Craniofacial cephalometric morphology in six-year-old girls with submucous cleft palate and isolated cleft palate

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Ninety-three girls with cleft palate (53 submucous cleft palate (SMCP) and 40 isolated cleft palate (ICP)), mean age 6.2 years (range 5.5–7.5), were compared retrospectively from lateral cephalograms. Forty-three patients with SMCP had had surgical treatment at the mean age of 3.4 years (range 0.9–6.8), 10 of the SMCP patients were unoperated. Twenty-six patients with ICP had clefts of the hard and soft palate, and 14 had clefts of the soft palate only. Palatal closure of ICP had been done at the mean age of 1.5 years (range 1.0–2.1). The skeletal craniofacial morphology was similar in SMCP and ICP. The maxilla and mandible were well related to each other but slightly retrusive in relation to the cranial base. The slight skeletal retrusion was significantly more masked by soft tissue in patients with SMCP. The patients with SMCP showed higher values for soft tissue maxillary and mandibular prominence. □ *Cephalometry; isolated cleft palate; morphology; submucous cleft palate*

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Classic submucous cleft palate (SMCP) consists of bifid uvula, notching of the posterior border of the bony palate, and palatal muscle diastasis (1). In occult SMCP there is only an abnormal levator muscle insertion into the posterior border of the palate (2). The classic SMCP may be diagnosed on oral examination but the occult variant is recognizable on nasopharyngoscopy, and cannot be detected on physical examination alone. Submucous cleft palate is felt to be of clinical relevance only in the presence of velopharyngeal insufficiency (VPI) or persistent problems with feeding or secretory otitis media. It has been estimated that the incidence of SMCP is roughly 1:1000, and only approximately 10% of these patients are symptomatic for VPI (3–5). Surgical procedures that have been used to treat VPI resulting from SMCP (2, 4, 6, 7) include excision of the submucous cleft palate and primary closure, pharyngeal flap, palatal pushback techniques, intravelar veloplasty, a combination of two or more techniques consisting of pharyngeal flap, pushback, and/or intravelar veloplasty and Furlow double reverse Z-plasty. The goal of surgical treatment in patients with submucous cleft palate and velopharyngeal inadequacy is to provide velopharyngeal competence either by improving velar function or by restricting the passage between oropharynx and nasopharynx (7). There is still controversy about the type and timing of surgery as well as the effect of palatal and velopharyngeal surgery on craniofacial growth. Optimally, surgery should achieve velopharyngeal closure so that facial growth is not inhibited and structures are not under tension.

Previous cephalometric studies on craniofacial morphology of patients with cleft palate deal mainly with isolated cleft palate (8–14). In patients with ICP the maxilla is short and retrusive in relation to cranial base (8–14). Also, the mandible is small and retrusive with an obtuse gonial angle and steep mandibular plane (8–11). Because both maxilla and mandible are small and retrusive, the relationship between the jaws is often satisfactory (15). It has been shown that boys have consistently larger craniofacial dimensions than girls (13, 14, 16).

The purpose of this study was to evaluate cephalometrically the craniofacial morphology in girls with submucous cleft palate and isolated cleft palate.

Materials and methods

The patients comprised 53 girls with submucous cleft palate and 40 girls with isolated cleft palate who had attended the Cleft Center, Department of Plastic Surgery, Helsinki University Central Hospital during 1980–1995. In all patients, diagnoses of submucous cleft palate were verified at the Cleft Center either clinically or by nasopharyngoscopy. Patients with combined clefts or syndromes were excluded. The comparability of the groups is given in Table 1.

