Clin Oral Invest (2012) 16:1261–1266
DOI 10.1007/s00784-011-0601-4

ORIGINAL ARTICLE

Predictors of poor dental arch relationship in young children with unilateral cleft lip and palate

Yuh-Jia Hsieh · Yu-Fang Liao · Akshai Shetty

Received: 2 July 2010 / Accepted: 3 August 2011 / Published online: 12 August 2011

Abstract The aim of this cross-sectional outcome study using retrospective data capture of treatment histories was to examine the characteristics of young children with unilateral cleft lip and palate who had poor dental arch relationship (i.e., Goslon 5). The study sample comprised 120 children born with nonsyndromic complete unilateral cleft lip and palate between 1995 and 2003, and were aged between 5.0 and 7.0 years (mean age, 5.1 years) at the time of data collection. The dental arch relationship was assessed using the Goslon yardstick from intraoral dental photographs. An independent investigator recorded treatment histories from the clinical notes. The inter- and intra-examiner agreements evaluated by weighted kappa statistics were high. There was no association between dental arch relationship and the type of presurgical orthopedics or pharyngeal flap. Dental arch relationship was associated with the initial cleft size (odds ratio, OR=1.3; 95% confidence interval, CI=1.1−1.5, $p<0.01$), surgeon grade for palate repair (OR=5.0, 95% CI=1.2−19.9, $p<0.05$), and primary gingivoperiosteoplasty (OR=2.8, 95% CI=1.0−8.1, $p=0.05$). These data suggest that intraoral dental photographs provide a reliable method for rating dental arch relationship. Wide initial cleft, high-volume surgeon, and primary gingivoperiosteoplasty are predictors of poor dental arch relationship outcome in young children with unilateral cleft lip and palate. These findings may improve treatment outcome by modifying the treatment protocol for patients with unilateral cleft lip and palate.

Keywords Cleft size · Dental arch relationship · Gingivoperiosteoplasty · Presurgical orthopedics · Treatment outcome · Unilateral cleft lip and palate

Y.-J. Hsieh · Y.-F. Liao (✉)
Department of Craniofacial Orthodontics and Craniofacial Center,
Chang Gung Memorial Hospital,
199 Tung-Hwa North Road,
Taipei 105, Taiwan
e-mail: yfliao0125@yahoo.co.uk

Y.-J. Hsieh · Y.-F. Liao
College of Medicine, Chang Gung University,
Taoyuan, Taiwan

A. Shetty
Department of Orthodontics, DAPM RV Dental College,
Bangalore, India

Introduction

Despite consensus on the need for centralized treatment of patients with unilateral cleft lip and palate (UCLP), there is still no generally accepted protocol [1, 2]. One of the aspects of treatment protocols on which no consensus exists is the primary infant management. Some investigators believe that presurgical orthopedics (PSO) before lip repair is advisable [3]. Effects of passive PSO (i.e., dental plate) on facial growth have been recently evaluated through randomized controlled trials [4, 5]. The findings do not substantiate the claim that fitting a dental plate permits favorable growth of the maxilla. Some studies demonstrated that active PSO (i.e., dental plate with extraoral strapping) had a harmful effect on the growth of the maxilla [6, 7]. To improve outcomes, clinicians have used other procedures, such as extraoral lip taping or elastic traction for nasoalveolar molding [8, 9]. Whether these procedures influence growth outcome is unknown.

Surgery at the alveolar cleft may also have the potential to disrupt facial growth. Primary bone grafting or Skoog's gingivoperiosteoplasty (GPP) in infancy has been shown to disrupt maxillary growth [6]. Millard and Latham [10]

modified Skoog's GPP technique by incorporating active PSO (i.e., pin-retained dental plate) to narrow the alveolar gap before the GPP, reducing the amount of periosteal undermining needed to perform the GPP. The effect of Millard's GPP on maxillary growth remains controversial. One group found no difference in maxillary growth following GPP [11, 12]. In contrast, others found poor maxillary growth in patients who had GPP [13–17]. However, it is impossible to clarify whether the growth disturbance was attributable to the active PSO or Millard's GPP.

