Vertical maxillary growth in unilateral cleft lip and palate.

A comparison of two surgical protocols

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Licentiate Thesis

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This work is dedicated

to **Jan Lilja,**

and to the memory of my sister, **Rahma**

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Abstract

Objective: The aim of the present study was to compare vertical maxillofacial growth in patients born with unilateral cleft lip and palate (UCLP) who were treated with one of two different surgical protocols.

Design: A retrospective cohort study.

Subjects: One hundred seventy-six consecutive patients with complete UCLP treated at Sahlgrenska University Hospital in Gothenburg, Sweden, were divided into two groups: (1) the W-K group, consisting of 60 patients born 1965 to 1974 who were treated surgically according to a Wardill-Kilner (W-K) protocol, and (2) the Gothenburg DHPC group, consisting of 116 patients born 1975 to 1995 who were treated surgically according to the Gothenburg delayed hard palate closure (DHPC) protocol.

Methods: Cast models and lateral cephalograms obtained at 10 years of age were analyzed.

Results: Patients treated according to the Gothenburg DHPC protocol had significantly increased palatal vault height, anterior upper facial height, anterior maxillary height, overbite, and maxillary inclination than patients treated according to the W-K protocol. There were no differences in posterior upper facial height or in posterior vertical maxillary height between the two groups.

Conclusion: There is increased palatal vault height, anterior upper facial height, anterior maxillary vertical height, and overbite - and therefore increased maxillary inclination at 10 years of age - in patients with complete UCLP who were treated surgically according to the Gothenburg DHPC protocol rather than the W-K protocol. The Gothenburg DHPC protocol can therefore be considered to result in more favorable anterior vertical maxillary growth compared to the W-K protocol.

List of publications

The thesis is based on the following publications:

I Bakri S, Rizell S, Saied S, Lilja J, Mark H. Height of the palatal vault after two different surgical procedures: study of the difference in patients with complete unilateral cleft lip and palate. J Plast Surg Hand Surg. 2012 Sept;46(3-4):155-8.

II Bakri S, Rizell S, Lilja J, Mark H. Vertical maxillary growth after two different surgical protocols in unilateral cleft lip and palate patients. Accepted by Cleft Palate Craniofac J., August 2013.

Abbreviations

CL/P	Cleft lip, with or without cleft palate
CLP	Cleft lip and palate
CP	Cleft palate
UCLP	Unilateral cleft lip and palate
W-K	Wardill-Kilner
DHPC	Delayed hard palate closure

Introduction

Cleft lip with or without cleft palate (CL/P) comprises 65% of all orofacial malformations and is one of the most frequent congenital anomalies (Calzolari et al., 2004). The birth prevalence of CL/P is higher than that of Down's syndrome or of neural tube defects, but is still lower than that of cardiovascular malformations (Bianchi et al., 2000; Cox, 2004).

Children with CL/P have higher morbidity and mortality than non-cleft children, and they need continuous multidisciplinary care throughout their life from birth to adulthood (Bender, 2000; Chuo et al., 2008).

Categories of clefts

Cleft means "split", "separation", or "fissure". Depending on the characteristics of the embryology, anatomy, and physiology of the defect, clefts of the lip and palate can be divided into four general categories: (1) those involving the lip and alveolus, (2) those involving the lip and palate, (3) those in which the palate alone is affected, and (4) those involving congenital insufficiency of the palate. The term "palate" includes both the hard palate and the soft palate (Fig. 1) (Berkowitz, 2013). A cleft can vary from a minor notch in the lip, or a bifid uvula, to complete bilateral cleft lip and palate that extends through the alveolar ridge and involves the whole palate bilaterally (Carroll and Mossey, 2012).

Figure 1. Schematic drawing showing the different types of clefts. A. Cleft lip and alveolus. B. Incomplete unilateral cleft lip and palate. C. Cleft palate. D. Complete unilateral cleft lip and palate. E. Complete bilateral cleft lip and palate.

Epidemiology

It is accepted that CL/P occurs in about 1 per 700 live births, but there are significant variations depending on geographic location, racial and ethnic background, and socioeconomic status (Hagberg et al., 1998; Calzolari et al., 2004; Mossey et al., 2009). The WHO global registry suggests a variation in prevalence at birth of CL/P of 3.4–22.9 per 10,000 births, and an even more pronounced variation for CP, with prevalence of 1.3–25.3 per 10,000 births (Mossey and Castillia, 2003). In addition, CLP is twice as common in males whereas CP is twice as common in females (Mossey and Little, 2002). About 70% of CLP cases are non-syndromic and are not associated with other malformations. However, 30% of the cases are associated with other anomalies, and more than 500 syndromes are associated with CLP (Milerad et al., 1997; Schutte and Murray, 1999; Cobourne, 2004).

Etiology

The etiology of CL/P is still largely unknown. The majority of CL/P cases are believed to have a multifactorial etiology, with several genetic and environmental factors interacting to shift the complex process of morphogenesis toward an abnormality where a cleft can occur (Amaratunga, 1989; Kohli and Kohli, 2012). Recently, a meta-analysis showed that maternal factors most often associated with CL/P are: tobacco, alcohol, obesity, stressful events, low blood zinc levels, and fever during pregnancy. On the other hand, substitution of folic acid during pregnancy has been found to reduce the risk of CL/P (Molina-Solana et al., 2013).

Furthermore, some genomes have been found to have several regions containing loci that may lead to CLP (Brito et al., 2012; Pegelow et al., 2013). Several genes have been suggested as candidates for clefts, e.g. the genes for transforming growth factors alpha and beta, which are expressed during the palatine arch development, and genes express folic acid receptor, that is shown to be linked to CLP pathogenesis (Bianchi et al., 2000).

Overview of embryonic craniofacial development

A precise coordinated cascade of developmental processes involving cell migration, growth, differentiation, and apoptosis results in the development of craniofacial structures; thus, the first term of pregnancy is the most sensitive period for development of craniofacial malformations. At this early stage, interaction with teratogens can also lead to alterations in embryogenesis (Molina-Solana et al., 2013).

Development of the face

The development of the face is complex. Neural crest cells from the neural folds migrate through mesenchymal tissue into the developing craniofacial region by the fourth week of embryonic development. Five facial prominences are formed surrounding the primitive mouth: the frontonasal prominence on the cranial side, a pair of maxillary prominences laterally, and a pair of mandibular prominences caudally, surrounding the primitive oral cavity (Fig. 2 a). The formation of nasal placodes (ectodermal thickenings) then divides the lower portion of the frontonasal prominence into paired medial and lateral nasal processes (Fig. 2 b). By the end of the sixth week, the medial nasal processes merge with the maxillary processes on each side, leading to formation of the upper lip and the primary palate (Fig. 2 c) (Sperber, 2002; Jiang et al., 2006).

Development of the palate

The primary and secondary palatal shelves develop as outgrowths of the medial nasal and maxillary prominences, respectively, and are remodeled and fused to form the intact roof of the oral cavity. During the sixth week of embryogenesis, the paired palatal shelves grow vertically down the sides of the developing tongue (Fig. 2 d). By the seventh week, the palatal shelves rise to a horizontal position above the tongue and fuse in midline. Palatal mesenchyme then differentiates into bony and muscular elements. In addition, the secondary palate fuses with the primary palate and the nasal septum (Fig. 2 e). The fusion process is complete by the tenth week (Fig. 2 f) (Jugessur and Murray, 2005; Mossey et al., 2009). The complexity of the palatogenesis is perhaps reflected by the high incidence of clefts in humans (Bush and Jiang, 2012).

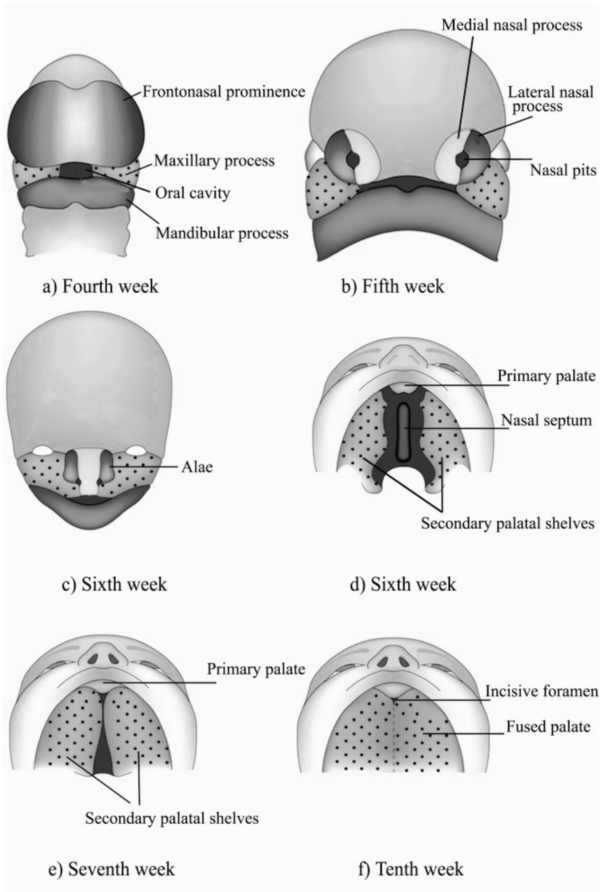

Figure 2. Schematic drawings showing the embryological development of the lip and palate.

