Oral anatomical defects associated with cleft palate and cleft lip

E. Jiménez-Castellanos[2], A. Carmona[1], C.J. Catalina-Herrera[1] and J. Jiménez-Castellanos[1]

1- Departamento de Anatomía y Embriología Humana
2- Departamento de Estomatología. Facultad de Medicina, Universidad de Sevilla, 41009 Sevilla, Spain

SUMMARY

Patients suffering from a cleft palate and/or cleft lip present evident anatomical defects in both formations. However, these developmental disorders are often accompanied by important disturbances in other anatomical structures of the mouth and adjacent tissues that may affect normal dental occlusion and, consequently, the basic functions of mastication and phonation. The objectives of the present work were to describe and discuss the above structural modifications, presenting several clinical cases in which the anatomical defects are described together with the functional implications. The anomalies found include important variations in the number of teeth, the position, disposition and relation of the alveolar processes, and the impairment of dental occlusion. In the cases studied, these anatomical defects have elicited prominent alterations in the normal functions of mastication and phonation, and also in facial aesthetics. Photographs of mouths, dental casts, and X-rays are used to illustrate our report.

Key words: Cleft palate – Cleft lip – Oral cavity – Dental occlusion – Mastication

INTRODUCTION

During the fifth and sixth weeks of prenatal life, the primary palate is formed below the primitive posterior nares. From this structure, the upper lip, the anterior portion of the maxillary alveolar processes, and the rostralmost portion of the palate will develop later. While the nasal septum is developing, each mass of maxillary mesoderm originates yet another medially directed extension: the palatal processes (lateral palatal shelves). These processes extend medially below and behind the primitive posterior nares on a level with the primitive palate. With further growth, the free edges fuse: first with the posterior margin of the primitive palate, and then progressively with each other at the mid-line to form the secondary definitive palate. Thus, the oral and nasal cavities become separated and the hard and soft palates are formed (Sperber, 2001). Successful fusion of the three embryonic components of the palate involves a complicated synchronization of shelf movements with growth and withdrawal of the tongue and with growth of the mandible and head.

Cleft palate and cleft lip represent multifactorial developmental anomalies involving both genetic predisposition (Bender, 2000; Aldred, 2001; Vieira and Orioli, 2001) and environmental agents (Presscott et al., 2001).

Clefting of the palate and/or lip may be caused by failure at any of the stages of palatogenesis: defective growth or delayed elevation of the palatal shelves, defective shelf fusion, failure of medial-edge epithelial cell death, possible postfusion rupture, etc. (Berkovitz et al., 1995).

Patients with a cleft lip and/or cleft palate present evident anomalies in both structures. However, the development of these defects is often associated with anatomical deficiencies in

Submitted: July 12, 2004
Accepted: September 25, 2004

Correspondence to:
Prof. J. Jiménez-Castellanos. Departamento de Anatomía y Embriología Humana, Facultad de Medicina, Avda. Sánchez Pizjuán 4, 41009 Sevilla, Spain. Tel.: 34 94552865; Fax: 34 94381662. E-mail: jjcb@us.es

other oral and perioral structures, frequently involving normal dental occlusion and hence the main masticatory and phonatory functions.

Here we used data from clinical cases to focus on the description of different anatomical anomalies linked to abnormal palatal development.

Material and Methods

Five cases of cleft palate and/or cleft lip were used in this study. Photographs of mouths, dental casts and X-rays are used to illustrate our findings.

Results

Case 1.- Male aged 19, presenting a complete cleft palate without previous surgical treatment (Fig. 1, B). Corresponding to the location of the palatal defect, the left upper lateral incisor tooth was absent. A versioned palatolingual occlusion and an anterior open-bite had been partially corrected with orthodontic treatment (Fig. 1, A).

Case 2.- Female aged 25, showing cleft lip and cleft palate with previous surgical treatment. In relation to the palatal anomaly, the right upper incisor was absent (Fig. 1, C). As a consequence of the premaxillary bony defect, a narrowing of the maxillary bone and an inadequate support exerted by soft tissues, especially the lip, were observed, together with a mandibular protrusion (Fig. 1, D).

Case 3.- Female aged 46, showing cleft lip and a premaxillary defect. A maxillary compression with bilaterally cross-bite was also seen (Fig. 1, E). Alveolar borders were thick and high and were associated with defects in supporting tissues (upper lip) (Fig. 1, F, G). A mandibular protrusion was also evident (Fig. 1, H).

