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Oral Synechia–A Case Report with Review

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Abstract:
Background:*.Synechiae is rarely seen as an isolated disease. It is generally observed together with various syndromes such as Van der Woude and cleft palate lateral alveolar synechiae syndrome, and is concomitant with other anomalies in the maxillofacial or other regions of the body.*
Case:*This article reports a case of isolated congenital alveolar synechiae in a 3 year old baby girl. Release of the synechia was performed without complications. We review the current literature and discuss diagnosis and surgical management of this rare condition.*
Keywords*: Adhesion,Cleft palate, Congenital, Floor of the Mouth, Synechia,Syngantia,*

I. Introduction

Cleft palate with intraoral synechia is a rare congenital condition. It usually occurs sporadically, although familial cases are known in the literature (Furhmann et al., 1972). The presence of a single midline synechia is encountered less frequently than the more common lateral interalveolar adhesions, which may be single or multiple (Ogino et al., 2005)[1]. Although it may be present in isolation with cleft palate, alveolar synechiae are usually associated with Van der Woude syndrome, oromandibular limb hypogenesis syndrome, microglossia, micrognathia and popliteal pterygium syndrome[2,3]. This fusion can be unilateral or bilateral and includes synechiae or syngnathiaOral synechia presents in many different types which usually involving the intraoral maxillary and mandibular areas that include the gum pads ,lower lip,Tounge or oropharengyal isthmus.[5]Congenital oral synechia is a rare phenomenon with only a few documented literature.[4] Restricted mouth opening causes difficulty in feeding, swallowing, and respiration thus affecting the growth of the infant. In long-standing cases, termporomandibular joint (TMJ) ankylosis often occurs because of immobility and lack of function, necessitating more complicated surgical treatment. [3]The sooner the treatment is rendered the lesser the possibility of mandibular growth disturbance and facial deformities.

II. Case report

A 3 days old baby girl infant was referred to department of Oral and maxillofacial surgery in K.D Dental College And Hospital,Mathura, for examination and treatment. Initial examination shows upper and lower fusion (figure 1) i.e. A diffuse mucosal band extending from the floor of the mouth to the palate on left side and hard palateand impossibility of mouth opening for infant. Initial consultations with pediatricians and anesthetist were carried out and usual examinations were taken before the operation. General health of infant was confirmed by pediatrician. Also, no abnormality was reported in her family history. Surgical transection of the band was performed under inhalation anesthesia since endotracheal intubation was deemed too difficult.The mucosal banding was separated under conscious sedation with the aid of an electrocautery (figure.2). This releases the mandible and resulted in improved mouth opening. The band tissue was excised and sutured . Patient will be on long-term follow-up. The hard palate defect will be reconstructed at a later stage.

