

Two-Stage Palatal Surgery with Early Veloplasty and Delayed Hard Palate Repair: A Balanced View on Speech and Midfacial Growth Outcome

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Abbreviations

A-P	Anterior-posterior
CLP	Cleft lip and palate
HPR	Hard palate repair
RCT	Randomized controlled trial
SPR	Soft palate repair
UCLP	Unilateral cleft lip and palate
VPC	Velopharyngeal competence
VPF	Velopharyngeal flap
VPI	Velopharyngeal incompetence

18.1 Introduction

The surgical protocol is the most important factor for subsequent outcome of cleft lip and palate (CLP) treatment. Today, there is general consensus that, particularly, the protocol for repair of the palate is crucial for normal speech development and adequate long-term midfacial growth. In this chapter, we will describe experiences from protocols, where the palate has been repaired in two stages: early soft palate repair (SPR), at ages from 3 to 24 months, followed by delayed hard palate repair (HPR) at varying ages during the first decade of life up to adolescence. Statistics from the year 2000 indicated that this surgical regimen was used in Europe by more than a third of the cleft teams (Shaw et al. 2000). (Another type of two-stage palatal surgery would be initial HPR, followed by later SPR, but this variant of two-stage surgery is not considered here.)

This chapter is written by three members of the cleft team from Gothenburg, Sweden, where two-stage palatal repair has been advocated for about 35 years. During this rather long time, we have gained considerable clinical experience in addition to significant knowledge through research covering many different aspects of the surgical method. The major reason for our change to this protocol was dissatisfaction with occlusion as well as midfacial growth in CLP patients treated according to our previous regimen (Friede and Johanson 1977). In a later follow-up study, we learnt that also the patients' speech had not developed as optimally as expected after our earlier protocol (Lohmander-Agerskov et al. 1993).

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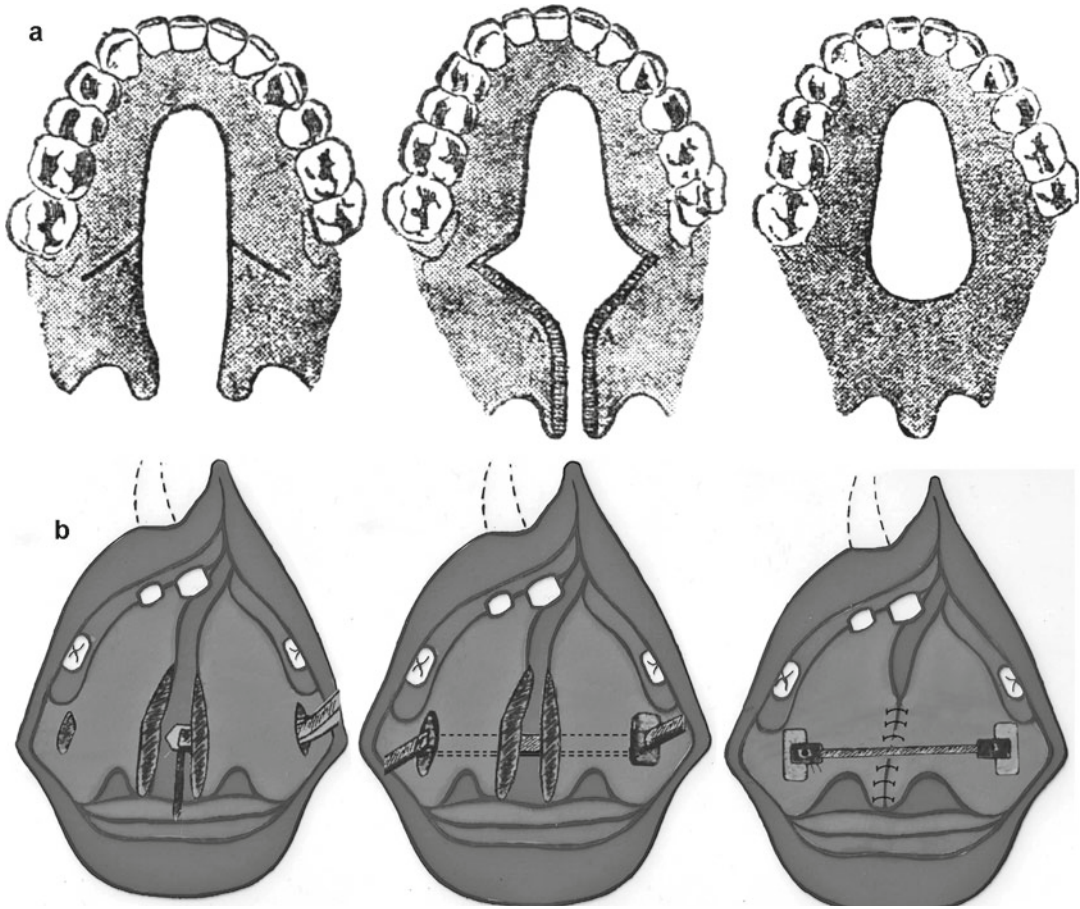


Fig. 18.1 Illustrations of early methods for SPR. (a) Obtained from Gillies and Fry (1921) (With permission from BMJ Publishing Group Ltd) and (b) drawings illus-

trating details in the SPR method used by Schwegendiek (1978). See text for further explanations

18.2 Historical Background

In 1921, Gillies and Fry published a paper, crucial to the development of the two-stage palatal repair method. They outlined a new, what they called “revolutionary principle” to improve treatment outcome in patients born with wide clefts of the palate. They suggested that the two halves of the soft palate, after being partly separated from the hard palate, should “be united in as far back a position” as possible (Fig. 18.1a). This procedure enlarged the remaining cleft of the hard palate, which in their opinion, should not be surgically repaired. They felt that a dental prosthesis, covering the hard palatal defect, would enhance possibilities to achieve

treatment goals such as “perfect speech, perfect mastication, normal nasal respiration, and normal bony contour” of the midface. A later long-term outcome study reported encouraging results for facial and occlusal development, while speech outcome was judged as less satisfactory (Walter and Hale 1987).

18.2.1 The First Pure Surgical Two-Stage Protocols for Palatal Repair

It was Herman Schwegendiek (1955) from Marburg, Germany, who, in the early 1950s, first described a true surgical two-stage palatal repair

method (Fig. 18.1b), which, over time, was employed in a great number of CLP patients. Even today, the two-stage protocol sometimes is referred to as the “Schweckendiek method,” though, presently, important details of the original description are no longer advocated and new features have been added. Follow-up reports from the German cleft center by the originator’s son, Wolfram Schweckendiek (1978, 1981a, b), described great satisfaction with the devised regimen, both regarding the patients’ speech development and their long-term maxillary growth. However, when an outside team examined some of the Marburg patients, they could only confirm the highly acceptable facial growth (Bardach et al. 1984). Regarding speech outcome of the investigated sample, an unusually high incidence of velopharyngeal incompetence (VPI) was found, most likely due to a short soft palate with poor mobility.

In the 1950s, Slaughter and Pruzansky (1954) from the United States also reported use of two-stage palatal surgery, particularly in patients where the palatal cleft did not lend itself to one-stage repair. They outlined several factors for the team to consider before deciding to perform velar surgery as an initial procedure. Examples of such determinants were width of the cleft, length and mobility of velum, and relation of velum to contiguous areas in nasopharynx. No outcome studies of their method were published, however, which might suggest that the results did not reach up to the authors’ expectation. Although a few American teams began advocating the two-stage palatal surgery protocol in the mid-1960s (Blocksmas et al. 1975; Cosman and Falk 1980; Dingman and Argenta 1985), it was mostly in Europe the method gained acceptance. Interest in the protocol was boosted here, in particular after the Zürich cleft team reported favorable outcome following change to the two-stage method in 1967 (Hotz and Gnoinski 1976). When members of the Gothenburg cleft team in the mid-1970s also contemplated change to the two-stage regimen with early SPR followed by later HPR, it was the excellent short-term result from Zürich, which was the precipitating factor for us. At this time, several other Swedish teams began advo-

cating the two-stage protocol as well, while other cleft centers in Scandinavia did not join the group using the protocol until more recently.

18.2.2 American Rejection of the Two-Stage Protocol

In the early 1980s, particularly, speech pathologists from a limited number of American cleft centers questioned the advisability of introducing the two-stage method for palatal repair (Witzel et al. 1984). They maintained that the few papers published on this subject had demonstrated severe speech problems, both before and after HPR. Some surgeons also expressed concerns about the protocol, which they felt not only jeopardized the patients’ speech development, but in addition, it resulted in inferior surgical results. The incidence of fistulas in the repaired cleft region increased significantly, and furthermore, the patients’ occlusion did not improve as much as hoped for (Cosman and Falk 1980; Jackson et al. 1983). Though this criticism was built only on short-term observations with minimal scientific analyses, many surgeons, especially in the United States, chose to abandon the two-stage method. Today, very few American teams appear to advocate the protocol with early SPR and delayed HPR (Katzel et al. 2009).

18.3 Surgical Details Introduced During Development of the Two-Stage Palatal Protocol

Though Gillies and Fry (1921) had employed surgical separation of part of the soft palate from the posterior edge of the hard palate, Schweckendiek (1955) did not include this important surgical step at the reintroduction of the two-stage protocol. With no detachment of velum at SPR, the repaired soft palate became both short and tight. The younger Schweckendiek (1978) presented schematic drawings, illustrating how tension in the sutured velum could be alleviated (Fig. 18.1b). Before uniting the velar halves, small incisions were made laterally on both sides. The dissections

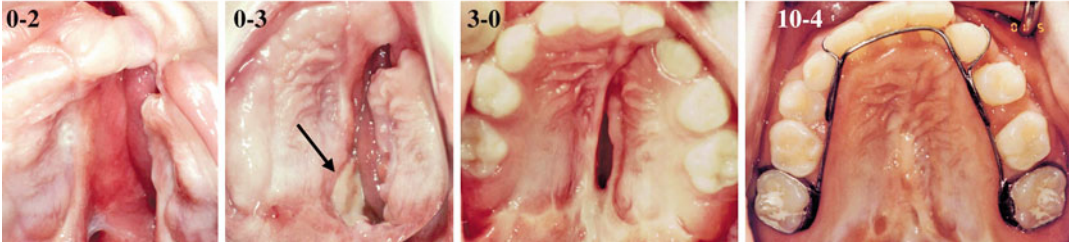
Patient #1; male

Fig. 18.2 Series of palatal views of a male patient from preoperatively to age 10 years and 4 months. The picture from 3 months shows early healing from where the poste-

rior vomer flap was raised (*arrow*) and turned backward. Note narrowing of the residual cleft in the hard palate. The HPR added only midline scars

penetrated the soft palate, where a transverse rubber band was inserted. At the end of SPR, it was tightened to reduce tension at the midline stitches. The device was kept in place after surgery for 1–2 weeks. Releasing incisions around the maxillary tuberosities and cutting of hamulus on both sides were other ways trying to deal with the increased tension in the repaired velum.