Case 4.- Male aged 19, presenting clefting of the palate and lip with several prior surgical corrections. Both left upper incisors were absent (Fig. 1, I), with important malocclusion mainly due to the right upper dental hemiarcade (Fig. 1, J). Maxillary compression, bilateral cross-bite, defective support from soft tissues and a protrusive mandibular position were also detected (Fig. 1, K).

Case 5.- Male aged 29, with cleft palate and lip and previous surgical treatment (Fig. 1, L). Both upper lateral incisors were absent (orthopantomographic study performed at age 5) (Fig. 1, M). There was an important malocclusion with maxillary compression followed by a prominent overgrowth of alveolar margins in height and width (Fig. 1, N). Other clinical signs were bilateral cross-bite and mandibular protrusion (Fig. 1, O).

Discussion

There is disagreement in the literature about the association of cleft palate/lip and hypodontia. Thus, some authors have pointed to a prevalence of hypodontia linked to clefting in 77% of cases; six times higher than in the normal population (Roth and Hirschfelder, 1991; Jiroutova and Mullerova, 1994; Shapira et al., 1999). In contrast, other authors have considered that hypodontia is due to dental loss as a consequence of the surgical operations performed on these patients. These authors argue that the prevalence of hypodontia in the postcanine region is similar to that found in the normal population (Lekkas et al., 2000; Budai et al., 2001; Wangpichit et al., 2001). Although the prevalence of hypodontia in postcanine regions might be expected to be similar to that of the normal population, in zones where the palatal defect is located it is frequent to find, in conjunction with the anterior palatal malformation, an associated agenesis of one or more teeth of the maxillary dental arcade, as occurred in our cases 1, 2, and 3. Most authors agree about the existence of considerable variations in the size, position, and disposition of the alveolar processes; in particular, in terms of a reduced width and depth of the dental arcades (Dericke et al., 1994). Consequently, such patients, in addition to presenting important speech problems (Lohmander-Agerskov et al., 1995; Laitinen et al., 1998; Nandukar, 2002), respond poorly to intensive phonatory articulation therapies (Van Demark and Hardin, 1986; Chapman, 1993).

Once the phonatory function had begun, these patients used as a sounding board not only their mouths but also their nasal cavities. Therefore, bearing in mind the considerable added variations of the alveolar processes seen in our cases 3, 4, and 5, we agree with the above authors as regards the poor response of these patients to phonatory articulation therapies, even after the anatomical defect has been solved using prothesic obturators. The patients still maintain a nasal tone of voice with marked difficulties to emitting certain palatolingual phonemes.

The disturbances of dental occlusion, even in previously treated patients, were present in more than 2/3 of this palatal malformation. The alterations most frequently found were posterior cross-bite, open-bite, and lingualized occlusion, together with a false prognathism (Da Silva Filho et al., 1998; Tomioka, 1998; Kruk-Jeromin et al., 1999; Johnson et al., 2000; Lambrecht et al., 2000; Prasad et al., 2000; Morris et al., 2000; Braumann et al., 2001). In many cases, these anomalies gave rise to severe deficiencies in masticatory function.

