In the control group, all foster mothers (100%, 39/39) were satisfied with the children's speech. The difference between the experimental group and the control group was significant (p<0.001).

Hearing

Regarding hearing, 98% (43/44) of the parents and guardians of children with CLP were satisfied and 2% (1/44) were dissatisfied. Age (p=0.091), gender (p=0.334), age at lip and palatal closure (p=0.181), cleft type (p=0.536) and maternal vs. paternal judgment (p=0.474) did not differ significantly within the cleft group. Furthermore, no significant differences

were observed between the experimental and control group (p=0.315), because all foster mothers (100%, 44/44) were satisfied with the hearing of the orphan children.

Appearance of the teeth

In the experimental group, 56% of the participants (18/32) were satisfied with the appearance of the children's teeth, whereas 44% (14/32) were dissatisfied. No significant differences could be established concerning age (p=0.561), gender (p=0.393), age at lip and palatal repair (p=0.574), time period between cleft closure and survey (p=0.486), cleft type (p=0.541) or maternal vs. paternal judgment (p=0.268). In contrast, all foster mothers in the control group (100%, 38/38) were satisfied with the children's appearance of the teeth. The difference between the experimental and control group reached significance (p<0.001).

Appearance of the lip

In the experimental group, two missing data regarding appearance of the lip were registered. Ninety-three percent (39/42) of the experimental group was satisfied with the children's appearance of the lip. Regarding age (p=0.345), gender (p=0.607), age at lip and palatal repair (p=0.204), cleft type (p=0.688) and maternal vs. paternal judgment (p=0.585), no significant differences were observed. In contrast, time period between cleft closure and survey significantly influenced the level of parental satisfaction (p=0.027). In dissatisfied participants, the time period was significantly larger (mean 3;7 years) compared to satisfied participants (mean 2;0 years). In the control group, all foster mothers (100%, 44/44) were satisfied with the children's appearance of the lip. The difference between the experimental group and the control group was not significant (p=0.071).

Appearance of the nose

Regarding the appearance of the children's nose, 86% (38/44) of the parents and guardians of children with CLP were satisfied and 14% (6/44) were dissatisfied. No significant differences could be observed in the experimental group, neither regarding age (p=0.559) or gender (p=0.448), nor regarding age at lip and palatal closure (p=0.868), time period between cleft closure and survey (p=0.171), cleft type (p=0.720) or maternal vs. paternal judgment (p=0.455). Given that all foster mothers (100%, 44/44) were satisfied with the appearance of the children's nose, the experimental group was significantly more dissatisfied in comparison to the control group (p=0.011).

Appearance of the face

All parents and guardians of children with CLP (100%, 44/44) and all foster mothers of the orphan children (100%, 44/44) were satisfied with the children's overall appearance of the face. Consequently, no further statistical analysis needs to be performed.

Discussion

The main aim of the present study was to assess satisfaction with speech and facial appearance for caretakers of Ugandan children with repaired isolated complete UCLP or BCLP. The cleft was surgically closed during a synchronous lip and palatal repair at a mean age of 1;0 year. In addition, these results were compared to a control group. Assessment of parental satisfaction with speech and facial appearance in young children with clefts is important, as the caretakers' view substantially influences the children's attitude toward their own appearance and speech *(Bull & Rumsey, 1988)*. Aesthetics affect the patient's social acceptance early in life *(Onah et al., 2008)*, particularly in the Ugandan society where children with clefts are at risk for social isolation due to the congenital malformations *(International Labour Organization, 2009)*. Furthermore, success of surgical treatment and persisting problems can be evaluated by the level of parental satisfaction *(Van Lierde et al., 2012)*. In the present study, overall high levels of satisfaction were observed in the experimental group, ranging from 56% regarding speech and the appearance of the teeth to 100% for the appearance of the face. These findings confirm previous studies focusing on parental satisfaction with speech and facial appearance in children with CL/P *(Nelson et al., 2012)* and indicate that parents and guardians considered the surgery to be worthwhile. Nevertheless, significantly more parents in the control group were satisfied with speech and appearance of the teeth and the nose. Moreover, the time period between cleft closure and survey was significantly larger regarding appearance of the lip in dissatisfied vs. satisfied participants. However, age and gender of the children with CLP, age at lip and palatal repair, cleft type (UCLP or BCLP) and parent that performed the rating (mother or father) did not significantly influence the level of parental satisfaction with speech and facial appearance. Absence of a significant difference regarding satisfaction with speech between parents of patients with UCLP or BCLP was a remarkable finding. The extent to which this comparable perception of parents can be related to similar results on detailed speech analyses is subject for further research.

Regarding appearance, all caretakers of the patients with CLP were satisfied with the face, and respectively 93% and 86% were satisfied with the lip and the nose. This high degree of satisfaction with aesthetics might be caused by the relief that the cleft lip (CL) is repaired. Normal aesthetics are of particular importance in the Ugandan society *(Human Rights Watch, 2010)*. Children with congenital malformations and facial disfigurement are often hidden and socially isolated. Surgical closure of the CL ensures an improvement of the facial appearance and, therefore, decreases social pressure and demonizing. This improvement of facial aesthetics will often lead to failure to complete the surgical procedure when the CL is closed during a first operation. Therefore, a synchronous CLP closure is adopted in CoRSU. Another possible explanation for the high level of parental satisfaction with facial appearance in these Ugandan patients is the positive influence of color and texture of the black African skin on the appearance of a scar, as this can drastically alter the way the scars are perceived *(Bush et al., 2011)*. However, these results should be interpreted cautiously, as the time period between cleft closure and survey in the three dissatisfied participants (1;6 years, 3;10 years and 5;6 years) regarding the appearance of the lip was significantly larger in comparison with satisfied participants. Therefore, critical judgments of facial appearance by an independent panel of surgeons, nurses and layman are necessary to confirm this hypothesis.

Similarly to appearance, high levels of satisfaction (98%) were observed regarding hearing. Dysfunction of the Eustachian tube in patients with clefts of the palate increases the frequency of recurrent acute otitis media and otitis media with effusion, causing a hearing loss of 30 dB on average *(Sharma & Nanda, 2009)*, which may negatively influence the speech-language development. As *Meyerson & Weddington (1986)* reported an overall decreased occurrence frequency of middle ear diseases among black vs. white American children, hearing problems might have been uncommon in the children with CLP of the present study. However, hearing has never been tested in Ugandan children with CL/P due to the financial costs and the lack of necessary professional equipment in most Ugandan hospitals. Therefore, parents of young Ugandan children with clefts might not be aware of an existing hearing impairment and, subsequently, highly rate their satisfaction with hearing.

In contrast to the high degree of satisfaction regarding overall facial appearance and hearing, significantly more participants from the experimental group were dissatisfied with the appearance of the teeth (44%) in comparison with the control group (0%). In patients with CLP, agenesis of the maxillary lateral incisor, a supernumerary tooth and rotated or displaced teeth are frequently seen dental anomalies *(Priya et al., 2011)*. Consequently, most children with CL/P need additional dental and orthodontic treatment. However, in the current study, patients were too young for orthodontic treatment and were assessed prior to any possible correction.

The degree of satisfaction regarding speech was significantly lower for the experimental group (56%) in comparison with the control group (100%). Compared to levels reported in satisfaction studies in the USA (12-17%) or the UK (5%) regarding children with CL/P from 5 years of age *(Broder et al., 1992; Williams et al., 2001)*, the level of dissatisfaction observed in the current study (44%) was considerably higher. Children with clefts are at risk for developing delayed or disturbed communication *(Richman, 1997)*. Compensatory articulation errors (e.g. glottal stops) and resonance disorders (e.g. hypernasality, nasal emission) are commonly reported in this population *(McWilliams & Witzel, 1994)*. In contrast to countries in Europe and the USA, speech therapy is generally not included in the treatment practice of Ugandan children with CL/P due to the limited number of Ugandan SLTs on the one hand *(Fagan & Jacobs, 2009)* and financial costs and absence of a medical insurance on the other hand. Furthermore, three-quarter of the Ugandan school-aged children with clefts in the current study (74%, 28/38) did not go to school. These factors both might have negatively influenced the satisfaction scores of the experimental group, considering that speech therapy and going to school usually ameliorate the patient's speech quality and intelligibility *(Broder et al., 1992)*. Further research regarding influence of race and ethnicity on parental satisfaction with treatment outcome is therefore recommended.

A few remarks need to be made regarding the present study. Firstly, a 2-point rating was used to assess parental satisfaction with function and appearance ('satisfied' and 'dissatisfied'), while generally a 7-point Likert scale is applied in the CEP (indicating a number from 1 to 7, ranging from very satisfied to very unsatisfied). Having only two possible outcomes could fail to detect degrees of satisfaction or dissatisfaction. A more discriminatory 7-point outcome scale might have picked up more subtle variations in levels of parental satisfaction. However, the current option was chosen, because most participants were not familiar with

the use of gradation with a 7-point scale. Since the same cleft-related items were rated by all participants, the content validation of the original CEP has most likely been maintained. In future research with African participants, the use of gradation is, however, advised, including the use of smiling and grumpy faces to reflect degrees of satisfaction. Secondly, parental satisfaction with speech and facial appearance was assessed during an interview. Respect for professionals and intimidation by being within a clinical environment might therefore have positively influenced the responses of parents and guardians of children with CLP *(Thomas et al., 1997; Turner et al., 1997)*, especially since these Ugandan participants were not familiar with surveys and scientific research. Nevertheless, using a questionnaire was not feasible, given that most parents or guardians were illiterate. Finally, the control group included foster mothers of orphan children instead of biological parents. Although daily life in the orphan home is similar to that of a common well-to-do Ugandan family living in a rural area and although the foster mothers care for these children as if they were their own, the foster mothers may differ from biological parents in their judgments.

Conclusion

In conclusion, the results of this study demonstrate that parents and guardians of Ugandan children with complete UCLP or BCLP who underwent a synchronous lip and palatal closure, are generally satisfied with the children's hearing and overall facial and lip appearance. Decreased satisfaction levels are observed regarding speech and appearance of teeth and nose. Synchronous lip and palatal repair is preferred to lip and palatal closure performed by multiple surgical interventions in Ugandan children with CLP given that failure to complete palatoplasty after lip repair is highly frequent in this cleft population, potentially resulting in speech and hearing problems. Raising public awareness and counseling parents concerning causes and consequences of clefts are indispensable in order to decrease prejudices and increase quality of life for patients with CL/P. Furthermore, establishing services for speech therapy, orthodontic treatment as well as systematic screenings for hearing disorders and diseases are essential for increasing parental satisfaction with appearance and function in Ugandan patients with clefts.

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Conflict of Interest Statement

The authors of this manuscript are employed by their respective institutions and have no relevant financial or non-financial relationships to disclose.

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Appendix 7.1: Continuing education questions

1. What is the primary goal of performing a synchronous lip and palatal repair as soon as possible in Ugandan babies born with cleft lip and palate?:
   a. To improve speech development in cleft children.
   b. To prevent babies from dying due to malnutrition.
   c. To decrease anxiety in cleft children due to hospitalization.
   d. To reduce surgery costs.
2. Which of the following cleft-related features differed significantly between the experimental group and the control group?:
   a. Parental satisfaction with appearance of the lip.
   b. Parental satisfaction with appearance of the face.
   c. Parental satisfaction with appearance of the teeth.
   d. None of the above-mentioned.
3. Which variables significantly influenced the levels of parental satisfaction regarding speech and facial appearance in the experimental group?:
   a. Age at surgical lip and palatal closure.
   b. Children's age at the time of survey.
   c. Cleft type.
   d. None of the above-mentioned.
4. Parents of cleft children are significantly more dissatisfied with the children's speech compared to foster mothers in the control group. This might be due to:
   a. Chronological age at cleft closure.
   b. The lack of speech therapy in the treatment protocol of Ugandan cleft children.
   c. The surgical technique used for cleft closure.
   d. The lack of speech and language stimulation by parents.
5. What is especially important to prevent Ugandan patients with clefts to be demonized and socially isolated?:
   a. Having normalized aesthetics and speech.
   b. Having a job.
   c. Having several children.
   d. Having a good education.

CHAPTER 8

IMPACT OF EARLY SYNCHRONOUS LIP AND PALATAL REPAIR ON SPEECH

Based on:

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Abstract

Aims: The purpose of this study was to describe articulation and resonance characteristics of Ugandan English-speaking children with cleft lip and palate (CLP) after synchronous lip and palatal closure (Sommerlad technique) prior to 6 months in comparison with an age- and gender-matched control group.

Methods: Eleven Ugandan patients with CLP (mean age 4;9 years), repaired during a synchronous lip and (soft and hard) palatal closure at a mean age of 3.4 months, were included as well as a control group (n=22) consisting of 2 Ugandan age- and gender-matched non-cleft children for each patient (mean age 4;10 years). Objective and perceptual speech assessment techniques were applied.

Results: Consonant inventories were significantly smaller in the CLP group. Moreover, phonetic disorders and phonological processes occurred in respectively 91% and 100% of the CLP group. Perceptual consensus evaluation revealed absence of hyponasality and cul-de-sac resonance in all patients. Hypernasality and nasal emission/turbulence occurred respectively in 18% and 27% of the patients. No significant group differences were observed regarding the mean nasalance values of oral speech samples.

Conclusion: Various deviations from normal speech development seem to occur in patients with CLP following synchronous lip and palatal closure before 6 months of age.

Key Words

Cleft • Speech • Resonance • Articulation • Synchronous lip and palatal closure • Timing of palatoplasty • Sommerlad technique

Introduction

The primary goal of surgical palatal closure in patients with cleft lip and/or palate (CL/P) is to restore the oronasal anatomy and velopharyngeal function in order to achieve normal feeding abilities, speech and hearing without disturbance of the maxillofacial growth *(Rohrich et al., 2000)*. Clefts of the palate (CP) are regularly characterized by obligatory (e.g. hypernasality, nasal emission) and compensatory (e.g. backing) speech disorders that might significantly decrease speech intelligibility *(Kummer, 2011)*. Literature reveals that speech results after surgical closure of the CL/P are determined by several variables such as surgical technique, experience of the surgeon, cleft type and age of cleft repair *(Denk & Magee, 1996)*. Since the previous century, many authors have studied the influence of timing of CP closure on speech *(Peterson-Falzone, 1996)*. The majority of the studies comparing speech results following early and late palatal repair, concluded that early closure of the CP was less associated with articulation *(Willadsen, 2012; Ysunza et al., 1998)* and resonance disorders *(Hardin-Jones & Jones, 2005; Rohrich et al., 1996)*. Therefore, one can question whether development of such speech disorders can be maximally avoided when successful palatal repair is performed prior to the first consonant productions, thus prior to the start of canonical babbling. Differences have been reported regarding onset, frequency and quality of canonical babbling productions of babies with untreated CP compared to typically developing infants *(Stout et al., 2011)*. Moreover, consonant inventories of babies with untreated CP have been noted to be smaller and to contain more consonants which require little or no intraoral air pressure *(Peterson-Falzone et al., 2006a; Stout et al., 2011)*. Considering that consonants frequently occurring in canonical babbling will also arise in the children's first words *(Peterson-Falzone et al., 2006a)*, deviations in canonical babbling might be a precursor for speech problems at a later age. Consequently, repair of the velopharyngeal sphincter prior to this prelinguistic stage (i.e. prior to 6 months of age) is assumed to prevent compensatory speech disorders to get well-established in the children's neuromuscular speech patterns and to emerge later in childhood *(Kaplan, 1981)*.

Technical difficulties of such surgical procedure and potential risks such as increased blood loss, airway obstruction and potential anesthetic problems have restrained many surgeons from performing a one-stage soft and hard palatal repair prior to 6 months of age *(Denk &*

Magee, 1996; Pradel et al., 2009; Rohrich et al., 2000), resulting in few opportunities to verify the afore reasoning. However, in the Comprehensive Rehabilitation Services in Uganda (CoRSU) hospital, very early synchronous repair of the cleft lip and palate (CLP) is considered to be a lifesaving procedure for Ugandan patients with CLP. The synchronous repair is advocated as failure to complete palatoplasty after lip repair is highly frequent in African patients with CLP due to operation costs, travelling costs, travelling distance and recovered aesthetics (Onah et al., 2008; Savaci et al., 2005) – in black African societies, clefts are often associated with evil spirits, curses and punishment for ancestral wrong doings (Dagher & Ross, 2004). Moreover, very early closure of the CP is advised as babies with unrepaired CP frequently lose weight in their first few months of life due to the inability to adequately breastfeed (Miller, 2011), resulting in a great risk for malnutrition (Cubitt et al., 2012). Consequently, synchronous CLP repair is performed once the patient has reached a target weight of 3kg, what is often prior to the age of 6 months, in order to improve nutrition and survival (Cubitt et al., 2012). Given that this cleft surgery is performed in safe circumstances by one experienced surgeon (A.H.) using the Sommerlad technique for palatal repair, this setting provides an excellent opportunity to assess the normality of speech characteristics after palatal repair prior to 6 months of age.

In literature, only few studies described speech after palatal repair prior to 6 months of age. The first reports were rather descriptive impressions and did not explain speech assessment strategies (Desai, 1983; Kaplan et al., 1982; Kaplan et al., 1974; Kaplan et al., 1980). Moreover, most later studies exclusively used perceptual (consensus) evaluations of one or more speech characteristics (Abdel-Aziz, 2013; Copeland, 1990; De Mey et al., 2006; Grobbelaar et al., 1995; Randall et al., 1983), except for Barimo et al. (1987), Ysunza et al. (1998) and Doucet et al. (2013), who included incompletely described phonetic analyses. In addition, in none of these studies, detailed phonological analyses or objective nasalance measurements were performed.

The studies noted divergent results for articulation and resonance after early palatal repair (≤ 6 months). Reports of normal articulation ranged from 40% (De Mey et al., 2006), over 57% (Copeland, 1990) to 65% (Randall et al., 1983) of the included patients, whereas Ysunza et al. (1998) noted articulation to be 'below normal limits' for all patients. Barimo et al. (1987) found absence of glottal stops and pharyngeal fricatives in all children aged 1;10 to 2;6 years, while Doucet et al. (2013) observed compensatory articulation (i.e. glottal stop, pharyngeal fricative or posterior nasal fricative) in 20% as well as velar substitution in 25% of the included patients. Regarding resonance, absence of hypernasality varied between 20% (Doucet et al., 2013) and 93% (Abdel-Aziz, 2013) of the study groups. Furthermore, nasal emission occurred in none of the patients described by Barimo et al. (1987) and Abdel-Aziz (2013), while Copeland (1990) and Doucet et al. (2013) observed nasal emission in 9% and 75% of the patients, respectively. These divergent results for articulation and resonance characteristics, ranging from close to normal to abnormal, might be associated with the various included surgical techniques, cleft types and/or speech evaluation strategies and do not provide a clear view on whether speech disorders can be majorly avoided when successful palatal repair is performed prior to the first consonant productions.

The purpose of this study is, therefore, to assess articulation and resonance characteristics in Ugandan English-speaking children with CLP after a synchronous lip and palatal closure prior to the age of 6 months using the Sommerlad technique. In order to verify normality, speech results of the CLP group will be compared to an age- and gender-matched non-cleft control group, as, to our knowledge, no information is yet available about speech development in typically developing Ugandan children. Given that the velopharyngeal sphincter would be repaired prior to the first consonant productions and given that good speech outcomes have been reported following palatal closure with the Sommerlad technique (Andrades et al., 2008), limited deviations from normal articulation and resonance are hypothesized.

Materials and methods

The research protocol was approved by the Ethical Committee of the Ghent University Hospital, Belgium (EC2011/269).

Participants

Seventy-five patients with CLP returned to the Speech-Language Therapy department of CoRSU after a synchronous lip and palatal closure at the age of 6 months or less. In 15% (11/75) of these patients, articulation and resonance assessments were performed. Patients with incomplete articulation assessments (n=3) or associated anomalies (n=5) and patients who were too young (< 2;6 years) (n=52) or uncooperative (n=4) were excluded. The experimental group consisted of 4 males and 7 females with a mean age of 4;9 years (range 3;2 to 7;2 years). Seventy-three percent (8/11) of the Ugandan patients attended school and the 3 youngest children would start in the near future. They were all native speakers of one of the Bantu languages, but the majority spoke Luganda (6/11, 55%). These subjects were however able to speak and understand English, since this is their second language. Considering that English is one of the official languages of Uganda according to the Ugandan constitution *(Uganda Law Reform Commission, 2006)*, the language is deeply rooted in media, administration and education *(Mpuga, 2003)*. Ten patients presented with unilateral CLP; bilateral CLP was diagnosed in 1 patient. All clefts were closed by one experienced surgeon (A.H.). The palatal repair was carried out using the Sommerlad technique; a modified Millard repair (unilateral CLP) or a modified Mulliken technique (bilateral CLP) was applied to perform lip closure. Cleft closure was carried out at a mean age of 3.4 months (range 2 to 6 months). No major peri- or post-operative complications such as anesthetic morbidity, were observed. However, secondary surgery for minor post-operative complications occurred in 6 patients (5/11, 45%), including one closure of wound dehiscence (1/11, 9%) and 5 fistula repairs (5/11, 46%). None of the patients received speech therapy or treatment for hearing problems prior to speech assessment.

The control group included for each patient two age- and gender-matched English-speaking Ugandan children, who attended school and had Luganda as native language and English as their second language. Inclusion of two children for each patient was chosen to increase the power of statistical group comparisons. Consequently, this group consisted of 22 Ugandan children with a mean age of 4;10 years (range 3;0 to 7;1 years). None of the children showed craniofacial anomalies or pathologies of oral, nasal or velopharyngeal structures and children with a temporary nasal obstruction were excluded. An Independent Samples t-test showed no significant age differences between the experimental group and the control group (p=0.912).

Speech assessment

Objective and subjective assessment techniques were applied to assess the articulation and resonance characteristics in this population. All participants were daily exposed to

English and were sufficiently able to comprehend and produce this language to complete an English speech assessment. Therefore, speech evaluations were carried out in English.

