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Congenital Intra-Oral Adhesions: Lateral Palatal Synechia Associated with Cleft Palate: A Case Report and Review of the Literature

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Abstract

Background: Lateral palatal synechiae are rare congenital adhesions running from the free borders of the cleft palate to the lateral parts of the tongue or the oral cavity floor, typically found in cleft palate lateral synechiae syndrome. In this study, we present a case of a cleft palate associated with lateral synechiae (intraoral fibrous band) extending from the margin of the cleft palate to the floor of the mouth on the left side. Management of the oral synechia should aim to allow for airway stability, successful oral nutrition and oro-mandibular development. The surgical treatment started with Surgical transection of the band under general anesthesia to facilitate the palatoplasty procedure. Oral synechia may present as an isolated abnormality or as a component of the syndrome. Only a few isolated cases of oral synechia have been previously reported in the literature. In this study we discuss variations in intraoral synechiae associated with cleft palate and the etiology of lateral palatal synechiae through a literature review.

Keywords:- Cleft palate, palatal synechiae, congenital malformation..

INTRODUCTION

Synechia is a broad term which describes a soft tissue or fibrous connection between anatomical structures. Congenital oral synechia is a rare phenomenon usually involving the intraoral maxillary and mandibular structures with only a few documented cases in the literature.[1] The condition is defined as synechia if the connection consists of fibromucosal tissue or fibrous bands which is more common than the synostosis where there is a bony connection.[2]

Cleft lip and palate are the a common craniofacial abnormalities reported while synechiae or intraoral epithelial bands are rare.[1] Presence of lateral synechia and cleft palate together is very rare and is known as cleft palate-lateral synechia (CPLS) syndrome.[3] Oral synechia may occur in isolation or associated with other syndromes such as van der Woude syndrome (VWS), popliteal pterygium syndrome (PPS), cleft palate lateral synechiae (CPLS) syndrome, and oromandibular-limb hypogenesis syndrome.[1,2,4,5]

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The purpose of treatment is to achieve a normal mouth opening to facilitate: patent airway, Feeding process, oral and facial development as well as surgical management of the cleft. The present study reports a one-year old patient who was diagnosed with isolated cleft palate associated with intraoral fibrous bands and also a review of the literature.

CASE REPORT

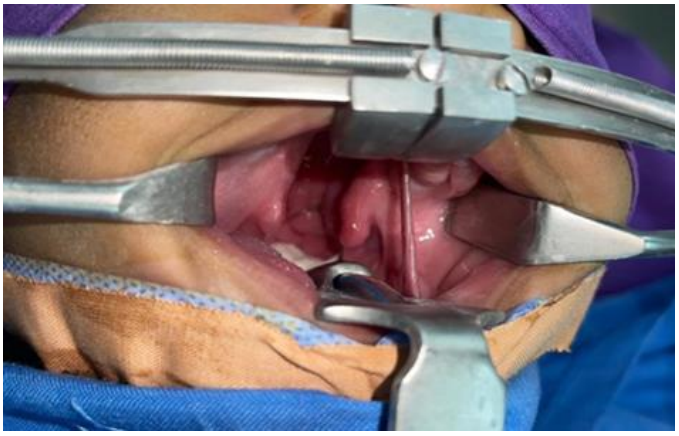

Figure 1: Intraoral view revealing the complete cleft palate with fibrous bands connecting the cleft margins to lateral tongue base.

A one-year old male patient was referred from ear, nose and throat (ENT) department to the maxillofacial department with incomplete isolated cleft palate and intraoral fibrous band which was present since birth. The family history was negative for oropharyngeal anomalies or cleft lip and palate, the child was vaccinated up to date. Physical examination revealed a cleft palate associated with lateral synechiae (intraoral fibrous band) extending from the margin of the cleft palate to the floor of the mouth on the left side [Figure 1A].

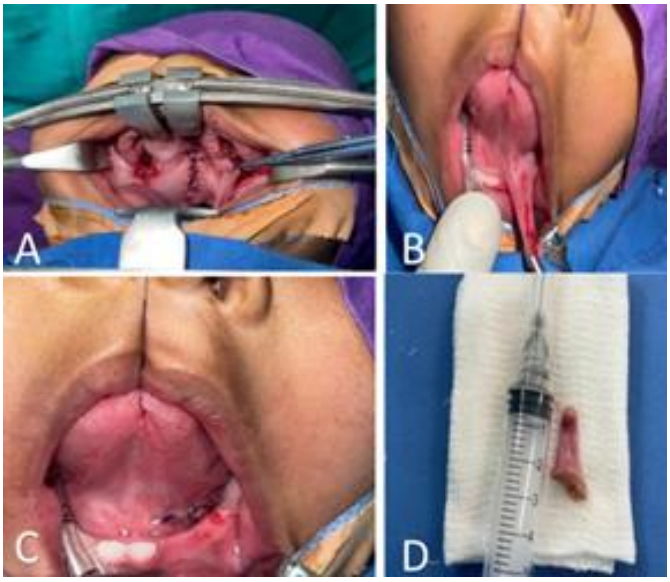

Figure 2: shows (A) The fibrous band released from the cleft palate margin and the palatoplasty procedure. (B) The fibrous band sill attached at the floor of the mouth. (c) Total excision of the band . (d) The excised fibrous band.