18.3.1 Modern Methods for Velar Repair

To avoid problems with a short and tight soft palate, surgeons began realizing that velum had to be released from the posterior hard palate (Braithwaite and Maurice 1968). This idea was supported by studies of Kriens (1970), showing that if there is a cleft of the soft palate, the velar muscular complex on both sides is running in an abnormal, anterior-posterior (A-P) direction. Therefore, it seemed logical, not only to detach but also to redirect those muscles to their normal transverse direction. To get access to the attachment of the velar muscles, mucosal/mucoperiosteal flaps were dissected from various positions at or within the posterior hard palate. The muscles were then cut from the palatal shelves, reoriented to a transverse course, and could be joined in the midline in a more posterior position than before. The procedure would be enhanced by addition of a posterior vomer flap (Malek and Psaume 1983), which was sutured to the nasal mucosa of the anterior soft palate (Fig. 18.2). With anterior velum attached

to the lower edge of the posterior nasal septum, the soft palate was lifted up to the level of the palatal shelves, which, during healing, helped reduce the size of the remaining cleft of the hard palate.

18.3.2 Methods for Repair of Remaining Cleft of Hard Palate

Regarding HPR, Schweckendiek (1955, 1978) did not suggest any particular method for this surgery and only mentioned that, preferably, it should be delayed until around puberty, i.e., when most of maxillary growth was completed. With such late repair of the residual cleft, the chosen method was not as crucial, as if this surgery had been performed at an early age. When later on some surgeons elected to close the remaining cleft already during development of the primary dentition, or sometimes even earlier, different repair methods were utilized. Without any direction from the originators of the two-stage protocol, most surgeons chose to use the same surgical method at HPR, as they were accustomed to in a one-stage palatal procedure. Examples of methods varied from use of uni- or bilateral mucoperiosteal flaps according to methods of Veau, Wardill-Kilner, von Langenbeck, Delaire, or others. Particularly, after use of methods where extensive mucoperiosteal flaps were shifted medially to cover the palatal cleft, areas of bone in the hard palate were left denuded. Growth-restricting palatal scars would then develop, which, depending on the position and size of

these scars, had a varying negative effect on maxillary development.

With delay of HPR, the residual cleft usually would narrow considerably (Owman-Moll et al. 1998), which the surgeon should have taken advantage of. The reduced width of the remaining cleft would, in many cases, allow primary repair of the residual opening in the hard palate after mobilization of the cleft edges without leaving any palatal bone denuded. In wider residual clefts, the repair could be accomplished with a turnover vomer flap or by use of bilateral flaps taken from the thin palatal mucoperiosteum close to midline. According to Delaire, inclusion of the thick palatal mucoperiosteum more laterally would cause bare bone in areas with increased risk for development of growth impairing scars (Markus et al. 1993). If the remaining cleft in the hard palate was very wide, it was suggested to postpone HPR until age 2 or 3 years. These surgical details were decisive factors for maxillary development during subsequent growth.

18.3.3 Timing of Palatal Surgery

Timing of repair of the two palatal procedures has also been characterized by great variation. Reports from literature have suggested ages varying from 3 to 24 months for SPR, and for HPR, different papers have proposed ages ranging from 6 months to 16 years. The timing preferences of the surgeon and the cleft team most often have been guided by subjective estimations of how the operations might affect speech and/or maxillary growth outcome.

18.3.3.1 Surgical Timing and Speech Development

Regarding speech development, the controversial debate on optimal age for palatal closure has been hampered by questionable comparisons between studies with different timings of surgery without any consideration to other factors, i.e., staging, sequence, or technique for the repair, which will influence the outcome. From theoretical perspective of speech-language development and particularly in relation to the sensitive period or state of readiness for speech development between the ages of 4 and 6 months, there is *no* controversy that an early, *complete* pala-

tal closure is preferable (Kemp-Fincham et al. 1990). The early age would mean before or at onset of pertinent canonical babbling and possibility to close the oronasal coupling for relevant development of oral pressure sounds. Both were found to be significant predictors of later speech and language performance (Oller et al. 1998; Chapman et al. 2003; Lohmander and Persson 2008; Scherer et al. 2008). Recent studies indicate that these factors can be reached to a higher degree, if the soft palate is repaired early, even if the cleft in the hard palate still is unoperated (e.g., Willadsen and Albrechtsen 2006). According to opinions published by one of the few American teams, currently advocating the two-stage repair (Rohrich et al. 2000), a protocol, with velar repair at around 3–6 months and delayed hard palate closure at age 15–18 months, would provide the best opportunities for normal speech development as well as favorable maxillary growth outcome.

18.3.3.2 Timing of Surgery and Maxillary Growth

Considering the growth influence from palatal surgery, our view is that possible effects from various timings, for SPR as well as HPR, depend upon whether the employed surgical methods will impair palatal areas, important for subsequent maxillary development. If using a method with definite propensity for growth restriction, an early repair should be delayed or preferably not be used at all. But, if surgery can be accomplished with minimal denudation of palatal bone in sensitive regions (Markus et al. 1993), the growth outcome of the procedure can be quite satisfactory even if performed during the patients' first year of life. These circumstances have seldom been considered, which has contributed to controversies about the benefits of the two-stage method for palatal repair.

18.4 Reported Speech and Growth Outcome After Some Variants of the Two-Stage Palatal Protocol

After introduction of a new philosophy for solving a specific surgical problem, such as repair of cleft palate, many surgeons tend to “jump on the

bandwagon,” and furthermore, some of them might devise their own treatment variants. When the two-stage palatal repair protocol was reintroduced in the 1960s and 1970s, many cleft teams converted to this regimen. Unfortunately, the majority of those early teams, including the operating surgeons, did not disclose, whether the new regimen had fulfilled their expectations or not. A few short-term reports were published in the 1980s, where the outcome generally was rated as poor.

18.4.1 Speech Outcome

Speech development, sometimes appraised before HPR, was judged as inferior to what was expected. These young children had significantly poorer articulation skills than their noncleft peers. Posterior substitutions had often developed and so had frequent VPI (Cosman and Falk 1980; Jackson et al. 1983; Noordhoff et al. 1987). From a methodological point of view, it has to be remembered that these evaluations were clinical, live judgements with no possibility for control of the data. If we believe that, even so, these early assessments were valuable, we suspect that many of the speech errors might have had their origin in missing important surgical details at closure of the soft palate. For instance, one of the papers stated, “the soft palate was closed directly with only minimal division of nasal mucosa and palatine aponeurotic fibers” (Cosman and Falk 1980). With no definite separation between the soft and hard palate, we suspect the surgeon had been unable to bring back the repaired velum to a position needed for achievement of velopharyngeal competence (VPC) on a regular basis. In other studies (Noordhoff et al. 1987), it was mentioned that SPR had been performed according to the original method of Perko (1979), but nothing was reported about use of a posteriorly based vomer flap. Omission of this crucial surgical step is likely to have caused reduced velar length in many cases and also a wider residual cleft in the hard palate due to less narrowing during early palatal growth. Tentatively, the short soft palate would increase the risk for VPI and posterior

substitutions, such as pharyngeal and/or glottal articulations. In the report by Noordhoff et al. (1987), all patients treated with the two-stage method were said to have increased articulation errors, particularly those individuals with wide remaining cleft of the hard palate. A later follow-up paper (Liao et al. 2010) confirmed increased hypernasality and compensatory articulation disorders in these patients.

18.4.2 Maxillary Growth Results

More recently, a number of teams have, in particular, reported the patients’ maxillary growth outcome with limited focus on their speech development. The majority of the papers described favorable midfacial growth (Noverraz et al. 1993; Tanino et al. 1997; Nollet et al. 2005, 2008; Sinko et al. 2008; Liao et al. 2010), while a few of the reports did not find any maxillary growth advantage of the two-stage protocol (Gaggl et al. 2003; Mølsted et al. 2005; Holland et al. 2007; Stein et al. 2007). The first group of papers generally advocated methods for SPR and especially for HPR, where closure of the cleft resulted in minimal denudation of bone in the palate. Examples of such procedures comprised employment of a turnover vomer flap, suturing of the cleft edges, mobilization of mucoperiosteal flaps dissected close to the cleft, or use of a modified von Langenbeck operation. On the other hand, in studies describing no maxillary growth benefits or inferior maxillary development, the methods used in these patients had created growth-restricting scars, mostly from HPR. Examples of surgical methods included employment of a Veau pedicle flap, a mucoperiosteal pushback procedure, or use of “unipedicled mucoperiosteal flaps.” All of them will give rise to significant areas of bare palatal bone, which will heal secondarily, leading to scar tissue development. Thus, these different opinions about definite growth advantage or insufficient maxillary development after the two-stage palatal protocol can be explained by the performed repair methods and, only to a limited extent, by *when* surgery was done, as described above. Such explanations

are easier to embrace than speculation about effects, e.g., from cleft team organization (Shaw et al. 1992a, b) or surgeons' different operating skills (Ross 1987a, b), as reasons for the patients' growth results. However, we have to remember that acceptance or rejection of the two-stage regimen might be influenced not only by how well the repaired maxilla will develop but also by other factors, such as increased risk for fistula formation, poor speech development, and need for more velopharyngeal flaps (VPFs); etc.

18.5 Outcome of Maxillary Growth as Well as Speech from Teams with Long-Term Records

Only a limited number of publications exist, where the patients' maxillary growth as well as their speech development after two-stage palatal surgery has been studied up to adolescence or early adulthood. From this very small group of papers, we have chosen to report treatment outcome in unilateral cleft lip and palate (UCLP) patients from two European cleft teams still practicing the protocol.