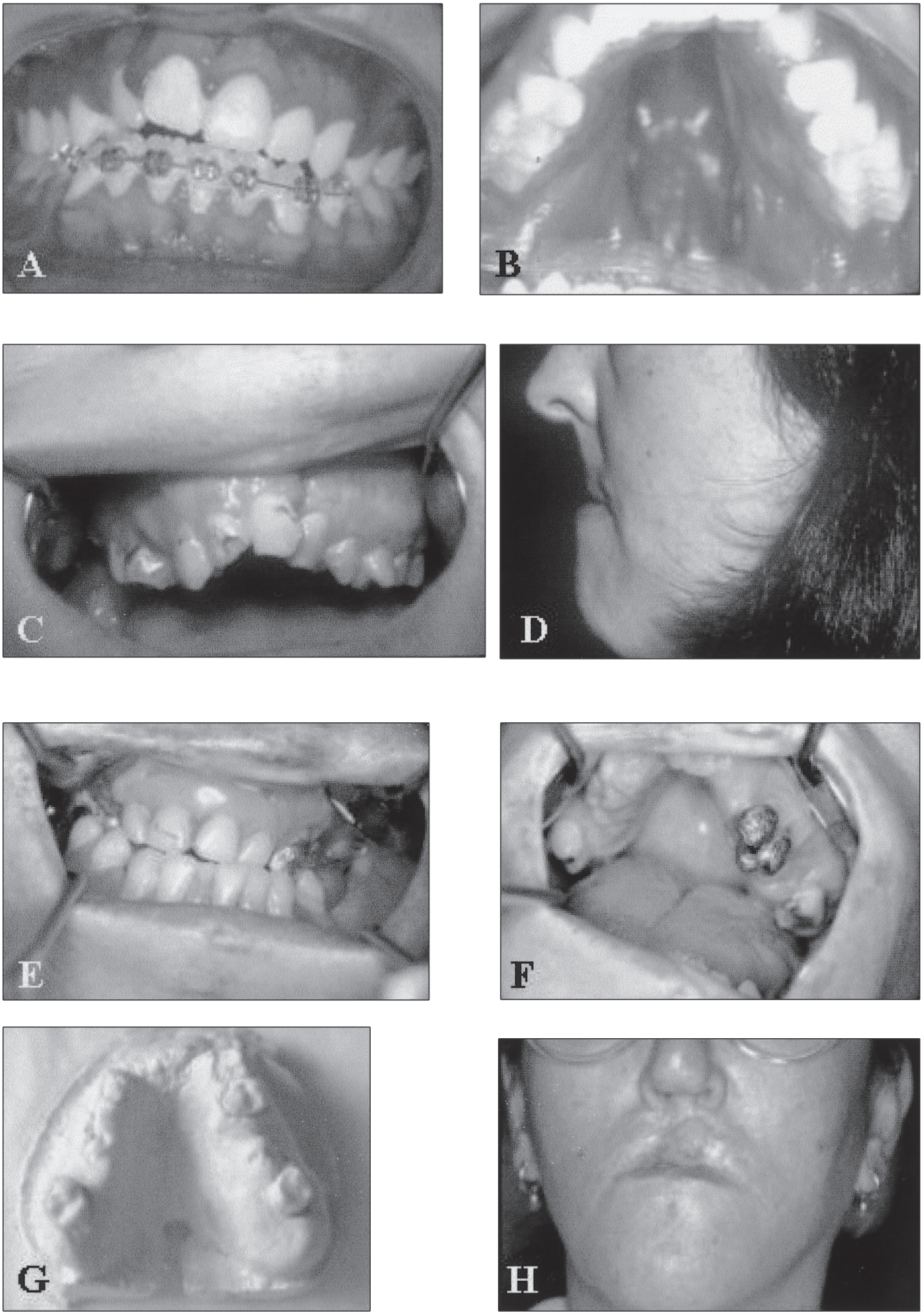

Fig. 1.- Part 1: **A**) Photograph showing an anterior open bite with absence of the upper left lateral incisor seen in case 1. **B**) Photograph showing a complete cleft palate found in case 1. **C**) An anterior open bite and absence of the right upper incisors (case 2). **D**) Photograph showing an inadequate support of the upper lip, together with a false prognathism found in case 2. **E**) Photograph of case 3 showing an anterior crossed bite and at the same time a bilateral posterior bite. **F**) Oral photograph showing high and wide alveolar rims with a narrowing and marked concavity of the palatal vault as seen in a palatal view of the upper dental arcade in case 3. **G**) Stucco model of the maxilla in case 3. **H**) Aesthetic defect and fibrosis of the upper lip accompanied with prognathism in case 3.

