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Declaration of Interest:

The authors declare no conflicts of interest.

Author Contributions (CRediT):

Benito K. Benitez: Conceptualization, Methodology, Validation, Formal Analysis, Investigation, Writing – Original draft preparation. **Andrzej Brudnicki:** Conceptualization, Methodology, Writing-Review and Editing. **Zbigniew Surowiec:** Conceptualization, Methodology, Writing-Review and Editing. **Ravi K. Singh:** Writing-Review and Editing. **Prasad Nalabothu:** Writing-Review and Editing. **Dieter Schumann:** Supervision, Writing-Review and Editing. **Andreas A. Mueller:** Conceptualization, Methodology, Formal Analysis, Investigation, Resources, Visualization, Writing – Original draft preparation.

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Continuous Circular Closure in Unilateral Cleft Lip and Palate Repair in One Surgery

Summary

The study aims at assessing wound healing and safety of single-stage two-layers continuous closure in patients with unilateral cleft lip and palate (UCLP).

Patients and methods: In this retrospective, descriptive cohort study, we assessed wound healing without fistula formation at 1, 3, and 6 months after a single-stage two-layer UCLP repair, in which the midline suture is continuously circular all along the oral and nasal sides. We examined lengths of hospital stay and the incidence of intra- and postoperative adverse events. Furthermore, we compared the cleft width at birth and on the day of surgery, after presurgical orthopaedics.

Results: Eleven UCLP patients underwent one cleft surgery between July 2016 and June 2018 at the age of 8 to 9 months. Full primary healing occurred in all patients without fistulas. Median length of post-operative hospital stay was 5 days (range = 4–9 days). No intra- or postoperative adverse events above Grade I (according to ClassIntra and Clavien-Dindo, respectively) occurred. Median and interquartile range (IQR) of the palatal cleft width decreased significantly from birth to surgery, i.e., from 12.0 mm (10.8–13.6 mm) to 5.0 mm (4.0–7.5 mm) anteriorly and from 14.0 mm (11.5-15.0 mm) to 7.3 mm (6.0-8.5 mm) posteriorly ($p=0.0033$ in both cases).

Conclusion: Given these preliminary results, the concept of single-stage continuous circular closure in UCLP has potential for further investigation. In particular, it remains to be proven that there are no relevant adverse effects such as inhibition of maxillary growth.

Registered in clinicaltrials.gov:NCT04108416

Keywords: unilateral cleft lip and palate; craniofacial malformation; orofacial cleft; treatment burden; surgical repair,

Introduction

There are wide variations in surgical methods to repair complete unilateral cleft lip and palate malformations (UCLP). A survey conducted in 2000 in 201 centers revealed that 194 different UCLP treatment protocols were applied (Shaw et al., 2001; World Health Organization, 2001). Treatment protocols involving two surgeries are the most common, followed by those involving three surgeries. In rare cases (5%), a single surgical intervention for complete closure is performed (Shaw et al., 2001). A goal of single-stage surgery is to reduce the global healthcare burden of craniofacial anomalies. The World Health Organization recognized the need for "the initiation of clinical trials concerning the specifics of surgery in a developing country setting, one-stage operations, optimal late primary surgery, anesthesia protocols (e.g. local anesthetic, inhalation sedation), and antisepsis" (World Health Organization, 2001). Moreover, a simplified surgical strategy would reduce the treatment burden for children suffering from orofacial clefts, psychosocial stress to the families and caregivers, as well as associated healthcare expenditure.

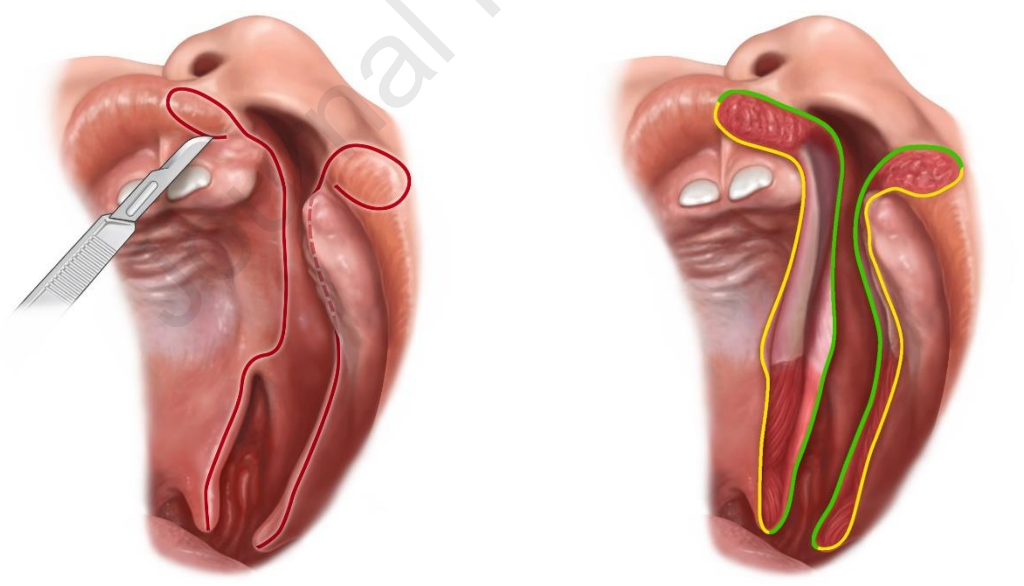
The first techniques for simultaneous repair of UCLP combined lip repair, unipedicled hard-palate repair, and soft-palate repair in adult patients (Farina, 1958). Simultaneous repair is nowadays safely applied in children below 10 months of age also in developing countries (Hodges, 2010). Most surgeons use unipedicled flaps with lateral releasing incisions to close the cleft palate. However, medial transposition of the flaps leads to undesirable raw bone surfaces laterally, with secondary healing (Deng et al., 2002; Guneren et al., 2015; Hodges, 2010; Honigmann, 1996). Even anteriorly, a raw bone surface remains if unipedicled hard-palate flaps are fixed in a pushback position (Savaci et al., 2005).