The problems of maxillary growth generally are reflected in the vertical, transverse, and anteroposterior dental arch relationship. The Goslon yardstick [18] is one of the most commonly used methods to assess dental arch relationship of patients with UCLP. The system initially was developed for late mixed dentition and early permanent dentition. According to Noverraz et al. [19], the yardstick is also useful at all stages of dental development and is suitable for longitudinal research. In Chang Gung, the treatment protocol for primary infant management has changed over time. Before 1999, passive PSO (i.e., dental plate) was used. From 1999 to 2001, passive PSO was replaced with two kinds of active PSO for nasoalveolar molding, either with extraoral lip taping alone (active 1) or taping combined with elastic traction (active 2). Primary GPP was performed at times until 2001. Since the treatment protocol has been evolving, the aim of this study was to examine the independent influence of different aspects of primary infant management on dental arch relationship in children with UCLP.

Methods

Patient population

Consecutively treated patients were selected on the basis of the following criteria: Taiwanese patients with nonsyndromic complete UCLP who were born between 1995 and 2003 and were treated at the Chang Gung Craniofacial Center, Taipei, Taiwan; no Simonart's band; PSO before lip repair; modified rotation–advancement lip repair at the age of 3–6 months; primary Millard's GPP at the time of lip repair, if any; one-stage two-flap palatoplasty at age 1 year; pharyngeal flap surgery for secondary velopharyngeal insufficiency, if any; and no orthodontic treatment or other craniofacial surgery such as alveolar bone grafting, orthognathic surgery, or distraction osteogenesis prior to photographic assessment at around the age of 5 years. The diagnosis was confirmed by neonatal photographs and a chart description written by a plastic surgeon, an orthodontist, or a clinical geneticist.

A total of 120 patients met the above criteria. Table 1 provides the characteristics for all patients. There was a preponderance of males. Most patients had active PSO. Only 19% of the patients underwent primary GPP at the time of lip repair. Thirty-one different surgeons (4 senior attendings, 27 fellows) were involved in all primary repairs. Most patients received one-stage palate repair by fellow surgeons.

Treatment history

One investigator (YJH) reviewed each patient's clinical notes. Details of the treatment history were recorded, including initial cleft size as defined by the anterior cleft width, age at the time of the operation, and whether a GPP was undertaken in conjunction with primary lip repair. The technique used for primary repair and the details of pharyngeal flap surgery for secondary velopharyngeal insufficiency were noted. The grade of the surgeon who undertook the lip repair and the palate repair was also

Table 1 Demographics and treatment characteristic of the patients

	Patients (n=120)
Gender, n (%)	
Male	80 (33)
Female	40 (67)
Distribution of cleft, n (%)	
Right	39 (32)
Left	81 (68)
Mean age (SD) at assessment, years	5.1 (0.3)
Mean size (SD) of initial cleft, mm	10.5 (3.8)
Presurgical orthopedics, n (%)	
Passive (dental plate)	31 (26)
Active 1 (dental plate/tape)	40 (33)
Active 2 (dental plate/tape/elastic)	49 (41)
Lip repair	
Mean age (SD) at repair, months	3.4 (1.1)
Surgeon grade, n (%)	
Attending	120 (100)
Primary GPP, n (%)	
Yes	23 (19)
No	97 (81)
Palate repair	
Mean age (SD) at repair, months	12.3 (1.5)
Surgeon grade, n (%)	
Attending	47 (39)
Fellow	73 (61)
Pharyngeal flap, n (%)	
Yes	15 (12)
No	105 (88)

detailed. The surgeons were divided into attendings, those who were able to operate independently, and fellows, those who were surgeons in training.

Outcome assessment

The dental arch relationship of the intraoral dental photographs (Fig. 1) was scored on two separate occasions by two calibrated examiners (YJH and YFL) with the use of the Goslon yardstick [18, 20]. The yardstick contained a 5-point scale. A score of 1 represents the most favorable, with positive overjet and overbite that would be treated only by conventional orthodontics; patients with a score of 5 would generally require orthognathic surgery because of the severe skeletal class III malocclusion. No conferring between examiners was allowed, and the final score for each set of photographs was agreed by consensus between the two examiners.