Normal maxillary growth

The development of the facial skeleton and the cranium begins a few weeks after conception, by intramembranous ossification. At the end of the sixth week, the maxilla develops by this process in the membranous tissue lateral to the cartilage of the nasal capsule. Between the intramembranous bones, there are sutures of fibrous tissue. These consist of bands of connective tissue joining the periosteal surfaces between the bones. The bone growth can proceed on each side of these sutures (Thilander and Rönning, 1985).

Further increase in the dimensions of bone also occurs by appositional growth on the external periosteal or internal endosteal surfaces. These processes are accompanied by a selective breakdown and resorption of bone tissue on other surfaces. This remodeling process is a combination of deposition and resorption. A continuous remodeling process occurs to develop the shape and proportions of the bone through the growth period. This process causes migration of bone in relation to fixed structures, and this movement process is called drift. The appositional activity usually exceeds the resorption activity during the growth period, and then a balance occurs throughout the rest of life (Enlow, 1982).

The midface generally grows in a downward and forward direction relative to the anterior cranial base (Björk, 1961). This sliding and more active movement of the maxilla complex (pre-maxilla, both maxillary bones and palatal bones) is called displacement or translation. In this process, adjacent bones are pushed away from each other, opening up the space at sutures, allowing different degrees of enlargement of separate bones. The remodeling and displacement occur simultaneously in order to develop the complex anatomy of the craniofacial skeleton (Fig. 3) (Enlow and Bang, 1965; Björk and Skieller, 1977).

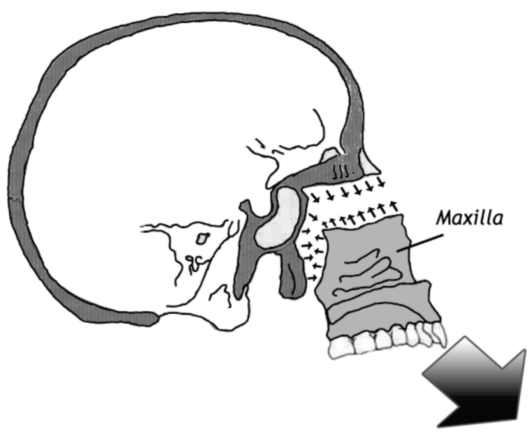

Figure 3. Schematic drawing showing that growth of the surrounding soft tissues displaces the maxilla downward and forward, opening up the space at the superior and posterior sutural attachments, allowing bone deposition on both sides of the sutures.

In the maxillary anteroposterior direction, growth partly takes place in the transverse palatine suture. This sutural activity is supplemented by bone deposition, mainly at the posterior palate and the tuberosities (Melsen and Melsen, 1982; Ross and Johnston, 1972; Enlow, 1982). Regarding the transverse direction, growth in the midpalatal suture occurs and this activity is most pronounced in the posterior part of the suture (Björk and Skieller, 1974). Regarding the vertical direction, an increase in the maxillary height occurs by bone deposition along the alveolar process and roof of the palate. The vertical growth of the alveolar process is rapid during tooth eruption, and exceeds the lowering of the roof of the palate by about threefold, therefore increasing the curvature of the palate. Simultaneously, bone resorption occurs on the nasal floor (Fig. 4) (Björk and Skieller, 1977; Thilander and Rönning, 1985).

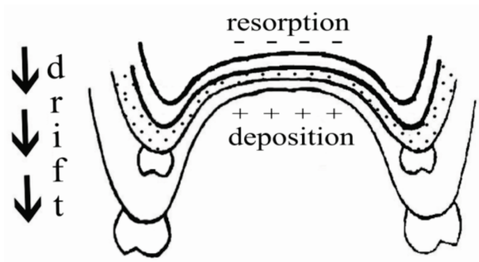

Figure 4. Schematic drawing showing the development of the palate by remodeling.

The surgical protocol in Gothenburg

Patients with CLP can be treated by a variety of surgical procedures that attempt to correct the facial deformity and associated functional impairment, including lip repair, palate repair, bone grafting to the alveolar cleft, procedures to correct speech problems and orthognathic procedures, and also final nose correction (Marsh, 1990). However, there is no generally agreed timetable for the repair of the cleft lip and palate. Early repair of the palate is considered to allow a more normal speech development, and early lip repair may promote better healing of the lip. On the other hand, early repair has been found to have negative effects on facial growth (Robin et al., 2006).

Between 1965 and 1974, surgical management of the CLP patients in Gothenburg was started at the age of 2 months using a cranially based vomer flap. This was followed by closure of the soft and hard palate at 9 months using a Wardill-Kilner (W-K) pushback palatoplasty. The essence of this technique is a V to Y incision and closure of the hard palate (Fig. 5). This pushback technique lengthens the palate and repositions the levator muscles. However, long-term results of patients treated with W-K technique did not meet expectations regarding mid-facial growth and occlusion, and led to high frequency of correcting osteotomies to advance the maxilla. The timing and surgical technique are thought to be the critical factors in the restriction of the anteroposterior and transverse maxillary growth that was seen (Friede and Johanson, 1977).

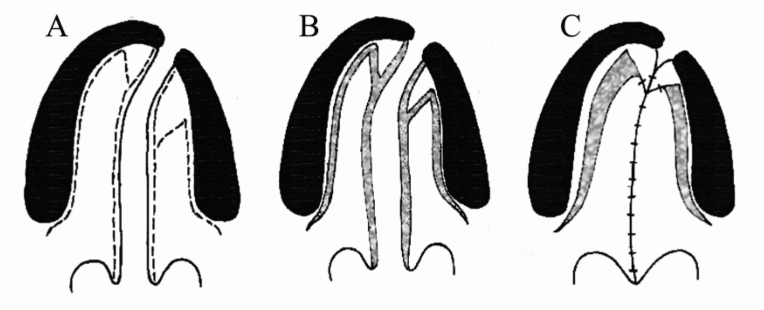

Figure 5. Schematic drawing showing the V to Y incision and closure in the Wardill-Kilner (W-K) pushback technique. A. the margins of the cleft have been marked. B. Medial incision along the junction of oral and nasal mucosa. The lateral incision has been made inside the alveolar ridge from the canine anteriorly to a point just behind the hamulus posteriorly. An oblique incision joins the anterior end of the lateral incision to the cleft margin. C. The tips of the oral mucoperiosteal flaps are sutured, indicating the degree of palatal lengthening.

Thus, in 1975 the Gothenburg delayed hard palate closure (DHPC) was introduced. Closure of the hard palate was then delayed until the stage of mixed dentition at about 8 years of age, leaving a residual cleft in the hard palate open. A posteriorly based vomer flap was also used in order to reduce the amount of scar tissue formation by making the denudation of the bone as minimal as possible (Fig. 6 and 7) (Friede et al., 1980; Lilja et al., 1995). Long-term evaluation of this technique revealed a far more favorable maxillary growth in both anteroposterior and transversal directions, and significantly reduced the need for osteotomies to advance the maxilla (Friede et al., 2012).

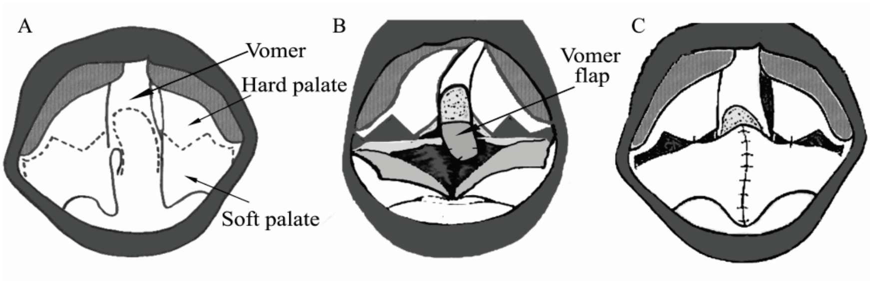

Figure 6. Schematic drawings showing the soft palate closure in the Gothenburg DHPC technique. A. The incisions follow a zigzag line between the soft and hard palate. The posterior vomer flap is also marked. B. Both sides of the soft palate are divided into two layers, the oral mucosa, and the nasal mucosa. Muscle bundles are attached together and are redirected to a transverse direction, and are also attached anteriorly to the vomer flap. C. The muscles and the raw surface of the vomer flap are covered with the oral flaps.