Articulation

The articulation skills were assessed using the standardized Photo Articulation Test – Third Edition (PAT-3) *(Lippke et al., 1997)*. This picture-naming test was developed for children from 3 years of age. While cued by colored pictures, the participants were asked to repeat 72 high frequency English words in which all English consonants occurred isolated in all permissible syllable positions as well as in common consonant clusters. The assessment was digitally videotaped using a Sony Handycam HDR-UX1 with a high quality built-in microphone to enable detailed consensus phonetic and phonological analysis. One speech-language therapist (SLT) (A.L.) experienced with cleft-related speech errors phonetically transcribed the speech samples using the International Phonetic Alphabet (IPA), IPA extensions *(International Phonetic Association, 1999)* and symbols for transcription of specific cleft-related articulation errors *(Peterson-Falzone et al., 2006b; Trost, 1981)*. Forty-eight percent (16/33) of the articulation assessments were independently transcribed by another SLT experienced with cleft-related speech disorders (K.B.). Mean consonant-for-consonant interjudge reliability was 80% (SD: 8.4%). The analyses concentrated on 18 consonants (/p/, /b/, /t/, /d/, /k/, /g/, /h/, /s/, /z/, /f/, /v/, /w/, /j/, /l/, /r/, /n/, /m/, /ŋ/) that occur in English and Luganda as experience shows that even Ugandan adults without clefts often show difficulties in correctly producing the English fricatives /ʃ,ɵ,ð/ and affricatives /tʃ,dʒ/. Articulation analyses included a consonant inventory and a phonetic and phonological analysis. The consonant inventory consisted of all native consonants that were correctly produced twice without making reference to the intended target sound. During the phonetic and phonological analysis, participants' productions were compared to the target sounds. In the phonetic analysis, error types at phoneme level were examined (omissions, additions, substitutions and distortions). The phonological analysis focused on error patterns at word level according to Ingram's classification (syllable structure processes, substitution processes and assimilation processes) *(Ingram, 1982)*. For each phonetic or phonological error type, the occurrence frequency (%) was calculated in function of the number of potential occurrences. When such error type could occur at least 4 times and presented in at least 20%, the error type was considered productive *(McReynolds & Elbert, 1981)*.

Resonance

Assessment of resonance included perceptual consensus evaluation and objective measurements of nasalance values.

PERCEPTUAL CONSENSUS EVALUATION

Digital audio-recordings of the first 30 words of the PAT-3 *(Lippke et al., 1997)* were perceptually evaluated by two SLTs (A.L., K.B.) who were blind for the participants' condition (cleft/non-cleft). Presence of hypernasality (absent, minimal, slight, moderate, severe), hyponasality (absent, mild, marked), cul-de-sac resonance (absent, mild, marked) and nasal emission/turbulence (absent, occasional, frequent) was rated in accordance with *John et al. (2006)*. Considering that nasal turbulence results from air flowing through an increased resistance in the nasal airway *(Peterson-Falzone et al., 2006b)*, nasal emission and nasal turbulence were judged as one *(Henningsson et al., 2008)*. Speech samples were first judged simultaneously, but independently. In case of disagreement, the sample was replayed until a consensus was reached. Mean interjudge reliability, calculated by the ratio of identical to total judgments was 97% (128/132).

NASALANCE VALUES

Objective assessment of resonance was performed using the Nasometer (model II 6450), a microcomputer-based system manufactured by *Kay Elemetrics Corporation (2010)*. This instrument uses two microphones on either side of a horizontal sound separator plate that rests on the upper lip. The signal from each microphone is individually filtered and digitized by custom electronic modules and, subsequently, the sound is processed by a computer. The resultant signal is a ratio of 'the nasal acoustic energy' to 'the nasal-plus-oral acoustic energy'. This ratio is multiplied by 100 and expressed as a 'nasalance score'. Prior to data collection, the Nasometer was calibrated according to the manufacturer's instructions. Placement of the headgear and necessary adjustments were conducted according to the manufacturer's specifications. Participants from the CLP and control groups were asked to repeat three types of speech samples from the MacKay-Kummer Simplified Nasometric Assessment Procedures (SNAP) test *(Kummer, 2005; MacKay & Kummer, 1994)*. The sustained vowels /a/, /i/ and /u/ were produced four times during two seconds at a comfortable pitch and loudness. Furthermore, the children were asked to repeat four times 8 consonant-vowel

syllable sequences containing the vowels /a/ or /i/ combined with a voiceless plosive (/p,t,k/) or fricative (/s/) consonant. The final speech sample included 12 English simple oral sentences, majorly containing bilabials, alveolars, velars or sibilants. Each sentence group was repeated twice, while the child was cued by pictures. For the nasometric assessment of syllable sequences and sentences, all patients speaking with nasal emission (n=3) and their control-counterparts (n=6) were excluded, as presence of nasal emission might incorrectly increase the nasalance values, although they should provide an indication of hyper/hyponasality *(Dalston et al., 1991)*.

Statistical analyses

Statistical analyses were completed using SPSS software (version 20.0). The CLP and control group were statistically compared. For categorical data (consonant inventory, amount of subjects with/without a certain error, perceptual consensus evaluation), 2x2 contingency tables were used. Chi-square tests or Fisher's exact tests were applied, depending on the expected cell counts. Normality of each continuous variable (total consonant inventory, phonetic analysis, phonological analysis) was tested separately for both groups using the Kolmogorov-Smirnov and Shapiro-Wilk test. In cases of normal distributed observations, Independent Samples t-tests were applied. When distributions deviated from normal, non-parametric Mann-Whitney U tests were performed. All significance levels were set at α=0.05.

Results

Articulation

Consonant inventory

A Mann-Whitney U test revealed a significant difference in completeness of the consonant inventory between the CLP (median 16/18, range 8-18/18) and control group (median 18/18, range 15-18/18) (p=0.004). The plosive /d/ (p=0.010) and the fricatives /s/ (p=0.030), /z/ (p<0.001) and /v/ (p=0.027) were significantly less present in the consonant inventory of the CLP group.

Phonetic analysis

Results regarding the phonetic analysis for the CLP and control group are presented in Figure 8.1. Distortions were the most commonly occurring phonetic errors in both groups. The CLP group produced significantly more consonant omissions (especially plosives and fricatives) (p=0.020), distortions (p=0.002) and substitutions (p=0.041) compared to the control group. No significant group differences were obtained for the occurrence frequency of additions (p=0.221).

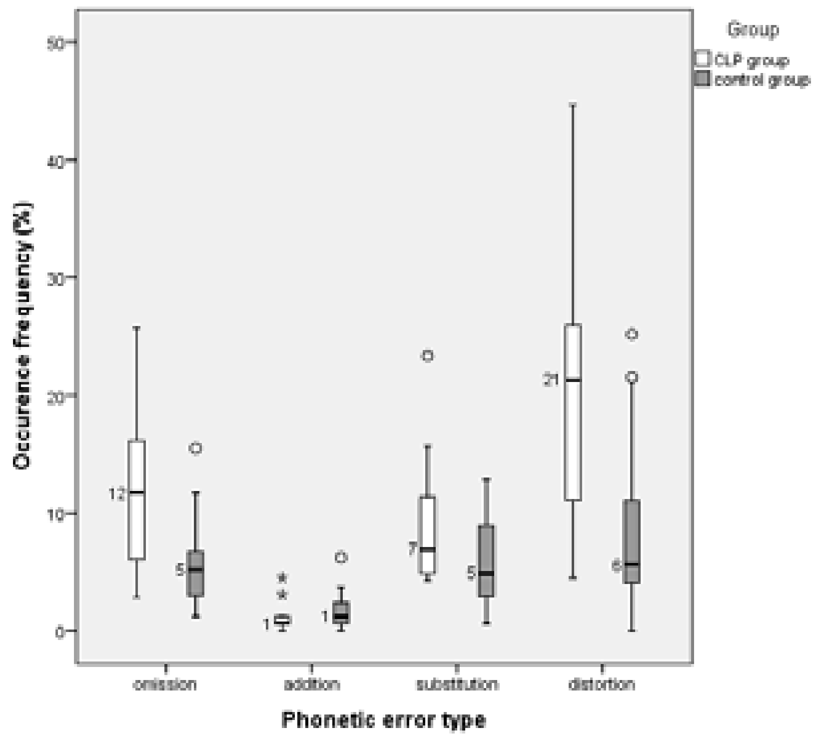

Figure 8.1 Occurrence frequency by overall phonetic error types for both the CLP (white) and control group (grey). Median occurrence frequencies are indicated next to the box plot.

Table 8.1 presents the results regarding the occurrence frequency of distortions for both groups. Group comparison revealed that the presence of (inter)dental productions of apico-alveolar consonants (p=0.270), lateral production of fricatives /s,z/ (p=0.157) and production of weak fricatives (p=0.388) or plosives (p=0.256) were similar for both groups. Significantly higher occurrence frequencies were observed in the CLP group compared

to the control group for compensatory articulation (i.e. produced with a palatal, velar, pharyngeal or glottal) of fricatives /s,z,f,v/ (p<0.001) and plosives /t,d/ (p<0.001) and for bilabial production of labiodentals /f,v/ (p=0.001).

According to the 20% frequency of occurrence criterion of McReynolds & Elbert (1981), at least one phonetic disorder was established in 91% (10/11) of the CLP group and 50% (11/22) of the control group. This group difference was significant (p=0.027).

Table 8.1 Results regarding occurrence frequency of distortions (expressed in %). Mean and median frequency of occurrence as well as standard deviation (SD) and range are presented for the CLP and control group. The p-values resulting from non-parametric Mann-Whitney U tests are indicated.

Distortion type	CLP group				Control group				p
	mean	SD	median	range	mean	SD	median	range	
(Inter)dental apico-alveolar consonants	13	14.4	9	0-41	8	12.1	3	0-50	0.270
Bilabial articulation of /f,v/	19	27.0	0	0-86	0	0.0	0	0-0	*0.001*
Lateral sigmatism	8	26.3	0	0-87	0	0.0	0	0-0	0.157
Weak plosives	6	6.8	4	0-23	4	3.5	3	0-13	0.256
Weak fricatives	10	8.1	7	0-23	8	9.9	6	0-40	0.388
Compensatory articulation of plosives	12	19.6	4	0-66	1	1.1	0	0-3	*<0.001*
- /p,b/	5	11.8	0	0-40	1	2.3	0	0-10	0.078
- /t,d/	27	31.3	13	0-89	1	2.0	0	0-5	*<0.001*
- /k,g/	8	23.2	0	0-77	0	1.2	0	0-4	0.381
Compensatory articulation of fricatives	31	26.3	21	0-83	4	9.4	0	0-39	*<0.001*
- /s,z/	41	33.1	43	0-94	6	13.1	0	0-54	*<0.001*
- /f,v/	9	18.5	0	0-57	0	0.0	0	0-0	*0.012*

Phonological analysis

Table 8.2 provides the occurrence frequency of phonological processes for the CLP and control group. In the CLP group, significantly higher occurrence frequencies were observed for cluster simplification (p=0.001), deletion of final consonants (p=0.009) and backing (p<0.001) compared to the control group.

Table 8.2 Results regarding the phonological analysis (expressed in %). Mean and median occurrence frequencies as well as standard deviation (SD) and range are indicated for the CLP and control group.

Phonological process	CLP group				Control group				p
	mean	SD	median	range	mean	SD	median	range	
SYLLABLE STRUCTURE PROCESSES									
Deletion final consonants	14	11.4	13	2-34	5	5.3	2	0-16	*0.034*[a]
Deletion initial consonants	2	2.8	0	0-8	2	2.9	0	0-8	0.687[a]
Deletion unstressed syllable	1	1.5	0	0-3	2	2.6	1	0-9	0.651[a]
Cluster simplification	38	17.9	41	9-62	16	10.8	15	3-48	*0.003*[b]
- Cluster reduction	35	18.8	38	6-62	15	9.8	13	3-42	*0.005*[a]
- Epenthesis	8	6.0	6	0-20	3	4.3	0	0-15	*0.008*[a]
Coalescence syllables	1	1.4	0	0-3	1	1.7	0	0-7	0.691[a]
Coalescence sounds	0	0.0	0	0-0	0	0.5	0	0-2	0.310[a]
SUBSTITUTION PROCESSES									
Fronting	8	8.1	4	0-26	5	4.8	4	0-18	0.420[a]
Backing	17	17.8	12	1-61	2	2.9	1	0-13	*<0.001*[a]
Stopping	3	4.8	0	0-16	1	2.3	0	0-10	0.159[a]
Liquid gliding	31	23.3	24	8-86	19	12.5	21	0-43	0.072[b]
Denasalization	0	0.0	0	0-0	0	0.0	0	0-0	1.000[a]
Devoicing	1	1.7	0	0-4	2	3.2	0	0-8	0.240[a]
ASSIMILATION PROCESSES									
Assimilation	3	3.6	2	0-11	3	2.6	2	0-10	0.893[a]
Reduplication	0	0.0	0	0-0	2	0.0	3	0-15	*0.025*[a]
OTHER PROCESSES									
Metathesis	1	1.1	0	0-3	2	1.6	2	0-5	*0.031*[a]
Substitution /n/ → /m/	11	22.7	0	0-77	4	5.1	0	0-17	0.595[a]

[a]Non-parametric Mann-Whitney U test; [b]Parametric Independent Samples t-test

According to the 20% criterion, at least one phonological process occurred in all patients with CLP (11/11, 100%) (i.e. deletion of final consonants, cluster simplification, fronting, backing, liquid gliding and substitution /n/ → /m/) and in 55% (12/22) of the controls (i.e. cluster reduction and liquid gliding). A Fisher's exact test revealed a significant group difference (p=0.013).

Resonance

Perceptual consensus evaluation

In the CLP group, hyponasality and cul-de-sac resonance were absent in all patients (11/11, 100%). Moreover, only few cases of hypernasality (82%, 9/11) and nasal emission (73%, 8/11) were observed. Minimal (1/11, 9%) and moderate (1/11, 9%) hypernasality occurred each in 1 patient. Nasal emission/turbulence was occasional (2/11, 18%) or frequently (1/11, 9%) observed in 3 patients. In the control group, none of the participants showed hypernasality, hyponasality, cul-de-sac resonance or nasal emission/turbulence. Group differences were only significant for nasal emission/turbulence (p=0.030).

Nasalance values

The mean nasalance values and standard deviations for the CLP and control group are presented in Table 8.3. None of the mean nasalance values of oral speech samples differed significantly between both groups (p<0.05).

Discussion

The purpose of the present controlled study was to describe articulation and resonance characteristics of Ugandan English-speaking patients with CLP (mean age 4;9 years) after synchronous lip and palatal closure (Sommerlad technique) performed before the age of 6 months (mean age 3.4 months) in comparison with an age- and gender-matched control group. In the middle of the speech-language development, speech was overall characterized by (1) incomplete consonant inventories. The most vulnerable consonants were the plosive /d/ and the fricatives /s,z,v/; (2) the presence of substitutions, omissions and distortions. In particular, distortions such as compensatory articulation of fricatives /s,z,f,v/ and plosives /t,d/ as well as bilabial production of /f,v/ were present; (3) at least one phonological

Table 8.3 Mean nasalance values (%) and standard deviation (SD) for the CLP and control group. Only patients speaking without nasal emission/turbulence (n=8) and their control-counterparts (n=16) are included for the syllable sequences and the sentence groups. Missing values for oral sentences were noted for two patients.

Speech sample	CLP Group			Control group			*p*
	n	Nasalance	SD	n	Nasalance	SD	
/a/	11	18.5	9.95	22	13.8	10.38	0.069[a]
/i/	11	33.6	6.98	22	26.7	16.72	0.444[a]
/u/	11	27.0	17.84	22	21.4	14.52	0.337[b]
/papapa/	8	11.3	4.62	16	10.2	7.85	0.417[a]
/pipipi/	8	20.3	11.18	16	17.3	15.92	0.238[a]
/tatata/	8	12.0	4.54	16	11.3	8.76	0.120[a]
/tititi/	8	20.0	9.70	16	20.7	16.67	0.653[a]
/kakaka/	8	10.6	4.75	16	10.8	6.35	0.742[a]
/kikiki/	8	20.4	8.59	16	21.2	15.05	0.697[a]
/sasasa/	8	18.9	18.23	16	12.2	9.60	0.120[a]
/sisisi/	8	30.6	18.96	16	22.3	15.49	0.153[a]
Bilabials	6	18.2	8.06	12	17.1	8.31	0.796[b]
Alveolars	6	18.3	8.29	12	17.7	8.88	0.880[b]
Velars	6	18.2	6.77	12	20.3	9.35	0.622[b]
Sibilants	6	26.7	14.86	12	23.0	11.43	0.569[b]

[a]Non-parametric Mann-Whitney U test; [b]Parametric Independent Samples t-test is applied.

process with an occurrence frequency ≥ 20% such as cluster simplification, liquid gliding, deletion of final consonants, fronting, backing or substitution of /n/ by /m/; (4) absence of hypernasality, hyponasality, cul-de-sac resonance and nasal emission in the majority of the patients; and (5) nasalance values within the normal range for all oral speech samples.

Optimal timing of primary repair of the soft and hard palate has been subject of discussion during the previous decades *(Peterson-Falzone, 1996)*. Many cleft and craniofacial teams advocate early primary repair of the soft and hard palate as this would provide better chances for good speech abilities *(Berkowitz, 2013)*, including prevention of compensatory articulation. In CoRSU, Uganda, repair of the soft and hard palate is performed prior to 6 months of age. Such timing protocol is rarely applied in clinical practice due to the technical difficulty of the surgical procedure and the potential risks such as increased blood loss, airway obstruction and potential anesthetic problems *(Denk & Magee, 1996; Pradel et al., 2009; Rohrich et al., 2000)*. However, in resource-poor countries, early synchronous lip and palatal closure can be

a life-saving procedure. Moreover, as mentioned in literature *(Denk & Magee, 1996; Hodges & Hodges, 2000)* and as seen in this study, generally no anesthetic morbidity and mortality are present.

When results of the current study are compared to those of previously published studies regarding speech following palatal repair prior to 6 months, few similarities and especially differences are obtained. These differences might be explained by variations in cleft type, surgical technique, age at speech assessment, speech therapy received prior to assessment and speech evaluation strategies.

In the present study, hypernasality was observed in 18% (2/11) of the CLP group and nasal emission/turbulence was found in 27% (3/11) of the patients. Similarly, *Abdel-Aziz (2013)* reported hypernasality in 14% (3/21) of the 4-year-old patients with cleft soft palate following palatal repair performed at 3 to 6 months of age with the Furlow Z-plasty, although none of them presented with nasal emission. In contrast, higher percentages for hypernasality have been reported by *De Mey et al. (2006)* (70%, 31/44) and *Doucet et al. (2013)* (80%, 16/20) for 3-year-old children with UCLP. Moreover, *Doucet et al. (2013)* reported higher rates of nasal emission (75%, 15/20) compared to the current Ugandan CLP group. These differences might be explained by the two-stage soft and hard palatal closure carried out in both studies, given that two-stage procedures have been reported to result in significantly more deviations of resonance compared to one-stage palatoplasty *(Van Lierde et al., 2004)*.

Regarding articulation, 91% (10/11) of the current CLP group showed at least one phonetic disorder with an occurrence frequency ≥ 20% compared to 50% (11/22) of the control group. Similarly, *Ysunza et al. (1998)* reported articulation scales below normal limits for all 4-year-old patients with UCLP after minimal incision palatopharyngoplasty at the age of 6 months. However, in other studies about speech following early palatal closure, percentages of patients with normal articulation varied from 40% (18/44) *(De Mey et al., 2006)* over 57% (57/100) *(Copeland, 1990)* to 65% (11/17) *(Randall et al., 1983)*. The higher number of patients with normal articulation reported by *Copeland (1990)* was expected as 42% of the patients received speech therapy before assessment. *De Mey et al. (2006)* and *Randall et al. (1983)* gave no information about this possible influencing variable. Furthermore, *Barimo et al. (1987)* and *Doucet et al. (2013)* noted absence of glottal stops and pharyngeal fricatives in respectively 100% (22/22) and 85% (17/20) of the included patients, while in the current study, compensatory articulation of plosives /t,d/ and fricatives /s,z,f,v/ were significantly more often observed compared to the control group. The potential occurrence of compensatory articulation of high-pressure consonants is well-known in patients with palatal clefts. These errors generally arise as a functional offset to the structural anomaly in order to improve velopharyngeal closure *(Peterson-Falzone et al., 2001)*. Moreover, they often remain after recovered anatomy and physiology *(Peterson-Falzone et al., 2001)*. Consequently, the significantly higher occurrence of compensatory articulation errors in the current CLP group might be a remainder of former minor post-operative complications, considering that 83% (5/6) of the Ugandan patients with repaired fistulae (n=5) or palate dehiscence (n=1) show compensatory articulation of at least one sound with an occurrence frequency ≥ 20% and given that secondary surgery was performed at a median age of 14 months (range 6 to 49 months). Nevertheless, secondary surgery seemed to have restored the potential to close the nasal airway during speech, given the lack of significant group differences for weak plosives and fricatives and considering the absence of hypernasality and nasal emission in most patients with CLP. Unfortunately, confirmation by objective nasoendoscopic or videofluoroscopic assessment of the velopharyngeal mechanism was impossible as the equipment was not available in this Ugandan hospital.

In addition to the phonetic errors, at least one phonological process was noted in all patients (11/11) of the current CLP group. Given that the majority of phonological processes is usually considered to be vanished in typically developing children by the age of 4;0 to 4;6 years (except for liquid gliding) *(Grunwell, 1981)*, the presence of phonological processes in the youngest participants of the present study might have been age-appropriate. However, deletion of final consonants, cluster simplification and backing occurred significantly more in the CLP group compared to the control group. Moreover, 83% (5/6) of the patients older than 4;6 years showed at least one phonological process different from liquid gliding. Consequently, phonological development of the included patients seems to be delayed, what might be explained by the above-mentioned former structural constraints of the patients' speech mechanism *(Chapman, 1993)*.

