Intra Oral Epithelial Bands in a New Born: Report of Two Cases and Review of Literature

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Received: 14 November 2010 / Accepted: 30 April 2011 / Published online: 17 May 2011
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Intra oral epithelial bands is a very rarely reported condition. It is sometimes associated with cleft lip and palate [1, 2]. Stenberg et al. suggested that these connective tissue bands stretch between the mandible and the maxilla [3]. Miskynyar reported that congenital adhesion occurs between maxilla and mandible (syngnathism) or between tongue and palatal margins (glossopalatal ankylosis) and he states that adhesions consist of membranous band of epithelium which is supported by connective tissue, muscles or bone [4]. Congenital fusion of jaws is also a very rare condition which may be unilateral or bilateral, and may or may not be associated with syndromes. In the long standing cases, temporomandibular joint ankylosis can occur [5].

Only 17 cases of intra oral bands were found by Spivack and Bennett [6] until 1968 reported as *palatoglossal ankylosis* in the literature. Laster et al. in 2001 [7] reported only 24 documented cases of congenital maxillo-mandibular fusion.

We at our centre Acharya Vinoba Bhave Rural Hospital Sawangi, Wardha, Maharashtra, India operated 1,134 cases

(Table 1) of cleft patients from February 2007 to July 2010, out of which 664 were males and 470 females. Out of which only two patients presented with synechiae and syngnathia associated with cleft palate. The syndromic patients were 710, the most common syndrome associated was Pierre Robins Sequence.

Case 1

A patient with intra oral epithelial bands (Fig. 1) along with ambiguous genitalia and club feet (Fig. 2). Ambiguous genitalia is a birth defect where the outer genitals do not have the typical appearance of either a boy or a girl [8].

A 2 day old neonate with a history of normal, full term delivery was referred to our out patient Department of Oral and Maxillofacial Surgery with the chief complaint of restricted mouth opening and difficulty in feeding.

On examination, multiple bands were present between upper and lower alveolus with cleft palate, bilateral congenital webs at the knees, bilateral club feet, and absence of scrotum. After complete routine haematological examination, patient was taken under dissociative anaesthesia and the fibrous bands were coagulated and cut with the help of bipolar electrocautery (Fig. 3). The mouth opening of 23 mm was achieved on table after releasing the bands. The post operative course was uneventful and patient was put to breast next day.

Case 2

A 6 day old neonate with a history of normal, full term delivery was referred to our out patient Department of Oral

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Table 1

Unilateral cleft lip	364
Bilateral cleft lip	77
Cleft palate	478
Alveolar bone grafting	38
Fistula repair	57
Secondary surgeries	120

Fig. 1 Patient with intra oral epithelial bands **Fig. 2** Photograph showing ambiguous genitalia and club feet

and Maxillofacial Surgery with the chief complaint of restricted mouth opening (Fig. 4) and difficulty in feeding.

On examination congenital bands were present between alveolus of both maxilla and mandible bilaterally and

Fig. 3 Photograph after excision of bands **Fig. 4** Photograph of patient with restricted mouth opening

between palate and tongue. After complete routine haematological examination and pediatric fitness, patient was taken under dissociative anaesthesia and adhesions were released with the help of bipolar electrocautery (Fig. 5). After releasing the adhesions, the mouth opening obtained was 20 mm. The post operative course was satisfactory and the patient was put to breast next day.

Discussion

A very few cases of congenital intra oral epithelial bands is reported in the literature [9, 10]. These alveolar adhesions were rarely seen in isolation, they are always accompanied by congenital defects [11, 12]. Donepudi et al. reported two

Fig. 5 Photograph of patient after adhesions were released with the help of bipolar electrocautery

cases of cleft palate lateral synechia (CPLS) syndrome in a single family and described surgical management of the synechia [13]. Albert K. Oh reported subglossopalatal synechia in association with cardiac and digital anomalies [14]. Snijman et al. reported congenital fusion of gums in a 3 week old child along with temporomandibular joint ankylosis [15]. Symptoms of the patient with intraoral bands are restricted opening of the mouth and feeding difficulties.