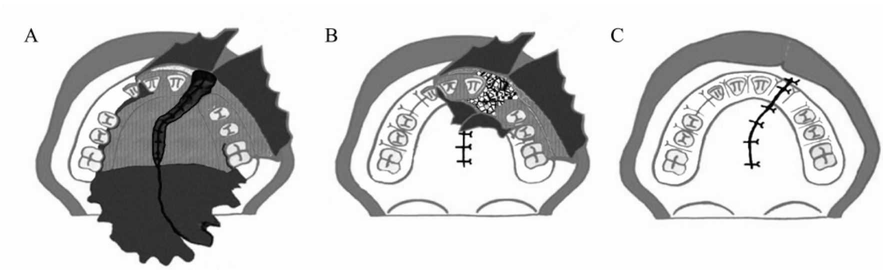

Figure 7. Schematic drawing showing the method for repair of the residual cleft in the hard palate in combination with alveolar bone grafting. A. Incisions are made along the necks of the teeth and along the edges of the residual cleft. The palatal and gingival mucoperiosteal flaps are raised. B. The palatal mucoperiosteal flaps are closed in the palate. Bone grafting is performed at the cleft in the alveolus. C. The grafted bone is covered by the gingival and anterior palatal mucoperiosteal flaps, which are sutured together.

Several studies on maxillary growth, including studies by the Gothenburg team, have mainly investigated the anteroposterior and transversal growth, using both cast models and lateral radiographs. However, there have been few studies on the vertical dimensions of maxillary growth.

Maxillary growth in CLP

Research work investigating the effect of surgery on facial growth in CLP has shown severe maxillary deficiency in all dimensions in patients who have been operated at an early age (Graber, 1949; Ross, 1970; Friede, 1995). In most surgical techniques, mucoperiosteal flaps are raised and displaced medially, and frequently posteriorly. The denuded palatal bone is then covered by scar tissue, which could join the maxilla, the palatal bones, and the pterygoid plates of the sphenoid, a condition termed "maxillary ankylosis" (Ross, 1970). Another effect of the palatal scar tissue is the influence of dentoalveolar structures. The maxillary tooth eruption and vertical development of the alveolar process could be reduced by the scarring. The severity of the maxillary dental arch constriction has been found to be closely related to the distribution of palatal scar tissue (Ishikawa et al., 1998).

The maxillary growth in CLP patients might also be negatively affected by the bony union in the midline of the maxilla seen after some cleft surgeries. This could be from a bone graft (Friede and Johanson, 1974) or from a periosteal envelope promoting bone formation (Prydso et al., 1974; Mølsted et al., 1987).

Follow-up of growth and dental occlusion

Dental casts

Dental casts are a standard procedure in orthodontic records, and they are fundamental for diagnosis, treatment planning, case presentations, and evaluation of treatment progress and results. Caliper and ruler are used in conventional dental cast analysis, which produces accurate, reliable, and reproducible measurements (Santoro et al., 2003). Several digital two- and three-dimensional methods have been introduced during the past decade (Braumann et al., 2001; Fleming et al., 2011). However, manual measurement still appears to be the golden standard (Thilander, 2009).

According to the treatment protocol in Gothenburg cleft team; dental casts are taken at the time of lip and palate repair, as well as at 5, 7, 10, 13, 16 and 19 years, using alginate and non-custom trays.

Lateral cephalometric radiographs

Cephalometric analysis is also a standard method for analysis of craniofacial deformities, for orthodontic treatment planning, and in evaluating growth and treatments (van Vlijmen et al., 2010). Cephalometry continues to be the most versatile technique because of its validity and practicality. In comparison with newer imaging techniques, the cephalogram gives high diagnostic value at a low physiological cost (Melsen and Baumrind, 1995).

In Gothenburg, lateral radiographs are taken using a cephalostat according to a standardized cephalometric guideline, with natural head position and teeth in centric occlusion and the velum at rest. This is done at 5, 7, 10, 13, 16, and 19 years of age.

Aims of the study

Overall aim

The overall aim of this study was to compare vertical maxillary growth in UCLP patients operated with the pushback technique according to Wardill-Kilner (W-K) protocol and in patients operated with the Gothenburg delayed hard palate closure (DHPC) protocol.

Specific aims

- To compare the palatal vault height after W-K and DHPC surgical protocols.
- To study the overbite, maxillary height, upper anterior and posterior facial height, and maxillary inclination in patients treated according to the two different surgical protocols.

Patients and methods

Patients

The study was conducted on 176 consecutive caucasian patients born 1964 to 1995 with UCLP. The patients were operated at the Department of Plastic Surgery, Sahlgrenska University Hospital, Gothenburg, Sweden.

Exclusion criteria were: secondary palatal surgical procedure (pharyngeal flap or pharyngoplasty), syndromic cleft, craniofacial or systemic malformation, or presence of Simonart's band (a band of soft tissue partially connecting cleft sides) of more than 0.5 mm. Fistula closure was not regarded as an exclusion criterion.

The patients were divided into two groups according to the surgical protocol used.

The Wardill Kilner (W-K) group : The surgical protocol can be summarized in following steps: (a) lip adhesion and closure of the nasal floor and the anterior part of the hard palate using a single-layer, cranially based vomer flap at 2 months, (b) closure of both hard palate and soft palate using a pushback method at 9 months, (c) final lip-nose repair at 18 months of age, and (d) bone grafting to the alveolar process at about 8–10 years (Friede and Johanson, 1977).

The Gothenburg delayed hard palate closure (DHPC) group: The surgical protocol can be summarized in the following steps: (a) lip adhesion at 2 months, (b) soft palate closure including posteriorly based vomer flap at 7 months, (c) final lip-nose surgery at 18–20 months, (d) closure of the residual cleft in the hard palate with bone grafting to the alveolar process at about 8–10 years (Lilja et al., 1995).

Lip-nose surgeries and bone grafting were performed using the same techniques and timing in both protocols. Preoperative orthodontic treatment (mainly maxillary expansion) was given to 78% of the patients who were treated according to W-K protocol and to 25% of those treated according to the DHPC protocol (Friede et al., 1987).

Cast model analysis

Cast models were obtained at 10 years of age from 176 consecutive caucasian patients born with UCLP. The W-K group consisted of 60 patients born between 1965 and 1974 (36 males and 24 females; 35 left side and 25 right side). The Gothenburg DHPC group consisted of 116 patients born between 1975 and 1995 (81 males and 35 females; 69 left side and 47 right side).

The palatal vault height was measured at four locations (A–D). At point A: the perpendicular distance from the midpoint of the line connecting the highest points of the mesolingual cusps of the first maxillary molars to the palate. At point B: the perpendicular distance from a point 10 mm anterior to point A to the palate. At point C: the perpendicular distance from a point 7 mm left of point A at the same line to the palate. At point D: the perpendicular distance from a point 7 mm right of point A on the same line to the palate (Fig. 8).

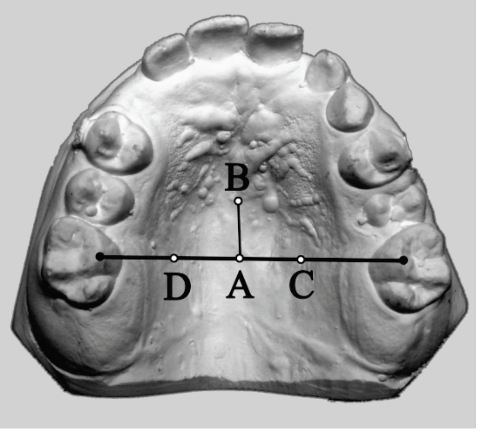

Figure 8. Picture showing the highest points of the mesolingual cusps of the first molars, and the four points at which palatal vault height was measured.

The measurements were performed through holes in a plastic sheet, using a digital calliper. The end of the digital caliper was pressed against the palatal contour. Wax blocks were used to fix the models and the covering plastic sheet in order to ensure good stability. The digital caliper was adjusted to subtract the thickness of the plastic sheet (2.2 mm) in all measurements. The same digital caliper and same plastic sheet were used for all measurements (Fig. 9).

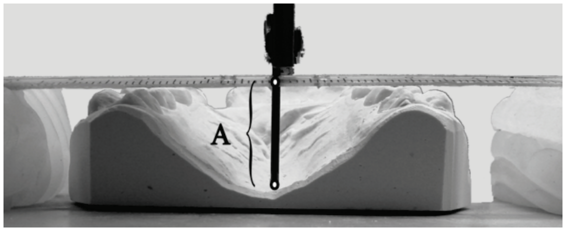

Figure 9. Picture showing measurement of the palatal vault height at point A.

Cephalometric analysis

Lateral cephalometric radiographs were taken at 10 years of age from 92 consecutive caucasian patients born with UCLP. The W-K group consisted of 46 patients born between 1965 and 1974 (27 males and 19 females; 27 left side and 19 right side). The Gothenburg DHPC group consisted of 46 patients born between 1982 and 1989 (34 males and 12 females; 24 left side and 22 right side).

The cephalograms were taken in maximal intercuspal position and with the head fixed in a cephalostat. The enlargement factor was adjusted for measurement of linear distances. The measurements were performed using a computerized cephalometric software program (Viewbox®; dHAL Software, Athens, Greece). The landmarks and variables measured in this study are shown in (Fig. 10) (Björk, 1947; Subtelny, 1957; Thilander et al., 2005). Two authors (SB and SR) localized each landmark by agreement.