The mouth opening was mildly restricted. There were no lower lip pits or other deformities in the limbs or genital organs. The echocardiogram showed a normal heart anatomy and function. The pre-operative investigations were within normal ranges. The surgical treatment started with Surgical transection of the band under general anesthesia to facilitate the palatoplasty procedure. [Figure 2]

DISCUSSION

Congenital intraoral fibrous bands are uncommon. They may occur in isolation or more frequently in association with cleft lip and/or palate and other anomalies.[1] In the present case, the fibrous band was not isolated it was associated with incomplete cleft palate.

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The treatment started with division of the adhesions to facilitate the surgical repairs for the cleft palate.

During oral development, the downward and forward movement of the tongue allow the fusion of the palatal in the midline during the 7th week of embryological development. With the absence of tongue protrusion, prolonged contact between the alveolar arches results in the fusions.[6] A number of theories regarding the pathogenesis of oral synechia have been proposed, which were all based on abnormalities occurring during embryological development.

A commonly accepted theory by Mathis in 1962 suggested that these fibrous bands to be remnants of the buccopharyngeal membrane. As a consequence of prolonged contact between the tissues a subsequent fusion of the oral epithelium and adjacent structures occur.[1] Other authors describe the creation of a sub-glosso-palatal membrane caused by limited tongue mobility, which leads to fusion of this layer to the palate.[7] Another possible cause is genetics where Fuhrman et al proposed the name CPLS syndrome in a case involving 5 members of a family, suggesting an autosomal dominant inheritance Genetics.[8] Teratogenic, and mechanical insults during the first trimester may halt tongue descent and lead to cleft palate and abnormal fusion that results in intraoral bands between the upper and the lower jaws or the cleft margins and the floor of the mouth.[8]

There are a variety of locations in the mouth where the fibrous bands attach from the maxilla to the mandible. Ogino et al. (2005) reviewed 60 cases of intraoral synechiae and found 52 cases of synechiae located laterally in the oral cavity.[9] Oral Synechiae can be categorized into 4 groups based on the anatomy of the fibrous bands.

- The posterior glossopalatal type (buccopharyngeal synechia) is soft tissue adhesion extending from the soft palate to posterior tongue.
- The anterior midline glossopalatal type (glossopalatine synechia) is a soft tissue adhesion extending from the base of the tongue or floor of the mouth in the midline to the hard palate
- The lateral subglossopalatal type is soft tissue adhesion extending from the hard palate, mostly of the palatal cleft, to the lateral part of the tongue or the floor of the mouth.
- The interalveolar type is soft tissue adhesion between the maxillary and mandibular alveolar ridges.

Only a few isolated cases of oral synechia have been previously reported.[10,11] When the patient has both cleft palate and lateral subglossopalatal synechia, it is diagnosed as cleft palate lateral synechia syndrome. Fuhrmann and co- workers described the Cleft palate- lateral synechiae syndrome (CPLSS) which composed of mucosal banding from the floor of the mouth to margins of the cleft and micrognathia with great variability in the expression of the banding, which ranges from a thin friable membrane to thick mucosal bands.[8] In the present case, the patient is more likely to have cleft palate lateral synechia syndrome. He had isolated cleft palate and a lateral sub-glossopalatal fibrous bands with no other abnormalities like cleft lip or lower lip pits.

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Oral synechia may present as an isolated abnormality or as a component of the syndrome. Common syndromes associated are Orofacial digital syndrome, oro-mandibular limb hypogenesis syndrome and Van Der Woude syndrome are similarly reported to be associated with intraoral fibrous bands.[5] Syngnathia is also found in association with Flynn syndrome, which presents with coarse facial features, cloudy cornea, small eyes, cleft palate, defects of the diaphragm, hypoplasia of the lungs and distal limb deformities.

Management of the oral synechia should aim to allow for airway stability, successful oral nutrition and oro-mandibular development. Given the low incidence of Cleft palate- lateral synechiae syndrome (CPLSS), the evidence on the surgical management of intra-oral adhesions is limited to case reports.[7] Intubation is challenging in such patients an alternative would be to provide inhalation anesthesia, while the surgeon expedites transection of the band. As the airway remains a priority in these patients, provision should always be made for an emergency surgical airway.[12]

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.[13] Despite this, controversy exists on the timing of excision, either to be done early-on or delayed months later. Several authors describe purposefully delaying surgery until 6 months of age in order to facilitate operative techniques.[4] Interestingly, Donepudi et al. have mentioned the simultaneous excision of oral adhesions with reconstruction of the hard palate. They used the intra-oral bands as local flaps to help reconstruct the floor of the nose.[7] Nonetheless, this would require that the patient awaits several months prior to surgery.

The prognosis of intraoral fibrous bands is good. Mild TMJ restriction can occur, especially in the cases where fibrous band release is delayed. Mostly, limitations resolve after band release and a period of physical therapy.[14]

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CONCLUSIONS

Early diagnosis of oral synechiae, isolated or as a part of syndrome is important for management and providing good care for the patient. The goal of surgical intervention mainly to ensure airway patency, successful oral nutrition and oro-mandibular development.

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