The presence of congenital intra oral epithelial bands is a very rare condition. Congenital intraoral bands may consist of epithelium supported by varying amounts of connective tissue. If only soft tissues are involved, the condition is termed as synechiae and the term syngnathia is used when bone is involved [16]. Failure of division (persistent oropharyngeal membrane) or abnormal epithelial behaviour are considered as the embryological explanation for synechia [17]. Different methods have been mentioned in the literature to excise these bands using surgical instruments [18], silk ligatures, scissors [19], electrocautery [20], and with scalpel [15]. We too used electrocautery for excision of epithelial bands in both the cases. The rationale behind the use of electrocautery was it prevents excessive bleeding and provides complete haemostasis.

Schonauer et al. reported endoscopic release of intra oral fibrous bands in a patient with popliteal pterygium syndrome [21]. A case of bony fusion of the jaws accompanied by microglossia, cleft palate and malformation of the hand has been described by Pettersson [22]. During the seventh to eighth week of intra uterine life when palatal shelves and tongue are in close contact, the palatal closure depends on the downward and forward movement of tongue and extension of neck. The failure of this predisposes to

alveolar fusion. Abnormal fusion can also be due to genetic, teratogenic or physical insults during this period, but most of the times they are syndrome related [7, 23]. Early intervention is necessary to avoid late temporomandibular joint related problems.

Conflict of Interest None.

References

- Shah RM (1977) Palatomandibular and maxillomandibular fusion, partial aglossia and cleft palate in a human embryo. Report of a case. *Teratology* 15:261
- Bagatln B (1990) Congenital intraoral epithelial bands: report of two cases. *J Oral Maxillofac Surg* 48:310–311
- Stenberg N, Sagher U, Golan J et al (1983) Congenital fusion of the gums with bilateral fusion of the temporomandibular joints. *Plast Reconstr Surg* 72:385
- Miskynar SA (1979) Congenital mandibulomaxillary fusion. *Plast Reconstr Surg* 63:120–121
- Mortazavi SH, Motamedi MH (2007) Congenital fusion of jaws. *Indian J Pediatr* 74(4):416–418
- Spivack J, Bennett JE (1968) Glossopalatine ankylosis. *Plast Reconstr Surg* 42:129
- Laster Z et al (2001) A complete bony fusion of the mandible to the zygomatic complex and maxillary tuberosity: a case report and review. *Int J Oral Maxillofac Surg* 30:75–79
- Lee MM, Donahoe PK (1997) The infant with ambiguous genitalia. *Curr Ther Endocrinol Metab* 6:216
- Hayward JR, Avery JK (1957) A variation in cleft palate. *J Oral Surg* 15:320
- Mathis H (1962) Über einen Fall von Emahrungsschwierigkeit bei connataler Syngnathise. *Dtsch Zahnrtl Z* 17:1167
- Goodarce TE, Wallace AF (1990) Congenital alveolar fusion. *Br J Plast Surg* 43:203–209
- Verdi GD, O'Neal B (1984) Cleft palate and congenital alveolar synechia syndrome. *Plast Reconstr Surg* 74:684–686
- Donepudi SK et al (2009) Cleft palate lateral synechia syndrome: an opportunity for unique surgical closure. *Int J Pediatr Otorhinolaryngol* 73(6):861–866
- Oh AK (2008) Subglossopalatal synechia in association with cardiac and digital anomalies. *Cleft Palate Craniofac J* 45(2):217–221
- Snijman PC et al (1966) Congenital fusion of the gums. *Am J Dis Child* 112(6):593–595
- Gartlan MG, Davies J, Smith RJ (1993) Congenital oral synechiae. *Ann Otol Rhinol Laryngol* 102(3):186–197
- Shaw WC, Simpson JP (1980) Oral adhesions associated with cleft lip and palate and lip fistulae. *Cleft Palate J* 17(2):127–131
- Verdi GD, O'Neal B (1984) Cleft palate and congenital alveolar synechia syndrome. *Plast Reconstr Surg* 74(5):684–686
- Discussion RP (1984) Cleft palate and congenital alveolar synechia syndrome. *Plast Reconstr Surg* 74(5):686
- Valnicek SM, Clarke HM (1993) Syngnathia: a report of two cases. *Cleft Palate Craniofac J* 30(6):582–585
- Schonauer I et al (2002) Endoscopic release of intraoral synechiae in popliteal pterygium syndrome. *Ann Plastic Surg* 49(5):550–552
- Pettersson G (1961) Aglossia congenital with bony fusion of the jaws. Report of one case. *Acta Chir Scand* 122:93
- Dalal M, Davidson PM (2002) Cleft palate congenital alveolar synechiae syndrome. Case report and review. *Br J Surg* 55:256–257