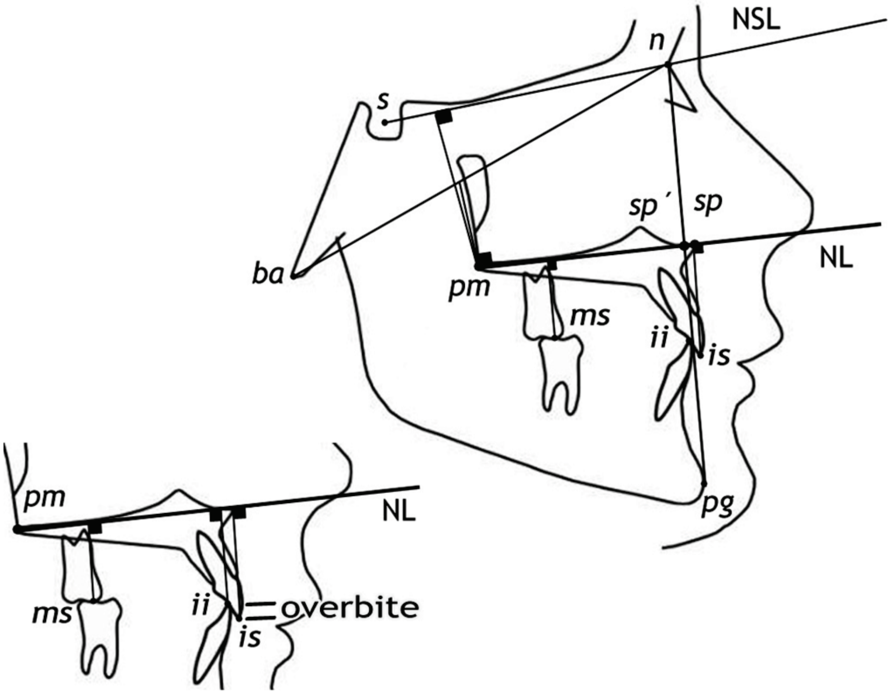

Figure 10. Schematic drawing showing reference landmarks and lines: n (nasion), the anterior limit of sutura nasofrontalis; s (sella), the center of sella tursica; ba (basion), the anterior-most point of foramen magnum; pm (pterygomaxillare), the point of intersection of palatum durum, palatum molle, and fossa pterygopalatina; sp (the spinal point), the apex of spina nasalis anterior; pg (pogonion), the most prominent point of the chin; sp´, the intersection between the nasal line (NL) and the n–pg line; is, the edge of the upper central incisor; ii, the edge of the lower central incisor; ms, the edge of the medial cusp of upper first permanent molar; NSL, nasion-sella line; NL, baseline of the maxilla; NBa line, cranial base line.

The following measurements were obtained: n-sp´, the distance between n and sp´; NL-is, the perpendicular distance from NL to is; overbite, the vertical difference between ii and is perpendicular to NL; NL-ms, the linear perpendicular distance from NL to ms; s-pm, the distance between s and pm; NSL-pm, the perpendicular distance from NSL to pm; NBa-pm, the distance from pm perpendicular on NL to NBa; NSL/NL, the angle between NL and NSL.

Precision of measurements

The precision of the registrations was tested by repeated measurements of randomly selected cases (30 cases in the cast model analysis and 20 cases in the cephalometric analysis) at intervals of more than 1 month. The error of the method was calculated according to Dahlberg (1940) and did not exceed 0.5 mm in either study.

Statistical analysis

Statistical analysis of the data was performed with two-sample Student's *t*-test to test for differences between the two surgical protocols (using IBM SPSS Advanced Statistics 19).

Ethical approval

Data collection and analysis were carried out according to ethical principles for medical research involving human subjects. Approval was obtained from the local research ethics committee (Regionala etikprövningsnämnden i Göteborg, Dnr: 1020-12).

Results

Study I showed that the palatal vault height was significantly greater at the four points measured (A–D) in the Gothenburg DHPC group than in the W-K group (Table 1).

Table 1. Results from cast model comparison of the palatal vault height at points A–D in the W-K protocol and the Gothenburg DHPC protocol

	W-K (60 patients)		DHPC (116 patients)		P-value
Measuring points	Mean palatal vault height (mm)	SD (mm)	Mean palatal vault height (mm)	SD (mm)	
A	12.45	1.66	15.63	1.81	*< 0.001*
B	11.57	2.27	13.47	2.31	*< 0.001*
C	8.71	1.73	11.21	2.16	*< 0.001*
D	9.71	2.27	11.63	2.14	*< 0.001*

Study II showed that the anterior upper facial height (*n-sp'*), anterior maxillary height (NL-*is*), overbite, and maxillary inclination (NSL/NL) were statistically significantly greater in DHPC group than in the W-K group (Table 2).

For the remaining cephalometric variables, no statistically significant differences were found. There were no statistically significant differences in cleft side or gender between the groups.

Table 2. Results from cephalometric comparison of the W-K and the Gothenburg DHPC protocols

	W-K (46 patients)		DHPC (46 patients)		P-value
Variable	Mean (mm)	SD (mm)	Mean (mm)	SD (mm)	
n-sp´	43.98	2.99	45.41	2.35	*< 0.05*
NL-*is*	24.89	2.64	26.33	2.55	*< 0.01*
Overbite	1.74 ¥	1.93	3.18	2.07	*< 0.01*
NL-*ms*	19.21 ¥	2.53	20.00	1.67	*n.s.*
s-pm	38.06	2.77	38.98	2.64	*n.s.*
NSL-*pm*	36.03	2.67	36.29	2.52	*n.s.*
NBa-*pm*	18.87	2.39	18.88	2.18	*n.s.*
NSL/ NL (°)	9.07	3.55	10.67	3.29	*< 0.05*

The number of registrations for each surgical group was 46, except for two variables in the W-K group (¥), where 44 registrations were obtained because of unclear landmarks in the lateral radiograph. *n.s.* = not significant.

Discussion

The present work revealed significantly greater values of palatal vault height, anterior upper facial height, anterior maxillary height, overbite, and maxillary inclination in patients operated according to the Gothenburg delayed hard palate closure protocol than in patients operated according to the Wardill-Kilner protocol. However, while the values for palatal vault height and anterior upper facial height approached non-cleft reference values in the DHPC group more than in the W-K group, maxillary inclination showed the opposite trend (Thilander et al., 2005; Thilander, 2009).

The palatal vault was significantly higher in the Gothenburg DHPC protocol group than in the W-K protocol group in all measuring locations investigated. This result is in agreement with previous findings showing that the DHPC protocol results in favorable anteroposterior and transversal maxillary growth (Friede and Johanson, 1977; Friede et al., 1980; Friede et al., 2012). After the soft palate closure in the DHPC technique, a narrowing of the remaining cleft in the hard palate occurs (Owman-Moll et al., 1998). The residual cleft in the hard palate is therefore easy to repair with minimal scar tissue formation, resulting in favorable maxillary growth in all dimensions (Friede and Enemark, 2001).

The lower palatal vault seen in W-K group can be explained by the excess scar tissue in the palate which inhibits the vertical eruption of the teeth by anchoring the periodontal fibers that are attached to the teeth (Ross, 1970, 1987). Still, the palatal vault height obtained by the DHPC protocol was far from that in a non-cleft reference group of same age and ethnicity (Thilander, 2009). In order to approach normal palatal vault height, it is therefore important to develop the surgical techniques of CLP closure further.

Reports have indicated that operated CLP patients have a narrower, shorter, and shallower hard palate than non-cleft controls (Okazaki et al., 1991; Smahel et al., 2004). Kharbanda and co-workers (2002) have shown that surgical protocols that give higher palatal vault are associated with better overall growth of the maxilla. The palatal vault height could therefore be considered as an indicator of surgical outcome. The palatal vault height in our W-K group was similar to the best values found in the last-mentioned study, so the values found in the Gothenburg DHPC group were even better.

The palatal vault height appears to be of importance for speech, and is thought to be correlated to the quality of speech (Berkowitz, 2013). It has been shown that low palatal vault height is associated with increased speech problems (Okazaki et al., 1991; Laitinen et al., 1998; Grunwell et al., 2000; Kharbanda et al., 2002). Furthermore, a change in the dimensions of the palate has been proposed to affect other functions such as swallowing, breathing, mastication, and Eustachian tube function (Kimes et al., 1991; Smahel et al., 2003, 2004).

The reduced palatal vault height is not associated with adaptive reduction in tongue size. This means that the relative tongue size in CLP patients is higher than in non-cleft controls (Kimes et al., 1991). Apart from this, the palatal vault height has been found to be the most important factor affecting the position of the tongue in the oral cavity (Bourdiol et al., 2010). An additional reason for speech problems in CLP patients is that the tongue is thought to press the mandible inferiorly, impairing the vertical jaw relations (Berkowitz, 2013).

Our work shows that the DHPC protocol gives significantly greater upper anterior facial height and anterior maxillary height than the W-K protocol. Still, both the DHPC and W-K groups showed lower figures than normal (Thilander et al., 2005). These results are in agreement with previous studies showing that repair of the CLP by almost any surgical technique results in restriction of the maxillary growth (Ross, 1970).