Statistical analysis

Patient characteristics and descriptive statistics were summarized. Continuous data were expressed as means ± standard deviation, and nominal data were expressed as frequency and percentage. The quadratic-weighted kappa statistics was used to evaluate inter- and intraexaminer reliability. The degree of agreement was interpreted as described by Altman [21]. Weighted kappa values less than 0.20 indicate poor agreement; 0.21–0.40, fair agreement; 0.41–0.60, moderate agreement; 0.61–0.80, good agreement; and 0.81–1.00, very good agreement.

Bivariate analysis was first performed to assess the relationship between the outcome of dental arch relationship and five potential variables, initial cleft size, type of PSO, GPP, surgeon grade for palate repair, and pharyngeal flap surgery. When a significant effect was found ($p \leq 0.2$), the variables were then incorporated into the multiple logistic regression model. The results from the logistic regression analysis were reported as odds ratios with 95% confidence intervals. The odds ratios were the ratios in the odds of poor dental arch relationship with one more unit in the explanatory variable than another one. The p values were two sided and considered to be significant if $p \leq 0.05$.

Results

The weighted kappa values for the interexaminer agreement ranged from 0.86 to 0.95, indicating very good agreements between the examiners. The kappa values for the intraexaminer agreement ranged from 0.95 to 0.99, indicating very good agreements. For Goslon score distribution, 1% had a Goslon score of 1, 3% had a score of 2, 17% had a score of 3, 58% had a score of 4, and 21% had a score of 5.

Fig. 1 Seven views of one set of intraoral dental photographs for one patient. (*Above*, *left*) A frontal view in occlusion. (*Above*, *center* and *right*) Right and left overjet views. (*Center*) Right and left buccal views in occlusion. (*Below*) Upper and lower arch occlusal views

This means that 21% of the patients had a poor treatment outcome that would require a combined surgical–orthodontic approach.

There was no association between dental arch relationship and the type of PSO or pharyngeal flap surgery. The multiple logistic regression analysis revealed that dental arch relationship was associated with the initial cleft size, surgeon grade for palate repair, and primary gingivoperiosteoplasty. Poor outcome was associated with wide initial cleft (odds ratio, OR=1.3, 95% confidence interval, CI=1.1−1.5, $p<0.01$), high-volume surgeons (OR=5.0, 95% CI=1.2−19.9, $p<0.05$), and primary gingivoperiosteoplasty (OR=2.8, 95% CI=1.0−8.1, $p=0.05$) (Table 2).

Discussion

Over the last two decades, the Goslon yardstick has usually been chosen as an outcome measure in studies evaluating dental arch relationship in patients with UCLP. Since then, increasing understanding of factors adversely affecting treatment outcome resulted in an improvement of protocols. The Goslon yardstick was originally applied to dental casts. Intraoral photographs have been proven as a viable alternative to the application of the Goslon yardstick on dental casts [20, 22]. The weighted kappa value of the present study approves good reliability of the photographic method. Although the Goslon yardstick is useful for longitudinal assessment, the original 10-year-old yardstick should be modified to provide better prediction of future outcome for 5-year-old children. Mars and coworkers [23] suggest that Goslon 3 should be rated as Goslon 2 and Goslon 4 as Goslon 3 since the edge-to-edge bite is normal in a 5-year-old and the lingual eruption of the mandibular permanent incisors to their predecessors. In this study, we did not modify the Goslon yardstick in this way. We categorized Goslon 5 as poor dental arch relationship, rather than considering Goslon 4 and 5 as poor outcomes.