Considering the presence of compensatory articulation disorders and phonological processes in the current CLP group, speech therapy might be recommended in some

patients with early synchronous lip and palatal repair. However, unlike in countries in Europe and the USA, speech therapy is generally not included in the treatment practice of Ugandan patients with CLP due to the limited number of SLTs *(Fagan & Jacobs, 2009)*, the financial costs and the lack of a medical insurance. Consequently, expansion of speech-language therapy training and specialization of SLTs in cleft-related speech disorders is necessary in Uganda. When speech therapy can be provided, the goals and approaches need to be adapted to the error patterns of the individual patient. Motor-oriented speech therapy should be applied in the majority of the patients in order to modify the atypical production of disordered speech sounds by teaching the correct place and manner of articulation for a single phoneme at a time *(Van Riper, 1978)*. Early initiation of the therapy is preferred, as it is easier to eliminate compensatory articulations initially than to treat them subsequently *(Kuehn & Henne, 2003)*. Correction of the vulnerable plosive /d/ and fricatives /s,z,v/ would be recommended in many patients of the current study. However, a phonological linguistic approach is preferred in case of phonological disorders, including introduction of sets of sounds to improve the child's understanding and production of the rule based phonological sound system *(Bessell et al., 2013)*. For some patients of the current study, treatment of cluster simplification, deletion of final consonants and backing might be appropriate.

Some limitations of the present study deserve mention. This study included a limited number of patients with CLP. The authors are aware that there are a lot of intrinsic problems with matched group designs when sample sizes are small, as it is the case here. Variability in the CL/P population is large and the amount of possibly influencing factors is huge. Therefore, a substantial number of patients is needed to avoid variability. However, these numbers are not yet available due to the young age of the Ugandan patients with early synchronous lip and palatal repair visiting the Speech-Language Therapy department of CoRSU. Moreover, many patients with CLP operated at CoRSU before the age of 6 months did not return to visit the Speech-Language Therapy department for a speech assessment. Perhaps, these patients might not experience considerable speech problems or post-operative complications which are worth the traveling costs and distance. Further investigation with a larger sample will ascertain whether the current results can be generalized to the entire Ugandan cleft population. Furthermore, it should be noted that multiple variables, both known and unknown, can affect children's speech performance. Isolated cleft type, surgical technique and post-operative complications are known variables in this study. Variables such as hearing levels, history of otitis media, cognitive capacities, closure pattern of the velopharyngeal sphincter assessed by videofluoroscopy and/or nasoendoscopy, children's and parents' (if still alive) self-efficacy beliefs, children's motivation and other family, school and community factors are however unknown in this poor country. In addition, it is unclear whether the use of the non-native English language during speech assessments may account for some of the registered speech disorders. However, the authors tried to minimize this influence by excluding non-native consonants from the articulation analyses. Moreover, no influence is assumed on the comparison between the CLP and control group, as speech assessments in both groups were carried out in the children's second language (English).

Conclusion

In conclusion, speech after early synchronous CLP closure with the Sommerlad technique results at a mean age of 4;9 years in a limited number of resonance disorders. However, more nasal emission, smaller consonant inventories and more phonetic errors and phonological processes were observed compared to an age- and gender-matched control group. The articulation errors might have been caused by former palatal openings and require adjusted speech therapy. Future comparison with patients who underwent later palatal closure should reveal whether the current Ugandan patient group nevertheless benefited from palatal surgery prior to 6 months.

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CHAPTER 9

THE IMPACT OF PALATAL REPAIR BEFORE AND AFTER 6 MONTHS OF AGE ON SPEECH CHARACTERISTICS.

Based on:

Luyten, A., Bettens, K., D'haeseleer, E., De Ley, S., Hodges, A., Galiwango, G., Bonte, K., Vermeersch, H., & Van Lierde, K. (2014). The impact of palatal repair before and after 6 months of age on speech characteristics. *International Journal of Pediatric Otorhinolaryngology*, 78, 787-798.

Abstract

Aims: Optimal timing of palatal repair is still subject of discussion. Although literature provides some evidence that palatal closure prior to 6 months positively influence speech outcome in children with clefts, only few studies verified this hypothesis. The purpose of this study was to describe and compare articulation and resonance characteristics following early (≤ 6 months) and later (> 6 months) palatal repair, performed using the Sommerlad technique.

Methods: Comparison was made between 12 Ugandan children with isolated cleft (lip and) palate following early palatal repair (mean age 3.3m) and 12 Belgian patients with later palatal repair (mean age 11.1m), matched for cleft type, age and gender. A Ugandan and Belgian age- and gender-matched control group without clefts was included to control for language, culture and other environmental factors. Articulation assessments consisted of consonant inventories and phonetic and phonological analyses that were based on consensus transcriptions. In addition, resonance was evaluated by perceptual consensus ratings and objective mean nasalance values.

Results: The Belgian and Ugandan control groups were comparable for the majority of the variables. Comparison of cleft palate groups revealed no clinically relevant significant group differences for consonant inventory or phonological processes. Phonetic analysis showed significantly more distortions in the Belgian cleft palate group due to higher occurrence frequencies for (inter)dental productions of apico-alveolar consonants. Neither perceptual consensus ratings of hypernasality, hyponasality, cul-de-sac resonance and nasal emission/turbulence, nor objective mean nasalance values for oral speech samples revealed significant group differences ($p>0.05$).

Conclusion: Articulation and resonance characteristics of young children following palatal repair before and after 6 months of age seem to be at least comparable.

Key Words

Cleft • Speech • Articulation • Resonance • Timing of palatoplasty • Sommerlad technique

Introduction

Timing of cleft closure, and particularly timing of palatal repair, was subject of discussion during the previous decades. While some authors advocated an early closure of the cleft palate (CP) to improve speech outcome, others advised delayed palatal repair to optimize midfacial growth. Today, craniofacial teams in northern countries such as Europe and North-America still show a huge diversity in timing sequences for cleft closure. Results from the Eurocleft project illustrate that the majority (135/201, 67%) perform lip closure at the age of about 3 months *(Berkowitz, 2013)*, followed by a one- or two-stage soft and hard palatal closure at some later age in infancy or childhood *(Shaw et al., 2001)*. In resource-poor countries, this treatment protocol is often adapted. High default rates were noted for palatoplasty after successful lip repair *(Hodges, 2010; Onah et al., 2008)* because of financial considerations *(Adeyemo et al., 2009)*, huge travel distances *(Schwarz & Khadka, 2004)* and removal of the social stigma due to recovered aesthetics *(Bradbury & Habel, 2008)*. Consequently, synchronous closure of cleft lip and palate (CLP) and one-stage closure of cleft soft and hard palate are advocated *(Lehman, 2008)*. Considering the inability of patients with CP to adequately breastfeed and the consequential risk for malnutrition and death *(Cubitt et al., 2012)*, the craniofacial team of the Comprehensive Rehabilitation Services in Uganda (CoRSU) hospital recommends this surgical sequence as soon the patient reaches a target weight of 3kg, often resulting in cleft closure before 6 months of age *(Hodges, 2010)*.

Such early repair of the velopharyngeal mechanism might prevent that articulation errors get well-established in the children's neuromuscular speech patterns *(Kaplan, 1981)*. In babies with unrepaired CP, delays in onset of canonical babbling as well as differences in frequency and quality of productions have been noted *(Stout et al., 2011)*. The canonical babbling stage is however a crucial step toward verbal vocabulary. Consonants frequently used in canonical babbles will arise in the children's first words *(Peterson-Falzone et al., 2006a)*. Therefore, deviations in canonical babbling will most likely be reflected in later speech-language development *(Scherer et al., 2008)*.

Literature currently provides evidence that closure of the CP before the age of 6 months positively influences speech outcome in children with clefts. To our knowledge, 9 studies have described speech outcome following palatal closure before the age of 6 months *(Abdel-*

Aziz, 2013; Barimo et al., 1987; Copeland, 1990; De Mey et al., 2006; Desai, 1983; Kaplan et al., 1982; Kaplan et al., 1980; Luyten et al., 2014; Randall et al., 1983). Four other studies compared speech results of children with palatal closure before and after 6 months of age *(Doucet et al., 2013; Grobbelaar et al., 1995; Kaplan et al., 1974; Ysunza et al., 1998)*. An overview of these studies is provided in Table 9.1. Descriptive studies reported overall small deviations from normality for speech intelligibility, articulation and resonance, despite some exceptions. Moreover, three out of four comparative studies reported significantly improved speech in children with early palatal repair *(Grobbelaar et al., 1995; Kaplan et al., 1974; Ysunza et al., 1998)*. The overall better speech outcome after later compared to early hard palatal closure observed by *Doucet et al. (2013)* was attributed to differences in surgical technique, in particular to the success of intravelar veloplasty (Sommerlad technique) performed in the later CP closure group.

Drawing overall conclusions from these studies is however difficult due to several reasons. Patients with various cleft types were included and several palatal closure techniques were performed. Furthermore, the materials and method section in some studies offers very few information regarding age *(Desai, 1983; Kaplan et al., 1974; Randall et al., 1983)*, cleft type *(Randall et al., 1983)*, assessed speech samples *(Desai, 1983; Doucet et al., 2013; Grobbelaar et al., 1995; Kaplan et al., 1974; Randall et al., 1983; Ysunza et al., 1998)* and perceptual rating strategies *(Barimo et al., 1987; Desai, 1983; Kaplan et al., 1974; Randall et al., 1983; Ysunza et al., 1998)*. In addition, objective assessment techniques to evaluate speech characteristics were generally not performed. Regarding study design, only four studies compared the experimental group with children without clefts *(Barimo et al., 1987; Kaplan et al., 1974; Luyten et al., 2014; Ysunza et al., 1998)*. Moreover, in studies comparing patients with early and later palatal closure, information on whether the two experimental groups were matched for cleft type *(Kaplan et al., 1974)*, age *(Grobbelaar et al., 1995; Kaplan et al., 1974)* and/or gender *(Grobbelaar et al., 1995; Kaplan et al., 1974)* was often missing.

As shown by this literature review, more comparative studies on speech characteristics after early (≤ 6 months) and later palatal closure (> 6 months) are necessary. In CoRSU, palatal repair is generally performed before the age of 6 months. In contrast, the Belgian craniofacial team of the Ghent University Hospital performs a one-stage palatal repair at the age of 12 months on average. In both teams, palatal closure is carried out by an experienced surgeon using the Sommerlad technique (A.H., K.B.), what provides an

Table 9.1 Overview of the studies reporting speech results of patients with CL/P after closure of the soft and hard palate before the age of 6 months.

Author	n and cleft type	Palatal closure: age and surgical technique	Age at speech assessment	Control group	Speech assessment	Speech results
Kaplan et al. (1974) (Vietnam)	(a) 13 UCLP/ BCLP (b) 13 UCLP/ BCLP/ CP	*Age:* 3-4m *Age* : 10-12m *Surgical technique:* von Langenbeck	n.a. n.a.	yes, matched for age	n.a.	Comparisons between Group A (Ga), Group B (Gb) and control group: ***Babbling***: Ga normal vs. Gb delayed; ***First sounds (nasals, sibilants)***: Ga normal vs. Gb delayed; ***One-word response***: Ga normal vs. Gb delayed; ***Two- to three-word response***: Ga delayed, but earlier than in Gb; ***Initial nasal sounds***: Ga 40% adequate and 60% comparable with Gb.
Kaplan et al. (1980) and **Kaplan et al. (1982)** (Vietnam)	28 UCLP/ BCLP	*Age:* 3-8m (mean 4m) *Surgical technique:* von Langenbeck	1-7y	no	n.a.	***Preverbal vocalization***: 71% (5/7) normal, 29% (2/7) dull mumbling; ***Nasality, phonation, intelligibility and fluency of speech***: 52% (11/21) excellent, 43% (9/21) satisfactory, 5% (1/21) poor speech because of delay and nasality.
Desai (1983) (UK)	100 UC(L)P/ BC(L)P	*Age:* ≤ 16w *Surgical technique:* 95 Wardill-Kilner 5 von Langenbeck	n.a.	no	n.a.	***Hypernasality***: 0% (0/100); ***Pharyngoplasty***: 0% (0/100)
Randall et al. (1983) (USA)	17 (cleft type n.a.)	*Age:* 3-7m *Surgical technique:* n.a.	>2y	no	*Perceptual analysis by one speech therapist:* - Resonance - Articulation - Vocal quality	***Resonance***: 24% (4/17) hypernasality and 12% (2/17) hyponasality; ***Articulation***: 65% (11/17) normal; ***Vocal quality***: 65% (11/17) normal; ***Secondary pharyngoplasty***: required in 12% (2/17).

Barimo et al. (1987) (USA)	22 BCLP/ UCLP/ CP	*Age:* 3-8m (mean 6m) *Surgical technique:* Complete reconstruction of the velopharyngeal sling with interdigitation of the muscles and minimal dissection of the mucoperiosteum	1;10-8;11y	yes, matched for age	*Speech samples:* 1;10-2;6y: Phonological data collected from spontaneous utterances 3;1-8;11y: Templin-Darley Screening Test of Articulation 3;1-5;11y: Bzoch Error Pattern Screening Articulation Test	*Articulation*: 1;10-2;6y (n=5): Absence of nasal emission, glottal stops and pharyngeal fricatives. Age appropriate phoneme type and frequency. 3;1-8;11y (n=17): 0% failed the screening articulation test. 3;1-5;11y (n=8): 89% of consonants correct articulation, 8% substitution errors, 0% distortion errors, 2% omission errors. Absence of glottal stops and pharyngeal fricatives.
Copeland (1990) (UK)	53 UCLP 17 BCLP 14 CP 16 soft CP	*Age:* 9-25w (mean 16.4w) *Surgical technique:* 92 Wardill-Kilner 8 von Langenbeck	3.8-6.3y (mean 5.5y)	no	*Speech samples:* Edinburgh Articulation Test, conversational sample based on picture description, counting from 1 to 10 *Perceptual rating by one speech therapist:* - Intelligibility - Nasal resonance - Articulation	***Intelligibility:*** 87% (87/100) acceptable speech, 13% (13/100) unacceptable speech; ***Nasal resonance and nasal emission***: 81% (81/100) normal resonance and no nasal emission or minimal degrees of one of these, 93% (93/100) normal/minimal abnormal resonance, 7% (7/100) moderate/severe abnormal resonance, 91% (91/100) normal/minimal nasal emission, 9% (9/100) moderate/severe nasal emission; ***Articulation***: 57% (57/100) normal or some immature features, 6% (6/100) very immature phonology, 36% (36/100) atypical errors.

Grobbelaar et al. (1995) (South-Africa)	184 soft CP	*Age:* 2w – 18m (median 6.2m) *Surgical technique:* 25 Dorrance 41 Wardill 19 Perko 20 Furlow Z-plasty 79 von Langenbeck	4-24y (mean 9.6y)	no	*Perceptual consensus rating:* - Intelligibility - Resonance - Articulation Perceptual evaluation of velopharyngeal competence confirmed by consensus evaluation of videofluoroscopy and/or nasoendoscopy	***Speech*:** Palatal repair ≤ 6 months: 95% (73/77) normal* ↔ palatal repair > 6 months: 92% (98/107) normal* (*normal resonance, articulation and intelligibility); ***Velopharyngeal inadequacy (VPI)*:** Palatal repair ≤ 6 months: 5% (4/77) VPI ↔ Palatal repair > 6 months: 8% (9/107) VPI; Significant differences in speech and less velopharyngeal incompetence after palatal repair before vs. after 6 months of age (except for the Dorrance repair).
Ysunza et al. (1998) (Mexico)	(a) 41 UCLP (b) 35 UCLP	*Age:* 12m *Age:* 6m *Surgical technique:* Minimal incision palatopharyngo-plasty	4y 4y	yes, no info about matching available	*Clinical assessment by two speech therapists:* - Nasal resonance - Spanish articulation test Consensus diagnose based on video-nasopharyngoscopy and multiview videofluoroscopy	***Articulation scale***: Below normal limits for the patient's age in all cases. Significantly better in Group B vs. Group A; ***Velopharyngeal insufficiency*** *(VPI, p>0.05)*: Group A: 20% (8/41) VPI ↔ Group B: 17% (6/35) VPI; ***Compensatory articulation errors (CAE) in patients with VPI*** *(p<0.05)*: Group A: 62% (5/8) CAE ↔ Group B: 0% (0/6) CAE.

De Mey et al. (2006) (Belgium)	(a) 26 UCLP (b) 18 UCLP	*Age:* Soft palate: 2.8-3.2m (mean 3m) + hard palate: 5.5-6.8m (mean 6.2m) *Age:* Hard & soft palate: 2.8-3.1m (mean 3m) *Surgical technique:* Malek protocol (soft palate) and vomer flap (hard palate)	3y and 6y 3y and 6y	no	*Speech samples:* Spontaneous speech, standardized speech assessment in French or Flemish *Perceptual rating by one senior speech therapist:* - Intelligibility - Nasality - Articulation	***Intelligibility*****:** *At 3 years*: significant group differences (*p*=0.05). Group A: 52% good, 32% mild deficiency, 18% major problem ↔ Group B: 63% good, 37% mild deficiency, 0% major problem. *At 6 years*: no significant differences between groups. 80% normal intelligibility; ***Nasality*****:** No significant group differences. *At 3 years*: 30% no nasality problems, 50% inconstant nasality, 20% constant nasality. *At 6 years*: 40% no nasality problems, < 10% constant nasality; ***Articulation*****:** No significant differences between groups. *At 3 years*: 20% important disorders; *at 6 years*: 40% normal articulation; ***Velopharyngoplasty required:*** Group A: 12% (3/26) at mean age of 5 years ↔ Group B: 6% (1/18) at 5.5 years. Speech satisfactory in both groups in the majority of children. However, earlier intelligibility of speech in group B.
Abdel-Aziz (2013) (Egypt)	21 soft CP	*Age:* 3-6m (mean 4m3w) *Surgical technique:* Furlow Z-plasty	4y	no	*Speech samples:* Counting from 1 to 20, phonetically selected speech sample *Perceptual rating:* - Hypernasality - Nasal emission Assessment of velopharyngeal closure by nasopharyngoscopy	***Hypernasality**:* 86% (18/21) absent, 14% (3/21) mild hypernasality; **Nasal emission**: 100% (21/21) absent; ***Velopharyngeal closure*** *(VPC):* 86% (18/21) competent VPC, 5% (1/21) borderline competent VPC, 10% (2/21) borderline incompetent VPC; ***Secondary pharyngoplasty***: none required

Doucet et al. (2013) (France)	(a) 20 UCLP (b) 20 UCLP	*Age:* Soft palate: 2.8-3.4m (mean 3.1m) + hard palate: 5.6-6.6m (mean 6.1m) *Surgical technique:* Sutured in midline without intravelar veloplasty (soft palate) + vomer flap (hard palate) *Age:* Soft palate: 5.7-6.3m (mean 6.0m) + hard palate: 16.9-18.7m (mean 17.8m) *Surgical technique:* Sommerlad (soft palate) + 2-layer hard palatal repair	3.0-3.6y (mean 3.3y) 2.9-3.7y (mean 3.3y)	no	*Speech samples:* n.a. *Perceptual rating by one senior speech therapist:* - Speech intelligibility - Hypernasality - Nasal emission - Nasal turbulence - Articulation Perceptual evaluation of velopharyngeal insufficiency confirmed by aerophonoscopy	***Speech intelligibility*****:** Significant group difference (*p*<0.05). Group A: 30% (6/20) adequate ↔ Group B: 75% (15/20) adequate; ***Hypernasality*****:** No significant group difference (*p*>0.05). Group A: 20% (4/20) absent, 35% (7/20) mild, 25% (5/20) moderate, 20% (4/20) severe; ***Nasal emission*****:** No significant group difference (*p*>0.05). Group A: 25% (5/20) absent, 35% (7/20) mild, 35% (7/20) moderate, 5% (1/20) severe; ***Nasal turbulence*****:** No significant group difference (*p*>0.05). Group A: 60% (12/20) absent, 40% (8/20) mild; ***Articulation*****:** Significantly more delays of ≥ 1y in group A (*p*<0.05). No significant group differences regarding consonant inventory, presence of bilabial consonants, velar substitution, compensatory articulations (i.e. glottal stops, pharyngeal fricatives, posterior nasal fricatives) (*p*>0.05); ***Velopharyngeal insufficiency*****:** Significant group difference (*p*<0.05). Group A: 55% (11/20) present ↔ group B: 15% (3/20) present.

Luyten et al. (2014)	10 UCLP 1 BCLP	*Age:* 2-6m (mean 3.4m) *Surgical technique:* Sommerlad	3;2-7;2y (mean 4;9y)	yes, matched for age and gender	*Speech samples:* 30 words (perceptual rating); vowels, oral syllables and oral sentences (Nasometry); Photo Articulation Test – 3rd edition (articulation analysis) *Perceptual consensus rating by 2 speech therapists:* - Hypernasality - Hyponasality - Cul-de-sac resonance - Nasal emission/turbulence Objective *assessment* of hypernasality by Nasometer and detailed articulation analysis	***Hypernasality***: 82% (9/11) absent, 9% (1/11) minimal, 9% (1/11) moderate; ***Hyponasality***: 100% (11/11) absent; ***Cul-de-sac resonance***: 100% (11/11) absent; ***Nasal emission/turbulence***: 73% (8/11) absent, 18% (2/11) occasional, 9% (1/11) frequent; ***Nasometry***: No significant differences in mean nasalance values compared to control group; ***Articulation***: 100% (11/11) showed at least one articulation error. Significantly smaller consonant inventories ($p<0.05$). Significantly higher frequencies of occurrence of phonetic errors such as compensatory articulation of fricatives /s,z,f,v/ and plosives /t,d/ and bilabial production of /f/ and /v/ ($p<0.05$). Significantly higher frequencies of occurrence for phonological processes (cluster simplification, deletion of final consonants, backing).

