Long-term effect of pharyngeal flap surgery on craniofacial and nasopharyngeal morphology in patients with cleft palate

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The craniofacial morphology of 48 consecutive adult males with isolated cleft palate was studied by means of lateral cephalograms at the mean age of 18.8 years. Twelve of the patients had received pharyngeal flap surgery between 4 and 12 years of age (mean age 6 years) to improve speech. No significant differences were noticed in craniofacial cephalometric relations between the patients who had not had velopharyngeal flap surgery (VPF−) and those who had (VPF+), although the latter showed a tendency toward a more vertical growth direction. In the pharynx, the VPF+ group showed larger sagittal depths of nasopharyngeal airway but smaller depths of oropharyngeal airway. The differences were significant at the levels of the upper nasopharynx and lower oropharynx. According to the hospital records, none of the patients demonstrated persistent airway obstruction. Cephalometry may be useful in evaluating the changes in pharyngeal airway dimensions that may be related to velopharyngeal flap surgery. □ *Cephalometry; cleft palate; pharyngeal flap; pharynx*

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It has been estimated that 20%–30% of patients with clefts develop hypernasal speech after primary palatoplasty (1). Velopharyngeal surgery is commonly used to correct the velopharyngeal insufficiency (VPI). As pharyngeal flap surgery reduces the nasopharyngeal airspace, there has been concern about the effect of velopharyngeal flaps (VPF) on facial growth and breathing. It has been shown that impairment of the nasopharyngeal pathway can lead to increased vertical growth of the lower face (2–4). Pearl & Kaplan (5) and Semb & Shaw (6) failed to substantiate a significant effect of pharyngeal flaps on midfacial growth, whereas Long & McNamara (7) and Ren et al. (8) found increased vertical growth direction following surgery. The latter found the changes to be temporary and stated that the influence of a pharyngeal flap on facial growth had no long-term clinical importance.

There are no long-term reports of VPF on pharyngeal cephalometric dimensions, but there are reports of immediate or persistent airway obstruction, snoring and sleep apnea secondary to surgery for velopharyngeal insufficiency in cleft as well as in non-cleft patients (9–15). Although postoperative airway obstruction has been reported, it has been indicated (12, 13) that VPF is a good method by which to improve speech, and harmful effects are rare. The pathogenesis of obstructive sleep apnea and associated snoring may involve a combination of reduced pharyngeal muscle tone and reduction in pharyngeal airway dimensions. In cleft patients, the nasopharyngeal area has been reported to be smaller than in non-cleft patients (16). Characteristic changes in facial morphology in patients with obstructive sleep apnea include mandibular deficiency, elongated soft palate, enlarged tongue, decreased posterior airway space, and inferior position of the hyoid bone (17–23). Despite the limitations of radiography, a good correlation has been reported between cephalometric and CT measurements and 3D CT scans for many pharyngeal structures (24).

As nasorespiratory function may be impaired after VPF, it is plausible that velopharyngeal surgery in young patients could have an effect on their later craniofacial growth. The purpose of this retrospective study was to compare cephalometrically the craniofacial morphology and the pharyngeal airway in adult male patients with isolated cleft palate with and without velopharyngeal flap surgery in childhood.

Subjects and methods

Patients

The patients comprised 48 consecutive adult Caucasian males with isolated cleft palate and who had attended regular follow-up clinics at the Cleft Center, Department of Plastic Surgery, Helsinki University Central Hospital during 1988. The initial consecutive series consisted of 56 patients, but 8 were excluded because of syndromes or combined clefts. The method of palatal closure at the age of 1.5 to 2 years was either Veau-Wardill-Kilner or Cronin push back. Twelve of the patients had received pharyngeal flap surgery at the age of 4–12 years (mean age 6 years) to improve speech (VPF+). Comparability of the groups is

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Table 1. Comparability of the groups of patients without (VPF-) and with previous velopharyngeal flap (VPF+)

