Sizes of dental arches in young adult patients with Pierre Robin sequence and isolated cleft palate

Sirpa H. Laitinen and Reijo Ranta

Cleft Center, Department of Plastic Surgery, Helsinki University Central Hospital, and
Department of Pedodontics and Orthodontics, Institute of Dentistry, University of Helsinki,
Helsinki, Finland

Laitinen SH, Ranta R. Sizes of dental arches in young adult patients with Pierre Robin sequence and isolated cleft palate. Acta Odontol Scand 1998;56:85–89. Oslo. ISSN 0001-6357.

The dental arch dimensions of 29 patients with Pierre Robin sequence (16.4–25.4 years of age) and of 31 patients with isolated cleft palate (16.9–19.9 years of age) were examined. All patients in the isolated cleft palate group and 21 patients in the Pierre Robin sequence group had had the Cronin modification V–Y pushback technique primary surgery performed. The Veau–Wardill–Kilner modification had been used in eight Pierre Robin sequence patients for primary surgery. With regard to patients whose height growth was nearly finished, both the upper and lower dental arch sizes were smaller in patients with Pierre Robin sequence than those in patients with isolated cleft palate. On comparison of the arch sizes, sex differences were more pronounced in the isolated cleft palate group. The high percentage of missing teeth and surgery was found to explain some of the small sizes of the dental arches in the Pierre Robin sequence group. ☐ *Craniofacial growth; hypodontia; micrognathia; oral surgery; Robin sequence*

Sirpa H. Laitinen, Department of Pedodontics and Orthodontics, Institute of Dentistry, PI 41, 00014 University of Helsinki, Helsinki, Finland

Pierre Robin sequence (PRS) patients have special respiratory problems that first appear when they are neonates. The association of newborn micrognathia with glossoptosis and cleft palate was first described by Pierre Robin in 1923 (1, 2). The etiology has been assumed to be heterogeneous (3, 4). U- and V-formed cleft palates occur with equal frequency, and the width and form of the cleft do not necessarily correlate to the respiratory distress (4). An insufficient descent of the tongue in the embryo before palatal shelves fusion has been explained to be a consequence of mechanical factors, such as a small and receded mandible and the head position of the fetus to produce cleft palate (5, 6). A delayed myofunctional maturation has also been associated with the etiology of PRS (7).

The partial postnatal catch-up growth of the mandible improves the airway dimensions dramatically (8). During the first 2 years the sizes of the mandible and tongue increase more in PRS children than in isolated cleft palate patients without micrognathia (ICP) or noncleft children (NONC) (8). The later craniofacial growth of the PRS patients improves the skeletal relationship between the maxilla and mandible, but the mandible size remains smaller and more retruded than in ICP patients (9, 10).

The size of the maxillary arch in PRS children is known to be similar to that of the ICP children before the palatal closure (11). The dental arch dimensions of the ICP patients are significantly smaller than those of NONC subjects both before and after palatal surgery (12–15). The dental arch dimensions seem to be independent of the timing (1–2 years of age) of primary palatal surgery (16) or the method of one-stage closure in ICP subjects (17). However, it has been reported that surgery can increase the contraction of the maxilla and frequency of anterior and lateral crossbite (18, 19). The extent of the cleft (18) and even the skill of the surgeon (20) are named among the influencing factors of the future development of dental arches.

The size of the mandibular arch of infants with PRS has not been accurately reported. Compared with ICP children, PRS children have shorter maxillary and mandibular arch depths at the age of 3 years and shorter maxillary and mandibular premolar widths at the age of 6 years (11).

The aim of this study was to analyze and to compare the dimensions and status of dental arches in young adult patients with PRS and ICP and determine whether the severity of the cleft, body size, and primary and secondary operations have an effect on the sizes of dental arches.