UCLP: unilateral cleft lip and palate, BCLP: bilateral cleft lip and palate, C(L)P: cleft palate with or without cleft lip, CP: cleft of hard and soft palate, soft CP: cleft of soft palate only, n.a.: not available

excellent opportunity for assessing speech characteristics after early vs. later palatal closure. However, comparison of both patient groups is complicated due to differences in language backgrounds. According to *Hutters & Henningsson (2004)* the level of difficulty to cope with a language for patients with CP is determined by phonetic characteristics of the sounds included in the language's consonant inventory. Considering that pressure consonants (i.e. plosives, fricatives and affricatives) are the most vulnerable sounds for patients with CP, the amount of these consonants in speech samples should be weighted in order to eliminate the variable 'language background' and increase the validity of comparisons *(Henningsson et al., 2008)*.

The aim of the current study is to describe and compare articulation and resonance characteristics of Ugandan patients with isolated cleft (lip and) palate (C(L)P) following early palatal repair (≤ 6 months) with Belgian patients having isolated clefts repaired after the age of 6 months. An identical surgical technique (Sommerlad) was used in both groups. The CP groups are matched for cleft type, age and gender. A well-defined speech assessment approach, including objective assessment techniques and perceptual consensus evaluations, is used *(Baudonck et al., 2010; Luyten et al., 2014; Van Lierde et al., 2013)*. In order to control for differences in language, culture and other environmental factors, two control groups are included. Each control group (Ugandan/Belgian) is matched with one of the CP groups (Ugandan/Belgian) for country, age and gender. In accordance with previous comparative literature about early vs. later palatal closure *(Grobbelaar et al., 1995; Kaplan et al., 1974; Ysunza et al., 1998)*, fewer articulation and resonance disorders are hypothesized in patients with early compared to later palatal closure.

Materials and methods

The research protocol was approved by the Ethical Committee of the Ghent University Hospital, Belgium (EC2011/269). All subjects gave informed consent to the work.

Participants

The current study included a Ugandan and Belgian CP group as well as a Ugandan and Belgian control group. The Ugandan CP group was recruited at the Speech Language Therapy department of CoRSU during four 10-day-long missions between January 2012 and January 2013 and included all Ugandan patients with isolated clefts of the soft and hard palate, who underwent one-stage palatal repair at the age of 6 months or earlier and who adequately performed English articulation and nasometric assessments. From the 84 patients with an early one-stage CP closure (≤ 6 months), 72 children were excluded due to incomplete articulation assessments (n=4), presence of associated anomalies (n=6), a young age (i.e. < 2;6 years) (n=56) or lack of cooperation (n=6). Twelve Ugandan patients were selected for further assessment. They all were able to speak and understand English, since this is their second language. English is one of the official languages of Uganda *(Uganda Law Reform Commission, 2006)* and is deeply rooted in media, administration and education *(Mpuga, 2003)*. The Ugandan CP group included 4 males and 8 females with a mean age of 4;9 years (range 3;2-7;2 years), presenting with CP (n=1), unilateral CLP (n=10, UCLP) or bilateral CLP (n=1, BCLP). UCLP and BCLP were closed during a synchronous lip and palatal repair, while CP was repaired by a one-stage soft and hard palatal closure. All surgical cleft closures were carried out by one experienced surgeon (A.H.) at 3.3 months on average (range 2-6 months). A modified Millard repair (UCLP) and a modified Mulliken technique (BCLP) were applied for lip closure. CP was repaired using the Sommerlad technique. Secondary surgery for minor postoperative complications was performed in 7 patients (7/12, 58%), including 1 closure of palate dehiscence and 6 fistula repairs. None of the patients received speech therapy or hearing treatment (e.g. ventilation tubes) prior to speech assessment.

For each participant of the Ugandan CP group, a Belgian Dutch-speaking patient with surgically closed isolated cleft matched for cleft type and age, was recruited from the craniofacial team of the Ghent University Hospital. Matching for gender was possible for 11 children (11/12, 92%). The Belgian CP group consisted of 5 males and 7 females with CP (n=1), UCLP (n=10) or BCLP (n=1). Mean age was 4;7 years (range 2;6-6;11 years). Dutch was the mother language of all patients. At 3.3 months on average (range 2-5 months), lip repair was performed using a modified Mulliken (n=1), modified Millard (n=7), Manchester (n=2) or Fisher (n=1) technique. All palatal closures were performed during one surgery by an experienced surgeon (K.B.) at a mean age of 11.1 months (range 9-15 months). A Sommerlad technique was applied in all but one patient. Untreated minor postoperative complications such as palatal fistulae (n=5) and palate dehiscence (n=1) were observed in 50% of the Belgian CP group (6/12). Ventilation tubes were inserted at least once in 9 patients (9/12,

75%) for treatment of recurrent otitis media. Six children with UCLP or BCLP (6/12, 50%) received speech therapy prior to speech assessment. The duration varied from 3.5 months to 3;6 years with a frequency between 30 min and 1h30 per week.

For each of both CP groups an age- and gender-matched control group (n=12) was included. None of these children showed craniofacial anomalies or pathologies of the velopharyngeal, oral or nasal structures and children with a temporary nasal obstruction were excluded. Participants of the Ugandan control group were recruited during 2 missions in September 2010 and January 2013. They had a mean age of 4;10 years (range 3;0-7;1 years) and spoke English as their second language. Dutch-speaking children with a mean age of 4;6 years (range 2;9-6;10 years), recruited between April 2012 and November 2012, were included in the Belgian control group. A one-way ANOVA could not reveal significant differences in chronological age among 4 groups (p=0.952).

Speech assessment

Objective and subjective assessment techniques were applied for comparison of articulation and resonance characteristics after early and later palatal closure *(Luyten et al., 2014)*. Considering that 7 different native languages were noted for the Ugandan participants, it was impossible to perform speech evaluation in these participants' mother language. Speech evaluation of Ugandan participants was carried out in English as they were daily exposed to this language and mastered it sufficiently to complete the assessment. In Belgian children, speech evaluation was performed in Dutch by the same speech-language therapists (SLTs). These SLTs were native speakers of Dutch and fluently spoke and comprehended the English language.

Articulation

Assessment of articulation characteristics in Ugandan participants was performed using the Photo Articulation Test – third edition (PAT-3) *(Lippke et al., 1997)*, a standardized picture-naming test. The participants were asked to repeat 72 high frequency English words containing all English consonants occurring isolated in all permissible syllable positions as well as in common consonant clusters (see Appendix 9.1). A similar picture-naming test consisting of 135 high frequency Dutch words was performed in Belgian participants.

The target words contained all Dutch singleton consonants and most clusters in all their permissible syllable positions (see Appendix 9.2). Digitally video-recorded (Sony Handycam HDR-UX1 with a high quality built-in microphone) speech samples were transcribed by an SLT (A.L.) experienced with cleft-related speech disorders using symbols of the International Phonetic Alphabet (IPA), IPA extensions *(International Phonetic Association, 1999)* and additional symbols for specific cleft-related errors *(Trost, 1981)*. A second experienced SLT (K.B.) transcribed independently 50% (24/48) of the English and Dutch articulation assessments. Mean consonant-for-consonant interjudge reliability was 79% (SD: 9.3%) for the English and 81% (SD: 8.4%) for the Dutch speech samples. In Ugandan participants, the 18 English consonants occurring in Luganda (i.e. the most common native language of all Ugandan participants: 18/24, 75%) were used for further analyses (i.e. /p/, /b/, /t/, /d/, /k/, /g/, /s/, /z/, /f/, /v/, /h/, /w/, /j/, /l/, /r/, /n/, /m/ and /ŋ/). Experience shows that even Ugandan adults without clefts often show difficulties to correctly produce the English fricatives /ʃ,ɵ,ð/ and affricatives /tʃ,dʒ/. In Belgian participants, all 21 consonants of the Dutch language were considered (i.e. /p/, /b/, /t/, /d/, /k/, /s/, /z/, /ʃ/, /ʒ/, /f/, /v/, /x/, /ɣ/, /h/, /w/, /j/, /l/, /r/, /n/, /m/ and /ŋ/).

Articulation analyses included a consonant inventory as well as phonetic and phonological analyses. The consonant inventory consisted of all native consonants that were correctly produced twice without making reference to the intended target sound. In contrast, comparison with the target sound was made during the phonetic and phonological analysis. In the phonetic analysis, consonant productions were analyzed for error types at phoneme level (omissions, additions, substitutions and distortions). Phonological analysis included analysis of consonant productions at word level according to Ingram's classification *(Ingram, 1982)* (syllable structure processes, substitution processes and assimilation processes). Occurrence frequencies (%) were calculated for each phonetic error and phonological process by the ratio of the number of occurrences to the number of potential occurrences. When an error type could occur at least 4 times and presented in at least 20%, the error type was considered productive *(McReynolds & Elbert, 1981)*.

Resonance

Assessment of resonance included perceptual consensus evaluation and measurement of objective nasalance values.

PERCEPTUAL CONSENSUS EVALUATION

Digital audio-recordings of the first 30 words of the English (Ugandan participants) and Dutch (Belgian participants) picture-naming tests were perceptually evaluated by two experienced SLTs (A.L., K.B.) who were blind for the participants condition (cleft/non-cleft). In both languages, the words contained a similar number of pressure consonants and nasal phonemes *(Hutters & Henningsson, 2004)*. Presence of hypernasality (absent, minimal, mild, moderate, severe), hyponasality (absent, mild, marked), cul-de-sac resonance (absent, mild, marked) and nasal emission/turbulence (absent, occasional, frequent) was determined in accordance with *John et al. (2006)*. Considering that nasal turbulence results from air flowing through an increased resistance in the nasal airway *(Peterson-Falzone et al., 2006b)*, nasal emission and nasal turbulence were judged as one *(Henningsson et al., 2008)*. Speech samples were first judged simultaneously, but independently. In case of disagreement, the samples were replayed until a consensus was reached. Interjudge reliability, calculated by the ratio of identical to total judgments for all resonance disorders, was 95% (91/96) for the Ugandan and 90% (86/96) for the Belgian participants.

NASALANCE VALUES

Objective evaluation of nasalance was performed using the Nasometer (model II 6450). The Nasometer is a microcomputer-based instrument, developed by *Fletcher & Bishop (1973)* and manufactured by *Kay Elemetrics Corporation (2010)*. This instrument contains a horizontal sound separator plate that rests on the upper lip, with a microphone on either side. The signal from each microphone is individually filtered and digitized. Subsequently, the data are processed by a computer. The resultant signal is the ratio of nasal to nasal-plus-oral acoustic energy, multiplied by 100 and is expressed as a nasalance score. Prior to data collection, the Nasometer was calibrated following the procedures outlined in the manual. Speech samples described in the MacKay-Kummer Simplified Nasometric Assessment Procedures (SNAP) test were used *(Kummer, 2005; MacKay & Kummer, 1994)* (see Appendix 9.3). Each participant was asked to sustain the oral vowels /a/, /i/ and /u/, and to produce twice

8 CV-syllable sequences and 12 simple oral English sentences (majorly containing bilabials, alveolars, velars or sibilants). For Belgian participants, a Dutch variant of the sentences was applied *(Van Lierde et al., 2002)* (see Appendix 9.3). Nasalance values of syllables and sentences were not obtained in Ugandan (n=3) and Belgian (n=4) patients with nasal emission/turbulence and their control-counterparts, since nasal emission/turbulence might increase nasalance values *(Dalston et al., 1991)*.

Statistical analyses

Statistical analyses were performed using SPSS software (version 20.0) with significance levels set at α=0.05. To exclude influence of gender, culture, language and other environmental factors, the age- and gender-matched Ugandan and Belgian control groups were statistically compared. When no significant differences could be established between both control groups, comparison of the Ugandan and Belgian CP groups was appropriate.

Comparison of categorical data was performed using chi-square tests or Fisher's exact tests, depending on the expected cell count. For all continuous variables, ANOVA was applied when the assumptions were fulfilled (homogeneity of variances, normality). A transformation (square root, logarithmic or reciprocal) was used in case of inequality of variances. When a transformation appeared insufficient, Mann-Whitney U tests were carried out as all such variables deviated from a normal distribution. In case of equality of variance, a non-parametric Kruskal-Wallis rank sum test was performed for variables that were not normally distributed. Post hoc testing consisted of LSD-tests (parametric) or Dunn's tests (non-parametric).

Results

Comparison of age- and gender-matched control groups

The Ugandan and Belgian control groups were overall comparable regarding percentage of acquired consonants, occurrence frequencies of specific phonetic errors and phonological processes, amount of participants with phonetic or phonological articulation disorders, perceptual consensus evaluations and mean nasalance values for all oral speech samples (p>0.05). However, significant differences were observed for the phonetic error 'additions' (p=0.038) and the phonological process 'devoicing' (p=0.011). Consequently, comparison of the Ugandan and Belgian CP groups was appropriate, except for additions and devoicing.

Articulation

Consonant inventory

For the Ugandan CP group, a median of 89% (16/18) acquired consonants was obtained (range 8-18/18, 44-100%) compared to a median of 81% (17/21) for the Belgian CP group (range 6-20/21, 29-95%). This group difference was not significant (p=0.271). In both CP groups, the consonant /r/ as well as sibilants and labiodental fricatives were the most frequently missing.

Phonetic analysis

Figure 9.1 displays the results regarding the phonetic analysis for all groups. Distortions were the most frequently occurring error types in both CP groups. Although no significant differences were observed between the Ugandan and Belgian CP group for substitutions (p=0.804) and omissions (p=0.375), significantly more distortions were found in the Belgian CP group (p<0.001).

An overview of the occurrence frequencies for various distortion types is provided in Table 9.2. Comparison of the CP groups revealed that (inter)dental production of apico-alveolar consonants occurred significantly more often in Belgian compared to Ugandan patients (p=0.006). No significant differences were obtained for bilabial articulation of /f,v/ (p=0.585), lateral sigmatism (p=0.616), labiolingual articulation of /p,b,m,w/ (p=0.317), compensatory articulation of plosives (p=0.354) or fricatives (p=0.347) and production of weak plosives (p=0.912) or fricatives (p=0.080).

According to the 20% criterion, all Belgian patients (12/12, 100%) showed at least one phonetic error compared to 92% (11/12) of the Ugandan CP group. This group difference was not significant (p=1.000).

Phonological analysis

Results regarding the phonological analysis for all groups are presented in Table 9.3. In both CP groups, the most frequently occurring processes were cluster simplification and liquid gliding, followed by backing, deletion of final consonants and fronting. In addition, devoicing was the third most frequent phonological process in the Belgian CP group. Comparison between both CP groups revealed no significant differences, except for coalescence of sounds (*p*=0.015) and reduplication (*p*=0.006). Both phonological processes occurred significantly more often in the Belgian CP group, despite small group differences.

In correspondence with the 20% criterion of McReynolds & Elbert (1981), at least one phonological process occurred in 100% (12/12) of the Ugandan and 75% (9/12) of the Belgian patients. This group difference was not significant (*p*=0.217).

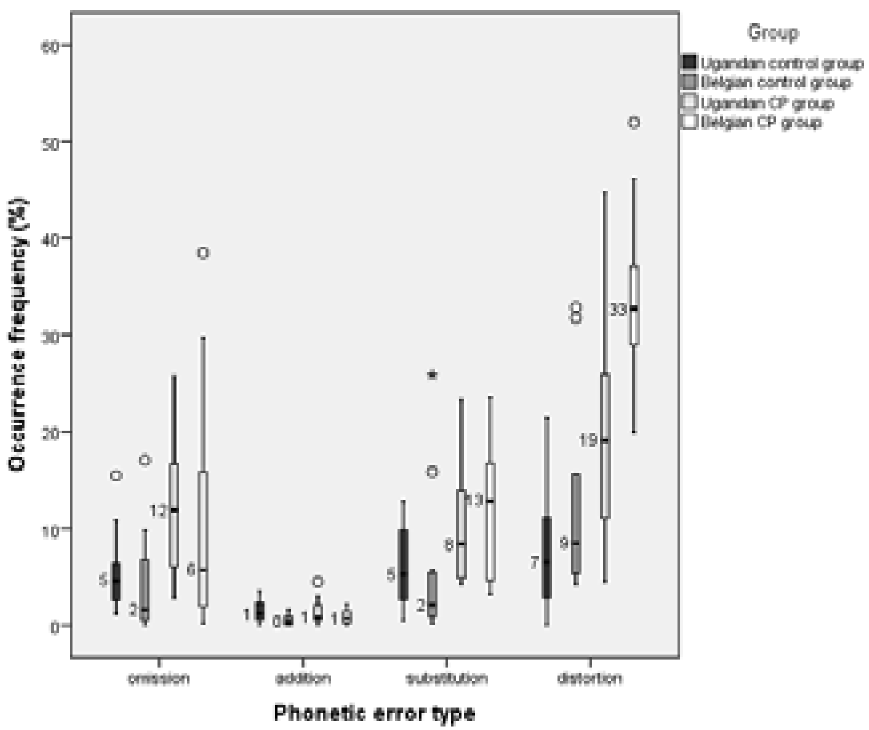

Figure 9.1 Occurrence frequency by overall phonetic error types for the Ugandan and Belgian control and CP groups. The median occurrence frequencies are indicated next to the box plot.

Table 9.2 Occurrence frequencies (%) of various distortion types are provided for the Ugandan and Belgian control and CP groups. Furthermore, the *p*-values resulting from the Kruskal-Wallis test ('Comparison 4 groups') as well as from the Dunn's test or Mann-Whitney U tests ('Post hoc tests') are indicated.

Distortion type	Ugandan control group				Belgian control group				Ugandan CP group				Belgian CP group				Comparison 4 groups	Post hoc test	
	mean	SD	median	range	mean	SD	median	range	mean	SD	median	range	mean	SD	median	range		Comparison control groups	Comparison CP groups
(Inter)dental apico-alveolar consonants	11	14	4	0-50	17	20	7	1-65	12	14	8	0-41	32	23	24	6-75	*0.010*	0.300	*0.006*
Bilabial articulation /f,v/	0	0	0	0-0	2	3	0	0-10	23	29	14	0-86	31	38	14	0-92	-	0.071	0.585
Lateral sigmatism	0	0	0	0-0	0	0	0	0-0	7	25	0	0-87	8	25	0	0-87	-	identical	0.616
Derhotacized /r/	n.a.	n.a.	n.a.	n.a.	38	28	28	4-83	n.a.	n.a.	n.a.	n.a.	64	28	67	19-100	n.a.	n.a.	n.a.
Labiolingual articulation of /p,b,m,w/	0	0	0	0-0	0	0	0	0-0	0	0	0	0-0	3	10	0	0-35	-	identical	0.317
Compensatory articulation of plosives	0	1	0	0-1	0	0	0	0-1	11	19	3	0-66	19	25	5	0-70	-	0.482	0.354
- /p,b/	0	0	0	0-0	0	0	0	0-0	4	11	0	0-40	12	17	1	0-50	0.402	-	-
- /t,d/	0	1	0	0-5	0	0	0	0-1	25	31	13	0-90	26	32	8	0-82	-	0.952	0.728
- /k,g/	0	1	0	0-4	0	0	0	0-0	7	22	0	0-77	12	28	0	0-76	-	0.317	0.684
Compensatory articulation of fricatives	2	3	0	0-11	2	5	1	0-18	32	25	29	0-83	25	28	18	0-91	*<0.001*	0.870	0.347
- /s,z/	3	5	0	0-17	4	8	0	0-30	41	32	43	0-94	43	43	33	0-100	*<0.001*	0.982	0.669
- /ʃ,ʒ/	n.a.	n.a.	n.a.	n.a.	0	0	0	0-0	n.a.	n.a.	n.a.	n.a.	29	44	0	0-100	n.a.	n.a.	n.a.
- /f,v/	0	0	0	0-0	0	0	0	0-0	11	19	0	0-57	9	29	0	0-100	-	identical	0.362
- /x,ɣ/	n.a.	n.a.	n.a.	n.a.	2	3	0	0-9	n.a.	n.a.	n.a.	n.a.	9	21	0	0-56	n.a.	n.a.	n.a.
Weak plosives	3	3	3	0-10	1	1	1	0-3	7	7	6	0-23	14	26	4	0-93	*0.031*	0.140	0.912
Weak fricatives	9	12	4	0-40	3	4	1	0-14	10	8	7	0-23	15	14	13	0-42	0.080	-	-

Table 9.3 Mean and median occurrence frequencies for various phonological processes as well as the range (%) are provided for the Ugandan and Belgian control and CP groups. The *p*-values resulting from the ANOVA or Kruskal-Wallis test ('Comparison 4 groups') as well as from the LSD test, the Dunn's test or the Mann-Whitney U test ('Post hoc test') are indicated.