	No. of patients	Mean age (years)	Range
VPF−	36	18.7	17.1–20.7
VPF+	12	19.1	17.0–20.6
Total	48	18.8	17.0–20.7

given in Table 1. Two of these patients had submucous clefts of the palate; 9 had VPF before school age at 4–6 years of age; 1 had the flap at 8 and 2 at 12 years of age. The pharyngeal flaps were either Sanvenero-Rosselli ($n = 9$) or modified Honig ($n = 3$) (Fig. 1). Both flaps are based superiorly. In the Honig velopharyngeal flap, the tip of the flap is positioned at the junction of the soft and hard palate. In addition, the base of the flap is positioned higher in the posterior nasopharyngeal wall. Surgery includes transverse division of the palatal aponeurosis and nasal mucosa with insertion of the distal fourth of a pharyngeal flap to line the nasal side. The rest of the unlined pedicle suspended in the nasopharynx contracts into a tube (25). The Sanvenero-Rosselli flap is attached to the whole length of the soft palate through longitudinal incision of the palate. The surgical method has been described in detail in another article (26).

Other secondary operations included closure of fistula in 3 patients in the VPF+ group and 2 in the VPF− group. In addition, 3 of the patients with the Sanvenero-Rosselli flap later required a flap revision, and in 1 patient an additional operation a.m Orticochlea was performed later to improve the velopharyngeal function.

Orthodontic treatment was received by 90% of the patients. None of the patients had undergone orthognathic surgery. In order to compare heights, the patients were

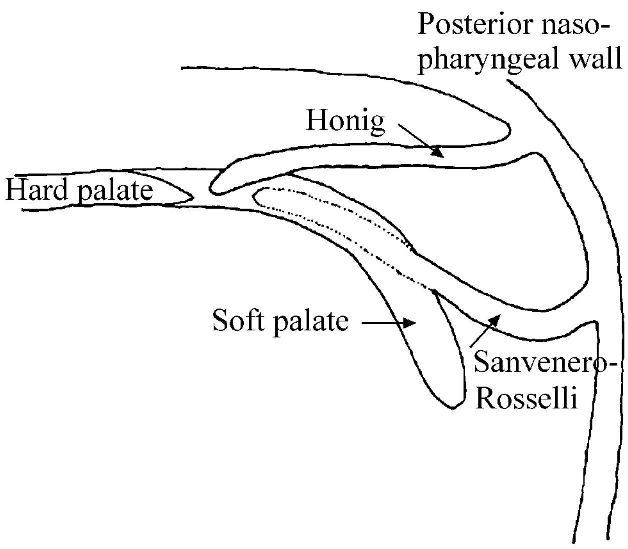

Fig. 1. Schematic illustration of the difference between the Sanvenero-Rosselli and Honig velopharyngeal flaps.