Bipedicled flaps for cleft palate repair were first described by von Langenbeck (von Langenbeck, 1972). The anterior tips of the bipedicled flaps remain attached to the anterior hard palate even with modern von Langenbeck techniques (Lindsay and Witzel, 1990). The resulting mobility restriction has prompted concerns that bipedicled flaps cannot cover anterior defects or a wide cleft (Losee and Lin, 2014). Furthermore, intentional anterior palatal openings remain after a von Langenbeck procedure (Lindsay, 1971). Nevertheless, bipedicled flap techniques have produced consistently good growth results, as shown in retrospective multicenter studies (Ross, 1987; Shaw et al., 1992) and in a randomized controlled study (Semb et al., 2017). Hence, a novel method of simultaneous lip and palate closure using bipedicled flap designs should allow safe closure of the anterior palate. This is possible using the method described by Dudkiewicz and colleagues (Brudnicki et al., 2014; Fudalej et al., 2010). This technique further allows for a gapless separation of the oral and nasal cavities and primary wound closure over the complete oral layer.

However, an open wound remains nasally with a single-layer closure at the transition between the hard and soft palate. This results from the need to transect the nasal mucosa and palatine aponeurosis along the posterior border of the hard palate towards the pterygoid process (Lindsay, 1971). However, complete closure without fistulae depends crucially on the healing of the mucosal layer of the nose.

To the best of our knowledge, there has been no description of simultaneous cleft lip and palate repair achieving a continuous two-layer separation of the oral and nasal cavities. The rationale of our technique was to avoid two known growth-inhibiting side effects: (1) open wounds as zones of secondary healing and (2) surgical manipulation of the alveolar segments. Figure 1 shows the single-stage continuous circular two-layer UCLP repair performed in the midline. This contrasts with current concepts of stepwise cleft closure with varying extents of open wounds, secondary healing and concomitant scarring.

We aimed to preliminary evaluate the wound healing and safety of one cleft surgery with a continuous circular two-layer wound closure in patients with UCLP.



a b

Fig 1. Visualization of a unilateral cleft lip and palate. (a) Incision outline for a single-stage continuous circular two-layer closure in the midline. (b) Visualization of the wound edges for continuous circular suture all along the oral (yellow) and nasal (green) sides. (Visualization Andreas A. Mueller and Markus Voll)

Patients and Methods

Study design and patient characteristics

We report according to the STROBE guidelines for cohort studies (von Elm et al., 2007). In this retrospective, descriptive cohort study, we assessed a single-stage two-layer continuous circular UCLP repair after passive plate therapy. All children had a nonsyndromic UCLP without Simonart's band. Patients were operated at our department by the last author (A.A.M.) between July 1, 2016, and June 30, 2018. All parents and guardians signed an informed-consent form for the surgical procedures and for releasing medical information and photographs for scientific purposes. The study was performed in accordance with the Declaration of Helsinki after obtaining approval from the Ethics Committee of Northwest and Central Switzerland (EKNZ; project IDs: EKNZ Req-2017-00902 and 2018-01561). The study was registered in clinicaltrials.gov (NCT04108416), in accordance with the IDEAL recommendations for surgical innovations (McCulloch et al., 2009).

For our first aim, we assessed wound healing without fistula formation by the absence of nasal food leakage and inspections at 1, 3, and 6 months postoperatively. For our second aim, we examined the length of hospital stay and the incidence of intra- and postoperative adverse events. For our additional aim we compared the cleft width between plaster casts at birth and on the day of surgery.

Surgical procedure

Surgical intervention took place when the infants were at least 8 months old and weighed around 8 kg. We placed the infants in supine position with their head elevated to reduce postural blood stasis in the operation field. We administered a single dose of methylprednisolone (2.5 mg/kg body weight, i.v.) to reduce surgical and laryngeal swelling. Infection prophylaxis consisted of amoxicillin and clavulanic acid (50 mg/kg and 5 mg/kg body weight, respectively) administered for 72 h postoperatively. Cuffed endotracheal tubes were used, with the cuff inflated as little as possible and accompanied by a throat pack. We used Octenisept® for extraoral, intraoral, and endonasal disinfection. At surgery, long-acting anesthetic blocks were administered behind the palatal tuberosity and the infraorbital nerves (0.25% levobupivacaine, maximum 1 mL/kg body weight), and 0.9% saline with adrenaline (10 µg/mL) was administered for hydrodissection underneath the mucosa and periosteum and prior to cleft muscle dissection to reduce bleeding. Appendix A describes the detailed surgical technique with continuous circular two-layer wound closure. Appendix B provides a video supplement documenting the surgical technique for hard-palate and soft-palate repair.

Appendix B: Video with surgical technique step-by-step part 1: hard-palate and soft-palate repair

We made an incision outline to mobilize bipedicled flaps on the cleft and healthy side. On the healthy side, the mucoperiosteal flap was designed to cover the palatal shelf and curved vomer to achieve a balanced split of the mucosa for closure of the nasal and oral layers (Benitez et al., n.d.). Medial pterygoid periosteal detachment assured complete mobility of the nasal mucosa at the hard-soft palate junction. Appendix C shows a three-dimensional model of the incision outline. We closed the nasal layer in the hard palate from posterior to anterior direction. Prior to reorientation of the cleft muscles, we sutured the nasal mucosa of the soft palate to the suture of the nasal layer of the hard palate without leaving a gap. The suture ran posteriorly to the uvula. Subsequently, the muscles were dissected, and the palatopharyngeus and levator muscles were reoriented and sutured transversely in the middle third of the soft palate. We sutured the oral mucosa of the soft and hard palates as well as the lateral surgical access incisions to allow primary healing. Figure 2 illustrates the incision outline, palatal wound closure, and mucosal conditions after primary wound closure.