The patients with ICP were grouped into two subgroups according to extent of cleft at birth. Twenty-six of the patients had clefts of the hard and soft palate, while 14 had clefts of the soft palate only. The severity of the cleft was classified from hospital records before primary surgery. In the ICP patients, one-stage hard- and soft-palate closure had been done at the mean age of 1.5 years (range 1.0–2.1 years) using the Veau-Wardill-Kilner V–Y pushback

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Table 1. Comparability of the groups of girls with submucous cleft palate (SMCP) and isolated cleft palate (ICP)

	SMCP	ICP	Total
No. of patients	53	40	93
Morphological classification:			
Hard and soft palate		26	
Soft palate alone		14	
Mean age (years)	6.2 (range 5.5–7.5)	6.2 (range 5.8–6.8)	6.2 (range 5.5–7.5)
Operated	43	40	83
Non-operated	10	0	10
Mean age at operation	3.4 (range 0.9–6.8)	1.5 (range 1.0–2.1)	

operation or the Cronin modification with additional mucosal flaps from the floor of the nose (17). Secondary operations included closure of fistula in 1 patient and modified Honig pharyngeal flap in 3 patients. The submucous cleft palate had been operated in 43 patients mainly because of VPI, and only rarely because of persistent problems with ears or feeding. The techniques for surgical treatment consisted of palatal repair ($n = 11$, mean age 1.8 years, range 0.9–3.9), pharyngeal flap surgery ($n = 13$, mean age 3.6 years, range 1.0–6.1) or a combination of 2 or more techniques consisting of pharyngeal flap, palatal repair, and/or intravelar veloplasty (reconstruction of the levator sling) ($n = 19$, mean age 4.6 years, range 3.4–6.1). Palatal operations were first done using the Veau-Wardill-Kilner or the Cronin modification, and later using the Bardach (18) 2 flap palatoplasty ($n = 3$) or Mendoza (19) minimal incision palatopharyngoplasty ($n = 2$). Superiorly based flaps were used for pharyngeal flap surgery; first Sanvenero-Rosselli ($n = 2$) and later modified Honig (17). Secondary operations included closure of fistula in 1 patient. Neither the patients with ICP nor the patients with SMCP had received orthodontic treatment.

Standardized lateral cephalometric radiographs were used, taken with the head positioned according to the Frankfort horizontal plane with molar teeth occluded and lips in repose. 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. 1. Student's *t* test was used in the statistical analysis.

Results

The results are given in Table 2. In patients with SMCP, no differences in craniofacial cephalometric measurements were found between the operated and non-operated patients. In addition, no significant difference in craniofacial morphology was found between the ICP patients with hard and soft palate clefts and soft palate clefts alone. Thus the groups were pooled for further analysis. The skeletal craniofacial morphology was similar in both SMCP and ICP. The maxilla and mandible were well