Material and methods

Patients

Forty-nine young adult patients with PRS participated in the final follow-up visit, when the cephalogram, panoramic roentgenogram, and dental casts were taken and height and weight measured. Twenty-nine acceptable plaster casts of both dental arches were found for this examination. The subjects were 14 males and 15 females born between 1965 and 1975 (mean age, 20.3 years; range, 16.4–25.4 years). The diagnosis for PRS had been made by a pediatrician with the criteria of micrognathia,

Table 1. Comparability of the groups

	Veau–Wardill–Kilner pushback operation			Cronin modification			
	Male	Female	Total	Male	Female	Total	Total
No. of operations							
PRS							
Primary alone	1	1	2	5	7	12	14
With secondary	3	3	6	5	4	9	15
ICP							
Primary alone	—	—	0	12	15	27	27
With secondary	—	—	0	3	1	4	4
Morphologic classification							
PRS							
Complete	—	1	1	3	6	9	10
Partial	4	3	7	6	5	11	18
Soft	—	—	—	1	—	1	1
ICP							
Complete	—	—	—	8	6	14	14
Partial	—	—	—	7	10	17	17
Soft	—	—	—	—	—	—	—

PRS = Pierre Roloin sequence; ICP = isolated cleft palate.

cleft palate, and various degrees of respiratory difficulties originating from glossoptosis during the first months after birth. Associated distinct syndromes were excluded, but some minor anomalies were associated with 5 patients (hypertelorism, $n = 1$; epicanthal folds and external ears, $n = 1$; intra-abdominal anomaly, $n = 1$; stenosis pylori, $n = 1$; mediastinal neuroblastoma, $n = 1$).

The control group consisted of plaster casts of dentitions of 31 young adults with ICP born between 1969 and 1971 (mean age, 17.9 years; range, 16.9–19.9 years). They were randomly collected from a larger ICP group of 116 patients on the basis of the extent of cleft, age, and sex. Three ICP patients had associated anomalies (heart defect, $n = 1$; anomaly of III cervical vertebra, $n = 1$; hypospadia, $n = 1$). The comparability of the groups is shown in Table 1.

All of the patients had been operated on at the Cleft Center, Department of Plastic Surgery, Helsinki University Central Hospital, Helsinki, Finland.

The primary operations had been done as one-stage hard and soft palate closure using the Veau–Wardill–Kilner procedure or modification of the Cronin mucoperiosteal palatal V–Y pushback technique (21). All of the patients in the ICP group and 21 of 29 patients in the PRS group had had the Cronin modification primary surgery performed. Secondary palatal operations such as velopharyngoplasty or repair of fistulas had been done for 11 (38%) PRS and 4 (13%) ICP patients. The patients' files were reviewed for the original size of the cleft, and the sizes were categorized as follows: complete when cleft extended to the anterior half of the palate; partial when cleft extended to the posterior half of the palate; or soft when cleft reached the soft palate only (Table 1).

The patients' height and weight were taken (to the nearest 0.5 cm and 1 kg, respectively) at the Cleft Center. The measurements were converted to standard deviation scores (SDS) for height and percentual weight for height (%Wt), and revised growth standards based on the study of Sorva et al. (22) were used as the normative data.

Methods

The 10 measurements of dental cast models were done using the method presented by Moorrees (23) (Fig. 1). The measuring was performed to the nearest 0.1 mm by one person (S. Laitinen) at least twice, with a 2-week interval, with a sliding digital caliper (Mitutoyo). The first measurements were used for the calculation of data. Intraexaminer error was measured with paired t test of all 60 subjects between the 2 measurements of 10 variables. No significant differences were observed (t values, 0.16–1.24). The measurements differed most in the maxillary first molar distance ($P = 0.11$) and least in the maxillary canine and mandibular first and second premolar distances ($P = 0.40$).

The missing teeth (excluding the third molars) were counted from the plaster casts and rechecked from the panoramic roentgenograms and patients' treatment history. Crowding between the mesial surfaces of the first molars was visually estimated from the plaster casts of the dentition segmentally.