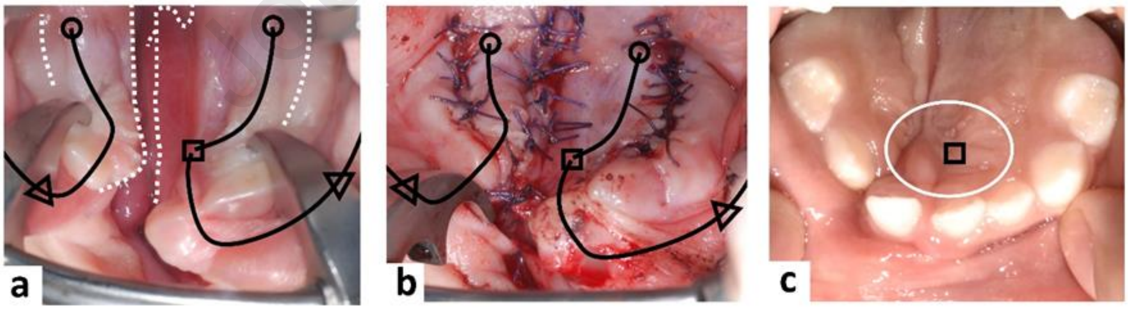


Fig 2. Cleft palate repair using bipedicled hard-palate flaps and continuous circular two-layer wound closure. (a) Complete unilateral cleft lip and palate at surgery at 8 months of age. The palatal vascular territory, supplied by the palatine arteries (**o**) and its nasopalatine artery (NPA) (**◘**) on the healthy side, connects (—) across the alveolar ridge with the labiofacial vascular territory (**Δ**) on both sides of the cleft. The incision outline (---) is shown for a two-layer closure of the hard palate using a vomer turnover flap and bipedicled palatal flaps. Preserving the anterior attachment of the palatal flaps allowed the anastomosing vascular connection between the palate and the labiofacial territory to be maintained. (b) Wound conditions at the end of palate repair and before lip repair. Lifting the bipedicled flaps without transposing them allows for complete primary wound closure in the midline and over the lateral surgical access incisions. Posteriorly, the palatine arteries are maintained as well as the nasopalatine artery and nasopalatine nerve at the incisive foramen on the healthy side. (c) Palate conditions at 2.5 years of age. No scarring in the anterior junction zone (white circle) around the area of the preserved NPA (**◘**).

After removing the mouth gag, we completed two-layer closure in the alveolar cleft area. Cleft lip dissection and reconstruction comprised primary rhinoplasty. Nasal shape definition was supported by nostril stenting by a silicon sheet (0.5 mm) and transmural fixation to eliminate dead space. Appendix D provides a video supplement with the surgical technique for the repair of alveolar, lip, and nose. Standard protocol included extubation in the operating room at the end of surgery. After the surgery, the children could be fed with milk or porridge immediately. No arm restraints or feeding tubes were used. Nostril retainers placed later than 1 week postoperatively were used for 4 months, but some patients or parents/guardians refused their use. All patients were followed up at 1, 3, and 6 months postoperatively and assessed for nasal food leakage and fistula formation.

Appendix D: Video with surgical technique step-by-step part 2: alveolar, lip, and nose repair

For our second aim we assessed patient records for intra- and postoperative adverse events classified above Grade I, as well as lengths of hospital stay. For classification of intraoperative events ClassIntra (version 1.0) a prospectively validated classification system was used (with a grading from 0 to V, Grade 0 defines no deviation from the ideal surgical course and Grade V defines a deviation leading to intraoperative death of the patient) (Dell-Kuster et al., 2020). For postoperative complications the Clavien-Dindo classification was used (with a grading from 0 to V, Grade 0 defines no complications from the normal postoperative course and Grade V leading to the patient's death) (Dindo et al., 2004).

Presurgical orthopedic treatment with passive plate

After birth, all children underwent passive palatal plate therapy with nasal extension as described previously (Koželj, 2000, 1999; Nalabothu et al., 2020). Lip taping was used in addition (DynaCleft®, Southmedic, Ontario, Canada). The plate typically became unstable after 3–5 months and was renewed. With an orthodontic caliper, we obtained linear measurements on the maxillary impression plaster casts at the beginning and end (day of surgery) of plate therapy (Zurich model®, Art. 215-33, Otto Leibinger, Mühlheim, Germany). Figure 3 illustrates the palatal cleft width (pc), true cleft width (tc), and curved vomer width (cv) measured in the anterior and posterior cleft areas.

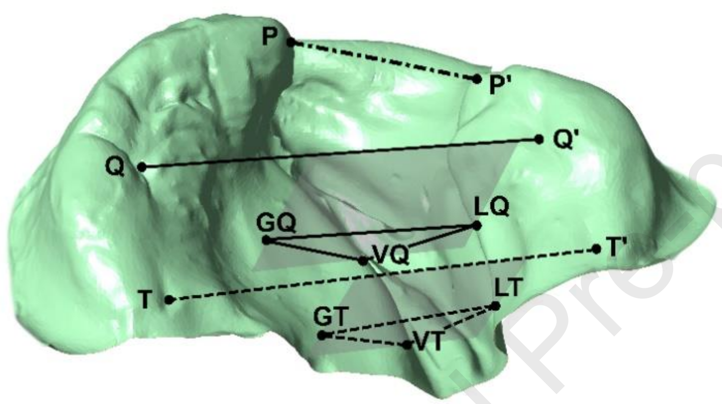


Fig. 3. Three-dimensional surface of a cast with reference points marked. Definitions of the reference points (Braumann et al., 2003, 2002; Nalabothu et al., 2020; Shen et al., 2015): Q and Q', gingival groove points (intersection of the gingival groove and lateral sulcus); T and T', posterior shelf pits (posterior end of the lateral sulcus); P and P', pole points (cleft edges of the alveolar ridges). A midpalatal-section plane through QQ' (perpendicular to QQ'T) defined: GQ by crossing the greater segment's junction to the vomer, VQ by crossing the vomer edge and LQ by crossing the lesser segment's shelf ridge. In the same way, the posterior-section plane TT' defined GT, VT and LT. In bilateral pairs of points, the prime (') indicates the point on the cleft side. T and T' were allocated in the depth of the lateral sulcus instead of the top of the alveolar ridge for better traceability (Brief et al., 2006; Seckel et al., 1995). The palatal cleft width (pc) was measured from GQ to LQ and from GT to LT, the true cleft width (tc) was measured from VQ to LQ and from VT to LT, and the curved vomer width (cv) was measured from GQ to VQ and from GT to VT.