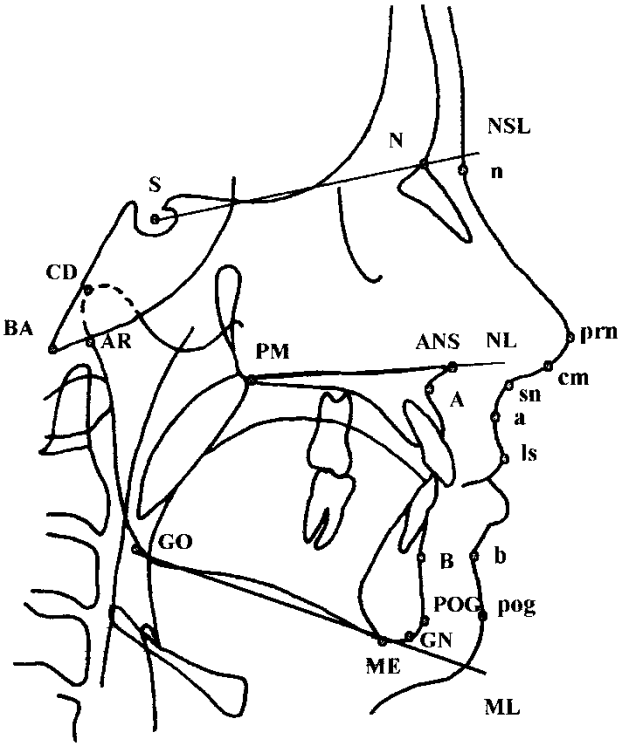

Fig. 1. Cephalometric landmarks. Abbreviations, full names and definitions. **A** (point A): deepest point on the anterior contour of the maxillary alveolar arch. **a** (soft tissue point a): deepest point on the soft tissue contour of the upper jaw. **ANS** (anterior nasal spine): tip of anterior nasal spine. **AR** (articulare): intersection between the external contour of the cranial base and the dorsal contour of the mandible. **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. **CD** (condylion): most posterior and superior point on the condylar head. **cm** (columella): most anterior point of the columella of the nose. **GO** (gonion): intersection between the external contour of the mandible and the bisector of the angle between the ramus line and mandibular line. **ls** (labrale superior): most anterior point of the upper lip. **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. **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. **prn** (pronasale): most prominent point of apex nasi. **S** (sella): centre of sella turcica. **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.

Table 2. The means, standard deviations, and *P* values of cephalometric variables in *t* test between girls with submucous cleft palate (SMCP) and with isolated cleft palate (ICP). Angles are reported in degrees and distances in millimeters

	SMCP		SMCP Operated/ Non-operated	ICP		ICP Hard and soft palate/ Soft palate alone	SMCP/ICP
	Mean ($n = 53$)	*s*	*P* value	Mean ($n = 40$)	*s*	*P* value	*P* value
N-S-BA	130.2	4.7	0.052	130.6	2	0.215	0.697
N-S	60.8	2.6	0.315	61	2.8	0.649	0.654
S-BA	36	1.7	0.082	36.5	3.7	0.151	0.138
N-BA	88.3	3.3	0.182	89.1	5.3	0.531	0.29
S-N-A	79.1	3.2	0.32	78.9	3.6	0.539	0.71
S-N-B	76	3.5	0.063	75.5	3.9	0.433	0.558
A-N-B	3.1	1.9	0.658	3.4	2	0.838	0.606
S-N-POG	75.7	3.6	0.081	75.3	3.9	0.092	0.542
ANS-PM	43.6	2.5	0.967	43.7	2.6	0.552	0.8
N-ANS	40.8	2.4	0.123	41.3	2	0.752	0.275
ANS-ME	54.7	3.6	0.931	54.3	3.1	0.1	0.575
S-GO	57.7	3.4	0.46	56.6	3.1	0.112	0.1
GN-CD	88.8	5.1	0.856	89.9	3.8	0.538	0.277
ME-GO	54.2	3.4	0.954	53.9	3.1	0.054	0.704
AR-GO	33.8	3.1	0.308	33	3.1	0.894	0.294
NSL/ML	37.3	4.6	0.268	38.6	5.1	0.692	0.19
NL/ML	29.3	4.5	0.976	28.2	5	0.149	0.264
S-n-a	92.2	4.2	0.435	89.4	3.7	0.664	0.001**
S-n-b	83.9	3.8	0.402	81.6	4	0.764	0.007**
a-n-b	8.3	2.4	0.829	7.8	2.3	0.399	0.49
S-n-pog	83.9	4	0.376	81.8	4.1	0.324	0.013*
n-sn-pog	161.1	5.1	0.697	162.2	4.9	0.564	0.295
n-prn-pog	132.7	4.8	0.894	133.8	4.3	0.899	0.26
cm-sn-ls	114.2	11.8	0.914	110.8	12.4	0.133	0.192

s = standard deviation.

related to each other but slightly retrusive in relation to the cranial base. No skeletal differences were observed. However, the slight skeletal retrusion was significantly more masked by soft tissue in patients with SMCP. In soft tissue profile, the patients with SMCP showed larger angles of soft tissue S-n-a, S-n-b and S-n-pog.

Discussion

A major finding of this study is that no skeletal differences existed between patients with SMCP and ICP. The fact that all patients were girls of the same age adds validity to the comparison. Even if submucous cleft palate is a milder form of overt cleft palate it seems to be associated with similar skeletal craniofacial morphology. The finding that the soft tissue profiles were different implies that the skeletal retrusion may be masked by soft tissue. It has been observed (14) that in adult patients with isolated cleft palate, soft tissue profile showed marked deviations in the patients with complete clefts when compared to incomplete or soft palate clefts. In the present study no difference could be observed in the ICP group between hard and soft palate clefts and clefts of the soft palate only. However, the patients were only 6 years old.