18.5.1 Zürich, Switzerland

In Zürich, Switzerland, the cleft team has used their version of the two-stage palatal repair protocol since the late 1960s. It was built on a systematic coordination between maxillary orthopedics and surgical interventions from early infancy (Hotz and Gnoinski 1976; Hotz et al. 1986). The team initiated preoperative maxillary orthopedic treatment shortly after birth, not only to help feeding but also for growth guidance of the maxillary segments. After lip repair at age 5–6 months, the infant continued with maxillary orthopedics until SPR, which was carried out at around 18 months. This surgery included reorientation of the velar muscles after their separation from the posterior hard palate (Perko 1979). The covering flaps of the oral mucosa were dissected from the posterior third of the hard palate, and, to reduce risks for maxillary growth impairment, the dis-

sections were made *supraperiosteally*. A posterior vomer flap was also raised, and after turning it backward, the flap was sutured as part of the anterior nasal layer at SPR (Hotz et al. 1986). With the nasal mucosa not separated from the posterior hard palate, inclusion of the vomer flap would not help elongate the soft palate, and therefore, a midline Z-plasty was added. At age 5–6 years, HPR was performed. Due to narrowing of the residual cleft, closure was accomplished by use of a turnover vomer flap for the nasal layer, and for repair of the oral layer, a mucoperiosteal flap was shifted medially from the noncleft side.

18.5.1.1 Follow-up Studies

The Zürich cleft team has published several follow-up investigations, particularly about maxillary growth. Roentgencephalometric results from a group of 10-year-olds born with UCLP indicated satisfactory A-P relationship between maxilla and mandible in about 80 % of the subjects (Gnoinski 1990). At age 15–20 years, these patients showed continuation of the favorable orofacial development, documented in the 10-year sample (Gnoinski 1991; Gnoinski and Haubensak 1997). Even if the cleft maxilla grew slightly in length also between 15 and 20 years of age, the average maxilla was shorter than in noncleft subjects. Only about 10 % of the patients needed maxillary orthognathic surgery to achieve an acceptable facial profile.

The team from Zürich has also reported cross-sectional, detailed speech data based on live assessments. After HPR and speech therapy, the patients' glottal or pharyngeal articulations were eliminated (Hotz et al. 1978). This outcome was in contrast to what was achieved with the previous treatment protocol. Nothing was mentioned about development of retracted articulations of certain consonants such as anterior plosives. Severe hypernasality decreased spontaneously with age, even before HPR was performed. After HPR, this speech error was claimed by the authors to have disappeared almost completely, often though, with help of intensive speech therapy. No pharyngeal flaps were judged by the team to be necessary in any of the Zürich patients.

18.5.1.2 Verification of the Zürich Speech Outcome

An outside investigator was recruited to verify the satisfactory speech development of the cleft children in Zürich (Van Demark et al. 1989). Thirty-seven UCLP patients were studied in a cross-sectional investigation at ages ranging from 6 to 16 years (mean 10.5 years). Oral examination generally revealed a mobile and fairly long soft palate, which was ascribed to the surgical method used for SPR. A comprehensive, independent speech analysis from audio recordings revealed that about 95 % of the patients displayed adequate or marginal VPC. The majority of subjects did not show speech errors such as glottal stops and pharyngeal substitutions. Even in the younger patients, the incidence of other compensatory articulations and “nasalizations” was low. The authors concluded that “good speech results for unilateral complete cleft lip and palate can also be achieved with late closure of the anterior palate, at least as done by the Zürich approach.”

Unfortunately, no further follow-up has been published.

18.6 Gothenburg, Sweden

When we planned the palatal surgery in our new protocol, the Zürich group had neither published any suggested ages nor detailed descriptions of their surgical procedures. For velar repair, we opted for an age around 6–8 months, while we wanted to delay closure of the remaining cleft in the hard palate to the mixed dentition. Regarding surgical details for SPR, we chose the approach to separate soft palate from hard palate right at the border region (Friede et al. 1980) (Fig. 18.3). The lateral incisions and denudation of palatal bone were to be made as minimal as possible. The velar muscle bundles should be united in the midline after complete release of the muscular fibers from their abnormal insertion at the pos-

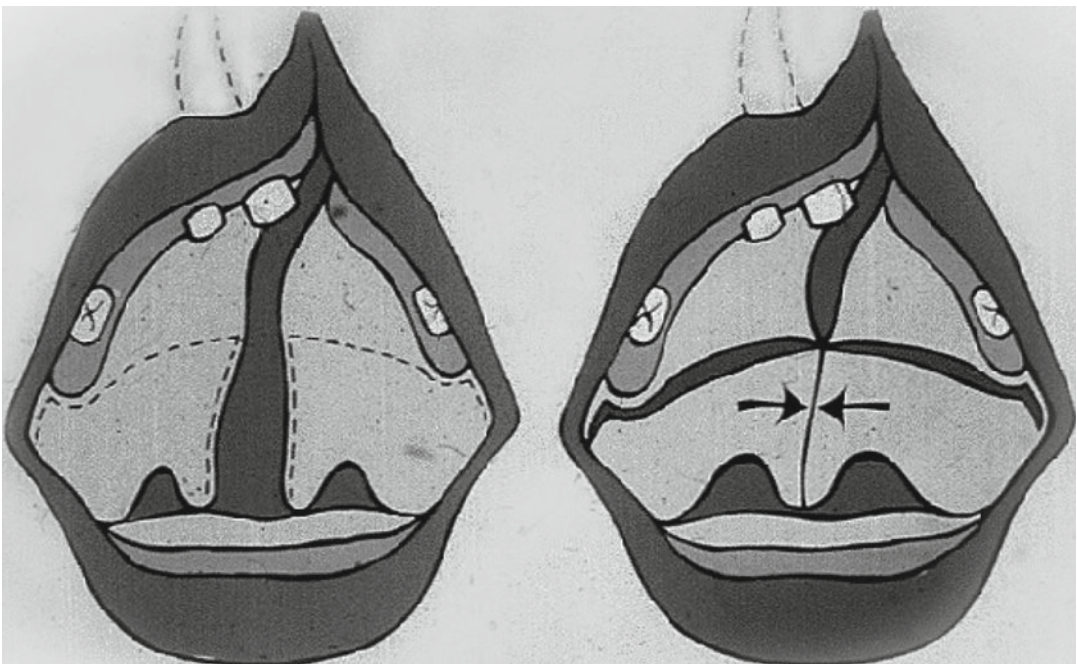


Fig. 18.3 Schematic illustration of SPR according to an early repair version used at Gothenburg Cleft Center. Incision lines run in the border region between velum and

hard palate. The two halves of the soft palate, with the muscle bundles reoriented to a transverse course, are united in the midline (*arrows*)

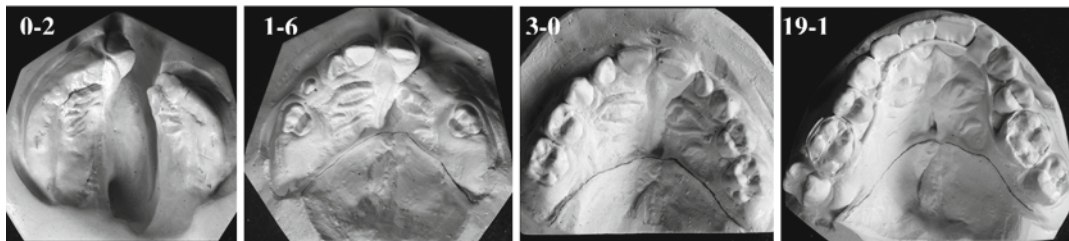
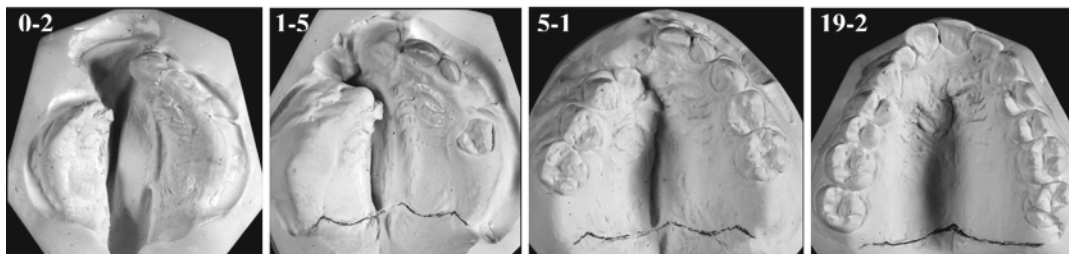
Patient #2; male**Patient #3; male**

Fig. 18.4 Anterior-posterior variation in position of scar line from SPR. In *upper patient*, the scar line is too far forward, harming growth in length of the maxillary dental

arch. In *lower patient*, the scar line is in correct position at posterior maxilla. The dental arch length is well developed with space for all teeth

terior border of the hard palate. The muscles remained attached to the nasal mucosa. At this stage of development of our method for velar repair, no posteriorly based vomer flap was raised and attached to the anterior soft palate. Therefore, the residual cleft of the hard palate did not narrow as much as expected. This made our surgeon dissect the palatal flaps further anteriorly, occasionally into the middle of the hard palate (Fig. 18.4). Such change would reduce the remaining cleft size and thereby enhance early speech development. However, after some time, we realized that this change in surgical detail at SPR also might restrict maxillary growth. It was first after introduction of the posterior vomer flap that the dissection line in the palatal mucosa returned to its previous position close to the edge of the hard palate. The Zürich surgeon Perko (1979) did not report in detail how their two-stage surgical procedures were carried out until 1979, and then, a posteriorly based vomer flap at SPR was not yet part of the Swiss surgical routine. It took another 1 or 2 years before many surgeons, including those from Zürich and Gothenburg, began utilizing this important addition to SPR.