Statistical analysis

We used Wilcoxon signed-rank test for within-group comparisons of cleft width measurements. Statistical significance was assumed at $p<0.05$. Statistical analysis was performed using Stata (version 15.1, StataCorp, College Station, TX, USA).

Results

From the medical records, eleven patients were assessed as eligible and could be included and analyzed. Table 1 shows the patient characteristics.

Table 1. Characteristics of patients with complete unilateral cleft lip and palate (n = 11)

Characteristic	Value
Sex	
Male	8 (73%)
Female	3 (27%)
Side of unilateral cleft lip and palate	
Right	6 (55%)
Left	5 (45%)
Gestational age at birth, weeks	40 (39–41)
Birth weight, g	3500 (3200–3765)
Age at start of plate therapy, days	1 (1–13)
Age at initiation of second plate, weeks	18.4 (15.0–22.4)
Age at surgery, weeks	35.4 (33.0–37.7)
Body weight at surgery, g	8300 (8000–8400)

Data are n (%) or median (IQR) values.

Surgical procedure

Full primary healing occurred in all patients during the early postoperative phase. No fistula was formed, as confirmed by inspections at 1, 3, and 6 months postoperatively and by the absence of any transient nasal food leakage after surgery. Figure 4 illustrates the healing stages in a representative patient.

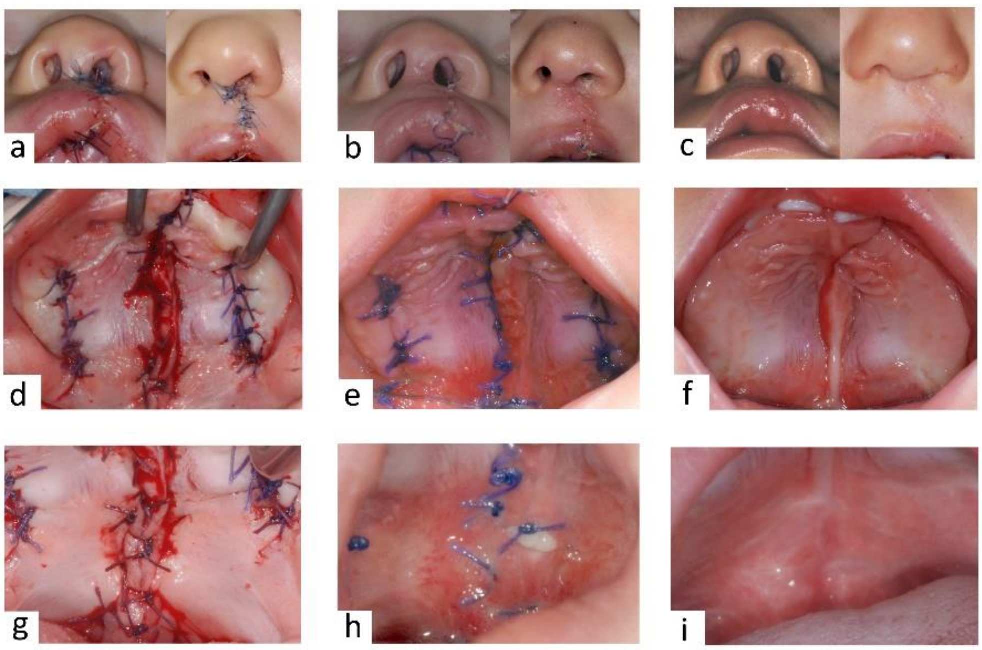


Fig 4. Postoperative findings after cleft repair in one single surgical intervention with continuous circular closure. Healing conditions at the end of surgery (a, d, g), at 7 days postoperatively (b, e, h), and at 6 months postoperatively (c, f, i). Alar convexity and nostril symmetry and patency were retained at 6 months postoperatively (a, b, c), accompanied by a history of night-time nasal breathing. The palatal vault convexity at 1 week (e) and 6 months (f) postoperatively was similar to that seen preoperatively. Palatal mucosa relief (rugae palatinae and papilla incisiva) of the anterior palate was fully maintained. Lateral surgical access incisions healed primarily and left inconspicuous scars. Soft palate (g, h, i) healed with a single linear scar.

Median length of post-operative hospital stay was 5 days (range = 4–9 days). No intraoperative adverse events above Grade I of the ClassIntra classification occurred (any deviation from the ideal surgical course: without the need for any additional treatment or intervention, patient with no or mild symptoms (Dell-Kuster et al., 2020). Postoperative complications showed a maximum of Grade I according to Clavin-Dindo, requiring no pharmacological or surgical treatment besides antiemetic, analgetic or antipyretic drugs (Dindo et al., 2004). Grade II complications such as the need for nasogastric feeding or blood transfusions did not occur. In particular, there were no adverse events requiring prolonged intubation or reintubation. Median hemoglobin level at end of surgery was 96.0 g/L (IQR = 92.0–98.0 g/L).

Presurgical orthopedic treatment with passive plate

From birth to surgery, median width of the anterior palatal cleft (pc) decreased by 7.0 mm, and the median width of the anterior true cleft (tc) decreased by 5.3 mm. Both changes were statistically significant (Table 2). In the posterior area, we also achieved statistically significant median reductions of palatal (6.7 mm) and true (3.5 mm) cleft width, respectively (Table 2). In contrast, the widths between the gingival groove points (Q to Q'; p=0.25) and posterior shelf pits (T to T'; p=0.30) remained almost unchanged. Table 2 summarizes the measurements illustrated in Figure 3.