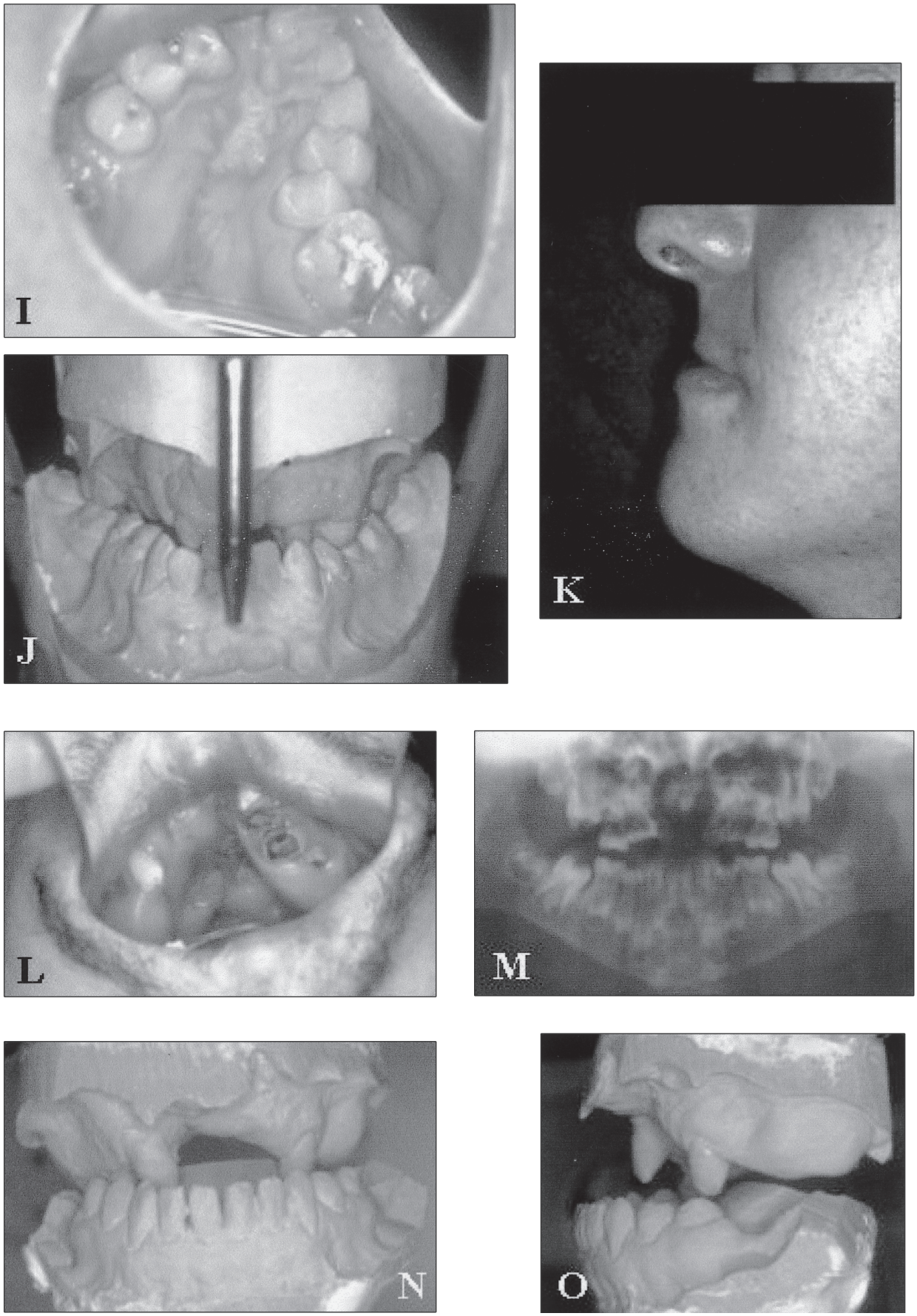

Fig. 1.- Part 2: **I**) Palatal view of the maxillary dental arcade showing the absence of the left upper incisors and a deformity of the upper left alveolar rim in case 4. **J**) Prominent malocclusion with posterior left cross-bite and maxillary compression in case 4. **K**) Aesthetic defect generated by inadequate support of the upper lip and false prognathism in case 4. **L**) Photograph showing an incomplete cleft palate found in case 5. **M**) Panoramic X-ray of the oral cavity in case 5 when the patient was aged 5. The premaxillary defect is seen, together with the absence of the permanent upper and anterior dental germs. **N**) Dental cast showing a prominent malocclusion, maxillary compression, and high and wide upper alveolar rims in case 5. **O**) Dental cast showing crossed anterior and bilateral posterior bites in case 5.

All the cases reported here are good representatives of disturbances of dental occlusion. Thus, they included an anterior open-bite (cases 1 and 2), anterior cross- bite (cases 3 to 5), lingual occlusion (cases 1, 3, and 5) and false prognathism (cases 2, 3, and 4). The most extreme situation was seen in our case 5, where tooth loss was accompanied by important malformations of the alveolar processes, with an absolute incongruence between the maxilla and mandible and the consequent impairment of masticatory function.

Also noteworthy in this issue is the existence of significant aesthetic effects, caused primarily by the weak support exerted by the upper lip as a consequence of the anterior bony and dental defects, as seen in our cases 2 and 4. This disturbance was also due to false prognathism (cases 2, 3 and 4) coupled to fibrosis and a poor elasticity of the upper lip, in many instances caused by repeated surgical interventions (case 3).

In conclusion, in addition to the developmental anomalies located in both structures, patients suffering from cleft palate/lip have important variations in the number of teeth, the position, disposition and relation of the alveolar processes, and an impairment due to dental occlusion. All these anatomical defects led to prominent alterations in the normal functions of mastication and phonation and in facial aesthetics.

References

Aldred MA (2001). Cleft lip and palate: new genetic clue. *Trends Mol Med*, 71: 539-540.

Braumann B, Rosenhayn SE, Bourauel C and Jager A (2001). Two- or three- dimensional cast analysis in patients with cleft lip and palate. *J Orofac Orthod*, 62: 451-465.

Bender DL (2000). Genetics of cleft lip and palate. *J Pediatr Surg*, 15: 242-249.