This study demonstrated that patients with UCLP who had a wide initial cleft are more likely to develop poor dental arch relationship by age 5. This observation is consistent with previous literature in this area [24]. Two hypotheses have been proposed to explain the tendency of patients with a wide cleft to have a poor outcome: (1) an intrinsic hypothesis implying an inherent tissue deficiency and (2) an iatrogenic hypothesis suggesting a surgical influence. Embryologically, there is not enough tissue to permit fusion of the segments. Postnatally, there is not enough tissue to permit "normal" surgical approximation of the segments. Obviously, too, surgery damages tissues and creates scarring that hampers subsequent growth.

There is increasing belief that surgeon experience may be a more important influence on the outcome than the timing or technique used for primary repair [1, 6], but it is currently less rigorously evaluated. Differences in outcomes achieved by centers in the Eurocleft study may be due, in part, to variation in surgeon experience [1]. Surprisingly, we found a strong association between poor dental arch relationship and high-volume surgeons (Table 3) in our sample. This is in direct contrast to the work of Williams et al. [25] who previously reported that high-volume surgeons achieved good speech outcome. The explanation for the discrepant results is unclear, although several additional studies have failed to find a relationship between the dental arch relationship and the volume of unilateral cleft lip and palate repairs undertaken by the surgeon [26–28]. Whether this relates to difficulty of case mix (i.e., the experienced surgeons had been assigned the difficult cases), sample size, or other variables is unclear at this time. Further prospective work is needed.

Primary Millard's GPP was also associated with poor dental arch relationship. Although a number of studies have addressed the issue of Millard's GPP predisposition to poor maxillary growth, no clear conclusion has been reached as all the GPP patients also had active PSO [13–17]. Similarly, all the nonGPP patients did not have PSO. Thus, it is impossible to differentiate the independent effects of the active PSO versus the GPP on growth. As a result, in this study, we combined assessments of GPP and active PSO, and we found that the GPP rather than the active PSO is a risk factor for poor outcome.

By the age of 5, a child with UCLP may have undergone pharyngeal flap surgery. The lack of association between the outcome dental arch relationship and pharyngeal flap in our sample is also noteworthy. In theory, pharyngeal flap could restrict facial growth by tethering the palate to the

Table 2 Multiple logistic regression for poor dental arch relationship

Covariate	Odds ratio	95% Confidence interval	*p* value
Initial cleft size (mm)	1.3	(1.1 to 1.5)	0.003
Surgeon grade for palate repair	5.0	(1.2 to 19.9)	0.02
Primary GPP	2.8	(1.0 to 8.1)	0.05

Table 3 Case load per surgeon for palate repair

	Case load
Median no. of palate repairs per attending (interquartile range)	5.5 (1.5−28.3)
Median no. of palate repairs per fellow (interquartile range)	2 (1−3)

posterior pharyngeal wall. However, the results of previous retrospective studies are contradictory [26, 29, 30]. Future research should be of clinical importance.

The finding that poor facial growth is associated with a wide cleft has some clinical implications. Treatment outcome in terms of growth could be anticipated according to cleft size. In the case of a child with a wide cleft, a high chance for poor growth outcome might be expected. The treatment protocol could vary according to the cleft size. In the case of a child with a wide cleft, prone sleep position, a later closure of the palate, or a staged palatal closure might be proposed. Infants sleeping in prone position could reduce cleft size [31]. Previous studies also showed that a later palate repair results in favorable maxillary growth because possible interference with maxillary growth is postponed to a later age when less growth remains [27, 28, 32–34] and that a staged palatal closure by starting closure of the soft palate with a posterior vomer flap incorporation may narrow the size of the remaining cleft spontaneously [35] or by starting closure of the hard palate with a single layer vomer flap may facilitate a smaller later palate repair [1].

It should also be mentioned that due to problems with facial growth, the surgical protocol of treatment of UCLP employed in Chang Gung has been modified. At present, primary GPP is no longer practiced.

From the results of this study, we conclude that intraoral dental photographs provide a reliable method for rating dental arch relationship. Wide initial cleft and primary gingivoperiosteoplasty are predictors of poor dental arch relationship outcome in young children with unilateral cleft lip and palate. In contrast, low-volume surgeon is protective against a poor outcome. Whether the predictors will change with time requires long-term follow-up.