Table 2. Palatal cast measurements after birth and after preoperative plate therapy at the time of surgery (n=11)

Variable	**Measure**	**Value after birth[+], mm**	**Value at time of surgery[‡], mm**	**Wilcoxon signed-rank test p-value**
Width of alveolar cleft ridges*	P to P'	12.0 (10.8–13.6)	5.0 (4.0–7.5)	0.0044
Width between gingival groove points	Q to Q'	27.0 (25.2–28.5)	26.0 (25.0–27.5)	0.2452
Anterior palatal cleft width*	GQ to LQ	12.0 (11.0–15.0)	5.0 (4.5–7.0)	0.0033
Anterior true cleft width*	VQ to LQ	7.3 (4.0–9.0)	2.0 (0.5–2.5)	0.0038
Anterior curved vomer width	GQ to VQ	6.5 (5.0–7.2)	5.0 (4.0–6.0)	0.0675
Width between posterior shelf pits	T to T'	30.0 (27.5–30.5)	30.0 (28.0–32.0)	0.3025
Posterior palatal cleft width*	GT to LT	14.0 (11.5–15.0)	7.3 (6.0–8.5)	0.0033
Posterior true cleft width*	VT to LT	6.0 (4.3–7.0)	2.5 (1.5–3.5)	0.0066
Posterior curved vomer width*	GT to VT	8.5 (7.5–9.0)	6.0 (5.0–7.0)	0.0044

Data are median (IQR) values. [+] Age 1 day (1-13). [‡] Age 35.4 weeks (33.0-37.7). * indicate that values after birth and at time of surgery differed at the 0.05 level of significance according to Wilcoxon signed-rank test. Q and Q', gingival groove points (intersection of the gingival groove and lateral sulcus); T and T', posterior shelf pits (posterior end of the lateral sulcus); P and P', pole points (cleft edges of the alveolar ridges). A midpalatal-section plane through QQ' (perpendicular to QQ'T) defined: GQ by crossing the greater segment's junction to the vomer, VQ by crossing the vomer edge and LQ by crossing the lesser segment's shelf ridge. In the same way, the posterior-section plane TT' defined GT, VT and LT. In bilateral pairs of points, the prime (') indicates the point on the cleft side.

The reduction of palatal cleft width from birth to the time of surgery mainly occurred anteriorly, due to a marked reduction of true cleft width, while the width of the curved vomer remained almost unchanged. The continuous support of the alar rim by the ovoid acrylic extension led to the alar cartilage maturing into a more convex shape. Figure 5 illustrates the morphologic changes resulting from presurgical plate therapy.

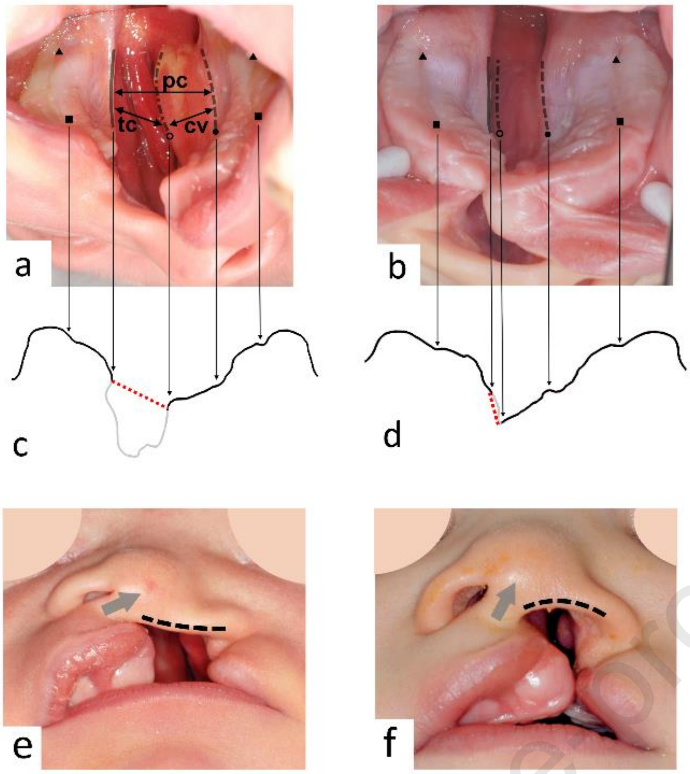


Fig 5. Morphologic changes during presurgical orthopedic therapy. Cleft morphology at birth (a) and at 8 months after passive plate therapy and lip taping (b). The palatal cleft width (pc) lies between the lesser segment's shelf ridge (—) and the greater segment's junction to the vomer (– –). This junction is indicated by the transition in the color of the mucosa from pink to red (Veau and Borel, 1931). The true cleft width (tc) lies between the lesser segment's shelf ridge (—) and the vomer edge (– ▪ – ▪). The width of the curved vomer (cv) comprises the area between the vomer edge (– ▪ – ▪) and the greater segment's junction to the vomer (– –). Thus, the true cleft denotes the cleft width of the fissure into the nose, whereas the palatal cleft denotes the gap in the palatal mucosa. The cv (○ to ●) and the separation between lateral sulci remained almost stable over time (▲ to ▲, ■ to ■). Coronal cross-section through the corresponding plaster model at birth (c) and at 8 months (d). The true cleft narrowed significantly, and its entrance plane changed from oblique (c, ···) to more vertical (d, ···). The shape of the ala on the cleft side changed from a concave (e, – –) to a convex (f, – –) curvature, and the tilted columella straightened up (e, f, →).

Discussion

UCLP repair is still mostly performed in multiple stages (Shaw et al., 2001). Inevitably, this leads to an opening of the created wound space between the operated and nonoperated areas. There, secondary healing takes place with a tendency to scarring. To prevent secondary healing, combined two-layer closure along the entire cleft lip-alveolar and palate border must be performed in a single surgery. However, with current one-stage techniques, it is not possible to achieve continuous circular two-layer closure of the oral and nasal cavities with primary healing (Brudnicki et al., 2014). Our hypothesis was that simultaneous cleft lip and palate repair can be accomplished in a single surgical intervention with continuous circular two-layer wound closure.