Phonological process	**Ugandan control group**				**Belgian control group**				**Ugandan CP group**				**Belgian CP group**				**Comparison 4 groups**	**Post hoc test**	
	mean	**SD**	**median**	**range**	**mean**	**SD**	**median**	**range**	**mean**	**SD**	**median**	**range**	**mean**	**SD**	**median**	**range**		**Comparison control groups**	**Comparison CP groups**
SYLLABLE STRUCTURE PROCESSES																			
Deletion final consonants	4	4	2	0-15	5	6	3	0-17	13	11	11	2-34	10	16	5	0-53	0.107	-	-
Deletion initial consonants	2	3	1	0-8	0	0	0	0-1	2	4	0	0-10	3	5	1	0-17	0.094	-	-
Deletion unstressed syllable	2	2	0	0-6	0	1	0	0-2	1	2	0	0-3	3	7	0	0-22	0.674	-	-
Cluster simplification:	16	14	15	3-48	17	16	13	1-58	40	19	43	9-64	39	26	33	3-73	*0.002*	0.910	0.870
- Cluster reduction	13	12	12	3-42	13	17	6	0-56	37	19	41	6-62	31	28	19	0-73	*0.009*	0.599	0.273
- Epenthesis	4	5	2	0-15	5	3	6	1-11	10	8	6	0-29	12	10	11	0-30	*0.037*	0.478	0.434
Coalescence syllables	1	2	0	0-7	0	1	0	0-3	1	1	0	0-3	1	1	0	0-4	0.875	-	-
Coalescence sounds	0	1	0	0-2	0	1	0	0-2	0	0	0	0-0	0	0	0	0-1	-	0.448	*0.015*
SUBSTITUTION PROCESSES																			
Fronting	6	6	4	0-18	4	5	3	0-19	8	8	5	0-26	8	16	3	0-59	0.376	-	-
Backing	1	1	1	0-3	1	2	0	0-6	16	17	11	1-61	19	21	9	0-61	*<0.001*	0.781	0.717
Stopping	1	3	0	0-10	0	0	0	0-1	3	5	1	0-16	6	9	2	0-28	-	0.464	0.313
Liquid gliding	19	14	21	0-43	8	18	1	0-58	34	24	25	8-86	20	21	15	0-60	*0.011*	0.051	0.160
Denasalization	0	0	0	0-0	0	0	0	0-0	0	0	0	0-0	0	1	0	0-2	-	identical	0.149
Devoicing	1	2	0	0-4	14	18	6	0-57	1	3	0	0-9	11	11	9	0-42	*<0.001*	*0.002*	n.a.
ASSIMILATION PROCESSES																			
Assimilation	4	3	2	0-12	2	3	1	0-8	4	4	2	0-12	3	4	2	0-12	0.713	-	-
Reduplication	0	2	0	0-0	1	1	0	0-3	0	0	0	0-0	2	2	1	0-6	-	0.733	*0.006*
OTHER PROCESSES																			
Metathesis	1	2	0	0-3	1	1	0	0-4	1	1	0	0-3	1	2	0	0-8	0.362	-	-
Substitution /n/ → /m/	11	5	3	0-77	0	1	0	0-3	11	22	3	0-77	9	14	3	0-44	0.058	-	-

Resonance

Perceptual consensus evaluation

Table 9.4 presents the results of the perceptual consensus evaluation for the Ugandan and Belgian CP group. In the Ugandan CP group, none of the patients showed hyponasality or cul-de-sac resonance (0/12, 0%), while 75% (9/12) had no hypernasality and 9 patients (9/12, 75%) had no nasal emission/turbulence. In the Belgian CP group, cul-de-sac resonance was absent in all patients (0/12, 0%). Ninety-two percent (11/12) showed no hyponasality. Moreover, absence of hypernasality and nasal emission/turbulence were respectively observed in 50% (6/12) and 67% (8/12). For none of these features, significant differences were found between CP groups (p>0.05).

Table 9.4 Perceptual consensus evaluation of hypernasality, hyponasality, cul-de-sac resonance and nasal emission/turbulence for the Ugandan and Belgian CP group. The p-values resulting from the Fisher's exact tests are indicated.

	Ugandan CP group (n=12)		Belgian CP group (n=12)		*p*
	n	**%**	**n**	**%**	
Hypernasality					
- Absent	9	75	6	50	0.400
- Minimal	2	17	1	8	
- Slight	0	0	3	25	
- Moderate	1	8	2	17	
- Severe	0	0	0	0	
Hyponasality					
- Absent	12	100	11	92	1.000
- Mild	0	0	1	8	
- Marked	0	0	0	0	
Cul-de-sac resonance					
- Absent	12	100	12	100	n.a.
- Mild	0	0	0	0	
- Marked	0	0	0	0	
Nasal emission/turbulence					
- Absent	9	75	8	67	1.000
- Occasional	2	17	3	25	
- Frequent	1	8	1	8	

Nasalance values

Mean nasalance values and standard deviations for the Ugandan and Belgian CP and control groups are provided in Table 9.5. No significant differences were obtained between both CP groups (p>0.05).

Discussion

The purpose of this study was to describe and compare articulation and resonance characteristics of 12 Ugandan patients with C(L)P after early palatal closure (≤ 6 months) and 12 Belgian patients (matched for cleft type, age and gender) with palatal repair performed after the age of 6 months by means of an identical surgical technique. A Ugandan and Belgian age- and gender-matched control group was included to control for language, culture and other environmental factors. Speech characteristics at the mean age of 4;8 years suggests that early one-stage closure of soft and hard CP (≤ 6 months) results in at least similar articulation and resonance characteristics compared with delayed palatal repair. The Ugandan and Belgian CP groups were comparable regarding consonant inventory, presence of phonetic disorders and phonological processes, perceptual consensus evaluations of resonance disorders and mean nasalance values for oral speech samples. However, the Belgian CP group showed significantly more distortions due to higher relative amounts (%) of (inter)dental productions of apico-alveolar consonants. Significant higher percentages for coalescence of sounds and reduplication in the Belgian CP group were not considered clinically relevant due to small occurrence frequencies in both CP groups (≤ 6% for all participants).

Comparing results of this study with literature, similarities and differences are found. Similar to the current study, *Doucet et al. (2013)* observed no significant differences at a mean age of 3.3 years between the early and late hard palatal closure group for hypernasality, nasal emission, nasal turbulence, consonant inventory, velar substitution or compensatory glottal stops and pharyngeal fricatives. However, *Grobbelaar et al. (1995)* reported significantly more normal articulation, resonance and intelligibility in patients with clefts of the soft palate (aged 4 to 24 years) following palatal closure before 6 months of age, despite small group differences. The power of statistical comparison in that study was higher compared

Table 9.5 Mean nasalance values (%) and standard deviations (SD) are provided for the Ugandan and Belgian control and CP groups. Only patients speaking without nasal emission/turbulence and their control-counterparts are included for syllable sequences and sentences. In each CP group, mean nasalance values of all sentences were absent for two patients. The *p*-values resulting from ANOVA or the Kruskal-Wallis test ('Comparison 4 groups') as well as from the Mann-Whitney U, LSD or Dunn's test ('Post hoc tests') are indicated.

	Ugandan control group			**Belgian control group**			**Ugandan CP group**			**Belgian CP group**			**Comparison 4 groups**	**Post hoc tests**	
	n	**Nasalance**	**SD**	**n**	**Nasalance**	**SD**	**n**	**Nasalance**	**SD**	**n**	**Nasalance**	**SD**		**Comparison control groups**	**Comparison CP groups**
/a/	12	13.5	6.63	12	10.3	6.24	12	19.6	10.26	12	19.2	9.24	*0.021*	0.341	0.902
/i/	12	28.3	19.67	12	24.6	9.17	12	37.1	25.10	12	43.1	18.72	0.116	-	-
/u/	12	21.8	15.85	12	14.8	7.66	12	29.8	19.64	12	32.3	22.12	0.070	-	-
/papapa/	9	12.2	9.64	8	9.4	2.13	9	11.4	4.36	8	17.3	10.58	0.305	-	-
/pipipi/	9	20.3	19.54	8	19.8	4.77	9	22.8	12.92	8	37.1	20.86	0.114	-	-
/tatata/	9	12.2	9.87	8	9.3	2.87	9	12.4	4.45	8	19.6	12.25	-	0.423	0.423
/tititi/	9	23.9	21.70	8	21.1	6.96	9	22.7	12.09	8	36.8	19.23	0.211	-	-
/kakaka/	9	11.6	7.32	8	11.4	2.56	9	11.1	4.68	8	15.8	7.15	0.430	-	-
/kikiki/	9	25.6	18.26	8	21.1	6.62	9	22.3	9.95	8	35.9	20.29	0.200	-	-
/sasasa/	9	12.7	10.99	8	8.6	1.60	9	18.6	17.08	8	19.3	10.94	*0.049*	0.745	0.745
/sisisi/	9	27.1	19.37	8	20.5	6.44	9	31.4	17.90	8	39.6	20.68	0.177	-	-
Bilabials	7	15.7	6.73	6	14.7	5.61	7	19.3	7.93	6	20.7	14.98	0.648	-	-
Alveolars	7	16.0	6.14	6	13.7	3.62	7	20.4	9.38	6	22.0	17.53	0.651	-	-
Velars	7	18.9	6.77	6	25.8	7.36	7	19.6	7.21	6	27.7	8.76	0.112	-	-
Sibilants	7	19.6	10.61	6	15.7	4.89	7	28.9	14.75	6	23.0	15.81	0.284	-	-

to the current study and the study of *Doucet et al. (2013)* due to the larger sample size. *Ysunza et al. (1998)* observed significantly better phonological development and less compensatory articulation disorders in 4-year-old children with UCLP following palatal closure at 6 vs. 12 months of age, while in the current study no significant differences were obtained for presence of phonetic and phonological articulation errors. This difference in outcome might be due to variability in patient groups. The patient groups in the study of *Ysunza et al. (1998)* were larger and more homogeneous as they included only Mexican Spanish-speaking patients aged 48 months with hearing levels within the normal limits. In the current study, important group differences were noted concerning speech therapy and hearing treatment. Fifty percent (6/12) of the Belgian patients received speech therapy prior to speech assessment and insertion of ventilation tubes was performed in 75% (9/12). In contrast, none of the Ugandan patients received either speech therapy or hearing treatment, given that, unlike in Europe and North-America, speech therapy and hearing treatment are usually not included in the treatment practice of Ugandan patients due to the limited number of SLTs, audiologists and otorhinolaryngologists *(Fagan & Jacobs, 2009)* as well as the financial costs and absence of a medical insurance. Although disordered speech was still noted in Belgian patients who received speech therapy, the therapy will probably have improved the patients' speech quality. In addition, failure of the Eustachian tubes in young patients with CP might cause middle ear problems *(Goudy et al., 2006)*, what might result in chronic or repeated long-lasting episodes of chronic conductive hearing loss and potentially influences speech-language development *(Schönweiler et al., 1998)*. Although no information about hearing levels was available for both CP groups due to lack of the necessary equipment, accommodation and trained audiologists in CoRSU and the performance of the majority of the speech assessments in Belgian patients and control subjects at their homes, group difference in amount of patients treated for middle ear diseases might have advantaged the Belgian CP group in terms of speech production skills.

One-stage palatal repair before the age of 6 months is rarely applied in clinical practice due to technical difficulty of the surgical procedure and potential risks such as increased blood loss, airway obstruction and anesthetic problems *(Denk & Magee, 1996; Pradel et al., 2009; Rohrich et al., 2000)*. However, *Hodges (2010)* reported neither mortality, nor significant anesthetic complications in a series of 106 Ugandan patients with CLP following synchronous

CLP repair between 6 weeks and 10 months of age. In resource-poor countries, early synchronous lip and palatal repair (CLP) and early one-stage closure of the soft and hard palate (CP) are advocated by some well-equipped cleft teams to prevent malnutrition and death and to ascertain closure of the CP. Moreover, early closure of the cleft yields several advantages, such as reduced psychosocial stress and anxiety for the child *(Evans & Renfrew, 1974; Savaci et al., 2005)* as well as reduced financial costs for patients and society *(Pradel et al., 2009)*. To what extent adaptation of the surgical timing protocol in northern countries might be considered, taken into account the overall lack of differences in speech characteristics following palatal closure before and after 6 months of age, needs to be balanced against the potential difference in maxillofacial growth outcome.

A limited number of patients is included in the current study. The authors acknowledge the disadvantages of matched group designs with small sample sizes like in the present study. However, a substantial number of patients with early synchronous CLP repair is not yet available in CoRSU due to the young age of Ugandan patients visiting the Speech-Language Therapy department. Moreover, many patients with CLP operated in CoRSU before the age of 6 months did not return to visit the SLTs for a speech assessment. Perhaps, these patients might not experience considerable speech problems or postoperative surgery-related complications (e.g. fistulae, palate dehiscence) which are worth the traveling costs and distance. Further investigation with a larger sample will ascertain whether the current results can be generalized to the entire Ugandan cleft population. Moreover, a larger sample will allow to describe differences in speech results by cleft type. In addition, patients with C(L)P show widely divergent results for articulation and resonance due to influence of several variables. Controlled factors in the current study are cleft type, timing and technique of surgical palatal closure, age, gender, language and culture. However, other unknown variables might have influenced the current results. First, although none of the patients showed syndromic clefts, no information on patients' cognition was available. When cognitive abilities in children are significantly decreased, difficulties and delays in speech development may arise *(Baylis et al., 2008)*. Second, as discussed above, the presence and amount of conductive hearing disorders and its influence on the patients' speech-language development in both CP groups was unknown. Third, cleft surgeries in Uganda and Belgium were performed by two different surgeons (A.H. and K.B.). Although both surgeons were experienced in cleft surgery for more than 10 years, learned the Sommerlad technique from the same surgeon and performed it already for an extended period, differences in surgical skill may to some extent account for some of the speech disorders observed. Fourth, Ugandan patients' speech was assessed in their second language (English), whereas Belgian patients' assessment was carried out in their mother tongue (Dutch). Although the same was true for both control groups (which showed no significant differences for hardly all comparisons) it is unknown to what extent this variable has influenced the speech results. Finally, Dutch-speaking SLTs performed all speech evaluations. Although they had a good knowledge of the English language and although most English consonants included for analysis were identical to those occurring in the Dutch language, possible influence of the listener's language background cannot be excluded. Due to the presence of these and other potentially confounding variables, caution is advised in generalizing the results of the current study.

Conclusion

In conclusion, comparison of speech characteristics at a mean age of 4;8 years between early (≤ 6 months) and later (mean age 11.4 months) palatal closure overall revealed no significant group differences for consonant inventories, presence of omissions, substitutions and phonological processes, perceptual consensus evaluation of hypernasality, hyponasality, cul-de-sac resonance and nasal emission or mean nasalance values for oral speech samples. However, significantly more distortions were observed in the Belgian CP group due to higher occurrence frequencies for (inter)dental productions of apico-alveolar consonants.

Based on these speech results and considering the financial, cultural and social aspects of clefts in resource-poor countries, early (≤ 6 months) synchronous lip and palatal closure for CLP and one-stage soft and hard palatal closure for CP, as applied in CoRSU, seems to be justified. Whether or not one-stage closure of the soft and hard palate before the age of 6 months should be applied in northern countries is subject for further research. To answer this question, other important variables such as influence on maxillofacial growth should be taken into account. More clarity might be provided by long-term follow-up assessments of the children included in the current study.

Conflict of interest statement

The authors of this manuscript are employed by their respective institutions and have no relevant financial or non-financial relationships to disclose.

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Appendix 9.1: Photo Articulation Test – third edition (PAT-3)

1. Pie
2. Apples
3. Cup
4. Monkey
5. Hammer
6. Comb
7. Witch
8. Flowers
9. Hanger
10. Book
11. Baby
12. Bathtub
13. Dog
14. Ladder
15. Bed
16. Nails
17. Bananas
18. Can
19. Cat
20. Crackers
21. Cake
22. Gum
23. Wagon
24. Egg
25. Table
26. Potatoes
27. Hat
28. Fork
29. Elephant
30. Knife
31. Hanger
32. Swing
33. Jars
34. Angels
35. Orange
36. Shoe
37. Station
38. Fish
39. Lamp
40. Balloons
41. Bell
42. Vacuum
43. TV
44. Glove
45. Chair
46. Matches
47. Sandwich
48. Saw
49. Pencil
50. House
51. Spoon
52. Skates
53. Stars
54. Zipper
55. Scissors
56. Keys
57. Blocks
58. Clock
59. Flag
60. These feathers
61. Bath brush
62. Crayons
63. Train
64. Thumb
65. Toothbrush
66. Teeth
67. Radio
68. Carrots
69. Car
70. Boy
71. Bird

Appendix 9.2: Dutch articulation test

1. Zeven
2. Kerk
3. Doos
4. Trompet
5. Garage
6. Radio
7. Oranje
8. Wortels
9. Sneeuwman
10. Wiel
11. Arm
12. Vis
13. Vinger
14. Groot
15. Ballon
16. Tafel
17. Ezel
18. Kabouter
19. Olifant
20. Cadeau
21. Kruis
22. Trommel
23. Tandenborstel
24. Zetel
25. Net
26. Gitaar
27. Huis
28. Giraf
29. Hond
30. Deur
31. Bloem
32. Vlag
33. Nest
34. Strijkijzer
35. Rood
36. Worst
37. Brief
38. Appel
39. Brievenbus
40. Drie
41. Fles
42. Slang
43. Acht
44. Auto
45. Boekentas
46. Suiker
47. Tent
48. Hoed
49. Koffiekan
50. Spons
51. Mes
52. Gieter
53. Bank
54. Frieten
55. Citroen
56. Neus
57. Tas
58. Wieg
59. Zon
60. Hamer
61. Scheerapparaat
62. Molen
63. Muts
64. Twee
65. Fiets
66. Stoel
67. Kasteel
68. Paddenstoel
69. Sleutel
70. Vogel
71. Klok
72. Geld
73. Geweer
74. Voet
75. Gordijn
76. Groen
77. Pruim
78. Boom
79. Vissen
80. Trein
81. Nacht
82. Ster
83. Kaars
84. Televisie
85. Kam
86. Lepel
87. Das

88. Hoofd
89. Een
90. Chocolade
91. Zwart
92. Potlood
93. Piano
94. Kapstok
95. Knoop
96. Boek
97. Jongen
98. Bal
99. Wolk
100. Banaan
101. Toilet
102. Knie
103. Blauw
104. Kameel
105. Paraplu
106. Paard
107. Borstel
108. Schaar
109. Peer
110. Glas
111. Meisje
112. Vlinder
113. Lamp
114. Muur
115. Sigaret
116. Bril
117. Schrijven
118. Vliegtuig
119. Zwaan
120. Telefoon
121. Trap
122. Leeg
123. Pluim
124. Soldaat
125. Stofzuiger
126. Uurwerk
127. Kraan
128. Kooi
129. Lachen
130. Hemd
131. Varken
132. Fototoestel
133. Zaag
134. Soep
135. Schilderij

Appendix 9.3: Speech samples for Nasometric assessment

Syllables

/papapa/
/pipipi/
/tatata/
/tititi/
/kakaka/
/kikiki/
/sasasa/
/sisisi/

English sentences

Bilabials

Pick up the book.
Pick up the pie.
Pick up the baby.

Alveolars

Take a tire.
Take a turtle.
Take a teddy bear.

Velars

Go get a cookie.
Go get a car.
Go get a cake.

Sibilants

Suzy sees the dress.
Suzy sees the scissors.
Suzy sees the horse.

Dutch sentences

Bilabials

Pak het boek.
Pak de pot.
Pak de baby.

Alveolars

Tover de tafel.
Tover de schildpad.
Tover de teddybeer.

Velars

Krijg ik een koek.
Krijg ik een kar.
Krijg ik een cake.

Sibilants

Suzy ziet de schort.
Suzy ziet de schaar.
Suzy ziet de staart.

CHAPTER 10

IMPACT OF DELAYED ONE-STAGE PALATAL CLOSURE PERFORMED WITH THE SOMMERLAD TECHNIQUE ON SPEECH IN UGANDAN YOUNGSTERS AND ADULTS WITH CLEFT PALATE

Based on:

Luyten, A., Bettens, K., D'haeseleer, E., Dhondt, C., Hodges, A., Galiwango, G., Vermeersch, H., & Van Lierde, K. Impact of delayed one-stage palatal closure performed with the Sommerlad technique on speech in Ugandan youngsters and adults with cleft palate. *Submitted in International Journal of Language and Communication Disorders, March 2014.*

Abstract

Background: Untreated clefts still regularly occur in resource-poor areas. Due to the aesthetics- and speech-related consequences, these anomalies are often associated with severe socio-economic problems. Therefore, when these patients eventually arrive in specialized health care services, the (remaining) clefts are generally closed under the assumption that repair will yield benefit. A few studies already systematically evaluated speech after delayed palatal closure and reported severe disorders. However, the extent to which radical muscle dissection (Sommerlad technique) in delayed palatal repair can reduce symptoms of velopharyngeal insufficiency in this patient population is still uncertain.

Aims: The purpose of the present study was to assess resonance and articulation characteristics and patients' satisfaction with speech following delayed one-stage palatal closure (≥ 8 years) performed with the Sommerlad technique. Additionally, the influence of age at palatal closure and the length of time interval between palatal repair and speech assessment were verified.

Methods: Fifteen Ugandan participants with various extents of palatal clefting 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 and speech evaluations were carried out afterwards (mean age 18;10 years). Objective nasalance values were calculated and perceptual consensus evaluations of various resonance disorders were performed. Moreover, detailed phonetic and phonological assessments as well as satisfaction surveys were carried out.

Results: Resonance disorders and articulation errors each occurred in 87% of the patients. Compared to the control group, significantly higher prevalence of hypernasality and significantly higher nasalance values for all oral and oronasal speech samples were obtained in the cleft palate group. Moreover, significantly smaller consonant inventories and significantly more phonetic and phonological disorders were observed. None of the speech characteristics correlated with the age at palatal closure. Furthermore, a negative correlation was only obtained for weakly produced fricatives /s,z/ and the length of time period between palatal repair and speech assessment. Finally, 80% of the cleft palate group was dissatisfied with their speech.

Conclusion: Despite the use of radical muscle dissection, palatal closure after 8 years leads most often to striking resonance and/or articulation disorders. Consequently, awareness about the availability and importance of as well as the access to high quality surgical cleft treatment in resource-poor areas should increase in order to prevent patients' late presentation at specialized centers.

What this paper adds?

A few studies have attempted to systematically evaluate speech following delayed palatal repair. Although the majority reported severe speech disorders in this patient population, the extent to which application of radical muscle dissection (Sommerlad technique, an advanced surgical technique that has been proven to be successful when performed in young children [≤ 12 months]) for delayed palatal repair can reduce symptoms of velopharyngeal insufficiency in this patient population is still uncertain.