Males and females were analyzed separately and together in the comparisons of differences. In the simple and multiple linear regression analyses, the dental arch measurements were regarded as dependent variables. The independent variables were number of missing teeth per dental arch, primary operation method, additional secondary operations, extent of cleft at birth, and sex. Simple and multiple linear regression analysis, the Student's t test, and the Mann–Whitney U-test were used

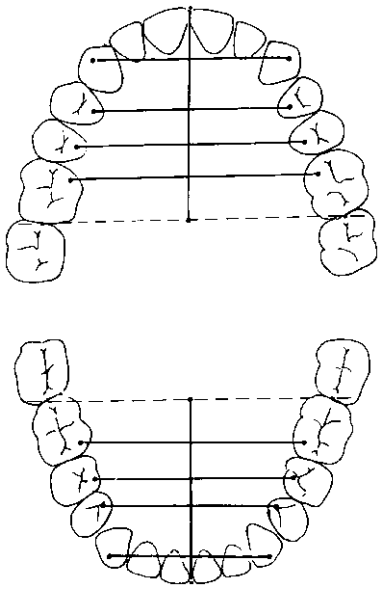

Fig. 1. Maxillary intercanine arch width 13–23: distance between cusp tips of the upper permanent canines. Maxillary interpremolar arch width 14–24: distance between palatal cusp tips of the upper first permanent premolars. Maxillary interpremolar arch width 15–25: distance between palatal cusp tips of the upper second permanent premolars. Maxillary intermolar arch width 16–26: distance between the mesiopalatal cusp tips of the upper first permanent molars. Upper arch depth: distance between a tangent to the middle area of the labial surfaces of the upper central incisors, and a line connecting the most posterior points on the distal surfaces of the upper first permanent molars. Correspondingly in the mandible. (Method of analysis used by Moorrees (23).)

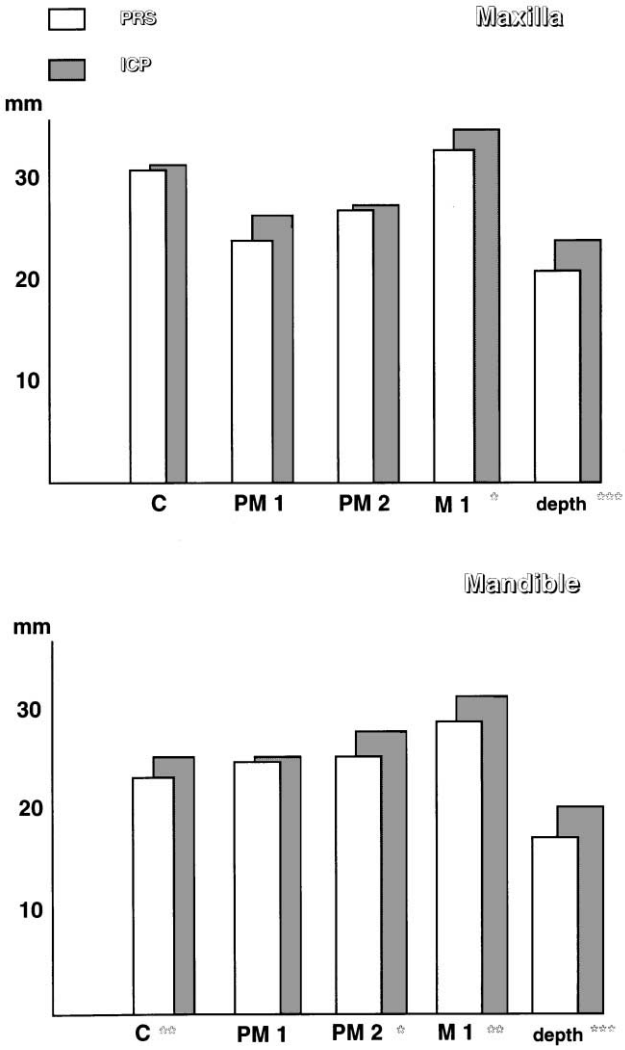

Fig. 2. The dimensions of the upper and lower arch width and depth in young adults with Pierre Robin sequence (PRS) and isolated cleft palate (ICP). C = canine; PM = premolar; M = molar. For definitions of the other variables, see Fig. 1. Results of the Student's *t* test are shown below the bars. $*P < 0.05$, $**P < 0.01$, $***P < 0.001$.

for the statistical analysis. Probabilities of less than 0.05 were considered significant.