UCLP deformity was reliably closed in one single surgery, followed by continuous circular two-layer closure along the entire oral and nasal surfaces, with preservation of the anterior palatal neurovascular supply. The surgical technique reliably produced a fully closed soft-tissue envelope at the end of surgery. The gingiva-periosteal layer of the alveolar process remained untouched, but oro-nasal communication in the alveolar cleft area was closed in two layers. At surgery, our study patients had a median age of 35.4 weeks and a median weight of 8.3 kg. We refrained from performing simultaneous closure of ULCP in the infants before 8 months of age although this is potentially feasible and safe (Hodges, 2010). This was done to facilitate developmental maturation of the child and tissue maturation to cope with surgery, healing, and recovery. Furthermore, between 8 and 10 months of age, the unmineralized, permanent tooth buds within the bone are well protected from surgery-related injury (Broomell, 1910; Lekkas et al., 2000).

The dissection plane in the hard and soft palates along the medial pterygoid plate lay in a subperiosteal plane. However, to simplify our palatoplasty, the soft palate muscle dissection may be further modified, using a small double-opposing z-plasty (Yamaguchi et al., 2016), which has shown favorable healing and speech outcome in a large patient sample. We made no transversal cuts in the anterior palatal region and at the junction between the hard and soft palates. Palatal vascular injections in fetuses (Bosma and National Institute of Dental Research, 1986) and neonates (Wilhelm, 1969, 1967) with and without clefts revealed that there are abundant vascular anastomoses between the nasopalatine and greater palatine artery as well as across the alveolar ridges between the greater palatine artery and vestibular branches of the superior labial artery (Figure 2a). Thus, our technique maintains the natural connection of the vascular territories between the lip (Mueller et al., 2012), alveolar, and hard-palate and soft-palate regions. In addition, we can assume that the sensitivity of the hard palate is preserved because sensitive nerves run parallel to the nasopalatine and greater palatine vessels. Moreover, the anterior part of the palate is the normal resting position for the tongue. Maintaining full sensation in the anterior palate might facilitate correct tongue position when speaking (Whitehill, 2002), at rest, and when swallowing, and further, the tongue's pressure on the palate is an important natural force for encouraging growth of the face. The anterior part of the palatal shelves has intrinsic tissue deficiency in patients with UCLP even if no surgery is performed (Latief et al., 2012) and is prone to growth inhibition after surgery (Berkowitz et al., 2005; Trotman et al., 1993). Moreover, downward remodeling of the anterior and posterior palatal regions to the same extent is essential for harmonious growth (Enlow, 1996).

Since the curved vomer lies in a more horizontal plane, it does not narrow the anterior region during plate therapy. In contrast, covering the curved vomer with palatal flaps would lead to a lack of tissue and problems for complete wound closure despite preoperative plate therapy. To select the optimal time point of hard-palate surgery solely on the basis of the ratio between palatal cleft area and total palate area must therefore be generalized with caution (Berkowitz et al., 2005). Suturing between the edges along the true cleft allows for complete wound closure, minimizes the need for tissue elevation and tissue shifting, minimizing the wound between the curved vomer bone and its overlying mucoperiosteum. Unnecessary scarring from repetitive surgery or secondary wound healing as well as vascular destruction in the anterior palate must be avoided to minimize interference with the natural growth potential.

Median length of hospital stay of 5 days (range = 4–9 days) after combined UCLP repair compares well with the mean of 5.82 days (range = 1–10 days) reported in a randomized, controlled study of variable two-stage protocols (Bannister et al., 2017) involving mostly lip and soft-palate closure. However, mean postoperative stay was 5.96 days, even in the group receiving isolated lip closure (Bannister et al., 2017). The healthcare system in which the first authors work reimburses cleft surgical procedures to the hospital based on diagnosis-related groups (DRG). Normal reimbursement after palatal surgery occurs if the patient is discharged between postoperative days 1 and 5 (expected mean of 4.2 hospital days). Reimbursement is the same regardless of whether the lip is operated in addition to performing the palate surgery. The healthcare system in which author R.K.S. works does not reimburse combined lip and palate surgery in patients younger than 8 years. Consequently, the DRG system does not reimburse the expenses for prolonged anesthesia due to single-stage UCLP repair. Additionally, single-stage surgery is associated with fewer reimbursements because the patient does not return for a second, third, or fourth step of UCLP repair. The number of reimbursed procedures is reduced by 50%, 66%, and 75% compared to two-stage (Semb et al., 2017), three-stage (Gundlach et al., 2013), and four-stage (Nadjmi, 2018) treatment protocols, respectively. Although the total treatment costs for combined UCLP repair are lower, poor reimbursement strategies clearly hamper implementation of single-stage UCLP surgery.

In total, 5 of the 11 patients came from a place outside our normal referral area. These parents specifically requested a single surgical intervention. The reasons expressed by the parents were to minimize surgical burden for the child and psychosocial stress on the family associated with the upcoming treatment. In our study, parents accepted lip repair at a later time than usual, with the benefit of their child having to undergo only a single surgical intervention.

In our study, patients underwent functional palatal plate therapy with a lower treatment burden compared to presurgical alveolar molding (Alfonso et al., 2020). Median width of the alveolar cleft ridges (P to P') decreased significantly from 12 mm to 5 mm in the period between birth and surgery onset (Table 2). However, there was a variable residual gap between the alveolar segments (IQR = 4.0–7.5 mm), since passive plate therapy relied solely on the functional interplay of the tongue, palate, and lip. Thus, the margins of the alveolar segments usually do not come into contact before surgery. However, contact of the alveolar segments was not necessary to achieve continuous and complete wound closure in two layers across the alveolar cleft region, since the alveolar mucosa was not implicated for closure.