There are only a few studies describing the cephalometric features of SMCP patients. According to Kaplan (2) the patients showed a minimal tendency toward maxillary hypoplasia, although no numerical values were given. Later Pearl & Kaplan (20) concluded that children with isolated cleft palate and submucous cleft palate demonstrated pre-operatively narrow SNA and SNB angles, but the SNA-SNB difference was normal. The cleft palate children who had had combined palatal pushback and superiorly based pharyngeal flap operations did not show growth retardation of the face (20). This is in agreement with our results. Park et al. (21) found few differences in cranial base (N-BA ans N-S-BA) in non-operated 4 to 7-year-old SMCP children with velopharyngeal competence and incompetence. The cranial base measurements were larger in children with VPC at 4 years of age but not later.

The age for surgical correction of submucous cleft palate depends on the diagnosis of the velopharyngeal inadequacy and therefore nearly always occurs later than the time for preferred treatment of patients with overt cleft palate (7). Many submucous clefts may remain undiagnosed into and through adulthood. Surgically treated children with isolated cleft palate have been shown to demonstrate great similarity in craniofacial morphology when compared to non-treated cleft palate children, but their morphology differs from non-cleft children and that of children with other cleft types (22). It has been difficult to prove that the type of palatal repair affects facial growth (11, 13, 14, 22–24). It has been speculated that pharyngeal flap surgery may induce changes in facial growth by increasing airway resistance with resulting adaptation in the lower face, and by direct restraint effect of maxillary growth (15). Long & McNamara (25) and Ren et al. (26)

found increased vertical growth direction following pharyngeal flap surgery, but the latter found the changes to be temporary. On the other hand, Pearl & Kaplan (20) and Semb & Shaw (27) failed to substantiate a significant effect of pharyngeal flap surgery on midfacial growth.

In this preliminary study no differences in craniofacial morphology were found between the operated and non-operated patients with SMCP. This suggests that the minor growth disturbance, causing the slight skeletal retrusion of maxilla and mandible in SMCP, may be associated with clefting per se. As the material is small, and different surgical methods had been used, caution is needed when interpreting the results. In addition, when judging the effect of surgery, speech, nasopharyngeal function and pharyngeal morphology as well as alveolar-dental dimensions should be included. It is also noticeable that the X-rays had been taken at the mean age of 6.2 years, before juvenile and pubertal growth spurts. In the treatment of cleft lip and palate there is always a time lapse between treatment and evaluation of the final outcome. The final facial morphology is the result of a sum of factors, such as the cleft itself, genetic factors, functional compensation for the anatomical deficiency, and the influence of surgical technique and additional therapy.

In conclusion, the skeletal craniofacial morphology was similar in both SMCP and ICP, but the slight maxillo-mandibular skeletal retrusion was significantly more masked by soft tissue in patients with SMCP.