Acknowledgments The authors thank Dr. Yu-Ting Chiu for her help in the preparation of the manuscript.

Conflict of interest The authors declare no competing interests.

Funding The authors declare no sources of funding for the study.

References

1. Shaw WC, Dahl E, Asher-McDade C et al (1992) A six-center international study of treatment outcome in patients with clefts of the lip and palate: part 5. General discussion and conclusions. Cleft Palate Craniofac J 29:413–418
2. Bearn D, Mildinhall S, Murphy T et al (2001) Cleft lip and palate care in the United Kingdom–the Clinical Standards Advisory Group (CSAG) Study. Part 4: outcome comparisons, training, and conclusions. Cleft Palate Craniofac J 38:38–43
3. Hotz MM, Gnoinski WM (1979) Effects of early maxillary orthopaedics in coordination with delayed surgery for cleft lip and palate. J Maxillofac Surg 7:201–210
4. Bongaarts CAM, Kuijpers-Jagtman AM, van 't Hof MA, Prahl-Andersen B (2004) The effect of infant orthopedics on the occlusion of the deciduous dentition in children with complete unilateral cleft lip and palate (Dutchcleft). Cleft Palate Craniofac J 41:633–641
5. Bongaarts CAM, van 't Hof MA, Prahl-Andersen B, Dirks IV, Kuijpers-Jagtman AM (2006) Infant orthopedics has no effect on maxillary arch dimensions in the deciduous dentition of children with complete unilateral cleft lip and palate (Dutchcleft). Cleft Palate Craniofac J 43:665–672
6. Ross RB (1987) Treatment variables affecting facial growth in complete unilateral cleft lip and palate. Cleft Palate J 24:5–77
7. Molsted K, Dahl E, Skovgaard LT et al (1993) A multicentre comparison of treatment regimens for unilateral cleft lip and palate using a multiple regression model. Scand J Plast Reconstr Surg Hand Surg 27:277–284
8. Grayson BH, Santiago PE, Brecht LE, Cutting CB (1999) Presurgical nasoalveolar molding in infants with cleft lip and palate. Cleft Palate Craniofac J 36:486–498
9. Grayson BH, Cutting CB (2001) Presurgical nasoalveolar orthopedic molding in primary correction of the nose, lip, and alveolus of infants born with unilateral and bilateral clefts. Cleft Palate Craniofac J 38:193–198
10. Millard DR Jr, Latham RA (1990) Improved primary surgical and dental treatment of clefts. Plast Reconstr Surg 86:856–871
11. Wood RJ, Grayson BH, Cutting CB (1997) Gingivoperiosteoplasty and midfacial growth. Cleft Palate Craniofac J 34:17–20
12. Lee CT, Grayson BH, Cutting CB, Brecht LE, Lin WY (2004) Prepubertal midface growth in unilateral cleft lip and palate following alveolar molding and gingivoperiosteoplasty. Cleft Palate Craniofac J 41:375–380
13. Henkel KO, Gundlach KK (1997) Analysis of primary gingivoperiosteoplasty in alveolar cleft repair. Part I: facial growth. J Craniomaxillofac Surg 25:266–269
14. Millard DR, Latham R, Huifen X, Spiro S, Morovic C (1999) Cleft lip and palate treated by presurgical orthopedics, gingivoperiosteoplasty, and lip adhesion (POPLA) compared with previous lip adhesion method: a preliminary study of serial dental casts. Plast Reconstr Surg 103:1630–1644
15. Berkowitz S, Mejia M, Bystrik A (2004) A comparison of the effects of the Latham-Millard procedure with those of a conservative treatment approach for dental occlusion and facial aesthetics in unilateral and bilateral complete cleft lip and palate: part I. Dental occlusion. Plast Reconstr Surg 113:1–18
16. Matic DB, Power SM (2008) The effects of gingivoperiosteoplasty following alveolar molding with a pin-retained latham appliance versus secondary bone grafting on midfacial growth in patients with unilateral clefts. Plast Reconstr Surg 122:863–870
17. Hsieh CH, Ko EW, Chen PK, Huang CS (2010) The effect of gingivoperiosteoplasty on facial growth in patients with complete unilateral cleft lip and palate. Cleft Palate Craniofac J 47:439–446
18. Mars M, Plint DA, Houston WJ, Bergland O, Semb G (1987) The Goslon yardstick: A new system of assessing dental arch relationships in children with unilateral clefts of the lip and palate. Cleft Palate J 24:314–322
19. Noverraz AE, Kuijpers-Jagtman AM, Mars M, van't Hof MA (1993) Timing of hard palate closure and dental arch relationships in unilateral cleft lip and palate patients: a mixed-longitudinal study. Cleft Palate Craniofac J 30:391–396
20. Liao YF, Huang CS, Lin IF (2009) Intraoral photographs for rating dental arch relationships in unilateral cleft lip and palate. Cleft Palate Craniofac J 46:415–419
21. Altman DG (1991) Practical statistics for medical research. Chapman and Hall, London, pp 404–408
22. Liao YF, Lin IF (2009) Dental arch relationships after two-flap palatoplasty in Taiwanese patients with unilateral cleft lip and palate. Int J Oral Maxillofac Surg 38:1133–1136