In the current study, none of the patients resolved both resonance disorders and articulation errors, despite radical muscle dissection. Therefore, earlier CP closure should be stimulated in resource-poor areas in view of avoiding or reducing development of cleft-related speech disorders.

Key Words

Cleft palate • Delayed one-stage palatal closure • Sommerlad technique • Resonance • Articulation • Resource-poor countries

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. In most countries in Europe and North-America, children born with CL/P are counseled by a craniofacial team shortly after birth. Multidisciplinary treatment adapted to the individual needs of the patient is then initiated. Surgical closure of the lip and the soft and hard palate is overall performed in early childhood during one to four operations *(Shaw et al., 2001)*. As a result, youngsters and adults with untreated clefts are rarely seen in Europe and North-America.

However, untreated CL/P in youngsters and adults regularly occurs in resource-poor areas. According to studies assessing reasons for late presentation in Nigeria *(Adeyemo et al., 2009)* and Nepal *(Schwarz & Khadka, 2004)*, explanations include lack of knowledge of availability of cleft repair, large distance to health services, and insufficient funds for travel, accommodation, and medical expenses. Considering that, in resource-poor countries, clefts are often associated with demonization, witchcraft, the curse of God and ancestral punishment *(Dagher & Ross, 2004)*, the deformed face and disordered speech predictably lead to psychosocial problems, social isolation, school absence, unemployment and celibacy *(Sommerlad, 2013)*.

In the past few decades, an increasing number of plastic and maxillofacial surgeons as well as craniofacial teams have visited resource-poor countries to treat patients with clefts and train local health care providers. In Uganda, for example, Christian Blind Mission (CBM) and Smile Train funded the establishment of a plastic and reconstructive unit in 2005, which moved in 2009 to the Comprehensive Rehabilitation Services in Uganda (CoRSU) hospital in Kisubi. In this hospital, a British plastic and reconstructive surgeon (A.H.) currently provides funded surgical treatment for patients with CL/P. As such, advanced surgical techniques, including the Sommerlad technique for palatal repair, are applied. Moreover, postgraduate trainings for plastic and reconstructive surgery are provided in collaboration with Mbarara University.

Although several surgeons reported on the surgical treatment of youngsters and adults with untreated clefts, speech assessment after delayed cleft repair have majorly been

descriptive impressions of professionals who were not speech-language therapists (SLTs) *(Adekeye & Lavery, 1985; Holdsworth, 1954; Hoppenreijs, 1990; Koberg & Koblin, 1973; Law & Fulton, 1959; Ortiz-Monasterio et al., 1974; Ortiz-Monasterio et al., 1966; Schwarz, 2006)*. Typically, these studies have provided scant information regarding patients' cleft type, palatal closure technique, assessed speech samples and perceptual (consensus) rating strategies. Moreover, all but two *(Law & Fulton, 1959; Schwarz, 2006)* of these studies broadly evaluated speech without distinguishing among speech intelligibility, resonance disorders and/or articulation errors.

Nevertheless, a few studies have attempted to systematically evaluate speech following delayed palatal repair. An overview is provided in Table 10.1. As seen in Table 10.1, 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)*. Moreover, many patients still showed hypernasality and nasal emission after delayed palatal closure *(Murthy et al., 2010; Sell & Grunwell, 1990; Zhao et al., 2012)*. As surgical techniques improve over years, this potential influencing factor needs to be taken into account. Conservative palatal repair was performed in all patients reported by *Sell & 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. Although this surgical technique has previously been reported to provide good speech results when performed in early childhood *(Andrades et al., 2008)*, the extent to which its application in delayed palatal repair can reduce symptoms of velopharyngeal insufficiency is still uncertain. Based on perceptual evaluations, *Murthy et al. (2010)* reported mild to severe hypernasality in all patients postoperatively, although *Zhao et al. (2012)* found absence of hypernasality in 36% (30/84) of the subjects. Moreover, *Murthy et al. (2010)* observed mild to moderate nasal emission in 94% (123/131) of the patients, while *Zhao et al. (2012)* noted this speech disorder in only 63% (53/84) of the experimental group. Unfortunately, none of both studies performed 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. Additionally, no information was provided regarding the patients' satisfaction with speech. Given the variation in speech results and the critical considerations, further research on speech outcome following delayed palatal repair performed with the Sommerlad technique seems required.

Table 10.1 Overview of the studies that systematically evaluated speech results after delayed palatal closure in patients with clefts.

Author	n and cleft type	Cleft closure: age and surgical technique	Age at speech assessment	Control group	Speech assessment	Speech results
Sell & Grunwell (1990) (Sri Lanka)	6 UCLP 2 BCLP 4 CP 6 soft CP	*Lip repair*: Age: n.a. Technique: n.a. *Palatal repair*: Age: n.a. Technique: - 5 Veau - 13 Langenbeck	*Age (preop)*: 11-19y (mean: 14y) all preop and 8m postop before speech therapy	n.a.	*Speech samples*: - Sihala: rote speech and modeled word list - imitation of sounds and trisyllables *Perceptual evaluation*: - nasality - articulation (place, manner)	***Nasality***: *Preoperative*: Severe or moderate hypernasality in all patients. *Postoperatively*: only 4 patients with soft CP improved (4/18, 22%); ***Articulation***: *Place*: Largely glottal and pharyngeal articulation before and after palatal closure. *Manner*: *Preoperative*: High frequency of nasal substitutions and approximations in patients with CLP. Some frication and plosion in patients with CP and soft CP. *Postoperative*: No spontaneous improvement.
Whitehill et al. (1996) (China)	1 BCLP	*Lip repair*: Age: infancy Technique: n.a. *Palatal repair*: Age: 13y Technique: n.a. (+ pharyngeal flap)	*Age*: 18y postop before speech therapy	n=1 without cleft	*Speech samples*: Cantonese: Connected speech and Cantonese Electropalatography Protocol - Revised *Perceptual evaluation* of resonance and phonetic transcription, quantitative and qualitative *analyses* for articulation	***Resonance***: Moderate hypernasality (score 4/6) and audible nasal emission; ***Articulation***: Moderate speech disorders. *Place*: Velar placement of alveolar plosives, fricatives and affricatives. Bilabial production of /f/. *Manner*: Nasal plosion of initial plosives /p,t,k/. Fricatives, affricatives and aspirated plosives accompanied with nasal emission.

Murthy (2009) and **Murthy et al. (2010)** (India)	99 UCLP 32 BCLP	*Lip repair*: Age: 6m after palatal repair Technique: - 99 (modified) Millard - 32 Mulliken *Palatal repair*: Age: ≥10y Technique: - 40 conservative muscle dissection with realignment of muscles at midline - 91 Sommerlad	*Age (preop)*: 10-35y (mean: 15.8y) all preop and 6 to 12m postop after home training	n.a.	*Speech samples*: - Tamil: 2 nasal, 2 oral, and 6 oronasal sentences - sustained vowels /a,e,i,u/ *Perceptual evaluation*: - speech intelligibility - resonance - nasal emission - articulation	***Intelligibility***: *Preoperative*: 22% (29/131) within first 3 (of 7) grades. *Postoperative*: 55% (72/131) within first 3 (of 7) grades; ***Hypernasality***: *Preoperative*: 17% (22/131) severe, 60% (79/131) moderate, 23% (30/131) mild. *Postoperative*: 36% (47/131) moderate or severe, 64% (84/131) mild; ***Nasal emission***: *Preoperative*: 25% (33/131) moderate, 69% (90/131) mild, 6% (8/131) normal. *Postoperative*: 16% (21/131) moderate, 78% (102/131) mild, 6% (8/131) normal; ***Articulation***: *Preoperative*: 38% (50/131) severe, 47% (61/131) moderate, 15% (20/131) mild articulation errors. *Postoperative*: 8% (10/131) severe, 48% (63/131) moderate, 44% (58/131) mild articulation errors. All speech parameters improved significantly after palatoplasty.
Zhao et al. (2012) (China)	31 UCLP 19 BCLP 32 CP 2 SMCP	*Lip repair*: Age: n.a. Technique: n.a. *Palatal repair*: Age: 5-24y	*Age*: n.a. all ≥6m postop	n.a.	*Speech sample*: - Mandarine: conversation and 10 short sentences - two/three-syllable words	***Hypernasality***: 6% (5/84) severe, 23% (19/84) moderate, 36% (30/84) mild, 36% (30/84) normal; ***Nasal emission***: 4% (3/84) severe, 27% (23/84) moderate, 32% (27/84) mild, 37% (31/84) normal;

Technique: - 40 two-flap palatoplasty with classic intravelar veloplasty - 44 Sommerlad	*Perceptual evaluation:* - hypernasality - nasal emission - velopharyngeal function	***Velopharyngeal function:*** 38% (32/84) needed secondary surgery for velopharyngeal insufficiency.

UCLP: unilateral cleft lip and palate, BCLP: bilateral cleft lip and palate, CLP: cleft lip and palate, CP: cleft palate, soft CP: cleft of soft palate only, SMCP: submucous cleft palate, preop: before palatal closure, postop: after palatal closure, n.a.: not available

The main purpose of the current study is to assess the resonance and articulation characteristics and patients' satisfaction with speech of Ugandan youngsters and adults with non-syndromic cleft palate (CP) following palatal closure performed with the Sommerlad technique after the age of 8 years. The age of 8 years is chosen given that phonetic and phonological development in typically developing children is considered to be completed at this age *(Peña-Brooks & Hegde, 2000)*. A well-defined speech assessment approach including objective assessment techniques as well as perceptual consensus evaluations, is applied. The patients' speech outcome and satisfaction with speech after delayed palatal repair is compared to age- and gender-matched Ugandan control subjects without clefts in order to control for the non-native language used during speech assessment (English). English remained one of the official languages of Uganda at the end of the colonial period in 1962 and is still deeply rooted in media, education and administration *(Mpuga, 2003)*. Although all participants included in the current study spoke English as their second language and were able to communicate fluently in English, potential influence of the non-native language on speech outcomes cannot entirely be excluded without comparison with a non-cleft control group. Furthermore, additional purposes were to verify the influence of age at palatal closure and length of time interval between palatal closure and speech assessment on resonance and articulation characteristics.

Materials and methods

This research project was approved by the Ethical Committee of the Ghent University Hospital, Belgium (EC2011/269).

Participants

The Ugandan CP group was recruited at the SLT department of CoRSU during four 10-day-long missions between January 2012 and January 2013 and consisted of 15 patients (7 males and 8 females) who underwent delayed repair of a non-syndromic CP. The CP group included 1 patient with a submucous CP, 3 patients with cleft soft palate and 4 with cleft soft and hard palate. Moreover, 6 patients showed unilateral cleft lip and palate (UCLP) and 1 presented with bilateral cleft lip and palate (BCLP). The native language of all patients was one of the Bantu languages (Luganda in 9/15, 60%). In all patients with cleft lip and palate

(CLP), surgical closure of the lip was performed in infancy by a local Ugandan surgeon at an estimated median age of 6 months (range 0;1-5;0 years). For all included patients, one-stage palatal closure was carried out at 15;10 years on average (range 8;0-27;8 years) by one experienced surgeon (A.H.) using the Sommerlad technique. Secondary fistula repair was performed in 2 patients (2/15, 13%). Four patients (4/15, 28%) received a secondary speech improving surgery. At the time of speech assessment, a mean age of 18;10 years (range 9;2-29;8 years) was noted for the CP group. The mean time period between palatal repair and speech assessment was 3;0 years (range 0;2-9;0 years). Two patients (2/15, 13%) received speech therapy prior to speech assessment: one male with a cleft of the soft palate (3x1h) as well as one female with a submucous cleft (1x1h/week for 8 months).

The age- and gender-matched control group was recruited at (1) the employees of CoRSU, (2) a local primary school and (3) the neighboring Swiss orphanage Kids of Africa. The group included 7 Ugandan males and 8 Ugandan females with a mean age of 18;4 years (range 8;11-29;2 years). The Independent Samples t-test could not identify significant age differences between the CP and control group (p=0.826). Inclusion criteria of the control group consisted of absence of (1) craniofacial or velopharyngeal anomalies, (2) self-reported problems with articulation and/or resonance, or (3) a cold at the day of testing. Moreover, all control subjects had a Bantu language as mother tongue (Luganda in 13/15, 87%).

Methods

Resonance

For the evaluation of resonance characteristics after delayed one-stage palatal closure, perceptual consensus ratings as well as objective assessment techniques were applied.

PERCEPTUAL CONSENSUS EVALUATION

Repetition of 15 simple English sentences, derived from the MacKay-Kummer Simplified Nasometric Assessment Procedures (SNAP) test *(Kummer, 2005)*, was digitally video-recorded (Sony HDR-UX1). These sentences, 12 oral and 3 nasal sentences, can be classified in 5 sentence groups based on the main consonants' characteristics: bilabials, alveolars, velars, sibilants, and nasals. Various types of resonance disorders (i.e. hypernasality, hyponasality, nasal emission/turbulence and cul-de-sac resonance) were perceptually evaluated by two

SLTs (A.L., C.D.) using a 4-point Likert scale (absent, slight, moderate, severe). Considering that nasal turbulence results from air flowing through an increased resistance in the nasal airway *(Peterson-Falzone et al., 2006)*, nasal emission and nasal turbulence were judged as one *(Henningsson et al., 2008)*. Similar to the procedure described by *Shriberg et al. (1984)*, the speech samples were first judged simultaneously, but independently. In case of disagreement, the samples were replayed until a consensus was reached. The interjudge reliability, calculated by the ratio of identical to total judgments for all resonance disorders, was 75%.

OBJECTIVE ASSESSMENT TECHNIQUES

Objective assessment of resonance included the measurement of nasalance values using the Nasometer (model II 6450). The Nasometer was developed by *Fletcher & Bishop (1973)* and manufactured by the *Kay Elemetrics Corporation (2010)*. The device consists of a headgear with a horizontal sound separator plate that is placed on the upper lip. A portable computer receives the oral and nasal acoustic energy collected by the microphones in front of the mouth and the nose. A nasalance score is calculated by dividing the nasal sound by the oral-plus-nasal-sound and multiplying this quotient with 100. Prior to the measurements, the appliance was calibrated according to the manual's instructions.

Nasalance values were obtained for 3 types of speech samples. At first, participants were asked to sustain the oral vowels /a/, /i/, and /u/ as well as the nasal consonant /m/ during 2 seconds at a comfortable pitch and loudness. Moreover, the participants twice repeated the 15 simple English sentences from the SNAP test *(Kummer, 2005)*, as cued by pictures. Finally, nasalance scores were obtained for an oronasal (Rainbow Passage) and oral (Zoo Passage) text. The passages were elicited via phrase repetition. Given that the presence of nasal emission might increase the nasalance values *(Dalston et al., 1991)*, all patients presenting with nasal emission/turbulence as well as their control-counterparts were excluded for the nasalance measurements of sentences and texts.

Articulation

Detailed consensus analyses of articulation characteristics were performed based on the Photo Articulation Test – third edition (PAT-3) *(Lippke et al., 1997)*. This standardized picture-naming test consists of 72 colored pictures of high frequency English words in which all

English consonants occur isolated in all permissible syllable positions as well as in common consonant clusters (see Appendix 10.1). The participants were asked to repeat these words as cued by the pictures, which was digitally video-recorded (Sony HDR-UX1). Narrow consensus transcriptions were performed by two SLTs (A.L., C.D.), using the International Phonetic Alphabet (IPA), the IPA extensions *(International Phonetic Association, 1999)* as well as additional symbols to describe specific cleft-related articulation errors *(Peterson-Falzone et al., 2006)*. The consonant-for-consonant interrater reliability was 87% on average (standard deviation 11.5%).

Given the widespread difficulties with correct pronunciation of the English fricatives /ʃ,ɵ,ð/ and affricates /tʃ,dʒ/ in Ugandans without clefts, further analyses were based on the 18 consonants (i.e. /p/, /b/, /t/, /d/, /k/, /ɡ/, /s/, /z/, /f/, /v/, /h/, /w/, /j/, /l/, /r/, /n/, /m/, and /ŋ/) arising in English as well as in Luganda, the most occurring native language of all participants (27/36, 75%). At first, a consonant inventory was composed, including all native consonants that were produced twice during the speech sample, regardless of whether those sounds were used correctly relative to the intended target sound. Subsequently, a phonetic analysis was performed at the segmental level by comparing the consonants with the target sounds. Four categories of errors were distinguished: omissions, additions, substitutions and distortions. The category 'distortions' was subdivided in various subcategories. In addition, phonological analysis consisted of analysis of consonant productions at word level. In accordance with the classification of *Ingram (1982)*, syllable structure processes, substitution processes as well as assimilation processes were distinguished. For all specific phonetic and phonological errors, the occurrence frequency (%) was calculated by the ratio of actual to potential occurrences. When such error type could occur at least 4 times and presented at least 20%, the error type was considered productive *(McReynolds & Elbert, 1981)*.

Satisfaction survey

A survey based on the Cleft Evaluation Profile *(Turner et al., 1997)*, used to assess satisfaction with cleft-related features, was performed as described by *Luyten et al. (2013)*. The topic of interest for the current study included the participants' satisfaction with speech. Therefore, the subjects were asked in one English yes-no question whether they were satisfied with their speech.

Statistical analyses

Statistical analyses were performed using SPSS software (version 19.0). In order to compare the CP and control group for categorical data, contingency tables were set and chi-squared or Fisher's exact tests were applied, depending on the expected cell counts. For all continuous variables, normality was assessed by the Kolmogorov-Smirnov and Shapiro-Wilk test. Group comparison was performed using parametric Independent Samples t-tests for normally distributed variables and non-parametric Mann-Whitney U tests in case of deviations of normality. Influence of age at palatal closure and length of time interval between palatal closure and speech assessment, both normally distributed variables, on articulation and resonance characteristics was assessed by calculating the Pearson (parametric) or Spearman (non-parametric) correlation coefficient for normally and non-normally distributed continuous variables, respectively. Within the CP group, a potential difference between patients with or without resonance disorders for both study parameters was assessed using Independent Samples t-tests. All significance levels were set at $\alpha=0.05$.

Results

Comparison of CP and control group

Resonance

PERCEPTUAL CONSENSUS EVALUATION

Perceptual evaluation ratings of hypernasality, hyponasality, cul-de-sac resonance and nasal emission/turbulence in the CP group by a 4-point Likert scale are provided in Table 10.2. Hypernasality occurred in 80% (12/15). Nasal emission/turbulence (4/15, 27%), hyponasality (3/15, 20%) and cul-de-sac resonance (2/15, 13%) were present in a minority of the CP group. Two patients (2/15, 13%) presented with normal resonance. In the control group, none of the participants showed resonance disorders. A chi-squared test revealed significant group differences for hypernasality ($p<0.001$). Group differences for nasal emission/turbulence ($p=0.100$), hyponasality ($p=0.224$) and cul-de-sac resonance ($p=0.483$) were not significant.

Table 10.2 Perceptual evaluation ratings (absent, slight, moderate, severe) of specific resonance disorders for the CP group.

	Hypernasality	Hyponasality	Cul-de-sac resonance	Nasal emission/ turbulence
Absent	20% (3/15)	80% (12/15)	87% (13/15)	73% (11/15)
Slight	33% (5/15)	20% (3/15)	7% (1/15)	7% (1/15)
Moderate	40% (6/15)	0% (0/15)	7% (1/15)	20% (3/15)
Severe	7% (1/15)	0% (0/15)	0% (0/15)	0% (0/15)

OBJECTIVE ASSESSMENT TECHNIQUES

Nasalance values in the CP and control group are provided in Table 10.3. Four patients of the CP group (4/15, 27%) with nasal emission/turbulence (i.e. 1 BCLP, 2 UCLP, 1 CP) and their control-counterparts were excluded from the calculation of mean nasalance values of sentence groups and texts. Moreover, nasalance values of all sentence groups were missing for 1 patient with CP (1/15, 7%). As seen in Table 10.3, significantly higher mean nasalance values were obtained in the CP group compared to the control group for all oral and oronasal speech samples (p<0.05). None of the nasal speech samples showed significant group differences (p>0.05).

Table 10.3 Mean nasalance values (%) and standard deviations (SD) for the CP and control group are provided. Only patients without nasal emission/turbulence and their control-counterparts were included for the sentence groups and texts.

Speech sample	CP group			Control group			*p*
	n	Nasalance	SD	n	Nasalance	SD	
/a/	15	26.9	11.41	15	17.1	11.17	*0.025*
/i/	15	57.0	17.83	15	24.3	10.87	*<0.001*
/u/	15	41.3	13.61	15	12.1	9.71	*<0.001*
/m/	15	92.7	3.35	15	91.5	6.87	0.835
Bilabials	10	38.5	16.66	10	15.7	7.41	*0.002*
Alveolars	10	36.1	17.39	10	16.2	9.22	*0.007*
Velars	10	40.4	15.93	10	21.8	10.34	*0.006*
Sibilants	10	40.4	21.79	10	16.7	8.10	*0.008*
Nasals	10	59.4	13.47	10	66.9	7.36	0.140
Oronasal text	11	47.4	12.73	11	35.8	6.51	*0.017*
Oral text	11	37.5	14.38	11	15.2	7.77	*<0.001*

Articulation

Articulation errors (i.e. incomplete consonant inventory and/or at least one phonetic or phonological error with a frequency of occurrence ≥ 20%) were present in 87% (13/15) of the CP group and in 27% (4/15) of the control group. This group difference was significant (p=0.001).

CONSONANT INVENTORY

In the CP group, the consonant inventories consisted of a median of 16 consonants (range 10-18/18). A complete consonant inventory was observed in 5 patients (5/15, 33%). The consonant inventories of the control group included a median of 18 consonants (range 17-18/18). One control subject (1/15, 7%) could not correctly produce the sound /z/ and obtained, consequently, an incomplete consonant inventory. The differences between the CP group and the control group were significant (p=0.001). The fricatives /s/ (p=0.006), /z/ (p=0.035), /f/ (p=0.042), and /v/ (p=0.006) were significantly more often absent in the CP group compared to the control group. The consonants /w/, /j/, /l/, /r/, /h/, and /m/ were included in the consonant inventories of all participants.