18.6.1 More Detailed Two-Stage Palatal Surgery

In 1996, our cleft team published a detailed report how the two-stage protocol was practiced in Gothenburg up to 1995 (Lilja et al. 1996). At soft palate surgery, incisions began around the posterior part of the maxillary tuberosities and then followed a zigzag route at the posterior border of the hard palate (Fig. 18.5). A posteriorly based vomer flap was dissected. Anteriorly, the incision was placed behind the vomero-premaxillary suture, and the flap had its base close to the junction between vomer and the cranial base. Mucosal flaps in the soft palate were raised by blunt dissection. Hamulus was identified but not broken. The insertions of velar muscles, including their attached nasal mucosa, were cut at the posterior border of the hard palate. A flap with the muscles connected to the nasal layer was then dissected free and mobilized to a posterior position. After that, the muscles including the levator were reconstructed to a transverse course, where suturing could be performed without tension. The vomer flap was raised and the nasal layer of velum was closed anteriorly to the level of the muscular sling by use

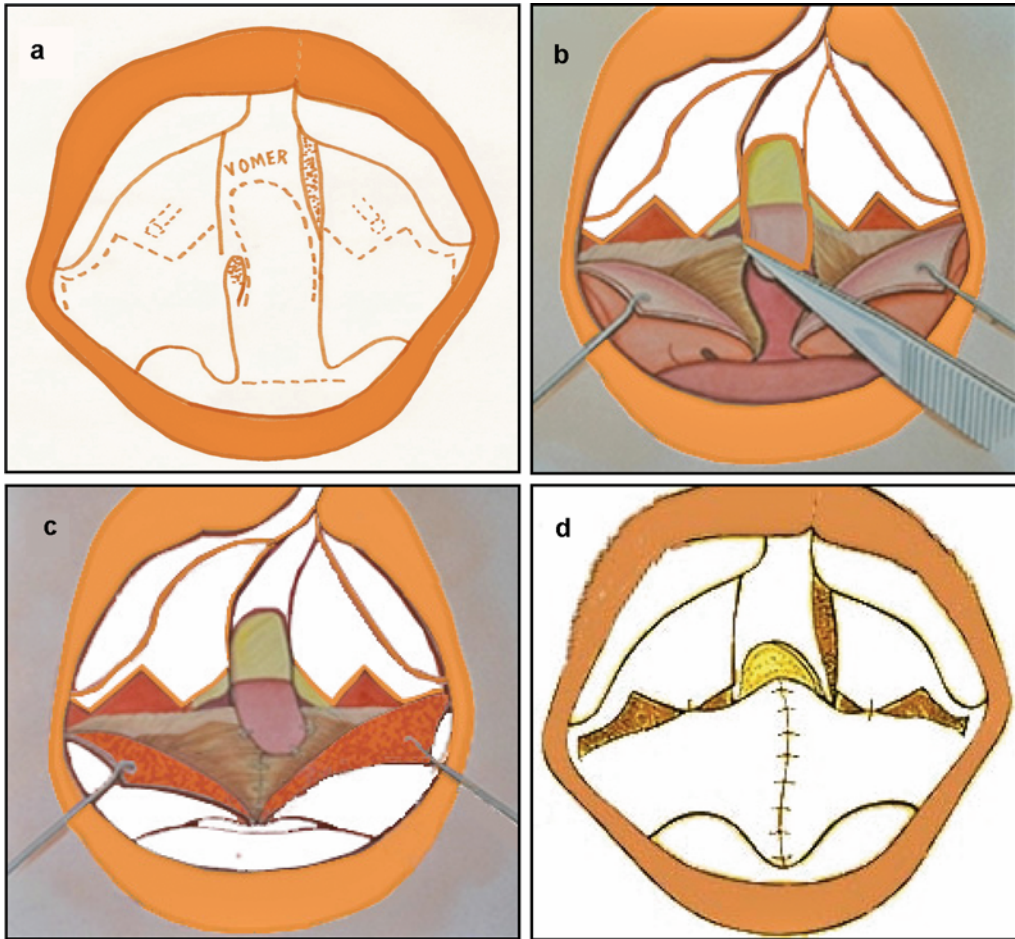


Fig. 18.5 Schematic drawings of the soft palate closure. (a) The incisions follow a zigzag line between the soft and hard palate. A posterior vomer flap is dissected, which has its base at the posterior-cranial part of vomer. (b) Both sides of velum are divided into two layers: the oral mucosa and the nasal mucosa with the forward inserting muscle bundles attached. The two layers are separated laterally and posteriorly to the uvula. Medially, the nasal layer with

the muscle bundles is cut bilaterally from the posterior hard palate. (c) The muscles are redirected to a transverse course, sutured together medially in a posterior position, and also attached anteriorly to the backward-turned vomer flap. (d) The muscles and the raw surface of the vomer flap are covered by the oral flaps, which are pushed in a medial-posterior direction

of the vomer flap. In this way, the vomer bone became connected to the anterior velum. The palatal closure was then continued over to the oral side, where a pushback procedure was performed within the oral layer of the soft palate. Inclusion of the vomer flap as well as the pushback surgery helped to increase the length of the repaired velum.

The HPR was delayed until the patient had reached the stage of early mixed dentition (7–9 years), because, thereby, the procedure could be combined with bone grafting to the

alveolar cleft. Surgery began with gingival incisions along the neck of the teeth on the palatal side and in part also labially (Fig. 18.6). In the area of the cleft, incisions were made along the cleft edges, and gingival and palatal mucoperiosteal flaps were raised. On the labial side, a back-cut was made in the cleft-side molar area. This facilitated mobilization of the gingival flap as well as the dissection in the cleft area. When the cleft had been dissected completely free on both palatal and labial side, suturing in the

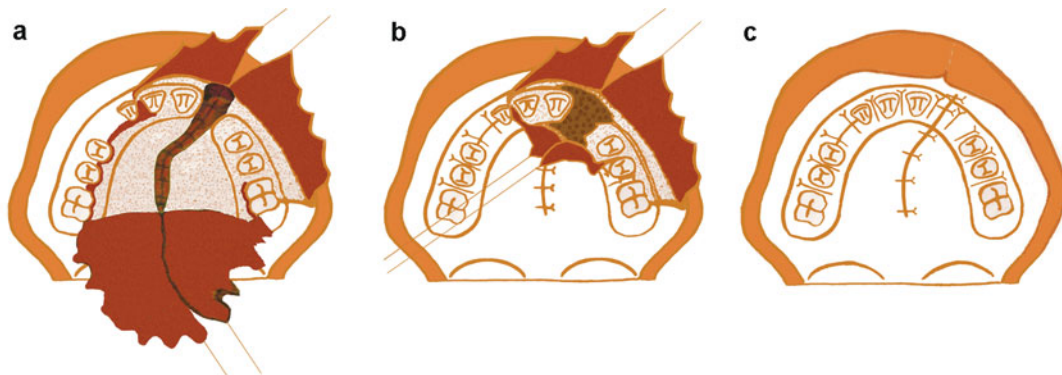


Fig. 18.6 Illustrations of the method for repair of the residual cleft in the hard palate in combination with bone grafting. (a) Incisions are made along the necks of the teeth and along the edges of the residual cleft. Palatal and gingival mucoperiosteal flaps are raised. The nasal layer is

closed. (b) The palatal mucoperiosteal flaps are closed in the palate. Bone grafting is performed to the cleft in the alveolus. (c) The grafted bone is covered by gingival and anterior palatal mucoperiosteal flaps, which are sutured together

midline began in the nasal layer and continued orally in the midline palate. Cancellous bone was then grafted to the cleft in the alveolar process and covered with gingival and palatal flaps. The operation was completed with suturing the gingival and palatal mucoperiosteal flaps together in some interdental spaces. We consider the palatal incisions along the necks of the teeth very important. Because of the reduced width of the residual cleft, suturing back of the combined palatal flaps along the dental arch could be done with none or only minimal palatal bone exposed close to the teeth. In 1996, timing for HPR was modified in an effort to prevent development of typical retracted oral speech deviations noted in some patients during preschool and early school age. The repair was then changed to be carried out around 3 years, which meant an extra operation, because HPR could no longer be performed together with the bone grafting procedure. If the residual cleft of the hard palate was narrow or of average size, it was closed in one layer by use of a simple turnover vomer flap (Fig. 18.7). This approach did not work so well for wider residual clefts of the hard palate due to increased risk for fistula development. In these cases, a two-layer repair was necessary. A vomer flap was then sutured to the nasal mucosa on the cleft side, and the suture line was closed with a mucoperiosteal flap from the cleft side of the palate (Fig. 18.8).

18.6.2 Follow-up Studies of Maxillary Growth

Over the years, the Gothenburg cleft team has reported several follow-up studies, both regarding maxillary growth and speech development. An early growth comparison at age 7 years, before HPR in the two-stage protocol, demonstrated significantly improved results in comparison to what was achieved with our previous regimen (Friede et al. 1987). In a more recent longitudinal follow-up study, we compared 30 consecutive UCLP patients, who had been subjected to our two-stage palatal surgery, to a similar sample from another center. Those patients had undergone HPR with a vomer flap at age 3 months, which was followed by SPR with a pushback method at 22 months (Friede and Enemark 2001). Comparison of roentgencephalograms, obtained during the age span from prepuberty to adolescence, demonstrated that our patients with delayed HPR had much better midfacial development than the other group of subjects. This difference was also noticed in the percentage of patients with need for later maxillary advancement surgery (10 % vs. 30 %). The favorable maxillary growth outcome in our patients was, some years later, confirmed in a cast study of 104 consecutive subjects with UCLP (Lilja et al. 2006). If just considering casts

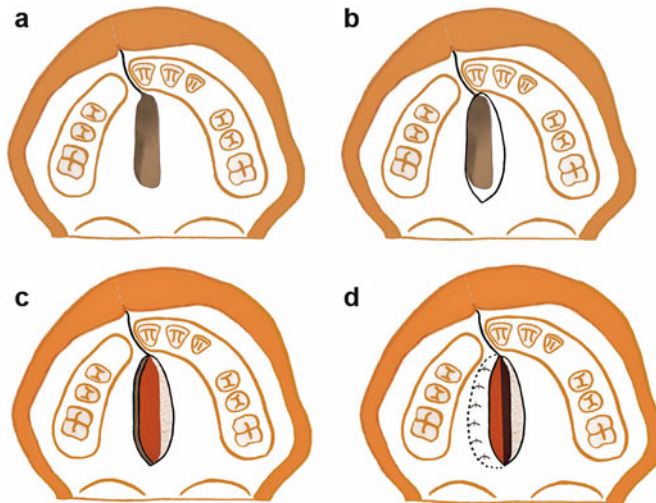


Fig. 18.7 Composite of drawings illustrating repair of a small- or average-sized residual cleft in the hard palate at age 3 years or preferably earlier. At this stage, bone grafting is not performed in connection with the HPR as shown in Fig. 18.6. (a) The residual cleft in the hard palate. (b) Incision lines. On the noncleft side, the incision goes into the flat medial part of the palate. On the cleft side, the incision is made at the cleft border between oral and nasal mucosa. (c) The vomer flap is raised. It contains some

millimeters of oral mucosa leaving a raw bone surface in the medial part of the palate on the noncleft side. On the edge of the cleft side palatal shelf, a subperiosteal dissection is performed and a pocket is created between the oral periosteum and the bone. (d) The vomer flap is tucked into the pocket and sutured, which finalizes HPR. Thus, this is a one-layer closure, and the raw surface of the vomer flap is left for secondary epithelialization