Results

The widths of dental arches were larger and the depths of the arches were longer in males than in females in both the PRS and ICP groups. The differences were significant in the ICP group for all the dimensions. In the PRS group, only the upper arch depth was significantly longer in males than in females ($P < 0.05$).

The comparison between the PRS and ICP males showed that the upper arch width at the first molars ($P < 0.001$) and the upper arch depth ($P < 0.05$) were significantly less in the PRS males. Except for the width between the first premolars, all lower arch dimensions were significantly shorter ($P < 0.01$) in PRS than in ICP males. The PRS females had shorter upper ($P < 0.001$) and lower arch depths ($P < 0.05$) and more narrow lower canine widths ($P < 0.05$) than the ICP females. The differences in the dental arch dimensions between the PRS and ICP groups are shown in Fig. 2.

Crowding of the dental arches occurred with the same frequency in the PRS (86%) and ICP (87%) groups. The mean of the number of missing or extracted teeth was 4 in the PRS and 3 in the ICP group. The constricting and shortening effect on lower dental arch was explained (20%–80%) by the lower number of teeth in PRS group with linear regression analysis. In the ICP group, only the lower arch depth was found to be significantly dependent (28%) on the number of missing teeth.

The sex influence explained the upper and lower dental arch width and lower arch depth differences better and more significantly in the ICP than in the PRS group. The cleft extent tended to have an inverse effect on upper and lower arch premolar and molar widths and depths—the wider clefts tended to have shorter dimensions—thus it did

not significantly explain the dental arch dimensions in either the PRS or the ICP group. The effect of the primary and secondary operations on upper arch premolar and molar width and depth was greater in the PRS than in the ICP group. The primary operation influence on upper arch first molar width and depth was nearly significant as compared with the other factors according to the multiple linear regression analysis. Furthermore, the PRS patients operated on with the Cronin method had shorter upper arch dimensions compared with those operated on with the Veau–Wardill–Kilner method. Significantly more secondary operations were performed on PRS than on ICP patients. The secondary operation subgroups showed a constricting tendency on upper arch premolar and first molar dimensions in both groups, but according to the multiple linear regression analysis the explanation values among other factors remained small.

The final height in standard deviation scores (SDS) was slightly, but not significantly, shorter in the PRS males (SDS, −0.17; range, −2.8–3.2) than in the ICP males (SDS, −0.003; range, −1.4–1.4). Correspondingly, PRS females were shorter (SDS, −0.74; range, −3.4–2.9) than the ICP females (SDS, −0.006; range, −2.1–1.2). The relative mean %Wt was >6% and did not differ significantly between the groups. The median %Wt was somewhat higher for the PRS males and females than the ICP males and females.

Discussion

The PRS group was older in the final records (males: mean, 20.6 years; range, 17.2–25.4 years; females: mean, 20.0; range, 16.4–23.0) than the ICP group (males: mean, 17.7; range, 17.1–19.0; and females: mean, 18.1; range, 16.9–19.9). This allows us to assume that the final growth had occurred in the first group while we were assessing the dental arch sizes and the heights and weights of the studied groups.

The results of the dental arch sizes suggest that, after height growth cessation, the PRS patients have smaller dental arches than the ICP patients. The dental arch size difference between the males and females was more pronounced in the ICP than the PRS group, which indicates that PRS is a more severe form of malformation. A previous study showed a reduction of the maxillary arch in Finnish PRS children from 0.2 to 6 years of age compared with ICP children (11). The maxillary arch became smaller in width and depth after the primary surgery was performed, and the reduction increased between the ages of 3 and 6 years. The same tendency was observed in the mandibular arch, even though no extractions of teeth were made in this period.

A growth study from birth to 12 years revealed that Finnish PRS children born at term without associated anomalies do not differ in any respect from Finnish children with ICP (24). The early failure in height growth was slight and was not observed after 1 year of age. The final height of the Finnish patients with ICP after the cessation of growth has been found not to differ from the normative standards of the Finnish population (17). It has also been suggested that cleft-lip and -palate children have their pubertal spurt about half a year later than normal children (25) and have a shorter final height (25, 26). The maturation delay mainly applies to boys (27, 28). However, the final heights of the PRS males in this study were similar to those of the ICP males, and within the normal variation of Finnish standards. The final heights of the PRS females were slightly shorter than those of the ICP females and the noncleft Finnish population standards. The final weights of both PRS males and PRS females did not differ from those of the ICP controls. As previously observed in ICP males (17), a tendency of the relative median %Wt increase (>6%) to result in obesity was observed in PRS males.