PHONETIC ANALYSIS

Phonetic analysis for the CP and control group is presented in Figure 10.1. The majority of errors produced by the CP group were distortions (mean occurrence frequency 29%, range 5-63%). Moreover, the CP group showed significantly more omissions (p=0.005), substitutions (p=0.005) and distortions (p=0.001) compared to the control group. No significant group differences were obtained for additions (p=0.535).

The occurrence frequency of various distortions for the CP and control group are provided in Table 10.4. In the CP group, some patients showed occurrence frequencies of at least 20% for apico-alveolar consonants (4/15, 27%) and bilabial production of /f,v/ (2/15, 13%). Moreover, such common occurrence was noted for the production of weak plosives /p,b,t,d,k,g/ and/or fricatives /s,z,f,v/ in 47% (7/15) of the CP group and for compensatory articulation of plosives (/p,b,t,d,k,g/) and/or fricatives (/s,z,f,v/) in 33% (5/15). Compared to the control group, significantly higher occurrence frequencies were observed in the CP group for weak plosives (p=0.002) and fricatives (p<0.001) as well as for compensatory articulation of plosives (p<0.001).

In accordance with the 20% criterion of McReynolds & Elbert (1981), 13 patients of the CP group (13/15, 87%) showed at least one phonetic disorder. In the control group, (inter)dental articulation of apico-alveolar consonants occurred with at least 20% occurrence frequency in 4 participants (4/15, 27%). This difference in presence of phonetic disorders between both groups was significant (*p*=0.001).

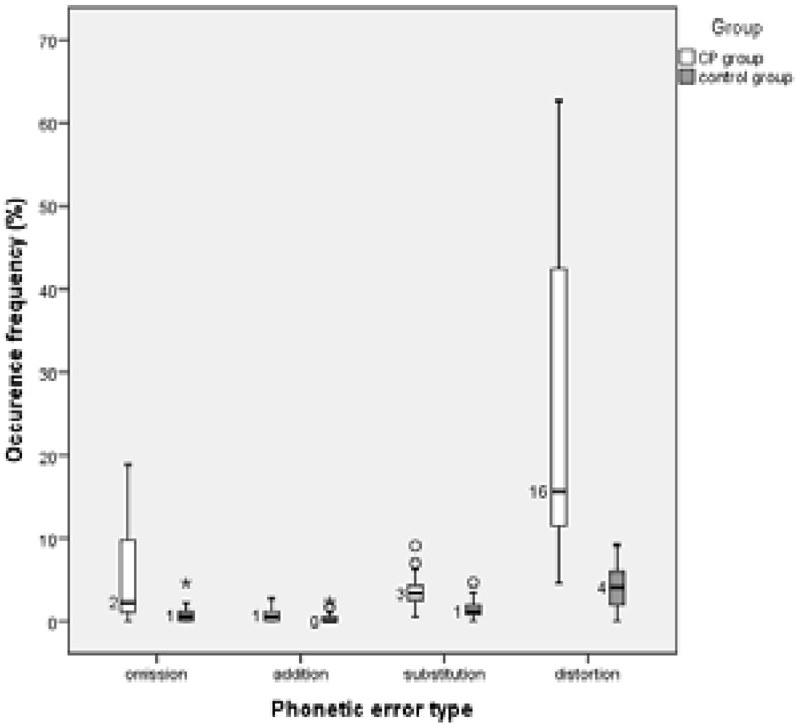

Figure 10.1 Box plots presenting the occurrence frequency of the various phonetic error types for the CP (white) and control group (gray). The median occurrence frequency is indicated next to the box plot.

Table 10.4 Overview of distortions occurring in the CP and control group. The mean and median occurrence frequency (expressed in %) as well as the standard deviation (SD) and the range are indicated.

Distortion type	CP group				Control group				*p*
	mean	SD	median	range	mean	SD	median	range	
(Inter)dental apico-alveolar consonants	20	29	5	0-83	5	6	4	0-20	0.300
Bilabial articulation of /f,v/	8	18	0	0-50	0	0	0	0-0	0.073
Weak plosives	23	21	16	3-64	3	2	3	0-7	*0.002*
Weak fricatives	29	32	13	0-94	2	3	0	0-10	*<0.001*
Compensatory articulation of plosives	10	10	6	0-32	0	0	0	0-0	*<0.001*
- /p,b/	6	1	0	0-4	0	0	0	0-0	0.150
- /t,d/	9	11	5	0-33	0	0	0	0-0	*0.004*
- /k,g/	22	26	12	0-76	0	0	0	0-0	*<0.001*
Compensatory articulation of fricatives	10	23	0	0-85	0	1	0	0-2	0.118
- /s,z/	12	27	0	0-97	0	1	0	0-3	0.118
- /f,v/	3	13	0	0-50	0	0	0	0-0	0.317

PHONOLOGICAL ANALYSIS

An overview of the most frequently occurring phonological articulation errors is shown in Table 10.5. The CP group, in comparison with the control group, exhibited significantly higher frequencies for deletion of final consonants (*p*=0.010), cluster simplification (*p*=0.019), fronting (*p*=0.009), backing (*p*<0.001), stopping (*p*=0.008), and assimilation (*p*=0.008). Taking into account the 20% criterion, 40% (6/15) of the CP group showed at least one phonological error such as cluster simplification, deletion of final consonants, backing, liquid gliding, and/or fronting. Considering that none of the control group presented with phonological errors, a significant group difference was observed (*p*=0.017).

Table 10.5 The most frequently occurring phonological errors in the CP and control group are presented. The mean and median occurrence frequency (expressed in %) are provided as well as the standard deviation (SD) and the range.

Phonological error	CP group				Control group				P
	mean	SD	median	range	mean	SD	median	range	
SYLLABLE STRUCTURE PROCESSES									
Deletion final consonants	10	13	4	0-35	1	3	0	0-12	*0.010*
Cluster simplification	14	16	6	0-45	2	3	0	0-10	*0.019*
- Cluster reduction	14	16	6	0-45	2	3	0	0-10	*0.013*
- Epenthesis	1	2	0	0-6	0	1	0	0-3	0.498
SUBSTITUTION PROCESSES									
Fronting	8	9	4	0-30	0	1	0	0-3	*0.009*
Backing	9	11	5	0-43	0	0	0	0-1	*<0.001*
Stopping	2	3	0	0-11	0	0	0	0-0	*0.008*
Liquid gliding	9	7	9	0-27	7	7	5	0-18	0.638
ASSIMILATION PROCESSES									
Assimilation	1	2	0	0-6	0	0	0	0-0	*0.008*

Satisfaction survey

Ten (10/15, 67%) patients of the CP group and their control-counterparts answered the question whether they were satisfied or dissatisfied with their speech. For the remainder patients (5/15, 33%: 2 UCLP, 1 CP, 1 soft palate only) the rating was performed by a relative and, as a consequence, they could not be included. Twenty percent (2/10) of the CP group and the entire control group (10/10) was satisfied with their speech. This group difference was significant (p=0.002).

Influence of age at palatal closure

Regarding resonance, no significant positive or negative correlations were observed within the CP group between age at palatal closure and nasalance values (p>0.10). Moreover, no significant differences in age at palatal repair were found between patients with or without

hypernasality (p=0.940), hyponasality (p=0.629), cul-de-sac resonance (p=0.185), or nasal emission/turbulence (p=0.964). Similarly, neither the extent of the consonant inventory (r=-0.018, p=0.949), nor any of the specific phonetic or phonological articulation errors (p>0.10), correlated significantly with age at palatal repair.

Influence of length of time interval between palatal closure and speech assessment

Within the CP group, the length of time interval between palatal closure and speech assessment did not correlate significantly with any of the mean nasalance values (p>0.05). In addition, no significant differences were observed between patients with or without hypernasality (p=0.877), hyponasality (p=0.528), cul-de-sac resonance (p=0.778), or nasal emission/turbulence (p=0.724) for the length of this time period. Regarding articulation, the time interval did not correlate significantly with the extent of the consonant inventory (r=0.104, p=0.711) or a specific phonological error (p>0.05). However, a significant negative correlation was observed for the length of time period between palatal closure and speech assessment and frequency of occurrence of weakly produced fricatives /s,z/ (r=-0.546, p=0.035).

Discussion

In resource-poor countries, CL/P is often left untreated in (early) childhood due to the lack of knowledge that clefts can be repaired, the substantial travelling distance to obtain care and/or the lack of resources *(Adeyemo et al., 2009; Schwarz & Khadka, 2004)*. Moreover, failure to complete palatoplasty after lip repair due to financial considerations and/or predominantly aesthetic concerns of parents seems to be widespread in these areas *(Onah et al., 2008)*. When youngsters or adults with CL/P eventually arrive in specialized health care services, the (remaining) clefts are generally closed under the assumption that repair will yield benefit. Lip repair will improve facial aesthetics, potentially enhancing self-confidence and improving prospects for education, employment and marriage *(Sommerlad, 2013)*. Palatal closure will reduce eating and drinking problems and will potentially improve hearing and/or speech abilities *(Sommerlad, 2013)*. However, the effect of delayed palatal closure with advanced surgical techniques on speech outcomes is still uncertain. Therefore, the current study aimed to assess resonance and articulation characteristics and patients' satisfaction with speech in Ugandan patients with non-syndromic CP following palatal closure with the Sommerlad technique after the age of 8 years.

Our primary finding was that none of the patients resolved both resonance disorders and articulation errors. This is in accordance with previously published studies reporting that palatal closure in youngsters or adults often will not normalize speech *(Murthy et al., 2010; Sell & Grunwell, 1990)*. In the current study, perceptual consensus evaluation ratings revealed absence of resonance disorders in only 13% (2/15). Slight hyponasality and slight to moderate cul-de-sac resonance were observed in 20% (3/15) and 13% (2/15) of the CP group, respectively, although mean nasalance values for nasal speech samples were still within the normal range. To our knowledge, no cases of hyponasality or cul-de-sac resonance were previously reported in literature after delayed palatal closure, except for one patient (1/4, 25%) with mixed hyper/hyponasality described by *Schwarz (2006)*. Furthermore, significantly higher mean nasalance values were observed in the current CP group for oral and oronasal speech samples compared to the control group and only 3 patients (3/15, 20%) showed no hypernasality. This prevalence was in between the percentages reported by *Murthy et al. (2010)* (0/131, 0%) and *Zhao et al. (2012)* (30/84, 36%), who both performed delayed palatal repair with the Sommerlad technique in the majority of the patients. However, in the current study, lower rates for nasal emission/turbulence (4/15, 27%) were observed compared to *Murthy et al. (2010)* (123/131, 94%) and *Zhao et al. (2012)* (53/84, 63%). This difference cannot be explained by differences in presence of residual fistulae, as these were absent in all patients of all three studies.

According to *Sell (2008)*, the high frequency of resonance disorders as a symptom of velopharyngeal insufficiency (VPI) following delayed palatal closure might be due to technical difficulties in late surgical palatal repair. Continuous upward pressure of the tongue into the unrepaired palate results in increased cleft width and vertically displaced palatal segments, what might hamper adequate restoration of the velopharyngeal valving mechanism *(Ortiz-Monasterio et al., 1966)*. Moreover, standard palatal repair in youngsters and adults often results in a palate which is way too short to reach the pharynx, leading as well to VPI and resonance disorders. Unfortunately, information regarding the velopharyngeal function of the CP group included in the current study could not be obtained given that

specialized equipment such as nasoendoscopy and/or videofluoroscopy were unavailable in CoRSU.

Articulation errors occurred in 87% (13/15) of the CP group included in the current study. Comparison with the control group revealed significantly smaller consonant inventories for the CP group as well as significantly higher occurrence frequencies for omissions, substitutions and distortions (particularly weak production of plosives and fricatives and compensatory articulation of plosives). Furthermore, the phonological processes deletion of final consonants, cluster simplification, fronting, backing, stopping, and assimilation occurred significantly more in the CP group compared to the control group. To our knowledge, detailed analysis of articulation, including a consonant inventory and phonetic and phonological analysis, following delayed palatal closure with the Sommerlad technique is not available in literature. Parallel to the current study, *Sell & Grunwell (1990)* reported largely glottal and pharyngeal articulation at 14 years on average after delayed palatal repair with conservative treatment in a Sri Lankan CP population and *Whitehill et al. (1996)* described velar placement of alveolar plosives, fricatives and affricatives in a Chinese patient with BCLP following palatal repair at the age of 13 years (surgical technique was not described). Delayed palatal repair might decrease obligatory articulation errors in some patients due to improved postoperative velopharyngeal functioning. However, technical challenges in palatal surgery will often result in inadequate restoration of the velopharyngeal valving mechanism and in persistence, to some degree, of velopharyngeal insufficiency and obligatory articulation errors. Moreover, compensatory articulation errors are typically well-established in youngsters and adults with untreated CP *(Ortiz-Monasterio et al., 1974)*. Motor skills associated with speech sound production are acquired over a long period of speech motor learning, which takes place in the first decade of life *(Ziegler & Ackermann, 2013)*. During this period, children develop a neural circuitry consisting of a feedback and feedforward system that integrates multiple vocal tract movement components into complex motor articulation patterns and that gains stability with age *(Guenther, 2006; Ziegler & Ackermann, 2013)*. Although successful primary surgical palatal closure in youngsters and adults might theoretically lead to spontaneous adaptation of the articulation patterns thanks to the brain's plasticity, spontaneous disruption of incorrect articulation patterns will often fail to occur. This negligible spontaneous recovery of speech was confirmed by the absence of significant negative correlations between most resonance and articulation disorders on the one hand and the time interval between palatal closure and speech assessment on the other hand. Consequently, postoperative speech therapy in this patient population is advocated *(Sell & Grunwell, 1990)*.

The success of postoperative speech therapy can be illustrated by the absence of articulation errors (i.e. complete consonant inventory as well as no phonetic or phonological errors with a frequency of occurrence ≥ 20%), despite the presence of moderate hypernasality, in the female patient with a submucous cleft who received speech therapy for 8 months (frequency: 1h/week). However, the requisite cost in absence of a medical insurance and the limited number of qualified SLTs challenge the routine provision of postoperative speech therapy in Uganda. Until recently, the only SLT department in Uganda was located at Mulago National Referral Hospital in the capital city, Kampala. However, in 2008, a bachelor program for SLTs was established at Makarere University, the only training program for SLTs in East and Central Africa. Consequently, the number of SLTs currently working in this country raised to 19. One of them is employed in CoRSU for one day per week since October 2012 in view of providing speech therapy to patients with CP. Considering that ambulant treatment is, however, only feasible for patients living not too far from the hospital, expansion of Speech-Language Therapy centers specialized in diagnosing and treating cleft-related speech disorders seems necessary.

To our knowledge, patients' satisfaction with speech following delayed palatal closure has never been reported in literature. However, according to the *World Health Organisation (2007)*, research should not only focus on body structures and functions, but should also include assessment of environmental and personal factors. In the current study, 80% (8/10) of the CP group was dissatisfied with their speech. Considering that *Turner et al. (1997)* noticed that satisfaction with the ability to verbally communicate highly influences the psychological well-being, these patients may be more at risk for development of psychological problems such as social withdrawal, depressions and anxiety disorders. Consequently, psychological counseling following delayed palatal closure may sometimes be required.

An additional aim of the current study was to verify the impact of age at palatal closure on resonance and articulation characteristics. Although it was hypothesized that more

resonance disorders and phonetic and phonological articulation errors would occur when the age at palatal closure increased, no significant positive or negative correlations were identified. These findings differ from those of *Koberg & Koblin (1973)*, who observed continuous worsening of functional speech with advancing age at time of palatoplasty. However, comparison is hampered by dissimilarities in surgical closure techniques and speech assessment protocol. Five different surgical techniques were included in the study of *Koberg & Koblin (1973)* and 'speech' was perceptually rated in consensus by a maxillofacial surgeon and an SLT. Therefore, the result of the current study, which suggests that success of surgical treatment of CP in subjects older than 8 years is not related to age at palatal closure, seems to be more reliable. The influence of other possible predictive factors for speech outcome after delayed palatal closure is subject for further research.

The strengths of the current study include measurement of objective nasalance values in addition to perceptual consensus evaluation of various resonance disorders, phonetic and phonological analyses after detailed phonetic consensus transcription, as well as comparison of the subjects with an age- and gender-matched control group to control for speech assessment in the non-native language. However, several limitations deserve mention. First, information of the velopharyngeal functioning was not available due to the lack of equipment such as a nasoendoscope or a multiview videofluoroscope in the resource-poor country. Second, the present study consisted of patients with various cleft types, including clefts of the soft palate only (n=3) and submucosal clefts (n=1). Improved speech outcomes might be expected compared to patients having CP, UCLP or BCLP *(Sell & Grunwell, 1990)*. However, the sample size was too small for stratification by cleft severity and composition of large homogeneous groups is difficult. Clefts of the lip that might influence the articulation of bilabial and labiodental consonants *(Landis & Cuc, 1975; Sommerlad, 2013)*, were repaired in infancy in all patients. The early presentation of these patients for lip repair suggests that recovery of facial aesthetics is an important motivation for Ugandan parents. However, the importance of palatal closure for functional outcomes such as feeding, hearing and speech might be underestimated in Uganda. Finally, several other unknown variables, including preoperative resonance and articulation characteristics, cognition, education, hearing levels, self-efficacy beliefs and socio-economic factors, might also have influenced the patients' speech outcome.

Conclusion

Similar to other studies regarding speech after delayed palatal repair in Sri Lankan *(Sell & Grunwell, 1990)*, Indian *(Murthy et al., 2010)* and Chinese patients *(Whitehill et al., 1996; Zhao et al., 2012)*, striking speech deviations were observed after delayed palatal repair in Ugandan patients with CP, despite the use of radical muscle dissection, an advanced technique that has been proven to be successful when performed in young children *(Andrades et al., 2008)*. Considering these distinct speech disorders in addition to the limited access to speech therapy for the majority of the patients, earlier CP closure should be stimulated in resource-poor countries in view of avoiding or reducing development of cleft-related speech disorders. Therefore, systematic medical examination of the neonate by trained medical staff should be promoted to trace CL/P. Moreover, more social workers should be employed to increase people's awareness regarding availability and importance of surgical cleft treatment. Furthermore, additional specialized treatment centers with trained local surgeons should be established to increase accessibility of care. Finally, in patients with CLP, synchronous lip and palatal closure should be carried out once the patient arrives at the hospital in order to prevent drop-outs for palatoplasty after successful lip repair.

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Appendix 10.1: Photo Articulation Test – third edition (PAT-3)

1. Pie
2. Apples
3. Cup
4. Monkey
5. Hammer
6. Comb
7. Witch
8. Flowers
9. Hanger
10. Book
11. Baby
12. Bathtub
13. Dog
14. Ladder
15. Bed
16. Nails
17. Bananas
18. Can
19. Cat
20. Crackers
21. Cake
22. Gum
23. Wagon
24. Egg
25. Table
26. Potatoes
27. Hat
28. Fork
29. Elephant
30. Knife
31. Hanger
32. Swing
33. Jars
34. Angels
35. Orange
36. Shoe
37. Station
38. Fish
39. Lamp
40. Balloons
41. Bell
42. Vacuum
43. TV
44. Glove
45. Chair
46. Matches
47. Sandwich
48. Saw
49. Pencil
50. House
51. Spoon
52. Skates
53. Stars
54. Zipper
55. Scissors
56. Keys
57. Blocks
58. Clock
59. Flag
60. These feathers
61. Bath brush
62. Crayons
63. Train
64. Thumb
65. Toothbrush
66. Teeth
67. Radio
68. Carrots
69. Car
70. Boy
71. Bird

PART 3

General discussion

CHAPTER 11

GENERAL DISCUSSION AND CONCLUSION

Cleft lip and/or palate (CL/P) arises worldwide in 1.7 per 1000 liveborn babies, including ethnical variations *(Mossey & Castilla, 2003)*. In Uganda, birth prevalence of CL/P is estimated between 0.73 *(Dreise et al., 2011)* and 1.34 *(Kalanzi et al., 2013)* per 1000 live births. Although this number seems low, a large amount of Ugandan children is born with a cleft each year due to the high annual number of births in this country *(UNICEF, 2003)*. In the Comprehensive Rehabilitation Services in Uganda (CoRSU) hospital, the only specialized center for CL/P repair in East-Central Africa, surgical timing of cleft closure for patients with CL/P has been changed compared to northern countries. In order to avoid defaults prior to completion of all necessary surgical procedures, nearly all clefts are repaired during a single surgery including synchronous lip and palatal closure for patients with unilateral (UCLP) or bilateral (BCLP) cleft lip and palate (CLP) and a one-stage soft and hard palatal closure in case of cleft palate only (CP). Moreover, to avoid (further) malnutrition and death in children with palatal clefts, very early cleft repair is preferred. Nevertheless, not all clefts are closed in infancy given that some patients present for the first time at the hospital in later childhood, youth or adulthood.

Prior to the start of this doctoral thesis, no information was yet available about satisfaction and speech outcomes following these adapted surgical approaches in Ugandan patients treated in CoRSU. Moreover, only few, if any studies systematically verified satisfaction and speech after such surgical treatment protocols. Considering the increased recognized importance of good speech and satisfaction outcomes following surgical cleft repair, the general purpose of the current doctoral thesis was to assess satisfaction and speech of Ugandan patients with surgically closed CL/P during a single surgery by one experienced surgeon using the Sommerlad technique for palatal repair. As such, a valuable contribution to the revelation of the optimal surgical treatment of patients with palatal clefts was aimed.