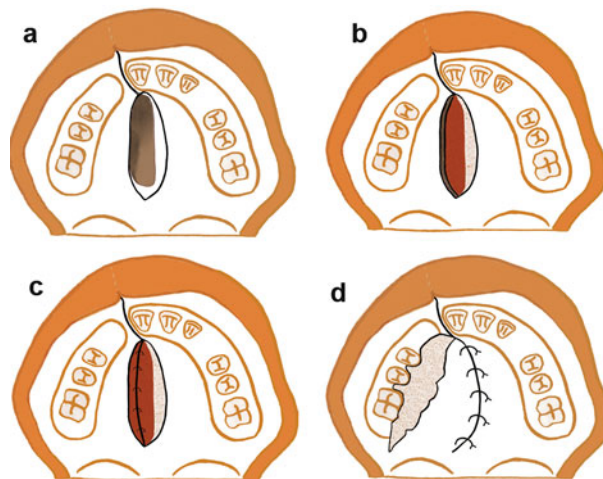


Fig. 18.8 Drawings illustrating HPR in an individual with a somewhat wide residual cleft. Preferably, the patient should have erupting/erupted upper deciduous molars, which usually means an age at around 3 years. (a) Incision lines. On the noncleft side, the incision goes into the flat medial part of the palate. On the cleft side, the incision is made at the cleft border between oral and nasal mucosa. (b) The vomer flap is raised. It contains some millimeters of oral mucosa leaving a raw bone surface in the medial part of the palate on the noncleft side. (c) On

the cleft side, a subperiosteal dissection is performed, and the nasal layer is lifted and brought in contact with the vomer flap and sutured. (d) Also on the cleft side, an oral mucoperiosteal flap is raised via an incision along the teeth. The flap is brought in contact with the incision on the noncleft side and sutured. The suture line between the vomer flap and the nasal layer is now covered. The raw bone surface is left for secondary epithelialization, which takes place on a surface with thin neighboring wound edges. Thereby, the risk for bad scar contraction is small

obtained at around 19 years, an acceptable dental relationship, reported as GOSLON scores 1, 2, and 3 (Mars et al. 1987), was found in 97 % of the subjects. This was rated as an exceptionally high incidence of satisfactory occlusion at early adulthood.

For our most recent longitudinal follow-up study (Friede et al. 2011), we used a consecutive series of 50 patients born with UCLP. All subjects had lateral roentgencephalograms from four selected age stages from preschool to early adulthood. Besides lateral cephalograms, we also studied maxillary casts from an age around 1.5 year to score the A-P position of the mucosal scar line from the dissection between soft and hard palate at SPR. Interestingly, we found that those patients with more anteriorly placed scar lines had definitely reduced maxillary lengths as appraised in their cephalograms from age 19 years (Figs. 18.9 and 18.10). This illustrates how small details at surgical reconstruction of the UCLP are important for optimal maxillary development during subsequent growth. However, the overall favorable midfacial development, noticed in our previous outcome reports, was confirmed in this study. In early adulthood, the patients' mean values for the skeletal profile convexity and for the sagittal jaw relationship were close to or within the 95 % confidence interval of the mean for noncleft Swedish subjects (Thilander et al. 2005). Generally, the average UCLP patient's facial morphology displayed a skeletal pattern in harmony with the retrognathic maxilla and mandible characterizing this group of patients (Segner 1989) (Figs. 18.11 and 18.12).

18.6.3 Follow-up Studies of Speech Outcome

Most of the reports on speech development from Gothenburg cleft center have been retrospective, longitudinal, follow-up studies of individuals born with UCLP and have, by and large, investigated consecutive series of patients. It should be mentioned though that particularly the early reports employed limited patient

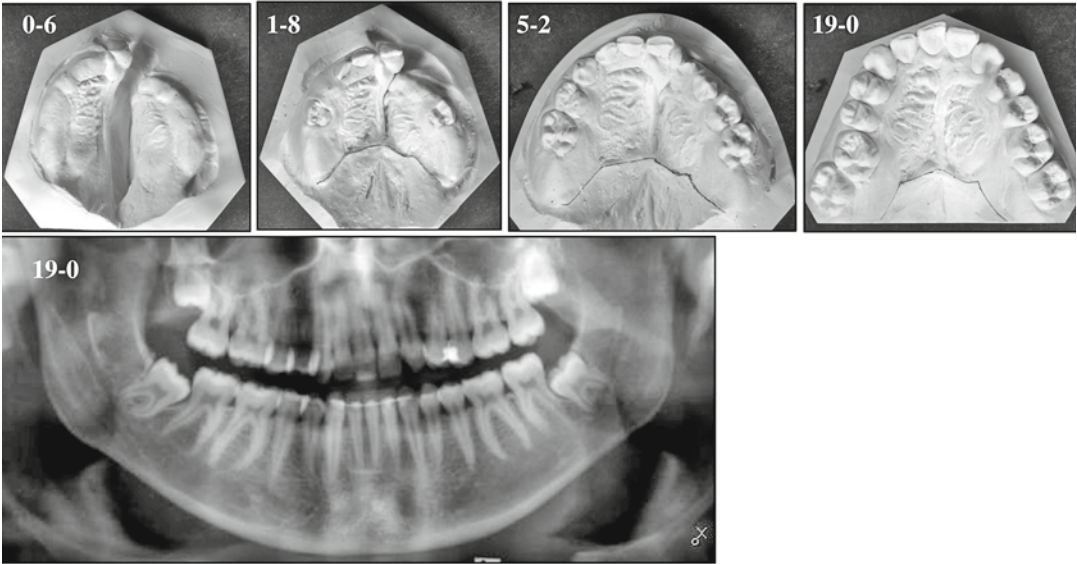
samples and, in addition, more than one surgeon had carried out the palatal repairs. We are aware that these facts might, to some extent, have reduced the reliability of our results. However, on the positive side, we must mention that all speech samples were rated using standardized audio recordings, which were identity blinded to the investigators and independently evaluated by two experienced speech-language pathologists, and in the later studies also by an external listener.

18.6.3.1 Early Results

Our initial speech results were reported in the beginning of the 1990s when our first patients, treated according to the two-stage palatal surgery protocol, were evaluated during their early childhood (Lohmander-Agerskov and Söderpalm 1993; Lohmander-Agerskov et al. 1995). *Hypernasality* and *audible nasal emission* occurred among the 5-year-olds before HPR in a moderate-to-severe degree in about 40 and 30 %, respectively. For hypernasality, the prevalence decreased to about 10 % at age 7, which still was an evaluation *before* HPR. For audible nasal emission, however, in most patients, the occurrence remained the same until about 1 year *after* HPR. The prevalence for both hypernasality and audible nasal emission were then reduced to occur in about 6 % of the patients. The gradual reduction in size of the remaining cleft was suggested as explanation for lowering the hypernasality rate even before hard palate closure. Supposedly, the smaller cleft size resulted in reduced influence on resonance from the nasal cavities (Lohmander-Agerskov et al. 1997). In contrast, there was no relationship between the aerodynamic-related variable audible nasal emission and the size of the residual cleft.

The early outcome studies on *articulation errors* revealed very rare occurrence of *glottal/pharyngeal misarticulations*. Low incidence of these speech deviances was considered an indication of adequate VPC in the vast majority of our patients. Instead, we discovered so-called retracted oral articulation of dental/alveolar plosives to a palatal or velar place, which, particularly before HPR, was a rather common finding (Lohmander-Agerskov 1998).

Patient #4; female



Patient #5; male

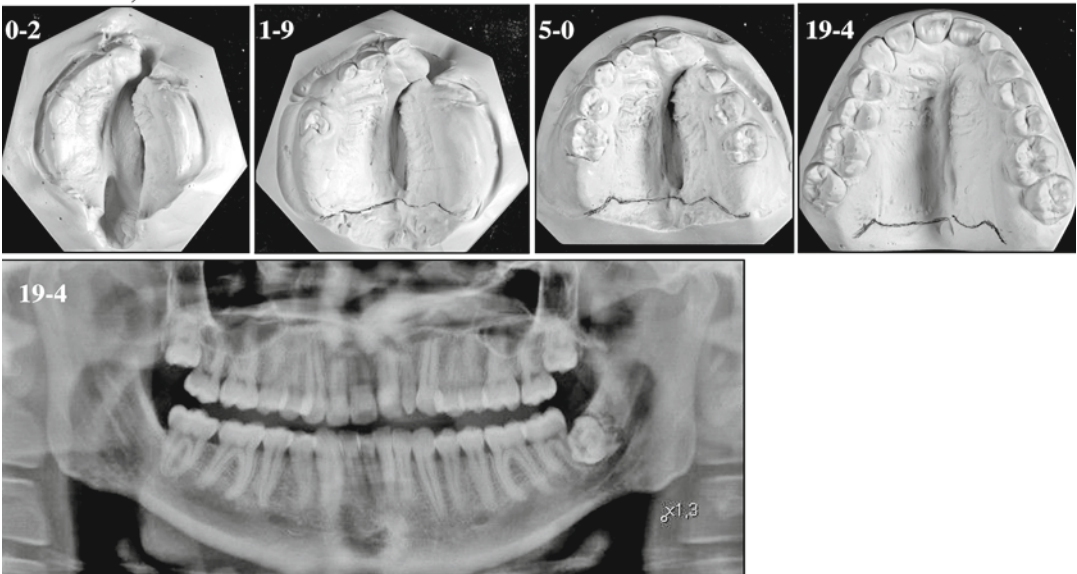


Fig. 18.9 Casts and orthopantomograms from two patients illustrating different development of maxillary dental arches. The *upper patient* demonstrates slight crowding at early adulthood in spite of no permanent lateral incisor on the cleft side. In the *lower patient*, the den-

tal arch is well developed with space for all permanent teeth. Note more posteriorly positioned scar line after SPR in this patient compared to the upper one. See Fig. 18.10 for midfacial development

In different reports, we recorded prevalences between 30 and 40 % with little reduction in rate before HPR and with weak, but significant, correlation to size of the residual cleft (Lohmander et al. 2002). The varying degrees of the retracted articulation errors after our two-stage protocol

occurred up to early childhood in about one third of the children. However, the speech deviation was gradually reduced to half of that at age 10 years, and around adolescence, the errors had disappeared. An interesting finding was that children who had had a spontaneous, early, func-