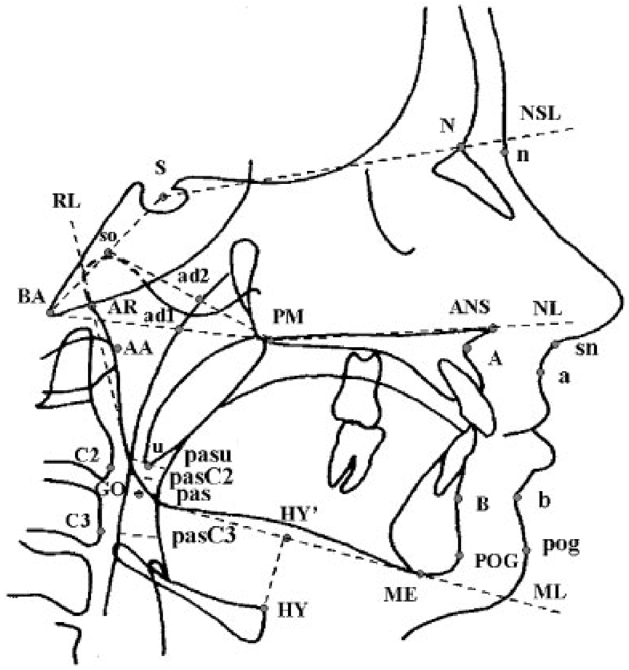

Fig. 2. Cephalometric landmarks. A (Point A): deepest point on the anterior contour of the maxillary alveolar arch; a (soft tissue point a): deepest point on the soft tissue contour of the upper jaw; AA (anterior atlas): most anterior point of the first vertebral corpus; ad1: intersection of the line PM-BA and the posterior nasopharyngeal wall; ad2: intersection of the line PM-so and the posterior nasopharyngeal wall; ANS (anterior nasal spine): tip of anterior nasal spine; AR (articulare): intersection between the external contour of the cranial base and the dorsal contour of the mandilble; B (Point B): deepest point on the anterior contour of the mandibular alveolar arch; b (soft tissue point b): deepest point of the soft tissue contour of the lower jaw; BA (Basion): most inferior point of the clivus of the occipital bone; C2 (second vertebra): most anterior inferior point of the second vertebral corpus; C3 (third vertebra): most anterior inferior point of the third vertebral corpus; GO (Gonion): intersection between the external contour of the mandible and the bisector of the angle between the ramus line and mandibular line; HY (Hyoid): most anterior and superior point of hyoid bone; HY′ (projection point of Hy): perpendicular distance of point Hy on the mandibular line; ME (Menton): most inferior point on mandibular symphysis; N (Nasion): most anterior point on the nasofrontal suture; n (soft tissue nasion): most concave point in the tissue overlying the area of the frontonasal suture; pas (posterior airway space at gonion level): sagittal depth of pharynx on the line through points B and Go; pasC2 (posterior airway space at C2 level): sagittal depth of pharynx on the line perpendicular to the posterior pharyngeal wall at the level of C2; pasC3 (posterior airway space at C3 level): sagittal depth of pharynx on the line perpendicular to the posterior pharyngeal wall at the level of C3; pasu (posterior airway space at uvula level): sagittal depth of pharynx on the line through perpendicular to the posterior pharyngeal wall through point u; PM (Pterygomaxillare): intersection between nasal floor and the posterior contour of maxilla; POG (Pogonion): most prominent point of the bony chin; pog (soft tissue pogonion): most anterior point of soft tissue chin; S (Sella): centre of sella turcica; so: midpoint of the distance from points S to Ba; u: most inferior tip of soft palate; sn (subnasale): point at which columnella merges with upper lip; ML (mandibular line): tangent to the lower border of mandible; through ME NL (nasal line): line through points ANS and PM; NSL (Nasion-Sella line): line through points N and S; RL (ramus line); tangent to the mandibular ramus through Ar.

measured to the nearest 0.5 cm at the cleft center. Symptoms of possible persistent airway obstruction, snoring, or sleep apnea were reviewed from the hospital records. No polysomnographic sleep recordings were available.

Cephalometric method

Standardized lateral cephalometric radiographs taken with the head positioned according to the Frankfort horizontal plane with molar teeth occluded and lips in repose were used. The cephalograms were traced twice by the same orthodontist during the same day using a computer-connected digitizer. The computer was programmed to calculate the mean of the two digitalizations, which were to be at an accuracy of 1 mm. The reference points and landmarks are shown in Fig. 2. Student's *t* test was used in the statistical analysis.

Evaluation of velopharyngeal function

The speech data were reviewed from hospital records. Speech evaluation included perceptual assessment and instrumental confirmation of velopharyngeal function by the speech pathologist at the Cleft Center. The degree of velopharyngeal insufficiency was assessed by combining the speech characteristics as follows: audible nasal air emission indicated mild insufficiency, hypernasality with or without audible nasal air emissions indicated moderate insufficiency, and compensatory articulation or weakness of plosives with or without hypernasality and audible nasal air emissions indicated severe insufficiency (26). In the instrumental confirmation of the need of velopharyngeal flap a still X-ray examination, the Nasometer (manufacturer Kay Elemetrics), and nasoendoscopy were used. Velopharyngeal function was assessed before and after the VPF.

Results

The patients in the VPF+ group showed larger sagittal depths of the nasopharyngeal airway and smaller sagittal depths of the oropharyngeal airway (Table 2). The differences were significant at the upper level of the nasopharynx (ad2-PM) and lower level of the oropharynx (pas C3). No significant differences were noticed in craniofacial cephalometric relations between the patients without (VPF−) and those with pharyngeal flap surgery (VPF+), although the VPF+ patients showed a tendency toward a more vertical growth direction (larger angles of NSL/ML, NL/ML, ML/RL and larger anterior face height (N-ME)). There was no difference between the heights of the patients in the two groups (VPF−, mean height 178 cm, range 163–191; VPF+, mean height 178 cm, range 170–178).