In the present study, a reduced overbite was found in the W-K group compared to the DHPC group. This result is also in accordance with previous findings showing that restriction of the upper anterior facial height and anterior maxillary height affect overbite (Ross, 1987; Lisson et al., 2005). Compared to the W-K protocol, the Gothenburg DHPC protocol gives significantly increased overbite. This finding may be one of the reasons why 78% of the patients in W-K group received orthodontic treatment as compared to 25% of the patients in DHPC group (Friede et al., 1987). This finding could also be a contributory factor to the fact that the DHPC protocol has been shown to result in very good dental arch relationship using the GOSLON Yardstick (Lilja et al., 2006).

Reduced upper facial vertical height has been also suggested to be of importance for the facial esthetics (Ross, 1987). For example, poor overall maxillary growth in operated UCLP patients has been shown to be correlated to least favorable nasolabial appearance (Asher-McDade et al., 1992).

We measured three variables to quantify the vertical dimensions in the posterior upper facial region *s-pm*, NL-*pm*, and NBa-*pm*, besides the posterior maxillary height NL-*ms*. No statistically significant differences in these dimensions were found between the two protocols investigated. The posterior facial height is, however, markedly less with both protocols than non-cleft values: the mean for NBa-*pm* was 18.88 mm in the DHPC group and 18.87 mm in the W-K group, as compared to 24.2 mm in non-clefts (Subtelny, 1957). This is in agreement with the findings of others, of reduced upper posterior facial height in operated CLP patients (Wermker et al., 2012). Comparisons between studies regarding upper posterior facial height in CLP are, however, difficult to make due to considerable variation in the cephalometric parameters used.

The restriction of the upper posterior facial height in the Gothenburg DHPC and the W-K protocols is not in agreement with previous studies indicating better facial growth with the Gothenburg DHPC than with the W-K protocols. However, it is in agreement with studies of other DHPC techniques, showing that upper posterior vertical growth is restricted in all early soft palate repair techniques (Ross, 1987). It is not entirely clear whether the reduced posterior maxillary development is due to the timing of the soft palate closure or to the particular surgical technique used (Swennen et al., 2002). We have suggested that the posteriorly based vomer flap used in the Gothenburg DHPC technique could be of significance for this finding, but more investigations are needed to clarify this matter.

In the present study, the Gothenburg DHPC protocol resulted in a greater maxillary inclination than the W-K protocol. The increased maxillary inclination result from the difference in restriction of the anterior and posterior vertical maxillary dimensions that lead to a change in the maxillary inclination angle. There have been many studies showing the same results in operated CLP patients (Paulin and Thilander, 1991; Ozturk and Cura, 1996; Swennen et al., 2002; Lisson et al., 2005; Fudalej et al., 2013).

Few studies have indicated that maxillary inclination is also of importance for speech. Maxillary inclination has, for example, been shown to be correlated to the level of velopharyngeal closure at the posterior pharyngeal wall. Increased maxillary inclination in operated CLP patients also results in velopharyngeal closure at a higher level and better speech (Satoh et al., 1999; Satoh et al., 2005). An increased maxillary inclination has also been found in operated CLP patients with normal speech (Semb and Shaw, 1990), and an increase in maxillary inclination is associated with less nasality (Stellzig-Eisenhauer, 2001).

We have suggested that the posteriorly based vomer flap reduces posterior facial vertical growth and therefore increase the maxillary inclination. The vomer flap is therefore important for adequate velopharyngeal competence

(Friede et al., 2012). However, further work is needed to investigate the importance of the posteriorly based vomer flap for speech.

Long-term speech outcome in patients who have been treated with the Gothenburg DHPC protocol has been found to be good, even before the hard palate repair (Lohmander et al., 2012). Better speech results have been found to result in surgical techniques leading to less growth restriction (Ito et al., 2006). Thus, the more normalized facial growth shown with the DHPC protocol would have contributed in a positive way to the findings of Lohmander and co-workers. Still, there is no clear evidence to support these conclusions.

In the work described here, we concentrated on investigating the vertical growth of the maxilla, which has not often been studied separately. The strength of this work was that the two groups under study shared the same ethnicity and cleft type, and they were treated by the same cleft team using the same surgical steps and techniques. The only difference between the two protocols was the palatal surgery technique. In some studies, linear measurements of cephalometry have been adjusted to an internal reference line to make different age groups more comparable (Ross, 1987, 1995; Swennen et al., 2004; Mishima et al., 2008; Wermker et al., 2012). In the present work, evaluation of a reasonable number of patients of the same age allowed us to instead compare real linear distances of all variables, therefore increasing the validity of the results.

However, one limitation of this work was that the patients were assessed before puberty, and it is feasible that growth restrictions may be more pronounced after the pubertal growth spurt. Further research is needed in order to investigate vertical maxillary growth in adult CLP patients.

Conclusions

There is greater palatal vault height, anterior upper facial height, anterior maxillary vertical height, and overbite - and therefore greater maxillary inclination at 10 years of age - in patients with complete UCLP who were surgically treated according to the Gothenburg DHPC protocol than in those treated according to the W-K protocol. The Gothenburg DHPC protocol can therefore be considered to result in more favorable anterior vertical maxillary growth than that obtained with the W-K protocol.

Clinical implications and future research

The vertical maxillary growth for either the W-K or the DHPC protocols has not been fully investigated. This work contributes new, important knowledge regarding the effect of these protocols that have been used in Gothenburg, on the vertical maxillary growth. The more normalized vertical maxillary growth found in the Gothenburg DHPC protocol could be a contributory factor to the good speech results and the improved facial esthetics that have been shown previously using this surgical protocol.

However, further studies are needed to improve the surgical protocols, aiming at normalizing the maxillary growth. In order to fully understand the effects of the present surgical protocols on vertical maxillary growth, the same patients should be investigated after completing their facial growth at 19 years of age. Future research should also investigate how maxillary growth is related to the velopharyngeal functions and to speech.

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ORIGINAL ARTICLE

Height of the palatal vault after two different surgical procedures: Study of the difference in patients with complete unilateral cleft lip and palate

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Abstract
The present study compared the height of the palatal vault in dental casts from 320 10-year-old children with unilateral cleft lip and palate (UCLP) operated on with the push-back technique according to Wardill-Kilner (W-K) with patients operated on with delayed hard palate closure (DHPC). The palatal height in patients operated on with the DHPC technique was found to be significantly higher than in patients operated on with the W-K technique. This coincides with better maxillary growth and better speech in the DHPC group.

Key Words: Delayed hard palate closure, UCLP, palatal vault height

Introduction

All cleft teams who care for patients with cleft lip and palate (CLP) have the obligation to follow-up their treatment results to find out whether the anticipated goals have been reached; if that is not the case, then action should be taken to improve matters [1–3].

The evolution of the surgical treatment used at the Cleft Palate Centre of Gothenburg, Sweden has been based on such follow-up evaluations [4].

Between 1965–1974 surgical management of the cleft palate included soft tissue closure using a cranially-based vomer flap combined with a Wardill-Kilner (W-K) push-back palatoplasty. Long-term results of patients treated with W-K technique did not meet the expectations regarding occlusion and mid-facial growth. The timing and technique used for closure of the hard palate were thought to be the critical factors in restriction of maxillary growth. This could particularly be true when extensive mucoperiosteal flaps were used and shifted medially to cover the palatal cleft. Areas of leaving denuded bone in the hard palate are then left for secondary healing resulting in growth restricting scars. These palatal scars have a negative effect on the maxillary development [5]. Follow-up on speech in CLP patients treated with W-K demonstrated a high incidence of VPI showing the limitations of the W-K technique also on speech outcome [6].

In 1975 closure of the hard palate (DHPC) was therefore delayed until the stage of mixed dentition. Leaving a residual cleft in the hard palate open revealed fear of less favourable speech development, however instead a speech improvement in relation to the previous protocol was experienced, especially after closure of the residual cleft [7–10].

In previous studies on the height of the palatal vault, a common finding is that the height is reduced in all patients with cleft lip and palate compared with normal controls [11–13]. A comparison of the height of the palatal vault in patients with UCLP from six different European cleft centres revealed significant differences in the height of the palatal vault from the different surgical protocols for palatal closure used in these centres. It was found that also patients with good maxillofacial growth had a higher palatal vault [14]. In our series of patients with complete UCLP, those operated on with the W-K technique had less favourable maxillary growth compared with those operated on with the DHPC technique, but the height of the palatal vault was not compared.

The aim of the present study was therefore to measure and compare the height of the palatal vault in patients with UCLP operated on with the push-back technique according to Wardill-Kilner (W-K) with patients operated on with delayed hard palate closure (DHPC).

Patients and methods

The study was conducted on dental casts from 320 consecutive Caucasian patients with UCLP born from 1965–1995, operated on at the Department of Plastic Surgery, Sahlgrenska University Hospital, Gothenburg, Sweden.

Inclusion criteria for the patients were to be Caucasian and have a unilateral complete cleft lip and palate, operated on by the cleft team in Gothenburg, Sweden, with available patient records and dental cast model.

Exclusion criteria were presence of syndromic clefts, craniofacial or systemic anomaly, non-eruption of first molars or low-quality cast models and presence of a soft tissue bridge of the lip of more than 0.5 mm (Simonart's band).

