Nonsyndromic palate Synechia with floor of mouth

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ABSTRACT

To discuss the embryological basis, sequela and management of intraoral synechia, and to report on the incidence of this condition at a facial cleft deformity clinic (FCDC), with specific attention to two rare cases of mucosal bands involving the floor of the mouth and palate. Review of the literature and a retrospective analysis of FCDC and case report of two cases. During the period of 30 years (1983–2013), the FCDC - University of Pretoria has managed in excess of 4000 cases. A review of the clinic statistics revealed only six cases in which intraoral synechiae occurred. The rarity of this condition at the FCDC is in keeping with the rare incidence in the international literature. Four syndromic cases were identified. Three cases were cleft palate lateral synechia syndrome, and one was an orofacial digital syndrome. Two nonsyndromic cases were identified, and both cases involved the floor of the mouth and palate. The attending physicians and surgeons should be aware of the most appropriate timing for management of this condition, in order to avoid unwanted sequelae. Supportive care should be provided, and emergency airway protocol should be available for all cases. A differential diagnosis should be considered which includes syndromic conditions.

Keywords: Floor of the mouth, palate, synechia

INTRODUCTION

Synechia is a broad term which describes a fibrous or soft tissue connection between anatomical structures. Oral synechia presents in many different configurations usually involving the intraoral maxillary and mandibular structures. Congenital oral synechia is a rare phenomenon with only a few documented cases in the literature. The soft tissue fusion may be complete or incomplete, and may present as an isolated malformation or occur in the presence of other abnormalities.

This condition may be associated with syndromes or may less commonly be nonsyndromic. The syndromes which are associated are Demarque-Van der Woude syndrome (DVWS), popliteal pterygium syndrome (PPS) [Figures 1 and 2], cleft palate lateral alveolar synechia syndrome, orofacial digital syndrome (OFDS) and Fryns syndrome.

Various pathogenetic mechanisms have been proposed, however no single suggestion can be validated. This article describes two cases of an uncommon presentation of nonsyndromic intraoral floor of the mouth synechia.

CASE REPORTS

Case 1
A 6-week-old baby was referred to the facial cleft deformity clinic (FCDC)-University of Pretoria with a diffuse mucosal band extending from the floor of the mouth to the palate [Figures 3 and 4]. The weight was 3.2 kg and the birth weight was reported to be 2.5 kg. The baby received nasogastric feeds since after birth.

There was an associated mandibular retrusion and limited mouth opening. It was elected to perform an endoscopic oral examination under inhalation anesthesia, to exclude other congenital abnormalities beyond the mucosal bands. An associated soft palate cleft and 40% hard palate cleft was noted, with no further abnormalities.

The mucosal banding was separated with the aid of an electrocautery. This releases the mandibula and resulted in
improved mouth opening with significant feeding benefits [Figure 5]. Mandibular catch-up growth was noted at the long-term follow-up. The soft and hard palate defect was reconstructed at a later stage.

Case 2
A 17-day-old female was referred to the FCDC from a rural hospital. The baby was born at 38 weeks gestation with Apgar scores 9/10 and 10/10. The child was born with low birth weight and as a result was managed with nasogastric feeds in neonatal Intensive Care Unit.

At the time of consultation, the baby received expressed breast milk through cup feeds. The baby presented with a mucosal band extending from the floor of the mouth associated with the left sublingual gland [Figures 6 and 7], to the edges of the complete soft and partial (25%) hard palate cleft, which obstructed anterior posturing of the tongue. Surgical transection of the band was performed under inhalation anesthesia since endotracheal intubation was deemed too difficult.

Immediately, after removal of the band, it was noted that the tongue was displaced into the nasopharynx [Figure 8].

A glossopexy [Figure 9] was performed to maintain the tongue in an anterior position so as to prevent airway obstruction.

The baby progressed well and was discharged to homecare after 1 week following the procedure.

**DISCUSSION**

During the 7th week of embryological development, normal oral development depends on the downward and forward movement of the tongue to allow for the palatal shelf fusion in the midline. The tongue protrudes through the oral cavity and as a result prevents fusion of the oral components. With the absence of tongue protrusion, prolonged contact between the alveolar arches results in the fusion.[2,3]

A number of theories regarding the pathogenesis of oral synechia have been proposed, which were all based on abnormalities occurring during embryological development. In the publication of Dinardo et al. it is mentioned that Hayward and co-worker postulated the connections to be a result of the close contact of
the palate to the epithelium of the alveolar ridge or floor of the mouth. A commonly accepted theory proposed by Mathis in 1962 suggested that these fibrous bands to be remnants of the buccopharyngeal membrane.[1]

Goodacre and co-worker are quoted to concur with the theory suggested by Mathis, but also implicated the presence of amniotic bands in the vicinity of the developing branchial arches as another plausible etiological factor. Environmental factors, such as meclozine and high dosage Vitamin A genetic insults and other teratogenic agents which result in failure of migration of mesodermal elements into the midline structures, should never be discounted as possible causative factors.[2,5] Gartland is quoted to have proposed two etiological theories for cleft palate lateral synechiae syndrome. The first is due a persistent buccopharyngeal membrane, which prevents closure of the palate and causing entrapment of the soft tissue between the cleft margins. The second theory is as a result of a subglossopalatal membrane which forms prior to the development of the cleft, and displaces the tongue into the nasal cavity, resulting in closure approximation of the associated structure with subsequent tissue fusion.[3]

Oral synechia may present as an isolated abnormality or as a component of the syndrome. Common syndromic associations...
Blade or electrocautery device. It is the authors preference to achieve by merely disrupting the band with the aid of a surgical treatment under general anesthesia.

When surgical transection of the band is elected, this may be achieved by merely disrupting the band with the aid of a surgical blade or electrocautery device. It is the authors preference to utilize an electrosurgery unit with a needle-tip Colorado needle (Stryker Leibinger Inc., USA) as it allows for precise surgical and hemostatic control throughout the procedure. The selection of anesthetic techniques must be given serious consideration. Intubation is challenging and would be needed to be performed in a blind fashion. An alternative would be to provide inhalation anesthesia, while the surgeon expedites transaction of the band. As the airway remains a priority in these patients, provision should always be made for an emergency surgical airway.

Timing of the surgical intervention depends on whether the patient presents with an airway problem. The bands need to be transected as soon as possible. Surgery may be delayed for 2–3 weeks if feeding is a problem. This window allows for nutritional supplementation and weight gain. Adequate mouth opening is usually achieved after excision of the band.

**CONCLUSION**

The buccopharyngeal membrane initially serves as a barrier between the primitive oral cavity and the oropharynx. Complete or partial persistence of the buccopharyngeal membrane results in oral banding. Management of these patients rests on securing a definitive airway, provision of nutritional support, and resection of the bands at an early age, in order to prevent growth abnormalities and ankylosis.

**REFERENCES**


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