Table 2. The means, standard deviations and *P* values of cephalometric variables in *t* test between patients without (VPF−) and with previous velopharyngeal flap (VPF+). Angles are reported in degrees and distances in millimeters

	VPF− (*n* = 36)		VPF+ (*n* = 12)		VPF− /VPF+
	Mean	*s*	Mean	*s*	*P* value
N-S-BA	132.3	7.1	132.2	6.5	0.994
S-N-A	78.6	4.2	78.4	3.1	0.795
S-N-B	78.4	4.1	78.5	5.5	0.941
S-N-POG	80.6	4.2	80.9	5.7	0.85
ANS-PM	48.1	3	47.2	3.8	0.51
NSL/ML	31.9	6.1	35.8	8.7	0.128
NL/ML	24.2	7.2	27.6	8.5	0.23
ML/RL	126.9	6.6	130.2	8.2	0.304
N-ME	113.5	6.7	116	6.9	0.284
S-GO	72.8	4.2	71.8	6.6	0.625
S-n-a	86.9	4.1	87.7	4	0.551
S-n-b	80.7	4.2	81.4	5.2	0.701
S-n-pog	82.4	3.9	82.9	4.7	0.771
n-sn-pog	164.6	6.2	164	7	0.777
PM-BA	43.8	4.2	44.9	4	0.456
ad1-PM	23	3.7	24.5	2.2	0.098
ad1-BA	20.8	2.9	20.4	3.7	0.796
ad2-PM	21.7	4.4	24.8	1.7	0.002**
ad2-so	17.4	4.2	15.9	2.7	0.118
pasu	15.7	3.6	13.4	3.4	0.066
pas	12.4	3.4	11.1	3.8	0.74
pas C2	12	3.5	10.2	3.6	0.135
pas C3	14.9	4.9	11.6	3.8	0.024*
HY-HY′	20.3	4.6	19.7	5.1	0.699
AA-HY	63.8	7	67.6	7.6	0.157

Preoperatively, all 12 patients who received VPF had VPI that was resistant to speech therapy. Preoperatively, VPI was severe in 10 patients, moderate in 1, and mild in 1. Postoperatively, 7 patients had velopharyngeal competence (VPC). In 4 patients with Sanvenero-Rosselli flap, VPI remained impaired postoperatively. In 3 of these patients a flap revision and in 1 patient an additional operation a.m Orticochlea were performed. Postoperative follow-up was not carried out in 1 patient. According to the hospital records, none of the patients had long-term symptoms of airway obstruction, snoring, or sleep apnea.

Discussion

Several factors can interfere with craniofacial and pharyngeal growth in cleft patients. These include congenital dysmorphology of the midface, functional adaptations, and surgical iatrogenesis. The present study is in agreement with earlier studies (5–6); pharyngeal flaps do not carry the risk of adversely affecting subsequent midfacial growth. However, there may be long-term effects on pharyngeal dimensions, because the patients with previous VPF showed larger sagittal depths of the nasopharyngeal airway and smaller sagittal depths of the oropharyngeal airway. It is significant that no differences were observed in the bony pharynx between the 2 groups. It is thus possible that in the VPF+ group compensatory narrowing had occurred in the oropharynx in order to control the airway pressure because of the larger nasopharyngeal airway.

It has recently been found that 6-year-old cleft boys operated on velopharyngeoplasty ad modum Honig showed wider sagittal depths of the nasopharyngeal airway (ad1-PM, ad2-PM) compared to those with velopharyngeal competence (27). On the other hand, in children the role of the adenoids and possible adenoidectomy has to be considered when evaluating the pharyngeal airway on a lateral cephalogram. The patients included in the present study were all adult males with cleft palate of the same age and similar height, which adds validity to the study. However, 2 surgical techniques of velopharyngeal flap were used. Three-quarters of the patients had had a Sanvenero Rosselli flap, whereas the other patients had had a modified Honig flap. In addition, 4 of the patients with the Sanvenero-Rosselli flap required revision of the flap or an additional operation to improve the velopharyngeal function. These additional operations may have influenced pharyngeal size.