A total of 176 patients fulfilled the inclusion criteria. The patients were divided into two groups according to the surgical technique.

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- Group A: 60 patients with UCLP were operated on with the W-K (36 boys, 24 girls and 35 left side, 25 right side). The patients were born between 1965–1974. The surgical procedure used can be summarised in the following steps: lip adhesion and closure of the nasal floor and the anterior part of the hard palate by using a cranially-based vomer flap at 2 months of age; final lip closure at 8 months of age; closure of both the hard palate and soft palate using a push back method at 16 months of age; and bone grafting to the alveolar process at ~ 8–10 years of age [5].
- Group B: 116 patients with UCLP were operated on according to Gothenburg protocol for DHPC (81 boys, 35 girls and 69 left side, 47 right side). The patients were born between 1975–1995. The surgical procedure used can be summarised in the following steps: lip adhesion at 1–3 months of age; soft palate closure at 6–8 months of age; final lip–nose operation at 18–20 months of age; and closure of the residual cleft in the hard palate with bone grafting to the alveolar process at ~ 8–10 years of age [7].

Dental casts and analysis

The height of the palatal vault was measured at 10-year cast models at four points (A–D) (Figure 1). Point A = the perpendicular distance from the mid-point of the line connecting the highest points of the mesolingual cusps of the first maxillary molars to the palate. Point B = the perpendicular distance from a point 10 mm anterior to point A to the palate. Point C = the perpendicular distance from a point 0.7 mm left to point A at the same line to the palate. Point D = the perpendicular distance from a point 0.7 mm right to point A at the same line to the palate.

The measurements were done through holes in a plastic sheet, with the use of a digital caliper; the end of the digital caliper was pressed to the palatal contour. Wax blocks to fix the models and the covering plastic sheet were used in order to secure good stability. The digital caliper was adjusted to subtract the thickness of the plastic sheet (2.2 mm) through all measurements (Figure 2). The same digital caliper and same plastic sheet were used through all measurements [15,16].

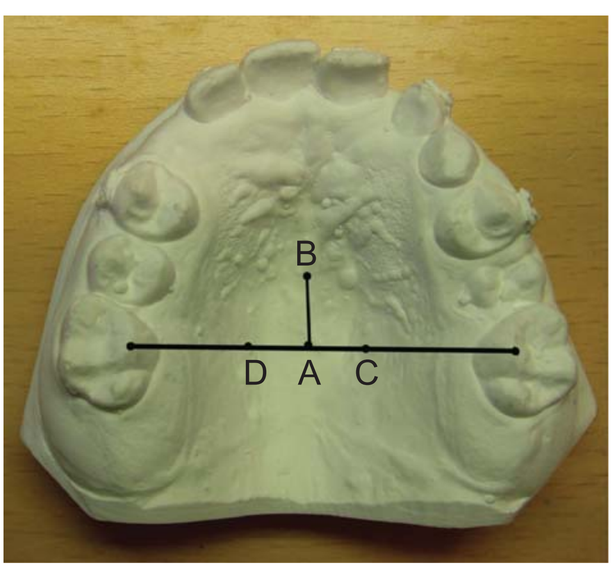

Figure 1. Showing markings of the highest points of the mesolingual cusps of the first molars, then the four points where we measured the height of the palatal vault.

Precision of measurements

The precision of the registrations was tested by double measurements of 30 randomly selected cases. The error of the method was calculated according to the Dahlberg's formula $SE = \pm\sqrt{\Sigma d^2 / 2n}$, where d is the difference between the two measurements and n is the number of measurements. The accidental error varies from 0.34 mm at point A, 0.41 mm at points B and 0.37 mm at point C to 0.39 mm at point D, indicating a high degree of precision and accuracy [16,17].

Statistical analysis

Statistical analysis of the data was undertaken using a Student's *t*-test (using IBM SPSS Advanced Statistics 19).

Results

The measurements showed that in all four locations (A–D) the height of the palatal vault was significantly higher for group B operated on with the DHPC technique compared with group A operated on with the W-K technique (Table I).

There was no significant difference in the height of the palatal vault between the right side UCLP and the left side UCLP and male and female patients operated on by any of the techniques. There was no significant difference in height between the sides of the palatal vault (points C and D) in any of the techniques.

Discussion

The present study concentrated on the height of the palatal vault in patients with complete unilateral cleft lip and palate. A comparison between patients operated on according to two different protocols, the W-K technique and the DHPC technique developed in Gothenburg, was performed. It was found that the palatal vault height was significantly higher in the patients operated on with the DHPC procedure compared with the W-K technique.

Casts were available at different ages (1, 3, 5, 7, 10, and 16 years). The 10-year cast models were chosen. This is the first available dental cast after completion of closure of the hard palate with the DHPC technique, it is also before the adolescence growth spurt in facial bones, and the differences at 10 years of age will become more significant with increasing age as the height of the palatal vault is related to overall maxillary growth.

The first molar was used as a landmark since it is the most posterior and most reliable at this age. In patients with cleft palate the palate is overall shallower and the anteroposterior location of the deepest point in the palate is more posterior than in non-clefts [12,18]. The technique using a manual calliper for measuring dentoalveolar development has earlier been used with high reliability [15,16].

The first time the question was raised about the height of the palatal vault as an indication of the surgical outcome was in 2002, when Kharbanda et al. [14] compared the results of the height of the palatal vault in six different European centres. It was found that the height of the palatal vault correlated significantly to the type of protocol used for cleft palate treatment and they found that the height of the palatal vault was also related to

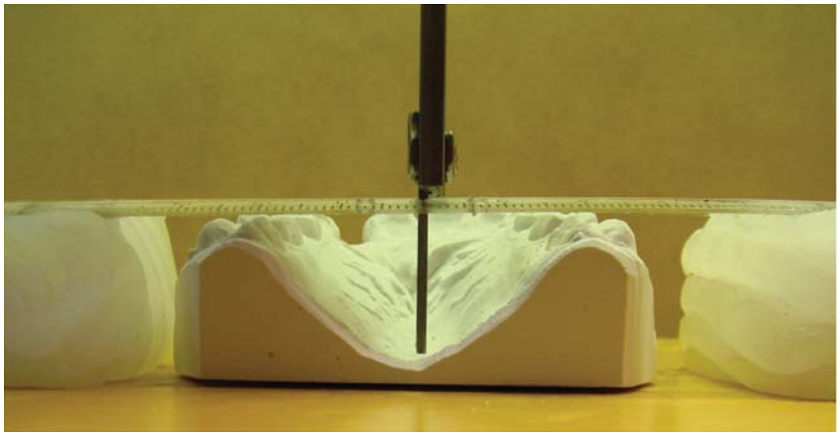

Figure 2. Showing measuring the height of the palatal vault at point A, and the stable plastic sheet with the use of the fixation wax block.

the overall growth of the maxilla. The same material also showed best speech results in centres that had the best height of the palatal vault [19].

In comparison with the previous study of Kharbanda et al. [14], the height of the palatal vault in our W-K group (12.45 ± 0.21 mm) was found to be similar to the best two centres (11.89 mm and 11.65 mm, respectively). The similarity may be explained by the superiorly-based vomer flap performed at the age of 2–3 months that was used in all centres.

However, with the HPC technique the height of the palatal vault was significantly higher (15.63 ± 0.17 mm) compared with these results, which is probably because the posteriorly-based vomer flap is reaching the junction between vomer and the cranial base, lifting the soft palate upwards without affecting the vomero-premaxillary suture anteriorly.

In order to estimate if our DHCP technique gives a normal palatal vault, the results were compared with normal individuals of the same age and ethnic group. However, at measure point A in the centre of the palate a much higher palatal vault was found in the normal group, 36.3 mm, compared with 15.36 ± 0.17 mm in DHPC [16]. This represents the challenge that cleft surgeons should face and raises the need to develop techniques resulting in normal maxillary dimensions.

Also more work should be focused on the effect of these dimensions on the speech. A significantly better height of the palatal vault at 10 years of age is probably of importance for speech production [14,16].

Previous studies have shown that children with UCLP at the age of 4–5 years have a narrower, shorter and shallower hard palate compared with normal controls. The speech quality was also correlated to the height of the palatal vault, suggesting the possible importance of the height of the palatal vault to the speech production as a separate factor other than the velopharyngeal function. It was also more evident in children with palatenised articulation [20].

There is no evidence that a smaller palatal volume is associated with adaptive reduction of the size of the tongue, instead the decrease in the height of the palatal vault suggests giving a considerable shortage of space for the tongue. The mandible may be pressed inferiorly into a posterior rotation leading to an open bite with impaired vertical intermaxillary relation and this may affect phonation by abnormal articulation and impaired formation of consonants. One must also consider that changes in the size and shape of the palate might affect other functions such as swallowing, mode of breathing, mastication, and Eustachian tube function [11,12,21].

The patients operated on with the Gothenburg DPHC protocol have been prospectively followed regarding speech development. The results show good speech without glottal articulation and very few patients with velopharyngeal insufficiency. Comparison of speech results remains to be done in the future. However, the number of secondary speech improving operations is significantly lower in the DPHC group (11%) [10,22]. From the present results it could be proposed that higher palatal vault in the patients operated on with the DHPC technique may contribute to more favourable speech in these patients.