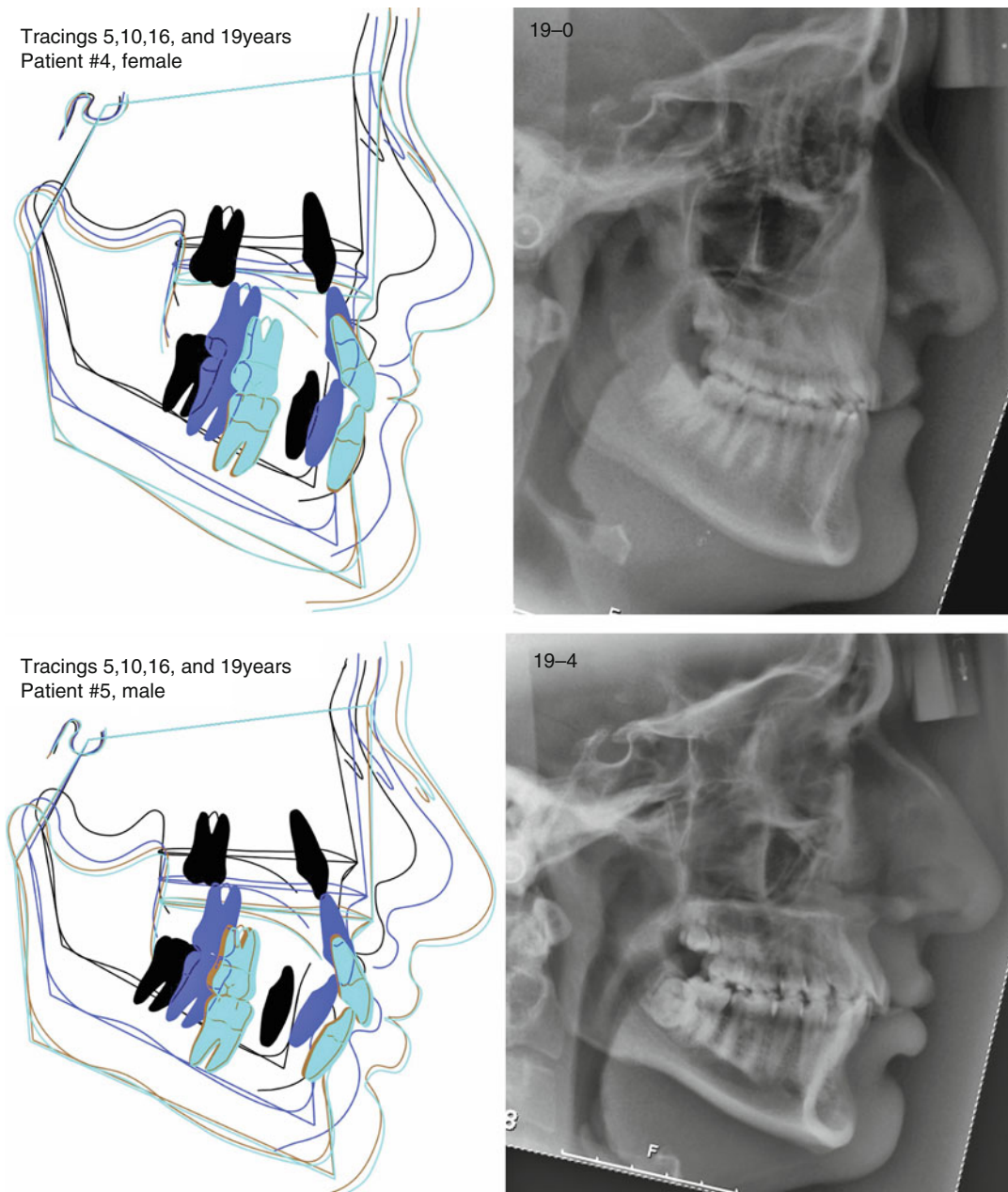


Fig. 18.10 Superimposed tracings at four age stages in addition to cephalograms at young adulthood of the two patients illustrated in Fig. 18.9. Notice better development of the maxilla of patient #5 compared to patient #4

tional closure of the residual cleft (Fig. 18.13) (evaluated before age 3 years) did not display any articulation errors at later ages (Lohmander-Agerskov et al. 1996). In another early study of possible factors related to development of retracted oral articulations, we found that chil-

dren with this speech error at ages 3 and 5 years were more likely to have been babies preferring to use a posterior place of articulation at babbling or maybe, more correctly, lacking anterior articulation at this stage of speech development (Lohmander-Agerskov et al. 1998).

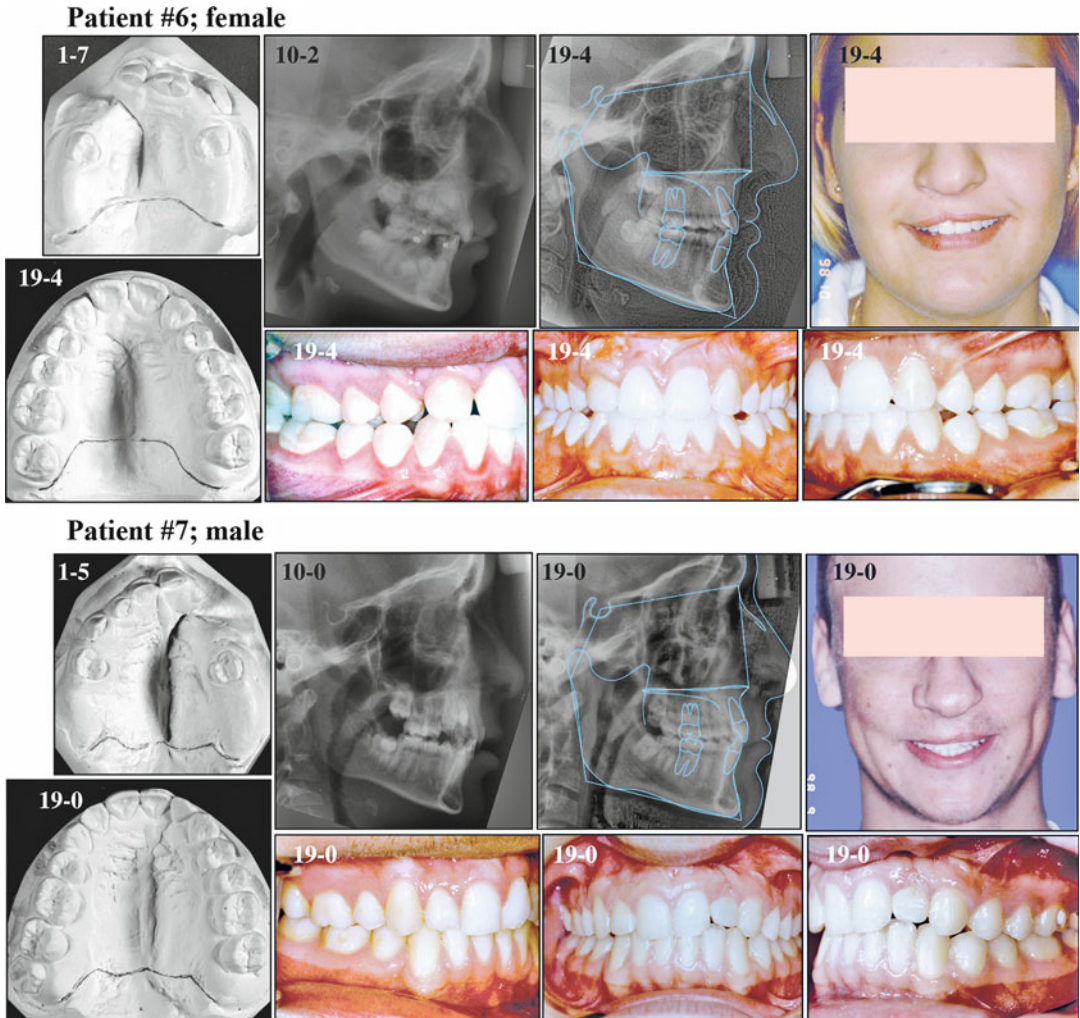


Fig. 18.11 Composite of casts, photographs, and cephalometric records from two typical patients treated according to the Gothenburg two-stage palatal surgery protocol. No orthognathic surgery have been performed. *Upper patient* showed satisfactory sagittal growth of the maxilla but, partly due to missing laterals, developed bilateral crossbite. Patient declined further surgery of her upper lip. Good speech developed already from early age. *Lower*

patient displayed excellent maxillary growth, both in sagittal and transverse dimension. Treated with a velopharyngeal flap at around 3 years due to short velum. In spite of narrow residual cleft, the patient developed retracted oral articulation. It turned out to be a phonological backing process, which disappeared with speech therapy after some years. Still some audible nasal emission at age 19 years

When we in 1996 made an effort to deal with noticed speech concerns, particularly the retracted oral articulation, we chose to lower the age for HPR. The timing of this surgery was gradually changed from a previous mean age of 8 years down to 3 years. Interestingly, a follow-up investigation did not reveal any significant speech improvements in spite of the earlier HPR (Lohmander et al. 2006).

The same prevalence of retracted articulation errors was found in these patients as in previous individuals, who had been subjected to HPR after age 8 years. Therefore, we concluded that if repair of the residual cleft was to have any potential for improvement of the patients' speech outcome, palatal surgery should be completed before age 3 years.

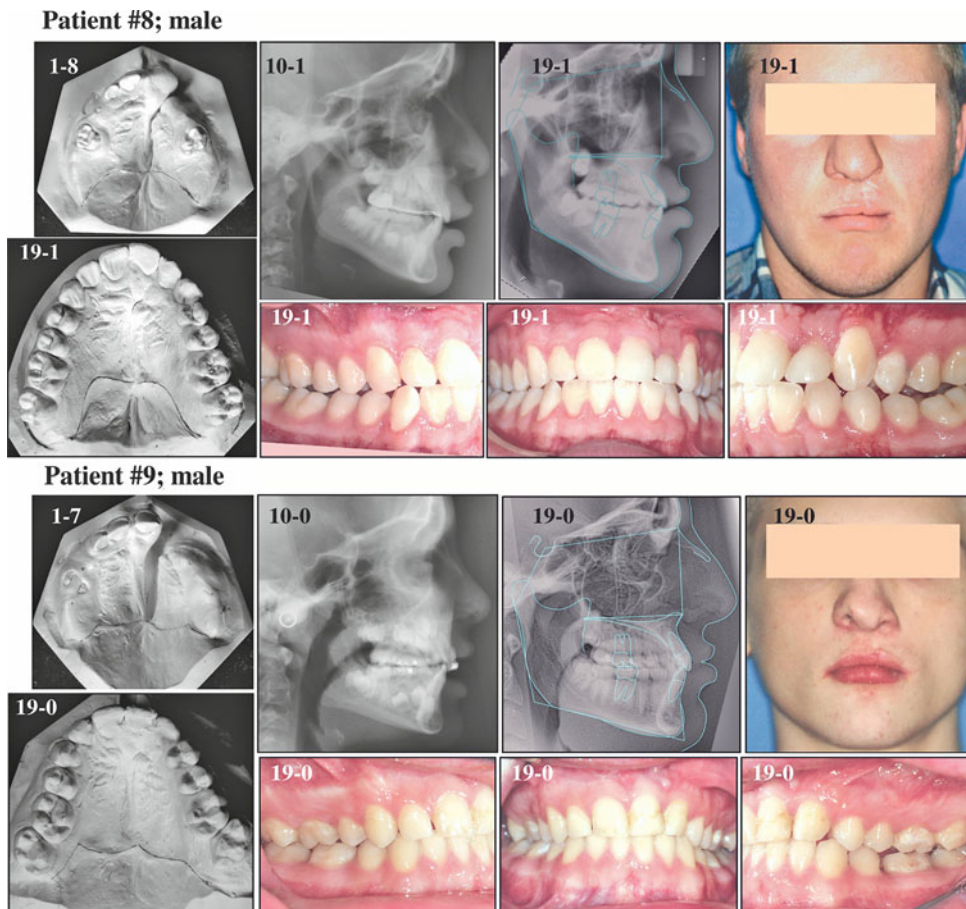


Fig. 18.12 Illustration of records from two representative patients treated with the two-stage method for palatal repair at Gothenburg cleft center. Outcome obtained without maxillary orthognathic surgery. *Upper patient* displays an overall acceptable intermaxillary relationship in spite of slight maxillary deficiency, compensated for by proclination of

the upper incisors, and also causing a tendency to bilateral crossbite. Good speech development since early ages. *Lower patient* demonstrates good maxillary development despite congenitally missing lateral incisor on the cleft side. Quite satisfactory early speech but has developed moderate hypermasality during postpubertal years