Usefulness of Ugandan-English normative nasalance values

Although perceptual listener's judgments are still the gold standard for evaluation of resonance disorders, the need for objective assessment techniques is increasingly recognized in clinics and research. The Nasometer has been proven to be a suitable, non-invasive, and objective instrument with good reproducibility *(Watterson & Lewis, 2006)* and significant correlations with perceptual judgments *(Brunnegård et al., 2012)*. Norms are however required for each language, given that significant differences between various normal populations have been proven *(Seaver et al., 1991; Van Lierde et al., 2001)*. Considering that norms for Ugandan children were unavailable, the Nasometer could not be used in Uganda. Therefore, normative nasalance values were established for Ugandan English-speaking males and females between 2;7 and 13;5 years (Chapter 6). No significant gender or age differences were observed.

The need for Ugandan normative nasalance values increased in recent years as speech-language therapy gradually became more prevalent in this country thanks to the education program established at Makarere University. Moreover, since the foundation of a Speech-Language Therapy department at CoRSU, an increased number of patients with resonance disorders is assessed and treated. These resonance disorders are generally caused by clefts or result from oral, nasal and/or pharyngeal surgery. In addition to the clinical usefulness, normative nasalance values of Ugandan children are also valuable in view of research on speech in patients with palatal clefts, as shown in Chapters 8 and 9. In Chapter 8, some children from the norm group were selected (based on matched age and gender) in order to objectively describe presence of hypernasality in Ugandan children with UCLP or BCLP following synchronous CLP repair in comparison with a non-cleft control group. Moreover, in Chapter 9, selected Ugandan children (based on matched age and gender) were compared to Belgian children without craniofacial anomalies to control for language differences. This comparison for identical and closely resembling oral speech samples revealed no significant differences in mean nasalance values between Ugandan-English- and Flemish-Dutch-speaking children.

Implications of satisfaction surveys

To determine the success of surgical treatment in patients with CL/P performed in CoRSU, satisfaction surveys were carried out in 44 parents or guardians of children with UCLP and BCLP (mean age 3;1 years) closed during a synchronous lip and palatal repair (Chapter 7) as well as in 10 patients (mean age 21;8 years) with palatal clefts repaired in youth or adulthood (Chapter 10). Such investigations are recommended by the *World Health Organisation (2002)*, given that environmental and personal factors in addition to body structures and functions contribute to the extent of the patient's disability. Overall high levels of parental satisfaction were observed for appearance of the face (44/44, 100%), lip (39/42, 93%) and nose (38/44, 86%). Moreover, increased satisfaction with lip appearance was noted when time intervals between surgery and survey decreased. Parents of children with CLP seem to be relieved by the decreased visibility of this congenital anomaly, particularly in the first few months after surgery. Ugandan patients with visible disabilities are often stigmatized and socially isolated *(International Labour Organization, 2009)*. They seem to be vulnerable for abuse, discrimination and denial of basic provisions *(Human Rights Watch, 2010)* and have decreased prospects for education, marriage and employment *(Bradbury & Habel, 2008)*.

Lower levels of parental satisfaction were found for teeth appearance (19/34, 56%) and speech (18/32, 56%) in patients older than 20 months and 18 months of age, respectively. Moreover, only 20% (2/10) of the youngsters and adults were satisfied with their speech following delayed palatal repair. In both studies, the percentages differed significantly compared to the age- and gender-matched non-cleft control group. These lower satisfaction levels might be explained by inappropriate preoperative parental and patients' expectations *(Oosterkamp et al., 2007)*. Therefore, even more comprehensive information should be provided prior to surgery about the potential (lack of) influence of surgical treatment on the patient's speech and teeth appearance in order to maximize patients' and parental satisfaction afterwards. Moreover, availability of and access to speech therapy, dental care and orthodontic treatment is limited in Uganda and should improve to provide a more comprehensive interdisciplinary treatment to all Ugandan patients with CL/P. Establishment of the Speech-Language Therapy department in CoRSU has been a first step forward to achieve this objective.

Early cleft closure during a single surgery: to do or not to do?

The surgical approach preferred in CoRSU for treatment of patients with CL/P consists of early closure of the entire cleft during a single surgery, including synchronous lip and palatal repair (CLP) or one-stage soft and hard palatal closure (CP). In view of improving feeding abilities and chances of survival, many clefts are closed prior to the age of 6 months. In order to verify if this surgical sequence and timing results in overall good or acceptable articulation and resonance characteristics, Ugandan patients with a mean age of 4;9 years were compared to a Ugandan age- and gender-matched control group (Chapter 8). This comparison revealed no significant group differences regarding presence of hypernasality, hyponasality and cul-de-sac resonance and concerning mean nasalance values for oral speech samples, despite a significantly higher occurrence of nasal emission/turbulence in Ugandan patients. Moreover, smaller consonant inventories, more phonetic disorders such as bilabial production of labiodentals and compensatory articulation of fricatives /s,z,f,v/ and plosives /t,d/, as well as more phonological processes such as cluster simplification, deletion of final consonants and backing were noted for Ugandan patients. This study showed that development of compensatory articulation errors had not been avoided, although the CP was presumably closed prior to the start of the canonical babbling stage (i.e. 6 to 10 months in typically developing children *(Chapman et al., 2001)*). The increased presence of compensatory articulation errors and phonological processes might be explained by the relatively high number of former structural irregularities such as fistulae and palate dehiscence in the current patient group *(Chapman, 1993; Peterson-Falzone et al., 2001)*.

This doctoral thesis provides, however, two arguments for applying this surgical timing protocol in resource-poor countries. First, no significant differences were obtained for parental satisfaction with facial appearance and speech of Ugandan children with UCLP or BCLP, repaired by a synchronous lip and palatal repair before or after the age of 6 months (Chapter 7). Second, comparison between Ugandan patients with early palatal closure (≤ 6 months) and a Belgian CP group (matched for cleft type, age and gender) with later one-stage soft and hard palatal repair (mean age 11 months) revealed at least comparable articulation and resonance characteristics (Chapter 9). No significant differences were observed for perceptual consensus evaluation of resonance, mean nasalance values for oral speech samples, consonant inventories or most phonetic errors and phonological processes. However, the Belgian CP group showed significantly more distortions due to higher occurrence frequencies for (inter)dental production of apico-alveolar consonants. Moreover, none of the included Ugandan patients received speech therapy or hearing treatment, while 50% of the Belgian CP group visited a speech-language therapist (SLT) and ventilation tubes were inserted at least once in 75% of the Belgian patients. Given the potential positive influence of speech therapy and hearing treatment on speech production, these group differences might have favored the Belgian CP group.

Multiple additional advantages for early closure of CL/P during a single surgery (≤ 6 months) in resource-poor countries have been reported. Although technically challenging, early synchronous closure of the entire cleft has been proven to be a safe procedure with minimal risks for minor peri- and postoperative complications when it is performed by an experienced surgeon and in presence of well-trained anesthetists and specialized anesthetic equipment, like it is the case in CoRSU *(Hodges, 2010)*. Moreover, cleft closure prior to the age of 6 months provides improved feeding abilities which decreases the risks for (further) malnutrition and death *(Cubitt et al., 2012)*, and reduces the feeding time lengths, the parental stress during feeding and the consequential negative effect on the parent-infant bonding process *(Miller, 2011)*. In addition, a single surgical procedure requires only one (long) journey to the hospital, one anesthesia and one hospitalization period, what results in decreased costs, lack of defaults for second surgery *(Hodges, 2010; Onah et al., 2008)*, lower medical risks *(De Mey et al., 2006)*, improved psychological well-being *(Kramer et al., 2008)* and less chances for developing learning disabilities *(Wilder et al., 2009)*. Although in CoRSU, surgical cleft closure is free for children and adults with CL/P, parents need to pay for travelling and basic provisions during hospitalization. Consequently, travelling to the hospital demands a great financial effort from parents and their surrounding family. Considering all the advantages of early CL/P closure during a single surgery, this surgical timing protocol seems to be the most appropriate approach for CoRSU and similar well-equipped reference centers for CL/P repair in resource-poor countries.

In the North, early synchronous lip and palatal repair (CLP) or early one-stage soft and hard palatal closure (CP) is not an indispensible life-saving procedure like in resource-poor countries. Nevertheless, many of the above-mentioned advantages of this surgical approach such as financial and psychological considerations, also apply for northern countries. Moreover, considering the availability for hearing treatment and speech therapy, even better speech outcomes can be hypothesized, especially for articulation. However, to form the complete picture on effectiveness of this surgical approach, information about patients' maxillofacial growth disturbances is necessary. When increased maxillofacial growth disturbances would be noted, the need for future orthodontic or surgical treatment for malocclusions and/or maxillary hypoplasia might increase as well. This would result in additional financial costs for the patient, his/her family and the society as well as in increased psychological difficulties such as depression or low self-esteem *(Millar et al., 2013)*. To date, no information on maxillofacial growth is yet available for Ugandan patients. However, maxillofacial growth outcomes after similar surgical approaches reported in other studies seem to be encouraging *(De Mey et al., 2009; Mueller et al., 2012)*.

Delayed cleft closure: is it still worthwhile?

In northern countries, presence of CL/P is generally detected immediately after birth or even prenatally *(Franco et al., 2013)* due to good access to affordable health care, the huge number of deliveries in presence of a doctor and/or midwife and systematical postnatal general health screening. When CL/P is diagnosed, interdisciplinary treatment by a craniofacial team is initiated and feeding support is provided. Different circumstances in resource-poor countries often cause failure to early diagnose and/or treat (the entire) CL/P resulting in high mortality rates in the first few months of life *(Cubitt et al., 2013)*. However, some patients with untreated CL/P manage to survive. Consequently, well-trained surgeons of temporary visiting teams and permanent specialized centers for CL/P (such as CoRSU) are faced with youngsters and adults with untreated CL/P *(Sell, 2008)*. Considering the large number of untreated patients, one can question whether cleft closure in youngsters and adults needs to be prioritized. In the current doctoral thesis, overall severely disordered resonance and articulation was observed after delayed palatal repair (mean age 15;10 years) in 15 Ugandan patients with various cleft types (Chapter 10). In comparison with an age- and gender-matched non-cleft control group, significantly more hypernasality as well as significantly higher objective mean nasalance values for oral and oronasal speech

samples were obtained in the CP group. Moreover, group comparison revealed smaller consonant inventories, more phonetic errors such as weak production and compensatory articulation of plosives and fricatives, as well as more phonological disorders such as cluster simplification, deletion of final consonants, backing, liquid gliding and fronting in Ugandan patients. Neither the age at palatal closure, nor the length of time interval between surgery and speech assessment overall correlated with speech performance. Whether articulation and/or resonance improved compared to the preoperative condition could not be assessed, given the lack of preoperative speech assessments in the majority of the patients. In literature, contradictory results have been reported when pre- and postoperative speech evaluations were compared *(Murthy et al., 2010; Sell & Grunwell, 1990)*. Particularly regarding compensatory articulation errors, potential spontaneous adaptation of the well-established articulation patterns will depend on the patient's reaction to the altered auditory and somatosensory feedback after successful palatal surgery *(Guenther, 2006)*. However, many patients will require speech therapy in view of improving speech abilities. Consequently, provision of speech therapy to Ugandan patients with delayed palatal repair in the Speech-Language Therapy department of CoRSU needs to be stimulated in future.

Despite postsurgical maintenance of decreased speech intelligibility, cleft closure in youngsters and adults can still be worthwhile. Closure of the cleft lip can remove the stigma and improve social acceptance within the community, what will result in enhanced prospects for education (in case of youngsters), marriage and employment *(Bradbury & Habel, 2008)* as well as in increased self-confidence and feelings of self-worth *(Sommerlad, 2013)*. Moreover, CP repair will improve the patient's feeding/drinking abilities and oronasal hygiene by decreasing or eliminating nasal regurgitation. Therefore, the benefits of delayed cleft closure seem to outweigh the risks associated with surgical interventions in youngsters and adults *(Sommerlad, 2013)*.

Strengths and limitations

The current doctoral thesis shows several strengths and limitations, which are summarized in Table 11.1.

Table 11.1 Summary of strengths and limitations of the current doctoral thesis.

Strengths	Limitations
- Socially relevant research - Economically relevant research at times of crisis - Contribution to rarely explored research fields - Normative nasalance values for Ugandan-English now available - Controlled study designs - Detailed speech assessments - Justification for surgical procedure in CoRSU in terms of satisfaction and speech outcome - Reflection on optimal cleft treatment in the North	- Small sample sizes - Speech assessments in English - No objective assessment of velopharyngeal function - Several uncontrolled potentially influencing factors - No information regarding maxillofacial growth

Searching for the optimal surgical treatment for Ugandan patients with CL/P in terms of good speech intelligibility, as aimed in the current doctoral thesis, is socially relevant considering that speech outcomes will partially determine the patients' disability *(World Health Organisation, 2002)*. Moreover, research topics of the current doctoral thesis are economically relevant, which is indispensable at times of crisis. In addition, rarely explored but essential research fields were subject of investigation in this doctoral thesis. Normative nasalance values for Ugandan English-speaking children were not yet available. Studies on satisfaction with facial appearance and speech in (parents of) patients with clefts were seldom conducted in non-white populations *(Noor & Musa, 2007; Reekie, 2011)* and have, to our knowledge, never been performed in a black-African population or following synchronous lip and (soft and hard) palatal closure. Moreover, only few studies (with some important methodological shortcomings) systematically described speech outcomes after palatal repair prior to 6 months of age *(Abdel-Aziz, 2013; Barimo et al., 1987; Copeland, 1990; De Mey et al., 2006; Doucet et al., 2013; Grobbelaar et al., 1995; Ysunza et al., 1998)* or after the age of 8 years *(Murthy et al., 2010; Schwarz, 2006; Sell & Grunwell, 1990; Whitehill et al., 1996; Zhao et al., 2012)*. Regarding study design, all studies on satisfaction or speech outcomes in Ugandan patients with CL/P included age- and gender-matched non-cleft control groups in order to control for ethnical or racial variations in parental satisfaction on the one hand and for typically developmental speech errors and/

or influence of dialect on the other hand. Furthermore, detailed speech assessments were carried out to verify speech outcomes following adapted surgical approaches. Perceptual consensus evaluations were performed by two SLTs for various resonance disorders. During these perceptual consensus evaluations, controlling the number of pressure consonants by using identical or comparable speech samples facilitated group comparisons *(Henningsson et al., 2008)*, although spontaneous speech would have leaned closer on reality. Moreover, perceptual judgments of hypernasality were confirmed by objective mean nasalance values. Regarding articulation, consensus transcriptions were made by two SLTs. In addition, detailed phonetic and phonological analyses were performed using occurrence frequencies. In contrast to perceptual ratings, occurrence frequencies allow to reveal subtle variations in articulation skills. Furthermore, they enable cross-linguistic comparisons considering that differences in language difficulty as determined by the number of high pressure consonants *(Hutters & Henningsson, 2004)* are taken into account by comparing relative amounts of articulation errors. Moreover, use of the 20% occurrence frequency criterion prevented that accidental slips of the tongues were called disorders. Finally, the current doctoral thesis provides justification for the surgical protocol currently applied in CoRSU in view of satisfaction and speech outcome. Moreover, reflection is made on the optimal timing for surgical treatment of patients with CL/P in northern countries.

A first major limitation of the current doctoral thesis are the small sample sizes included in studies on speech outcomes, which hamper generalization. Only few children with early synchronous lip and palatal repair (CLP) or early one-stage soft and hard palatal closure (CP) were qualified for inclusion, considering the young age of the majority of these patients seen at the Speech-Language Therapy department of CoRSU. Moreover, Ugandan children and adults with CL/P often not comply with appointments for follow-up assessments after surgical cleft repair. This is particularly expected in patients without postoperative complications or speech problems and might be explained by financial and/or cultural considerations. Second, all speech assessments in Ugandans were carried out in English, the participants' second language. Given the numerous languages currently spoken in Uganda *(Lewis et al., 2013)*, performing speech evaluations in the patients' mother language was impossible. Third, objective assessments of velopharyngeal function using nasoendoscopy and/or videoendoscopy were not carried out due to the lack of necessary equipment at

CoRSU. Fourth, we were unable to control many variables that might have influenced the patients' speech performance such as hearing levels, episodes of otitis media, cognition and socioeconomic factors, considering that audiologists, otorhinolaryngologists, equipment for hearing screenings and intelligence tests were unavailable in CoRSU. Moreover, no information was obtained regarding the patients' dental relationships and its influence on patients' speech results. Finally, lack of information regarding maxillofacial growth outcomes resulted in an incomplete picture on the effectiveness of the assessed surgical treatment approaches.

Future perspectives

In the current doctoral thesis, normative nasalance values have been established for Ugandan English-speaking males and females aged 2;7 to 13;5 years. Although no age differences were noted within this age range, literature currently provides evidence that normative nasalance values might differ significantly between children and (young) adults *(Abou-Elsaad et al., 2012; Hirschberg et al., 2006; Van Lierde et al., 2003)*. In Uganda, no normative nasalance values are yet available for youngsters or adults. However, such norms would be valuable considering that several patients present the first time at CoRSU after 13;5 years of age and given that some (often rather well-to-do) patients like to initiate speech therapy at (young) adult age to increase their socioeconomic prospects.

In addition, future studies regarding hypernasality in Ugandan patients with CL/P may focus on application of the Nasality Severity Index in clinics and research *(Bettens et al., 2013; Bettens et al., in preparation; Van Lierde et al., 2007)*. The Nasality Severity Index is a multiparametric approach that uses two different acoustic measurement techniques (i.e. nasalance values and spectral characteristics of /i/) to assess the degree of hypernasality. It is a non-invasive, easily repeatable index with high sensitivity and specificity which strongly correlates with perceptual judgments of hypernasality. Considering that the results of three different objective assessment techniques are reduced to one number, this tool seems to be easier to interpret and more powerful compared to the Nasometer.

In satisfaction surveys, treatment success was determined by interviewing parents/ guardians or patients regarding facial appearance and/or speech. Speech outcomes have

been additionally assessed in detail by experienced SLTs. However, judgments of facial appearance by an independent panel of professionals such as surgeons, nurses or other members of a cleft team, have not yet been performed. Nevertheless, this would be of value in order to obtain a more critical view on facial aesthetic outcomes after synchronous lip and palatal closure in Ugandan children and adults.

In this doctoral thesis, a limited number of young Ugandan children was assessed to describe articulation and resonance characteristics after early synchronous lip and palatal repair (CLP) or one-stage soft and hard palatal repair (CP) (≤ 6 months) and to compare these results with speech following early lip repair and later palatal closure (> 6 months). Therefore, only short-term speech results are provided for children in the middle of their speech-language development. Moreover, the small sample sizes hamper generalization. To ensure the effectiveness of the current surgical management in terms of speech outcome, long-term follow-up assessments are necessary. In addition, expansion of the sample size is required to allow generalization.

Many Ugandans present at CoRSU far beyond the optimal age for surgical CP closure. Verifying the reasons for such late presentation would allow to ameliorate earlier tracing of patients with clefts by social workers of CoRSU in order to increase the Ugandan patients' chances to survive and improve their speech later in life. Moreover, comparison of pre- and postoperative speech evaluations would clarify the effectiveness of delayed palatal closure in terms of speech outcomes.

Finally, the current doctoral thesis focused on satisfaction and speech outcomes of Ugandan patients treated in CoRSU. However, it should be realized that information on short-term and long-term maxillofacial growth outcomes in Ugandan patients is required as well to have a complete picture of effectiveness of these surgical approaches. Further research in this field is therefore advised.

Conclusion

Considering the infrequently used surgical sequence and timing approach for Ugandan patients with CL/P applied in CoRSU, the aim of the current doctoral thesis was to verify satisfaction and speech outcomes in these Ugandan patients treated by one experienced plastic and reconstructive surgeon using the Sommerlad technique for palatal repair. Insights on various areas have been provided:

- Normative nasalance values are now available for Ugandan English-speaking children.
- High levels of parental satisfaction with hearing as well as lip, nose and overall facial appearance were observed for Ugandan patients with UCLP or BCLP following synchronous lip and palatal repair, despite lower parental satisfaction levels for teeth appearance and speech.
- Although palatal repair prior to 6 months resulted in various deviations of normal speech development, at least similar articulation and resonance characteristics were observed compared to patients with palatal repair after 6 months of age.
- Overall, severely disordered speech characterized by hypernasality, incomplete consonant inventories and phonetic and phonological disorders was noted for patients with palatal cleft repair after the age of 8 years.

When these findings are placed within a broader framework, early closure of the entire cleft during a single surgery seems to be a suitable surgical approach for resource-poor countries in case the surgical conditions are adequate. Moreover, delayed cleft repair in youngsters or adults is considered to be valuable. In future, additional actions should be taken in CoRSU to decrease the amount of patients treated after infancy with the aim of increasing chances for survival and improving patients' speech production. Moreover, expansion of the Speech Language Therapy department of CoRSU is recommended in order to treat more patients with significant articulation and resonance disorders resulting from palatal clefts. Finally, otorhinolaryngologists, orthodontists and dentists should be included in CoRSU's permanent craniofacial team in view of providing comprehensive interdisciplinary treatment to all Ugandan patients with CL/P.

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Cleft lip and/or palate is a congenital craniofacial anomaly characterized by multifactorial inheritance. This anomaly is associated with atypical facial appearance, hearing problems, malocclusions and speech disorders, and arises worldwide on average in 1.7 per 1000 live births.

In the Republic of Uganda, 1128 to 2070 babies are born with a cleft each year. However, the facilities for surgical treatment are limited in this resource-poor country. In the well-equipped Comprehensive Rehabilitation Services in Uganda (CoRSU) hospital, surgical cleft closure by means of international accepted surgical techniques is offered for free. Considering the Ugandan cultural beliefs and circumstances, the treating surgeon prefers to perform synchronous lip and (soft and hard) palatal repair soon after the patient's arrival at the hospital.

The current doctoral thesis focuses on assessing satisfaction and speech outcome of these Ugandan patients with surgically closed cleft lip and/or palate treated during a single surgery by one experienced surgeon using the Sommerlad technique for palatal repair. As such, this doctoral thesis contributes to the search for the optimal surgical treatment for patients with palatal clefts.