In conclusion, the palatal height in patients operated on with the Gothenburg DHPC technique is significantly higher than in patients operated on with the Wardill-Kilner technique. This coincides with better maxillary growth and better speech in the DHPC group.

Table I. Increased height of the palatal vault with the DHPC technique compared with the W-K technique in each point measured as mean and SEM.

	W-K (60 patients)		DHPC (116 patients)		
	Mean (mm)	SEM (mm)	Mean (mm)	SEM (mm)	*p*-value
A	12.45	0.21	15.63	0.17	0.0001
B	11.57	0.29	13.47	0.21	0.0001
C	8.71	0.22	11.21	0.2	0.0001
D	9.71	0.29	11.63	0.2	0.0001

Declaration of interest: The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the paper.

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II

Vertical maxillary growth after two different surgical protocols in unilateral cleft lip and palate patients

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Disclosure:
There were no financial conflicts of interest on the part of any authors.

Objective: The aim of the present study was to compare vertical maxillofacial growth in patients born with unilateral cleft lip and palate (UCLP) who were treated using two different surgical protocols.

Design: A retrospective cohort study.

Subjects: We studied ninety-two patients with complete UCLP (61 males and 31 females) treated at Sahlgrenska University Hospital in Gothenburg, Sweden: 46 consecutive patients born 1965 to 1974 who were operated according to the Wardill-Kilner (W-K) protocol and 46 consecutive patients born 1982 to 1989 who were operated according to the Gothenburg delayed hard palate closure (DHPC) protocol.

Methods: we analyzed lateral cephalograms obtained at 10 years of age.

Results: Patients treated according to the Gothenburg DHPC protocol had significantly greater anterior upper facial height, anterior maxillary height, over bite, and inclination of the maxilla than those treated with the W-K protocol. Both techniques led to similar posterior upper facial height.

Conclusion: The Gothenburg DHPC protocol in patients with complete UCLP results in more normal anterior maxillary vertical growth and overbite and therefore increased maxillary inclination at 10 years of age.

Key words: cleft lip and palate, delayed hard palate repair, vertical maxillary growth, Wardill-Kilner.

Introduction

Cleft lip and palate (CLP) is one of the most common congenital anomalies. Treatment protocols for management of children with CLP differ markedly between cleft teams Mossey et al. (2009). Between 1965 and 1974, the protocol used by the cleft team in Gothenburg, Sweden, included hard palate closure using a cranially based vomer flap followed by a Wardill-Kilner (W-K) push-back palatoplasty. This technique led to poor midfacial growth and occlusion (Friede and Johanson, 1977). Based on follow-up studies, the protocol was changed in 1975 by introducing a delayed hard palate closure technique (DHPC). This method included closure of the soft palate with a posteriorly based vomer flap in the first year of life. Closure of the hard palate was delayed until the stage of mixed dentition. This DHPC technique showed significantly better long-term midfacial growth and occlusion (Friede, 1998; Friede et al., 1980; Friede et al., 2012), with favorable speech development (Lohmander-Agerskov, 1998; Lohmander et al., 2012).

Graber (1949) pioneered the research on factors influencing maxillary development in CLP patients, and stated that cleft surgery had a detrimental effect on maxillary growth (Graber, 1949). Restricted maxillary growth has been a constant finding in studies evaluating CLP patients treated according to different surgical protocols (Khanna et al., 2012; Ross, 1987; Semb and Shaw, 1998). Most previous work has been focused on craniofacial growth in the sagittal and transverse dimensions (Lisson et al., 1999; Mars et al., 1992; Molsted et al., 1992).

The vertical maxillary growth restriction has been shown to be a common finding in operated cleft patients, and it has also been shown to vary between different surgical techniques and their timing (Ross, 1987). Moreover, the growth restriction has been found to differ between anterior or posterior maxillary dimensions, and to change the maxillary inclination angle (Swennen et al., 2002). Reduced anterior vertical maxillary growth can be observed clinically as reduced overbite (Lisson et al., 2005; Ross, 1987), and reduced posterior vertical maxillary growth has been suggested to affect speech (Stellzig-Eisenhauer, 2001).

The effect of surgery on vertical maxillary growth in CLP is less well understood, and further investigation of the effect of different surgical protocols on these dimensions is still needed. The aim of the present study was to compare how vertical maxillary growth is affected by W-K with cranially based vomer flap and by the Gothenburg DHPC with posteriorly based vomer flap.

Patients and method:

The study was conducted on lateral cephalometric radiographs taken at 10 years of age from 92 consecutive Caucasian patients born with UCLP. The patients were operated at the Department of Plastic Surgery, Sahlgrenska University Hospital, Gothenburg, Sweden.

Exclusion criteria were: secondary palatal surgical procedure (as pharyngeal flap or pharyngeoplasty), syndromic clefts, craniofacial or systemic anomalies, and presence of Simonart's band of more than 0.5 mm. Fistula closure was not regarded as an exclusion criterion.

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The patients were divided into two groups according to the surgical protocol used.

The W-K group consisted of 46 consecutive patients born between 1965 and 1974 (27 males and 19 females; 27 left side and 19 right side).

The surgical protocol can be summarized as the following steps: (a) lip adhesion and closure of the nasal floor and the anterior part of the hard palate using a single layer cranially based vomer flap at 2 months, (b) closure of both hard palate and soft palate using a push-back method at 9 months, (c) final nose/lip repair at 18 months of age, and (d) bone grafting to the alveolar process at about 8-10 years (Friede and Johanson, 1977) (Table 1).

The DHPC group consisted of 46 consecutive patients born between 1982 and 1989 (34 males and 12 females; 24 left side and 22 right side).

The surgical protocol can be summarized as the following steps: (a) lip adhesion at 2 months, (b) soft palate closure including posteriorly based vomer flap at 7 months, (c) final lip-nose surgery at 18-20 months, (d) closure of the residual cleft in the hard palate with bone grafting to the alveolar process at about 8-10 years (Lilja et al., 1995) (Table 1).

Lip nose-nose surgery as well as bone grafting had same techniques and timing in both investigated protocols. Orthodontic treatment (mainly maxillary expansion) was received in 78% of Patients treated according to W-K protocol versus 25% in DHPC protocol (Friede et al., 1987).

Table 1. Timing of surgical procedures in the W-K and the Gothenburg DHPC surgical protocols.

Procedure	Time W-K	DHPC
Lip adhesion	2m	2m
Lip nose repair	18m	18m
Soft palate repair	9m	7m
Hard palate repair	9m†	8-10y
Bone grafting	8-10y	8-10y

m: month, y: year, (†) Vomer flap were used to close the anterior part of the hard palate at 2 month. The rest of the hard palate and the soft palate were closed at 9 month.

Lateral X-ray cephalograms and analysis

The cephalograms were taken at the ten-year follow-up in maximal intercuspal position and with the head fixed in a cephalostat. The enlargement factor was adjusted for measurement of linear distances. The measurements were performed using a computerized cephalometric software program (Viewbox®; dHAL Software, Athens, Greece). The landmarks and variables measured in this study are shown in Figure (1) (Björk, 1947; Subtelny, 1957; Thilander et al., 2005). Two authors (SB and SR) localized each landmark by agreement.

From these landmarks, the following measurements were obtained: *n-sp´*, the distance between *n* and *sp´*; NL-*is*, the perpendicular distance from NL to *is*; overbite, the vertical difference between *ii* and *is* perpendicular to NL; NL-*ms*, the linear perpendicular distance from NL to *ms; s-pm*, the distance between *s* and *pm*; NSL-*pm*, the perpendicular distance from NSL to *pm*; NBa-*pm*, the distance from *pm* perpendicular on NL to NBa; NSL/NL, the angle between NL and NSL.

Precision of measurements

The precision of the registrations was tested by a repeat measurement of 20 randomly selected cases, after more than 1 month. The error of the method was calculated according to Dahlberg (G, 1940) and did not exceed 0.5 mm.

Table 2. Results from comparison of the W-K and the Gothenburg DHPC protocols.

Variable		W-K mean (SD)	DHPC mean (SD)	*p*-value
n-sp´	(mm)	43.98 (2.99)	45.41 (2.35)	< 0.05
NL-*is*	(mm)	24.89 (2.64)	26.33 (2.55)	< 0.01
Overbite	(mm)	1.74 (1.93)†	3.18 (2.07)	< 0.01
NL-*ms*	(mm)	19.21 (2.53) †	20.00 (1.67)	n.s.
s-pm	(mm)	38.06 (2.77)	38.98 (2.64)	n.s.
NSL-*pm*	(mm)	36.03 (2.67)	36.29 (2.52)	n.s.
NBa-*pm*	(mm)	18.87 (2.39)	18.88 (2.18)	n.s.
NSL/ NL	(°)	9.07 (3.55)	10.67 (3.29)	< 0.05

The number of registrations for each surgical group was 46, except for two variables in the W-K group (†) where 44 registrations were obtained.
n.s. = not significant.