Patient #10; male

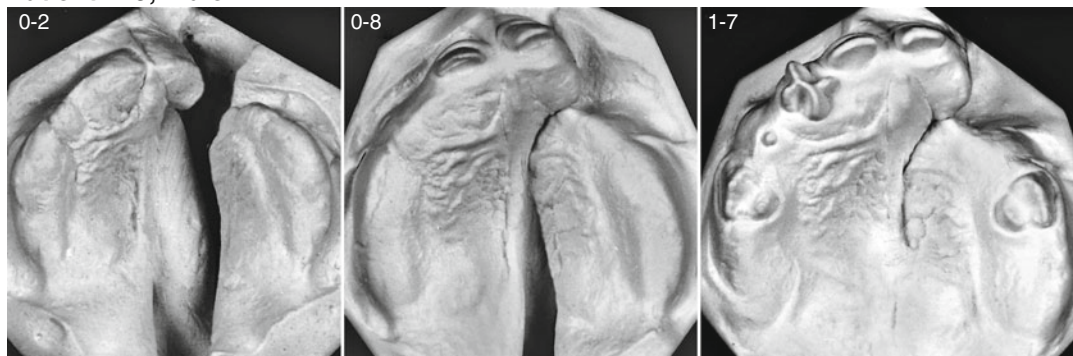


Fig. 18.13 Spontaneous functional closure of the cleft in the hard palate from a series of casts from the same patient. The first cast at age 2 months was obtained before lip

adhesion surgery, the second model from before velar closure, and the third cast from before final lip/nose repair

18.6.3.2 Longitudinal Long-Term Follow-up Results

A longitudinal study of a consecutive series of 55 patients with UCLP, obtained from a total cohort of 65 individuals, completed our follow-up of speech development after the Gothenburg original two-stage palatal protocol as used up to 1995 (SPR at 6–8 months and HPR at 7–9 years) (Lohmander et al. 2011, 2012). Based on obtained standardized audio recordings, blindly analyzed at ages 5, 7, 16, and 19 years, and clinically assessed at 10 years of age, we were able to verify previous findings, both the positive and the compromised ones, regarding our patients’ speech development. Hypernasality, audible nasal emission, and retracted oral articulation occurred in about 30 % of the 5-year-olds, i.e., when their cleft in the hard palate still was unoperated. Generally, all those three speech errors were, however, markedly reduced during the following years, and their prevalence was low at age 16 and 19 years. As a result of these enhancements and other spontaneous speech improvements, the prevalence of VPC was judged as quite satisfactory with a recorded prevalence of 82 % at age 16 and

87 % at age 19 years; at both ages, the patients’ speech intelligibility was judged as normal. It should be added that secondary VPF surgery had been performed in six of the 55 patients (11 %). Our main conclusion was that speech improved in children treated according to our two-stage palatoplasty protocol even before HPR. In addition, further small improvements occurred until early adulthood. An added advantage was that the prevalence of other, more severe cleft speech characteristics, such as symptoms related to VPI and particularly the occurrence of glottal misarticulations, was very low. An almost similar picture can be seen in Fig. 18.14, which displays cross-sectional data for the same ages but with inclusion of twice as many patients at ages 5, 7, and 10 years as investigated in the longitudinal, long-term follow-up. In addition, information at 3 years of age is also incorporated. Our longitudinal results (Lohmander et al. 2011, 2012) together with these cross-sectional data give a trustworthy picture of the long-term speech outcome after the original palatal repair procedures at the Gothenburg cleft center.

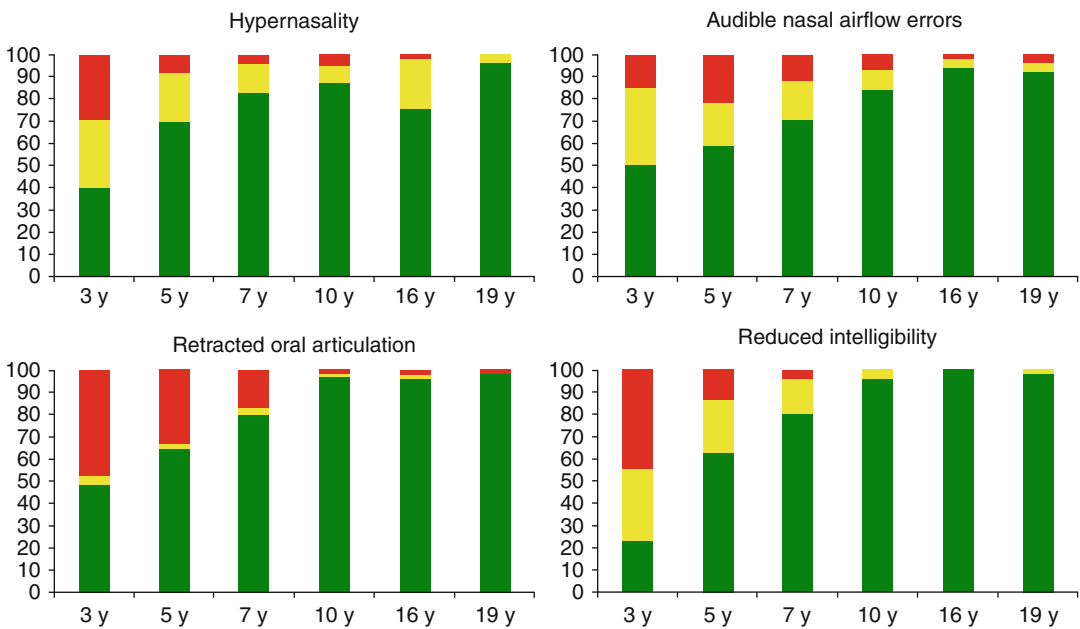


Fig. 18.14 Prevalences (%) of hypernasality, audible nasal airflow errors, retracted oral articulation, and reduced intelligibility in a cross-sectional series of patients born with UCLP. The number of patients included at each age were: 3 years=45; 5 years=93; 7 years=101;

10 years=88; 16 years=50; 19 years=46. A majority of the final patients (37) were included longitudinally from age 5 years (Lohmander et al. 2011). *Green* no/normal; *Yellow* Mild; *Red* Moderate-Severe

18.6.3.3 Retracted Oral Articulations

The speech deviation coined as “retracted oral articulation of anterior pressure consonants,” occasionally called “backing,” was the most common speech error affecting about one third of our preschool children born with UCLP (Lohmander et al. 2011, 2012). Similar speech problems have been reported from other centers and is sometimes considered an atypical phonological process (e.g., Timmons et al. 2001). However, if there still is a coupling between the oral and the nasal cavities, consonants normally produced in a place anterior to the communication may be retracted to a place behind the oronasal opening. With this in mind, the process should be regarded as a compensatory strategy for the inability to create sufficient intraoral pressure to produce high-pressure consonants (Fig. 18.15). This specific speech error is not necessarily incorporated in the child’s internal phonological system and will quite often recede without speech therapy.

18.6.3.4 Results Related to Early Speech Development

An important rationale for early SPR would be that it makes it possible for the patient to achieve increased velopharyngeal activity and oral place of articulation already during the early babbling stage. As an example of such achievements, it can be mentioned that a proportionally high occurrence of oral stops at 12 and 18 months has been reported in studies, which have described

outcomes associated with early SPR at our center (Lohmander et al. 2004, 2011, 2012) as well as in another Scandinavian center practicing the same surgical method (Willadsen and Albrechtsen 2006). The continuity between early consonant production and later speech has been described to occur in noncleft children (e.g., McCune and Vihman 2001) as well as in patients born with cleft palate and treated according to different surgical protocols (Chapman et al. 2003; Lohmander and Persson 2008; Scherer et al. 2008). A high number of consonant types and plosives, particularly with anterior placement during early speech development, were related to articulation accuracy, measured as percentage of correct consonants (Fig. 18.16) (Lohmander and Persson 2008), as well as with the vocabulary development (Scherer et al. 2008). Thus, in babbling, certain characteristics are important for later speech and language development. Early SPR seems to enhance this development.