Berkovitz BKB, Holland GR and Moxham BJ (1995). *Atlas en color y texto de Anatomía oral, histología y embriología*, 2nd edition. Mosby/Doyma, Madrid, pp 236-238.

Budai M, Kocsis SG, Korai E, Sagi I and Mari A (2001). Caries, gingivitis and dental abnormalities in patients with cleft lip and palate. *Fogorv Sz*, 94: 197-199.

Chapman KL (1993). Phonologic processes in children with cleft palate. *Cleft Palate Cran J*, 30: 64-72.

Da Silva Filho OG, De Castro Machado FM, De Andrade AC, De Souza Freitas JA and Bishara SE (1998). Upper dental arch morphology of adult unoperated complete bilateral cleft lip and palate. *Am J Orthod Dentofac,* 114: 154-161.

Dericke A, Kuijpers-Jagtman AM, Lekkas C, Hardjowasito W and Latief B (1994). Dental arch dimensions in unoperated adult cleft-palate patients: an analysis of 37 cases. *J Craniofac Genet Dev Biol*, 14: 69-74.

Jiroutova O and Mullerova Z (1994). The occurrence of hypodontia in patients with cleft lip and/or palate. *Acta Chir Plast,* 36: 53-56.

Johnson N, Williams AC, Singer S, Southall P, Atack N and Sandy JR (2000). Dentoalveolar relations in children born with a unilateral cleft lip and palate (UCLP) in Western Australia. *Cleft Palate Cran J,* 37: 12-16 .

Kruk-Jeromin J, Antoszewski B and Kot M (1999). Assesment of long-term results in cleft lip and palate. *Med Wieku Rozwoj*, 3: 393-405.

Laitinen J, Ranta R, Pulkkinen J and Haapanen ML (1998). The association between dental arch dimensions and occurrence of Finnish dental consonant misarticulations in cleft lip/palate children. *Acta Odontol Scand*, 56: 308-312.

Lambrecht JT, Kreusch T and Schulz L (2000). Position, shape and dimension of the maxilla in unoperated cleft lip and palate patients: review of the literature. *Clin Anat*, 13: 121-123.

Lekkas C, Latief BS, Terrahe SP and Kuijpers-Jagtman AM (2000). The adult unoperated clefp patient: absence of maxillary teeth outside the cleft area. *Cleft Palate Cran J,* 37: 17-20.

Lohmander-Agerskov A, Soderpalm E, Friede H, Persson EC and Lilja J (1995). Pre-speech in children with cleft lip and palate or cleft palate only: phonetic analysis related to morphologic and functional factors. *Cleft Palate Cran J*, 32: 353-359.

Morris DO, Roberts-Harry D and Mars M (2000). Dental arch relationships in Yorkshire children with unilateral cleft lip and palate. *Cleft Palate Cran J*, 37: 453-462.

Nandukar A (2002). Nasalance measures in Marathi consonant-vowel-consonant syllables with pressure consonants produced by children with and without cleft lip and palate. *Cleft Palate Cran J*, 39: 59-65.

Prasad CN, Marsh JL, Longre J, Galic M, Huebener DV and Bresina SJ (2000). Quantitative 3D maxillary arch evaluation of two different infant managements for unilateral cleft lip and palate. *Cleft Palate Cran J,* 37: 562-570.

Presscott NJ, Winter RM and Malcom S (2001). Nonsyndromic cleft lip and palate: complex genetics and environmental effects. *Ann Hum Genet,* 65: 505-515.

Roth P and Hirschfelder U (1991). Frequency of tooth agenesis in CLP patients with eruption of all four third molars. *Dtsch Zahnarztl Z*, 46: 734-736.

Shapira Y, Lubit E and Kuftinec MM (1999). Congenitally missing second premolars in cleft lip and cleft palate children. *Dentofacial Orthop,* 115: 396-400.

Sperber GH (2001). *Craniofacial development*. BC Decker Inc, Hamilton, pp 114-122.

Tomioka T (1998). Mandibular movements in dentulous cleft lip and palate patients with lingualized occlusion. *Kokubyo Gakkai Zasshi*, 65: 213-232.

Van Demark DR and Hardin MA (1986). Effectiveness of intensive articulation therapy for children with cleft palate. *Cleft Palate Cran J*, 23: 215-224.

Vieira AR and Orioli IM (2001). Candidate genes for nonsyndromic cleft lip and palate. *ASDC J Dent Child,* 68: 272-279.

Wangpichit K, Huntington NL and Kapala JT (2001). Comparison of three nonradiographic methods of mixed dentition analysis in cleft lip and palate patients. *Pediatr Dent,* 23: 476-480.