23. Mars M, Batra P, Worrell E (2006) Complete unilateral cleft lip and palate: validity of the five-year index and the Goslon yardstick in predicting long-term dental arch relationships. Cleft Palate Craniofac J 43:557–562
24. Peltomaki T, Vendittelli BL, Grayson BH, Cutting CB, Brecht LE (2001) Associations between severity of clefting and maxillary growth in patients with unilateral cleft lip and palate treated with infant orthopedics. Cleft Palate Craniofac J 38:582–586
25. Williams AC, Sandy JR, Thomas S, Sell D, Sterne JA (1999) Influence of surgeon's experience on speech outcome in cleft lip and palate. Lancet 354:1697–1698
26. Williams AC, Sandy JR (2003) Risk factors for poor dental arch relationships in young children born with unilateral cleft lip and palate. Plast Reconstr Surg 111:586–593
27. Liao YF, Cole TJ, Mars M (2006) Hard palate repair timing and facial growth in unilateral cleft lip and palate: a longitudinal study. Cleft Palate Craniofac J 43:547–556
28. Liao YF, Yang IY, Wang R, Yun C, Huang CS (2010) Two-stage palate repair with delayed hard palate closure is related to favorable maxillary growth in unilateral cleft lip and palate. Plast Reconstr Surg 125:1503–1510
29. Semb G, Shaw WC (1990) Pharyngeal flap and facial growth. Cleft Palate J 27:217–224
30. Oberoi S, Chigurupati R, Vargervik K (2008) Morphologic and management characteristics of individuals with unilateral cleft lip and palate who required maxillary advancement. Cleft Palate Craniofac J 45:42–49
31. Huang CS, Cheng HC, Chen YR, Noordhoff MS (1994) Maxillary dental arch affected by different sleep positions in unilateral complete cleft lip and palate infants. Cleft Palate Craniofac J 31:179–184
32. Lilja J, Mars M, Elander A et al (2006) Analysis of dental arch relationships in Swedish unilateral cleft lip and palate subjects: 20-year longitudinal consecutive series treated with delayed hard palate closure. Cleft Palate Craniofac J 43:606–611
33. Sinko K, Caacbay E, Jagsch R et al (2008) The GOSLON yardstick in patients with UCLP: review of a Vienna sample. Cleft Palate Craniofac J 45:87–92
34. Fudalej P, Katsaros C, Bongaarts C, Dudkiewicz Z, Kuijpers-Jagtman AM (2011) Dental arch relationship in children with complete unilateral cleft lip and palate following one-stage and three-stage surgical protocols. Clin Oral Investig 15:503–510
35. Owman-Moll P, Katsaros C, Friede H (1998) Development of the residual cleft in the hard palate after velar repair in a 2-stage palatal repair regimen. J Orofac Orthop 59:286–300