In this study the number of complete palatal clefts was greater among the PRS patients than in the ICP group, which may partly explain their narrow maxillary arch in the premolar and molar area. For this reason a crowding of the dental arches in that area is common, and guided premolar extraction is a favored orthodontic treatment, especially since the prevalence of hypodontia in PRS children is very high (29). The congenitally missing teeth are often the second premolars. Also, in this study the total number of missing teeth in both groups was high, affecting the reduction of the dental arch sizes. The sagittal maxillary length measured from the cephalograms was found to be shorter in the PRS than in the ICP patients of young adult age (9), which means that the small size is not only associated with the alveolar basis and dental arches in maxilla.

The primary operation seems not to affect the size of dental arches in ICP young adults according to Heliövaara et al. (17). In the present study the upper arch size of the PRS group showed some sensitivity to the performed primary operation, but because of the skew distribution of two operation methods, the finding may be a coincidence.

The PRS group had 12% more secondary operations than the ICP group. The number of additional operations was found to be a significant constricting factor on maxillary and mandibular intermolar widths (17). The same tendency was found in the PRS and ICP groups in this study, even though the explaining values of the statistics remained lower in our population. The need for velopharyngoplasty because of impaired speech development has been found to be 33% higher in PRS children than in ICP children (30). The skeletal growth direction of the maxilla and mandible may increase the need for velopharyngoplasty (8, 9, 30), but also the short maxillary arch and tissue tightness might enlarge the upper nasopharyngeal area and result in hypernasal speech. In addition, poor neuromuscular maturation has been suggested as a possible etiologic factor (7) and might even explain the unsatisfactory growth and movements of the tongue and pharyngeal soft tissues and, thereby as a secondary effect, the dental arch form and size.

It seems that the dental arches of the young adults with PRS are significantly smaller than those of young adults with ICP. This may be a consequence of different factors: intrinsic growth failure, cleft palate, reduced size and altered position of the tongue, increased number of palatal operations, and hypodontia with guided extractions as a chosen orthodontic treatment.