References

1. Calnan J. Submucous cleft palate. Br J Plast Surg 1954;6:264–82.
2. Kaplan EN. The occult submucous cleft palate. Cleft Palate J 1975;12:356–68.
3. Weatherley-White RCA, Sakura CY, Jr, Brenner LD, Stewart JM, Ott JE. Submucous cleft palate. Its incidence, natural history, and indications for treatment. Plast Reconstr Surg 1972; 49:297–304.
4. Bagatin M. Submucous cleft palate. J Maxillofac Surg 1985;13: 37–8.
5. Garcia Velasco M, Ysunza A, Hernandez X, Marquez C. Diagnosis and treatment of submucous cleft palate: a review of 108 cases. Cleft Palate J 1988;25:171–3.
6. Åbyholm FE. Submucous cleft palate. Scand J Plast Reconstr Surg 1976;10:209–12.
7. Gosain AK, Conley SF, Marks S, Larson DL. Submucous cleft palate. Diagnostic methods and outcomes of surgical treatment. Plast Reconstr Surg 1996;97:1497–509.
8. Shibasaki Y, Ross RB. Facial growth in children with isolated cleft palate. Cleft Palate J 1969;6:290–302.
9. Dahl E. Craniofacial morphology in congenital clefts of the lip and palate. Acta Odontol Scand 1970;28 Suppl 57.
10. Bishara SE, Iversen WV. Cephalometric comparisons on the cranial base and face in individuals with isolated clefts of the palate. Cleft Palate J 1974;11:162–75.
11. Jonsson G, Thilander B. Occlusion, arch dimensions, and craniofacial morphology after palatal surgery in a group of children with clefts in the secondary palate. Am J Orthod 1979; 76:243–55.
12. Smahel Z. Variations in craniofacial morphology with severity of isolated cleft palate. Cleft Palate J 1984;21:140–58.
13. Jakobsson OP. Repair of isolated cleft palate. A comparison of one- and two-stage surgery by dental arch measurements, cephalometry, and speech analysis. Uppsala: Acta Universitatis Upsaliensis; 1990.
14. Heliövaara A, Ranta R. One-stage closure of isolated cleft palate with the Veau-Wardill-Kilner V to Y pushback procedure or the Cronin modification. III. Comparison of lateral craniofacial morphology. Acta Odontol Scand 1993;51:313–21.
15. Semb G, Shaw WC. Facial growth in orofacial clefting disorders. In: Turvey TA, Vig KWL, Fonseca RJ, editors. Facial clefts and craniosynostosis. Principles and management. Philadelphia: W. B. Saunders; 1996. p. 28–56.
16. Jain RB, Krogman WM. Cleft type and sex differences in craniofacial growth in clefting from one month to ten years. Cleft Palate J 1983;20:238–45.
17. Millard DR. Cleft craft: the history of its surgery, vol. 3. Alveolar and palatal deformities. Boston: Little Brown; 1980.
18. Bardach J. Cleft palate repair: two flap palatoplasty, research philosophy, technique and results. In: Bardach J, Morris HL, editors. Multidisciplinary management of cleft lip and palate. Philadelphia: Saunders; 1990. p. 352–65.
19. Mendoza M, Molina F, Azzolini C, Ysunza Rivera A. Minimal incision palatopharyngoplasty. A preliminary report. Scand J Plast Reconstr Hand Surg 1994;28:199–205.
20. Pearl RM, Kaplan EN. Cephalometric study of facial growth in children after combined pushback and pharyngeal flap operations. Plast Reconstr Surg 1976;57:480–3.
21. Park S, Omori M, Kato K, Nitta N, Kitano I, Masuda T. Cephalometric analysis in submucous cleft palate: comparison of cephalometric data obtained from submucous cleft palate patients with velopharyngeal competence and incompetence. Cleft Palate Craniofac J 2002;39:105–9.
22. Bishara SE. Cephalometric evaluation of facial growth in operated and non-operated individuals with isolated clefts of the palate. Cleft Palate J 1973;10:239–46.
23. Palmer CR, Hamlen M, Ross RB, Lindsay WK. Cleft palate repair: comparison of the results of two surgical techniques. Can J Surg 1969;12:32–9.
24. Normando AD, da Silva Filho OG, Capelozza Filho L. Influence of surgery on maxillary growth in cleft lip and/or palate patients. J Craniomaxillofac Surg 1992;20:111–8.
25. Long RE, Jr, McNamara JA, Jr. Facial growth following pharyngeal flap surgery: skeletal assessment on serial lateral cephalometric radiographs. Am J Orthod 1985;87:187–96.
26. Ren YF, Isberg A, Henningsson G. The influence of pharyngeal flap on facial growth. Long-term results in the patients with isolated cleft palate. Scand J Plast Reconstr Surg Hand Surg 1994;28:63–8.
27. Semb G, Shaw WC. Pharyngeal flap and facial growth. Cleft Palate J 1990;27:217–24.

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