Presurgical palatal plate treatment led to significant narrowing of the anterior and posterior true cleft widths before surgery (anterior, $p=0.0038$; posterior, $p=0.0066$). In addition, plate therapy provided the possibility of using a nasal stent to improve nasal symmetry (Kozelj, 2007). However, long-term effects of the presurgical nasal molding remain controversial (Van Der Heijden et al., 2013). The anterior palatal cleft (GQ to LQ) was reduced significantly before surgery, but this was caused by the significant reduction of the true cleft (VQ to LQ), while the curved vomer (GQ to VQ) remained unchanged. The width of the true cleft was consistently reduced to less than 3 mm (IQR=0.5–2.5 mm). Thus, maximal benefit from presurgical plate therapy increased if surgical closure was restricted to the true cleft. Because the plane of entrance was almost vertical, only minimal transversal tissue shift was necessary (Fig 3b, d). Presurgical passive plate therapy reduced the need for tissue mobilization during palatal surgery and made it unnecessary to perform an early lip surgery to narrow the cleft palate. Therefore, we could perform lip surgery in conjunction with palatal surgery. This improved the benefit–burden ratio of UCLP management compared to staged protocols.

With our method, the fit of the plate was maintained well over several months without having to perform regular plate adaptations. This is in contrast to other forms of orthopedic plates applied presurgically, such as the Hotz plate (Hotz et al., 1978), dento-maxillary advancement appliance of Latham

(Latham, 1980), or nasoalveolar molding appliances (Grayson et al., 1999; Grayson and Cutting, 2001). These appliances and their modifications aim to actively mold the alveolar arches by performing regular grinding and adaptation of the plate every few weeks. This requires frequent consultations, which increases the overall treatment burden for patients and their families (Singer et al., 2018).

In our study, transversal width of the alveolar segments between QQ' and TT' remained constant during the period of plate therapy. Thus, three-dimensional position of the main contact zone of the plate remained stable. In terms of plate stability, the narrowing of the segments towards each other was compensated by expansive bone remodeling (Enlow, 1996). The plate prevented the tongue from entering the fissure of the true cleft. This led to new force equilibrium of the lip, tongue, and palate segments and the observed morphological adaptation. However, in the first months, the volume of the alveolar ridge itself increases. After 4 to 5 months, this resulted in instability of the plate, which required its renewal.

Our findings are consistent with those of investigations using the same type of preoperative therapy (Koželj, 1999). In patients with palatal cleft, transversal dimensions of the alveolar segments are wider than normal at birth. Koželj showed that without plate therapy, there is no spontaneous narrowing in the period up to 6 months of life.

To bring the alveolar segments into contact before primary surgery, additional extrinsic forces are required (Grayson et al., 1999). This leads to an increased treatment burden with frequent visits for plate adjustments, risk of tissue pressure sores (Levy-Bercowski et al., 2009), or interventions under general anesthesia (Shay et al., 2015). The attempt to bring the alveolar segments into contact before primary repair is meaningful if gingivoperiosteoplasty is planned at the same time (Hopper and Al-Mufarrej, 2014). However, gingivoperiosteoplasty (Wojtaszek-Slominska et al., 2010) and early alveolar ossification (Berkowitz et al., 2004; Eppley, 1996) have been reported to increase the risk of a negative growth effect. Furthermore, the effectiveness of gingivoperiosteoplasty for promoting bone formation remains uncertain (El-Ashmawi et al., 2018; Wang et al., 2016). We therefore refrained from performing gingivoperiosteoplasty, even in cases where the alveolar segments were in contact after passive plate therapy.

The Dutchcleft study tested the effects of a preoperative Hotz-type plate in a randomized controlled trial in 24 patients (Prahl et al., 2001). In contrast to traditional assumptions, plate therapy did improve neither feeding (Prahl et al., 2005) nor parent satisfaction (Prahl et al., 2008). Furthermore, in a protocol using staged repair of UCLP, plate therapy had neither a positive nor a negative influence on the maxillary form (Bongaarts et al., 2006; Noverraz et al., 2015). In a randomized, controlled study, using a nasoalveolar molding plate is expected to have a lasting positive effect on maxillary form (Shetty et al., 2017). Therefore, no negative permanent effect is to be expected from the plate itself. The Dutchcleft study concluded that plate therapy to improve the form of the maxillary arch can be abandoned because combined lip and palate surgery overrides the effect of preoperative plate therapy (Prahl et al., 2001). This recognizes, that preoperative plate therapy followed by isolated lip surgery does not contribute anything to the palate surgery. However, before lip surgery they found significant reductions of the alveolar, midpalatal, and posterior cleft widths when using plate therapy (Prahl et al., 2001).

Limitations and strengths of the study

The impact of our study is limited by the small number of patients, short follow-up period, and retrospective nature of this investigation. Comprehensive analysis of advantages and disadvantages of a specific treatment protocol requires assessment of all aspects of the final outcome (Allori et al., 2017) up to the end of growth and treatment. An intercenter study (Fudalej et al., 2019; Urbanova et al., 2016) using a similar single-surgery method without oral lateral raw surface but involving a raw surface in the soft-palate nasal layer showed a slightly more favorable growth outcome than staged lip and palate repairs at the patients' age of 10 years. Although the age of 10 years is too early to predict final growth outcome, relative growth ranking between the protocols used in intercenter studies remained stable between the ages of 9 and 20 years (Brattstrom et al., 2005; Semb et al., 2005). We assume that without preoperative plate therapy, the same surgical technique would necessitate undesirable broader tissue mobilization with a larger wound. However, it remains unclear as to what effect wider tissue mobilization, necessary to achieve tension-free closure of the cleft, will have on short- or long-term results.

In terms of study strengths, our surgical technique respected the blood microcirculation in the palate, especially in the anterior palate and labioalveolar junction. Further, it combined minimal tissue tension and primary healing. Long-term follow-up is needed to verify whether our surgical technique is consistent with the conclusion of Ross that "there is every indication that for facial growth the most simple treatment is as effective as any other"(Ross, 1987). As the lateral access incisions were completely closed at the end of surgery, it seems technically feasible to avoid these incisions (Brusati, 2016; Brusati and Mannucci, 1994; Li et al., 2021; Ogata et al., 2017) and replace them with submucosal periosteal incisions (Kobayashi, 2010). However, to date we have not implemented this technique in our standard protocol to avoid prolonging the surgical procedure.