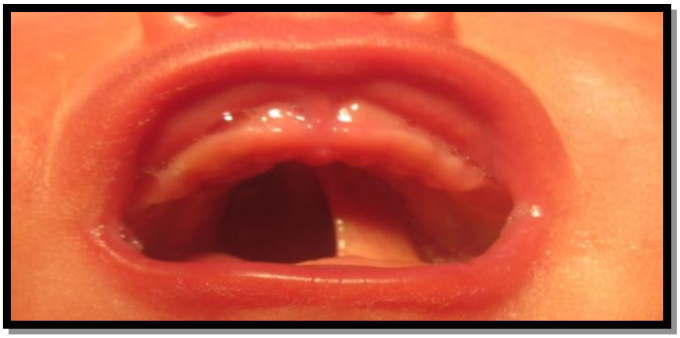

Figure1 :Intraoperative view of upper and lower fusion via fibours band

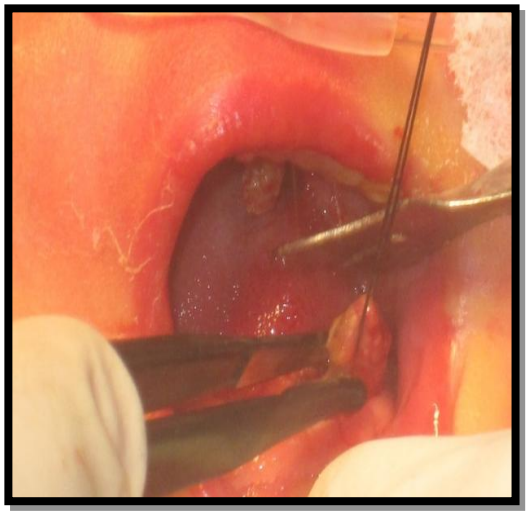

Figure 2: Excision of the bands and removal of each band

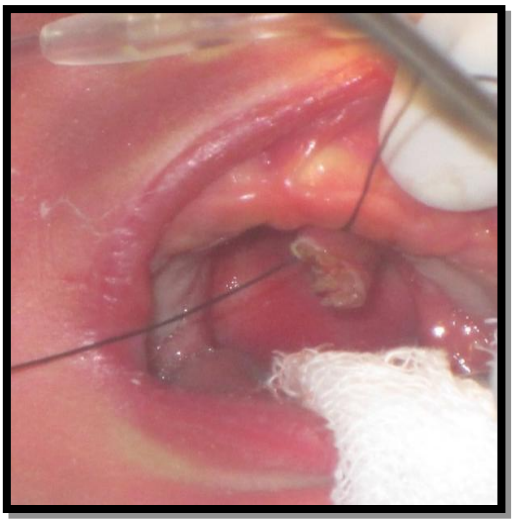

Figure 3:Excision of the band from upper arch and removal

III. Discussion

In the case presented here, there was coexistence of intraoral synechiae and cleft palate. Intraoral synechia accompanied by cleft palate was first described by Fhurmann et al3 in 1972 as cleft palate lateral synechia syndrome. It composed of mucosal banding from the floor of the mouth to margins of the cleft and micrognathia.[14]There is great variability in the expression of the banding, which ranges from a thin friable membrane to thick mucosal bands.Furthermore, intraoral synechia may be a component of many syndromes, such as van der Woude, popliteal pterygium, and Fryns syndromes. Alveolar synechiae in conjunction with a cleft palate is known as lateral alveolar synechiae syndrome.[2]Cleft palate lateral synechia syndrome (CPLS) was first described by Fuhrmann and colleagues (1972) in a case series of 5 members of a family who presented with cordlike adhesions running from the free borders of the palate to the lateral parts of the tongue and floor of the mouth (Preus et al., 1974; Haramis and Apesos, 1995).[1]

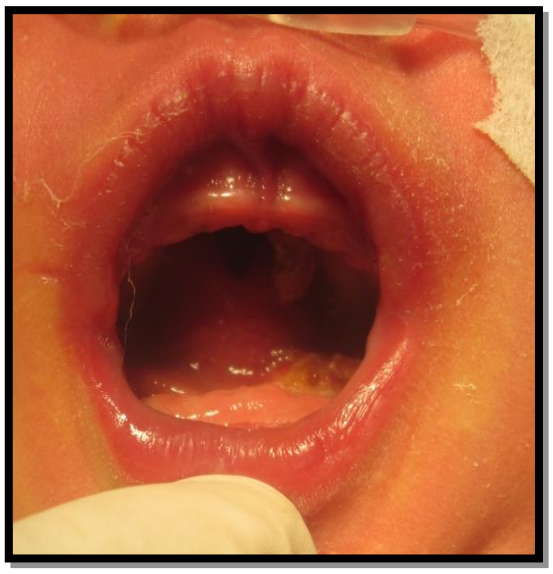

Figure4: Postoperative photograph

Demarque- Van Der Woude syndrome(DVWS) appears to be a less severe form of popliteal pterygium syndrome (PPS) based on etiological and genetic similarities. Abnormalities in the interferon regulatory factor 6 have been implicated in the DVWS- PPS disease spectrum. Demarque- Van Der Woude syndrome is

associated with lip pitting, cleft lip and/or palate, dental defects, limb abnormalities, cardiovascular defects and Hirschsprung disease[7]. PPS is a rare condition with an autosomal dominant mode of inheritance and is associated with popliteal webbing, syndactyly and nail defects. A variable involvement of oral structures may include features similar to Demarque- Van Der Woude syndrome.[16]

Orofacial digital syndrome is a genetic condition involving abnormalities of the face, oral cavity and digits of both upper and lower limbs. Oral features of this condition include hyperplastic frenums, pseudo- clefting of the upper lip, tongue abnormalities, cleft palate and high arched palate. Hard tissue features include abnormalities of the anterior teeth ranging from hypoplasia to supernumerary or missing teeth.[5-8]

Fryns syndrome was first described in 1979 as a "variable multiple congenital anomaly syndrome," and constituted of major features involving a coarse face with microformed eyes with clouded corneas, soft palate clefting, lung and diaphragm abnormalities. Deformities of the distal limbs were also noted. This condition has been identified to have an autosomal recessive inheritance pattern and is significantly lethal in the neonatal period of development.[9]

A number of theories regarding the pathogenesis of oral synechia have been mentioned in various literaturs., 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. [15]

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 brachial arches as another plausible etiological factor.[3] 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[10]. 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.It has been suggested that the presence of oral banding and cleft palate be regarded as a sequence. This was based on the premise that the pathological membrane prevented anterior and forward movement of the tongue, which in- turn prevented midline fusion of the palatal shelves and resulted in the formation of a cleft palate deformity.[11]

IV. 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. Not only treatment in early stages can be easier, but also it can help the growth and nutrition of the child; because by passing time, TMJ ankylosis happens in these people due to the immobility and lack of function which leads to difficulties in surgery.

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