18.7 The Scandleft Study

At the Zürich, Switzerland, International Symposium on Early Treatment of Cleft Lip and Palate in 1984, there were intense discussions regarding the advantages and disadvantages of different surgical procedures for treatment of patients with cleft lip and palate. Generally, these

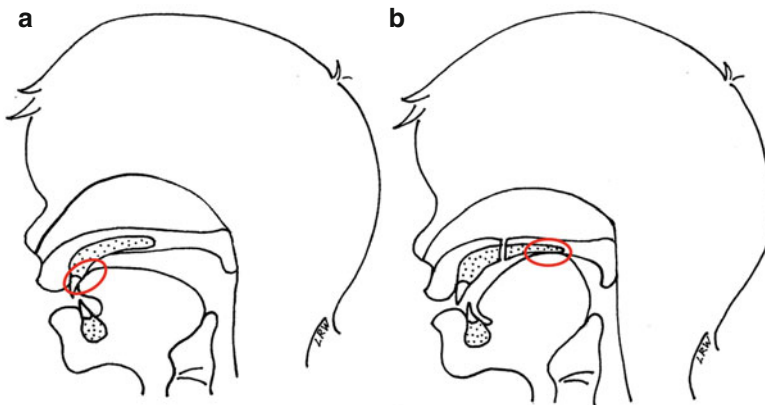
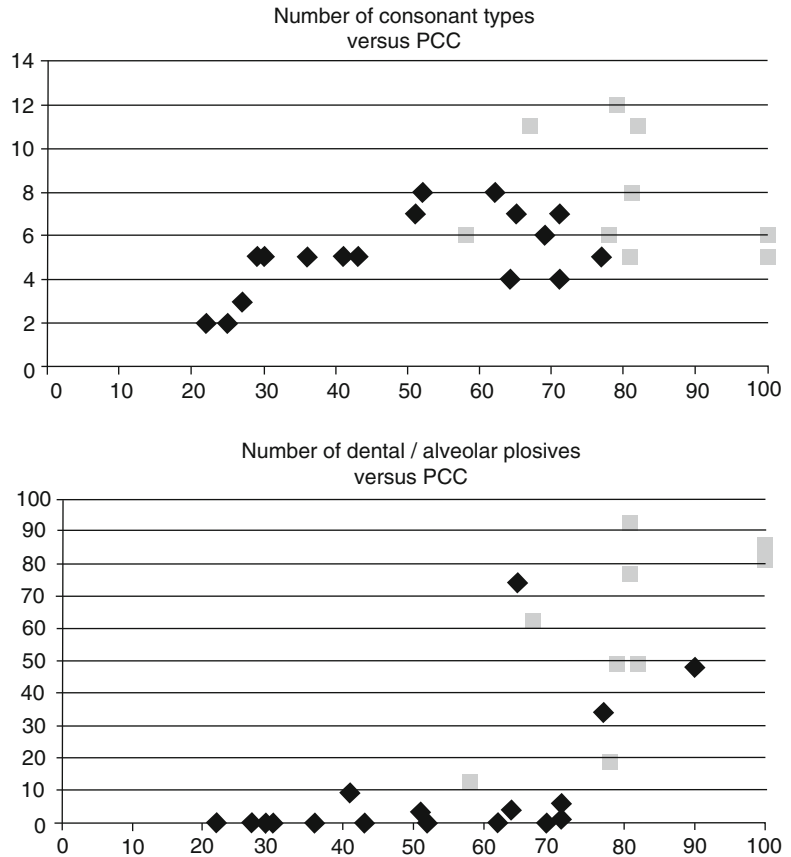


Fig. 18.15 (a) Correct place of articulation for dental/alveolar consonants and (b) retracted to velar placement, behind the oronasal opening. With adequate velopharyngeal function, the place of articulation is posterior to the

ornasal opening but in front of the velopharyngeal port, i.e., a compensatory strategy for the inability to create sufficient intraoral air pressure to produce high-pressure consonants in front of the opening

Fig. 18.16 (*Upper chart*) A high number of consonant types (inventory) and (*Lower chart*) a high number of dental/alveolar plosives at 18 months of age (=y-axis); both correlated significantly with a high percentage of consonants correct (PCC) at age 3 years (= x-axis) ($Rho = .57$, $p < .05$; $Rho = .68$, $p < .01$) in both UCLP (*black*) and comparison group (*gray*) (Lohmander and Persson 2008)



debates triggered growing interests in multidisciplinary inter-center research comparing outcome after different surgical protocols used for CLP treatment. As examples of the new trend, surgical results in four Scandinavian centers were analyzed, especially with regard to maxillofacial development (Friede et al. 1991; Enemark et al. 1993). Members of six European cleft centers made similar efforts (Shaw et al. 1992a,b; Mars et al. 1992; Asher-McDade et al. 1992; Mølsted et al. 1992, 1993a,b; Morrart and Shaw 1996), but here also, aspects on speech development were included in the outcome analyses (Grunwell et al. 2000). However, soon it became obvious that it would be impossible to separate and compare single elements of treatment protocols used at the different centers. Therefore, Scandinavian cleft teams from Helsinki, Stockholm, Linköping, Gothenburg, Copenhagen, Oslo, and Bergen together with two teams from the UK, Manchester

and Belfast, decided to start randomized controlled trials (RCTs) regarding effects of primary surgery (particularly palatal repair) in UCLP. These prospective RCT studies were designed as a set of three parallel trials, where groups of teams would test their local protocols against a common protocol.

This was how the Scandleft study was initiated (Semb 2001). The common protocol was a modification of the Gothenburg two-stage method for palatal repair. The modified surgical detail was that the palatal muscles were dissected free from the nasal mucosa and moved posteriorly. The muscles were sutured together in this position to form a posteriorly placed muscular sling. The nasal layer was left intact and closed with help of the posterior vomer flap. This is an early form of intravelar veloplasty, where the levator muscle was not identified and, consequently, not completely released. In the common protocol

used by all teams, the residual cleft in the hard palate was closed at age 1 year. Inclusion of new patients to the Scandcleft study was stopped 5 years ago and evaluation of the results began in 2012.

The local protocol in Gothenburg will be compared to the common protocol, and the parameter to be studied will be timing of surgery. Therefore, we had to use the modified procedure also in the local protocol, where the residual cleft was closed at 3 years. This leg of the study was performed in Copenhagen and Gothenburg, with the majority of the patients operated in Copenhagen.

18.8 Final Comments and Reflections

As a summarizing evaluation of the Gothenburg two-stage palatal surgery regimen, we have to judge the protocol as quite successful. Both from midfacial growth point of view and regarding speech development, the chosen surgical method has exceeded the expectations we had when the protocol was initiated in 1975.

During the early years after our change of palatal protocol, we introduced several surgical details, which proved important for later outcome. One such feature was where to separate velum from the hard palate at SPR. Preferably, it should be done right at the level of the posterior palatal shelves. Separation further anteriorly would often leave denuded areas of bone, where growth-restricting scar tissue would form. A surgical procedure related to HPR should also be mentioned. It has been described in many papers dealing with so-called pushback palatal surgery. Wide palatal flaps, raised for medial and posterior displacement at repair of the cleft, will often include parts of the thick oral mucoperiosteum covering the maxillary body. Scars in these lateral parts of the palate seem to be especially harmful for later maxillary growth. Such wide flaps should not be used at HPR in a two-stage protocol, because the remaining cleft in the hard palate has usually narrowed substantially after SPR. Therefore, it can be repaired, e.g., by suturing the cleft edges after their mobilization, by

employment of a simple turnover vomer flap, or, alternatively, by use of small flaps, raised in the thin mucoperiosteum close to the cleft. Shifting of these flaps medially, to cover the residual cleft, will result in scars with minimal maxillary growth restriction. Using such an approach would therefore allow for HPR at an early age (15–18 months) in most patients without jeopardizing subsequent maxillary development.

Inclusion of a posteriorly based vomer flap at velar repair seems to be an essential factor to facilitate narrowing of the remaining cleft during subsequent palatal growth and thereby enabling early HPR. In addition, the flap appears to improve the patient's ability to achieve adequate VPC. A likely explanation might be that the repaired velum can attain a more posterior and upward position than if this surgical detail is omitted.

A good velar function after early, successful veloplasty together with substantial reduction in size of the residual cleft in hard palate has been the most important factor to explain a satisfactory speech outcome after two-stage palatal surgery (Van Demark et al. 1989; Persson et al. 2002; Lohmander et al. 2006). Studies at Gothenburg cleft center of 3-year-old patients have revealed that speech developed typically (= as in individuals without a palatal cleft) in about 50 % of the children with UCLP in spite of an unoperated cleft in the hard palate. This outcome compares as good as or even favorably to what have been reported in the literature after *any* palatal surgery protocol (Lohmander 2011). The typical speech error among our patients at this early age was “retracted oral articulation,” which occurred in about 40 % of the 3-year-old subjects. However, occurrence of compensatory articulations related to VPI was very low and absent in the absolute majority of patients, which indicated satisfactory VPC, before the residual cleft in the hard palate had been repaired. Some researchers and experienced clinicians have expressed convictions that such a remaining open cleft would have a negative impact on the patient's velopharyngeal function and make it difficult or impossible to achieve VPC. These beliefs have not been verified though, neither in studies from Zürich nor from

Gothenburg. Actually, a high occurrence of plosives, although retracted to velar place beyond the opening in the hard palate, *presupposes* VPC. Although not wanted, we consider this oral articulation error (retracted oral articulation) much less severe than non-oral misarticulations (glottal and/or pharyngeal) because of its only minor impact on speech intelligibility. Furthermore, a significant number of children with retracted oral articulation improve their speech before school age spontaneously.

Early soft palate closure seems to give a better condition for early consonant development than if the entire palatal cleft is left unoperated until around 12 months of age. Early use of palatal obturators before and after early soft palate repair (6 months) in a two-stage regimen or before palatal closure in a one-stage protocol does not appear to enhance this process (Hardin-Jones et al. 2002; Lohmander et al. 2004). Early hard palate closure at 12 to 18 months of age would possibly reduce the prevalence of the oral speech errors and together with early soft palate closure give the best conditions for speech development taking other aspects of treatment into consideration.

The Gothenburg two-stage surgical protocol as described here is easy to teach and also easy to learn. The first stage, SPR, is based on open dissections of wide and thick flaps without any closed undermining. This makes the procedure a simple, fast, and predictable method with few or no dehiscences postoperatively. In developing countries with limited resources of cleft surgery, orthodontics, or speech therapy, we believe the 2-stage regimen, described by Kontos et al. (2001), will result in better outcome than what is achievable with other standard surgical protocols. If our initial surgery is performed during the patient's first year of life, the outcome will usually be very advantageous. Lip closure and SPR can be combined in one primary operation with good result in a great number of patients, even if the hard palate never is repaired. However, about one-third of the children are likely to need HPR to develop acceptable speech. As the residual cleft is surrounded by virgin tissue, this surgery will be easier and give a predictable, better result

than after closure of a palatal fistula. In most cases, the dental arch will develop with only minor irregularities, such as one or sometimes two deciduous teeth in crossbite on the cleft side, a narrow remaining cleft in the palate and alveolar ridge, and some rotation of the permanent central incisor on the side of the cleft.

The Gothenburg procedure with combined lip closure and SPR has been tested in cooperation with Universities of Campinas and Rio de Janeiro in Brazil, where patients with UCLP were operated on at different ages. The advocated Gothenburg protocol produced good occlusal development and acceptable narrowing of the residual cleft, if the Brazilian children had their surgery done early during their first years of life. However, if patients were older than 3 years, the residual cleft of the hard palate did not narrow very much over time. Therefore, in these cases, HPR could just as well be performed already 6 months after SPR.

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