Our findings confirm that presurgical passive plate therapy with low treatment burden resulted in a narrowing of the cleft palate. This led to morphologic conditions that facilitated continuous two-layer closure in a single surgery. Contact of the alveolar segments was not necessary for achieving continuous and complete wound closure in two layers. The biologically reasoned technique completely avoided secondary wound healing and surgical manipulation of the alveolar segments and respected the microcirculation of blood vessel supply of the palate. UCLP deformity was reliably closed in one cleft surgery followed by continuous circular two-layer wound closure along the entire oral and nasal surfaces. No surgical or anesthesia-related adverse events occurred.

Total treatment costs for primary repair combining lip and palate repair in a single surgery are lower than those for staged repair protocols. However, poor reimbursement conditions render single-stage surgical protocols economically unattractive for hospitals and hamper their implementation. Given our preliminary results, the concept of single-stage continuous circular closure in UCLP has potential for further investigation and requires long-term evaluation.

Appendix A. Detailed surgical procedure

Appendix B. Video with surgical technique step-by-step part 1: hard-palate and soft-palate repair

Appendix C. Three-dimensional incision outline, marked on a scanned model of a unilateral cleft lip and palate

Appendix D. Video with surgical technique step-by-step part 2: alveolar, lip, and nose repair

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Figure legends

Fig 1. Visualization of a unilateral cleft lip and palate. (a) Incision outline for a single-stage continuous circular two-layer closure in the midline. (b) Visualization of the wound edges for continuous circular suture all along the oral (yellow) and nasal (green) sides. (Visualization Andreas A. Mueller and Markus Voll)

Fig 2. Cleft palate repair using bipedicled hard-palate flaps and continuous circular two-layer wound closure. (a) Complete unilateral cleft lip and palate at surgery at 8 months of age. The palatal vascular territory, supplied by the palatine arteries (**o**) and its nasopalatine artery (NPA) (**◘**) on the healthy side, connects (**—**) across the alveolar ridge with the labiofacial vascular territory (**Δ**) on both sides of the cleft. The incision outline (**---**) is shown for a two-layer closure of the hard palate using a vomer turnover flap and bipedicled palatal flaps. Preserving the anterior attachment of the palatal flaps allowed the anastomosing vascular connection between the palate and the labiofacial territory to be maintained. (b) Wound conditions at the end of palate repair and before lip repair. Lifting the bipedicled flaps without transposing them allows for complete primary wound closure in the midline and over the lateral surgical access incisions. Posteriorly, the palatine arteries are maintained as well as the nasopalatine artery and nasopalatine nerve at the incisive foramen on the healthy side. (c) Palate conditions at 2.5 years of age. No scarring in the anterior junction zone (white circle) around the area of the preserved NPA (**◘**).

Fig 3. Three-dimensional surface of a cast with reference points marked. Definitions of the reference points (Braumann et al., 2003, 2002; Nalabothu et al., 2020; Shen et al., 2015): Q and Q', gingival groove points (intersection of the gingival groove and lateral sulcus); T and T', posterior shelf pits (posterior end of the lateral sulcus); P and P', pole points (cleft edges of the alveolar ridges). A midpalatal-section plane through QQ' (perpendicular to QQ'T) defined: GQ by crossing the greater segment's junction to the vomer, VQ by crossing the vomer edge and LQ by crossing the lesser segment's shelf ridge. In the same way, the posterior-section plane TT' defined GT, VT and LT. In bilateral pairs of points, the prime (') indicates the point on the cleft side. T and T' were allocated in the depth of the lateral sulcus instead of the top of the alveolar ridge for better traceability (Brief et al., 2006; Seckel et al., 1995). The palatal cleft width (pc) was measured from GQ to LQ and from GT to LT, the true cleft width (tc) was measured from VQ to LQ and from VT to LT, and the curved vomer width (cv) was measured from GQ to VQ and from GT to VT.

Fig 4. Postoperative findings after cleft repair in one single surgical intervention with continuous circular closure. Healing conditions at the end of surgery (a, d, g), at 7 days postoperatively (b, e, h), and at 6 months postoperatively (c, f, i). Alar convexity and nostril symmetry and patency were retained at 6 months postoperatively (a, b, c), accompanied by a history of night-time nasal breathing. The palatal vault convexity at 1 week (e) and 6 months (f) postoperatively was similar to that seen preoperatively. Palatal mucosa relief (rugae palatinae and papilla incisiva) of the anterior palate was fully maintained. Lateral surgical access incisions healed primarily and left inconspicuous scars. Soft palate (g, h, i) healed with a single linear scar.

Fig 5. Morphologic changes during presurgical orthopedic therapy. Cleft morphology at birth (a) and at 8 months after passive plate therapy and lip taping (b). The palatal cleft width (pc) lies between the lesser segment's shelf ridge (—) and the greater segment's junction to the vomer (– –). This junction is indicated by the transition in the color of the mucosa from pink to red (Veau and Borel, 1931). The true cleft width (tc) lies between the lesser segment's shelf ridge (—) and the vomer edge (– ▪ – ▪). The width of the curved vomer (cv) comprises the area between the vomer edge (– ▪ – ▪) and the greater segment's junction to the vomer (– –). Thus, the true cleft denotes the cleft width of the fissure into the nose, whereas the palatal cleft denotes the gap in the palatal mucosa. The cv (○ to ●) and the separation between lateral sulci remained almost stable over time (▲ to ▲, ■ to ■). Coronal cross-section through the corresponding plaster model at birth (c) and at 8 months (d). The true cleft narrowed significantly, and its entrance plane changed from oblique (c, ···) to more vertical (d, ···). The shape of the ala on the cleft side changed from a concave (e, – –) to a convex (f, – –) curvature, and the tilted columella straightened up (e, f, →).