References

1. Robin P. A fall of the base of the tongue considered as a new cause of nasopharyngeal respiratory impairment. Bull Acad Nath Med (Paris) 1923;89:37.
2. Randall P, Krogman WM, Jahina S. Pierre Robin and the syndrome that bears his name. Cleft Palate J 1965;2:237–46.
3. Gorlin R, Cohen M Jr, Levin LS. Syndromes of the head and neck. 3rd ed. New York: Oxford University Press; 1990. p. 700–3.
4. Rintala A, Ranta R, Stegars T. On the pathogenesis of cleft palate in the Pierre Robin syndrome. Scand J Plast Reconstr Surg 1984;18:237–40.
5. Siegel-Sadewitz L, Shprinzen RJ. The relationship of communication disorders to syndrome identification. J Speech Hear Dis 1982;47:338–54.
6. Diewert VM. Craniofacial growth during human secondary palate formation and potential relevance of experimental cleft palate observation. J Craniofac Genet Dev Biol 1986; Suppl 2:267–76.
7. Sher AE. Mechanisms of airway obstruction in Pierre Robin sequence: implications for treatment. Cleft Palate J 1992;29:224–31.
8. Figueroa AA, Glupker TJ, Fitz MG, BeGole EA. Mandible, tongue, and airway in Pierre Robin Sequence: a longitudinal cephalometric study. Cleft Palate J 1991;28:425–34.
9. Laitinen SH, Ranta RE. Cephalometric measurements in patients with Pierre Robin syndrome and isolated cleft palate. Scand J Plast Reconstr Hand Surg 1992;26:177–83.
10. Laitinen SH, Heliövaara A, Ranta RE. Craniofacial morphology in young adults with the Pierre Robin sequence and isolated cleft palate. Acta Odontol Scand 1997;55:223–8.
11. Laitinen SH. Sizes of dental arches in children with the Pierre Robin syndrome and isolated cleft palate aged from 0.2 to six years. Scand J Plast Reconstr Hand Surg 1993;27:285–90.
12. Athanasiou AE, Mazaheri M, Zarrinnia K. Longitudinal study of the dental arch dimensions in hard and soft palate clefts. J Pedodont 1987;12:35–46.
13. Nyström M, Ranta R. Sizes of dental arches and interdental space in 3-year-old children with and without cleft lip/palate. Eur J Orthod 1989;11:82–8.
14. Nyström M, Ranta R, Kataja M. Sizes of dental arches and general body growth up to 6 years of age in children with isolated cleft palate. Scand J Dent Res 1992;100:123–9.
15. Kilpeläinen P, Laine-Alava MT, Lammi S. Palatal morphology and type of clefting. Cleft Palate Craniofac J 1996;33:477–82.
16. Nyström M, Ranta R. Effect of timing and method of cleft palate repair on dental arches at the age of three years. Cleft Palate J 1990;27:349–53.
17. Heliövaara A, Pere A, Ranta R. One-stage closure of isolated cleft palate with the Veau–Kilner V to Y pushback procedure or the Cronin modification. II. Height, weight and comparison of dental arches. Scand J Plast Reconstr Hand Surg 1994;28:55–62.
18. Hellquist R, Pontén B, Skoog T. The influence of cleft length and palatoplasty on the dental arch and the deciduous occlusion in cases of clefts of the secondary palate. Scand J Plast Reconstr Surg 1978;12:45–54.
19. Friede H, Persson E-C, Lilja J, Elander A, Lohmander-Agerskov A, Söderpalm E. Maxillary dental arch and occlusion in patients with repaired clefts of the secondary palate. Scand J Plast Reconstr Hand Surg 1993;27:297–305.
20. Ross RB. Treatment variables affecting growth in cleft lip and palate. 6. Techniques of palate repair. Cleft Palate J 1987;24:64–77.
21. Heliövaara A, Rintala A, Ranta R. One-stage closure of isolated cleft palate with the Veau–Wardill–Kilner V to Y pushback procedure or the Cronin modification. I. Comparison of operative results. Scand J Plast Reconstr Hand Surg 1993;27:49–54.
22. Sorva R, Perheentupa J, Tolppanen E-M. A novel format for a growth chart. Acta Paediatr Scand 1984;73:527–9.
23. Moorrees CFA. The dentition of the growing child. Cambridge (MA): Harvard University Press; 1959.
24. Laitinen S, Heliövaara A, Pere A, Ranta R. Growth in children with Pierre Robin sequence and isolated cleft palate. Acta Paediatr 1994;83:1161–4.
25. Jensen BL, Dahl E, Kreiborg S. Longitudinal study of body height, radius length and skeletal maturity in Danish boys with cleft lip and palate. Scand J Plast Reconstr Surg 1983;91:473–81.
26. Day DW. Accurate diagnosis and assessment of growth in patients with orofacial clefting. Birth Defects 1985;21:1–14.
27. Menius JA, Largent MD, Vincent CJ. Skeletal development of cleft palate children as determined by hand–wrist roentgenographs: a preliminary study. Cleft Palate J 1966;3:67–75.
28. Geier M, Dahlman H. Unterschiede im Wachstum und Entwicklung zwischen Spaltträgern und Nichtspaltträgern. Fortschr Kieferopthop 1988;49:444–53.
29. Ranta R, Rintala A. The Pierre Robin anomalad—comparison of some disturbances in the formation of the teeth and the lower lip. Proc Finn Dent Soc 1983;79:155–61.
30. Haapanen M-L, Laitinen S, Paaso M, Ranta R. Quality of speech correlated to craniofacial characteristics of cleft palate patients with Pierre Robin sequence. Folia Phoniatr Logop 1996;48:215–22.

Received for publication 27 May 1997
Accepted 21 October 1997