Statistical analysis

Statistical analysis of the data was undertaken using Student's *t*-test to test for differences between the two surgical protocols (using IBM SPSS Advanced Statistics 19).

Data collection and analysis were carried out according to ethical principles for medical research involving human subjects. Approval of the local research ethics committee was obtained.

Results

Four variables showed statistically significant differences between the two surgical protocols. Anterior upper facial height (*n-sp´*), anterior maxillary height (NL-*is*), overbite, and maxillary inclination (NSL/NL) were all greater in the DHPC group (Table 2). For the remaining variables, no differences were found.

No statistically significant differences were found between cleft side or gender in any of the groups, which is why they were pooled.

Discussion

This study revealed significantly higher values for anterior upper facial height, anterior maxillary height, and overbite in the Gothenburg DHPC group than the W-K group (Table 2). However, while the *values for anterior upper facial height seemed to approach non-cleft reference values in the DHPC group more than in the W-K group, the maxillary inclination showed the opposite tendency (Thilander et al., 2005).*

The findings of a more normal anterior upper facial height, anterior maxillary height, and overbite in the DHPC than in the W-K group are in accordance with previous studies using the DHPC protocol, which that showed favorable maxillary growth for all dimensions investigated compared to the W-K protocol (Bakri et al., 2012; Friede and Johanson, 1977; Friede et al., 1980; Friede et al., 2012).

The normal growth of the maxillary complex has been extensively studied; regarding to vertical dimension, the maxilla is relocated downwards through appositional growth in the hard palate and the alveolar process. The bony surfaces of the maxilla are selectively resorptive or depository, to maintain the general shape of the midface during growth (Bjork and Skieller, 1974). Normal midfacial growth also involves displacement of the maxilla forwards and downwards in relation to the vomer. Through studies of CLP patients, the displacement has been documented to occur in the vomeropremaxillary suture (VPS) and mainly during the first year of life (Friede, 1978; H, 1977).

The vertical dimension of the maxilla is close to normal in unoperated cleft patients, indicating that surgery is the factor mainly responsible for growth restriction (Lambrecht et al., 2000). Thus, in the W-K technique, extensively denuded palatal bone results in scar tissue that negatively affect the maxillary growth in all dimensions (Ishikawa et al., 1998; Ross, 1970). The cranially based vomer flap in this technique is suggested to result in bone formation across the cleft, in addition to disturbing the VPS, resulting in increased restriction of maxillary growth (Friede and Johanson, 1977; Friede and Lilja, 1994; Prydso et al., 1974). However, the DHPC technique in the present study included early soft palate closure and the remaining cleft in the hard palate has been shown to narrow markedly until the hard palate closure, instead reducing cicatrization from hard palate repair (Friede and Enemark, 2001; Owman-Moll et al., 1998). Moreover, whatever technique is used, operating on the cleft palate at later age has also been shown to reduce the restriction of the maxillary growth whatever (Bardach et al., 1984; Xu et al., 2012). The DHPC techniques showed cephalometric values that were lower than in non-cleft individuals, indicating that restricted vertical growth still occurs (Thilander et al., 2005). This is in agreement with the statement of Ross (Ross, 1970) that repair of the cleft palate by any surgical technique will result in inhibition of the growth of the maxillary complex.

In the present study, we used three variables to quantify the vertical dimensions in the posterior upper facial region - *s-pm*, NL-*pm* and NBa-*pm*- besides the posterior maxillary height NL-*ms*. The DHPC group had slightly increased values in these four variables, but the differences were not statistically significant compared to the W-K group. Reports concerning upper posterior facial height in CLP patients have shown considerable variation regarding the cephalometric parameters used, resulting in difficulties in comparing results (Swennen et al., 2004; Wermker et al., 2012). However, in the present study the posterior upper facial height in both groups appears to have been less than in non-cleft controls (Subtelny, 1957).

The direction of maxillary growth is mainly vertical during the first 3 years of life (Donald H. Enlow, 1996), and early soft palate repair is believed to be related to an increased restricted posterior vertical growth (Ross, 1987; Swennen et al., 2002). However, it is not entirely clear whether the reduced posterior maxillary development is due to the early timing of soft palate closure or to the surgical technique (Swennen et al., 2002).

Increased inclination of the maxillary plane (NSL/NL) appears to be a characteristic feature of operated cleft patients (Ozturk and Cura, 1996; Paulin and Thilander, 1991; Swennen et al., 2002). The present study showed increased values for the maxillary inclination in both DHPC and the W-K groups compared to non-cleft reference values (Thilander et al., 2005). However, due to the differences in the anterior vertical growth, a significantly greater maxillary inclination was observed in the DHPC group than in the W-K group.

Restricted anterior maxillary vertical growth has been suggested to have negative effects on the facial aesthetics. Higher vertical position of the anterior nasal spine is shown to result in a shorter columellar base, reduced nasolabial angle, and a disproportion between the upper and lower facial heights (Ross, 1987).

In addition to the good sagittal growth previously shown for the Gothenburg DHPC technique, we also believe that the greater amount of anterior vertical growth shown in the present study is also favourable for the aesthetic outcome.

The more normal anterior maxillary vertical growth and overbite in DHPC protocol are in accordance with previous data of Friede and co-workers (1987) shown that fewer patients had received orthodontic treatment in DHPC protocol, 25% versus 78% in W-K protocol, in addition to the best dental arch relationships in DHPC protocol using the GOSLON Yardstick (Lilja et al., 2006).

The anterior maxillary growth was reduced in the W-K group and is thought to cause difficulties in establishing good functional jaw relations, even with orthodontic treatment (Isaacson et al., 1971; Ross, 1987), and it favors the development of a class-III skeletal abnormality (Markus et al., 1993). Still, a normal bite can be attained and maintained as a result of various compensatory mechanisms (Kuitert et al., 2006; Solow, 1980), for example, excess eruption of the maxillary posterior teeth (Ross, 1987).

It is unclear what clinical effects the posterior vertical growth and maxillary inclination have. Restricted posterior vertical development has been thought to affect the nasal aperture and consequently the air flow through the nose (Markus et al., 1993), but no clinical effect on breathing was found (Ross, 1987). Further studies are needed to understand the clinical implications of the vertical maxillary growth.

Both protocols have an increased maxilla inclination compared to non-cleft controls, and the maxillary inclination is thought to be a useful indicator for evaluation of the level of velopharyngeal closure (Satoh et al., 1999). Posterosuperior position and increased inclination of the maxilla have been suggested to be compensatory mechanisms facilitating the velopharyngeal closure (Satoh et al., 2005), and are associated with less nasality of speech (Stellzig-Eisenhauer, 2001). In addition, a significantly higher maxillary inclination has been found in CLP patients with normal speech than in patients who required pharyngeal flap due to speech compromise (Semb and Shaw, 1990).

It is important to note that the patients evaluated were assessed before puberty, why the results should be considered as preliminary, and it is feasible that growth restrictions may be more pronounced after growth spurt.

In conclusion, the present study shows that the Gothenburg DHPC protocol in patients with complete UCLP results in more normal anterior maxillary vertical growth and overbite, and therefore increased maxillary inclination at 10 years of age.

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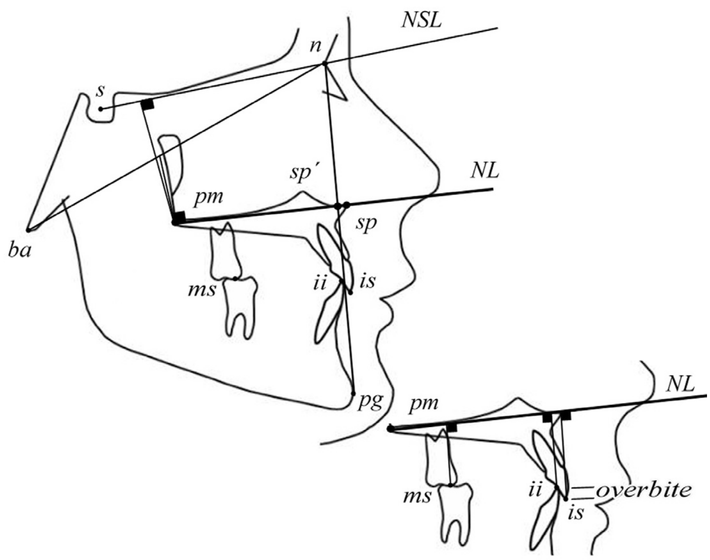

Figure 1. Reference landmarks and lines: *n* (nasion), the anterior limit of sutura nasofrontalis; *s* (sella), the center of sella tursica; *ba* (basion), the anterior-most point of foramen magnum; *pm* (pterygomaxillare), the point of intersection of palatum durum, palatum molle, and fossa pterygopalatina; *sp* (the spinal point), the apex of spina nasalis anterior; *pg* (pogonion), the most prominent point of the chin; *sp´*, the intersection between the nasal line (NL) and the *n–pg* line; *is*, the edge of the upper central incisor; ii, the edge of the lower central incisor; *ms,* the edge of the medial cusp of upper first permanent molar; NSL, nasion-sella line; NL, baseline of the maxilla; NBa line, cranial base line.