Velopharyngeal valving is dependent on the sensorimotor adequacy of the velum and its synergistic musculature and on the morphologic dimensions of the nasopharyngeal port (28). An increase in the width and depth of the nasopharynx, with a consequent increase in the pharyngeal cavity volume, has been observed in patients with cleft palate speech, as compared to patients with normal speech or nasality (29). In studying 17 to 21-year-old patients with isolated cleft palate, Haapanen et al. (30) found smaller soft tissue pharyngeal depth in velopharyngeally competent patients compared to the velopharyngeally incompetent patients. In addition, Wu et al. (16) observed that 6 to 32-year-old cleft patients with borderline velopharyngeal competency differed from the cleft patients with velopharyngeal incompetency in smaller pharyngeal depth and greater length of the hard palate. In the present study no preoperative series of X-rays were available. It is possible that preoperative cephalometric morphological differences have existed between the patients who received VPF and those who did not. On the other hand, according to Mazaheri et al. (31) it is impossible to predict cephalometrically at an early age those cleft patients who will later require pharyngeal flaps.

In cleft patients, maxillary growth deficits constrict the nasal floor, reduce airway size, and increase nasal airway resistance (32), which can lead to mouth-breathing. According to Hairfield et al. (33), almost 70% of the cleft lip and palate patients 'mouth-breathe' to some extent. Secondary procedures for velopharyngeal inadequacy, such as pharyngeal flap, increase the prevalence of mouth-breathing to 80% (33). In children, mouth-breathing may affect facial morphology (2–4). When studying the influence of pharyngeal flap on facial growth in patients with isolated cleft palate, Ren et al. (8) found differences in mandibular position and increased anterior facial height (as a reflection of habitual mouth-open posture) but found these changes to be temporary. In the present study, a more vertical growth direction was observed in the VPF+ group, although the difference was not statistically significant.

Cleft patients with pharyngeal flaps are at risk for acute airway obstruction in the immediate postoperative period and chronic airway obstruction manifested by obstructive sleep apnea (OSA) accompanied by mouth-breathing (34). It is possible for apnea to reappear following scar contracture of the pharyngeal ports (34). There is considerable variation in studies concerning methodology, time of follow-up, and incidence of OSA following pharyngeal flap surgery. In a polysomnographic sleep study (9), pharyngeal flap surgery was associated with sleep apnea in 9 of 10 patients 2 to 3 days following surgery. OSA was still present in 2 of 10 patients 3 months following surgery. In another investigation, Liao et al. (15) reported that 6 months postoperatively about 90% of cleft patients (Taiwanese adults and children) still had obstructive sleep apnea. On the other hand, Sirois et al. (12) found an incidence of 2% of persistent sleep apnea in a long-term follow-up of 2 to 22-year-old patients. Wells et al. (14) reported that 1% of patients (aged 2–44 years) developed sleep apnea at least 6 months postoperatively. According to the hospital records, none of the patients of the present study had long-term symptoms of airway obstruction. The characteristic morphologic changes leading to sleep apnea vary, but in non-cleft patients narrowing of the pharyngeal airway is reported consistently, although the site of the narrowing for obstruction is variable (35). It has also been found that, compared to controls, snorers exhibit cephalometrically narrower airways and reduced oropharyngeal areas (23).

The present paper was a preliminary retrospective study. The patients had been operated on between 10 and 20 years ago. It is obvious that additional studies are needed about the long-term effect of pharyngeal flap on pharyngeal dimensions with detailed patient history about possible airway obstruction. A VPF in childhood does not interfere with craniofacial growth but is associated with larger sagittal dimensions of the nasopharyngeal airway and smaller depths of the oropharyngeal airway in early adulthood. Cephalometry may be valuable in evaluating the changes in pharyngeal airway dimensions that may be related to velopharyngeal flap surgery.

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