CASE REPORT

Congenital alveolar synechiae with cleft palate

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Abstract

Congential maxillomandibular fusion or jaw adhesions is a rare entity. It is often associated with other congenital anomalies like cleft palate and is characterized by restricted mouth opening. This unusual presentation of congenital alveolar synechiae is highlighted in a 15 days old baby boy. Under inhalational anaesthesia his fibro-cartilagenous adhesions were excised and endotracheal tube was then placed. Good recovery without any residual scars or adhesions and a 25 mm mouth opening was achieved.

Keywords: Cleft palate, Alveolar synechiae, congenital fusim.

Introduction

Congenital adhesions of the jaws is rare. Less than 40 cases of alveolar synechiae have been reported in literature.[1,2] Although it may present in isolation with cleft palate, alveolar synechiae are usually associated with Van der Woude syndrome, oromandibular limb hypogenesis syndrome,[3] microglossia, micrognathia and popliteal pterygium syndrome.[4,5] The exact incidence of this condition is still unknown but the incidence of infantile congenital anomalies is approx. 2.5%.[6] Popliteal pterygium syndrome have associated alveolar synechiae in 33-43% cases.[5]

The fusion may be unilateral or bilateral. Restricted mouth opening causes difficulty in feeding, swallowing, and respiration thus affecting the growth of the infant. In long-standing cases, temporomandibular joint (TMJ) ankylosis often occurs because of immobility and lack of function, necessitating more complicated surgical treatment. The sooner the treatment is rendered the lesser the possibility of mandibular growth disturbance and facial deformities.

In this case report a child presented with alveolar band syndrome. It is unique in a way that there was no syndromic association, bands were fibrous and located anteriorly as opposed to common posteriorly placed bands.

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Case Report

A 15 days old baby boy suffering from inability to open the mouth since birth was brought to plastic surgery clinic for evaluation in July 2010. He had been fed on nasogastric feeding tube since birth. On examination it was evident that upper and lower alveolar ridges were fused bilaterally by the fibrous bands located at the molar areas restricting mouth opening with 7mm open bite (Figure-1). Radiologically there was no evidence of bony fusion. Laboratory blood exam values were within normal limits.

No significant information was obtained from the family's medical history. The mother was primigravida with no history of illness, trauma or drug use. The baby was delivered preterm by normal delivery.

Treatment

Following inhalational anaesthesia, xylocaine with epinephrine solution (1:150,000) was infiltrated for vasoconstriction. The fibro-cartilagenous adhesions of both sides were divided using surgical knife blade No. 15. Endotracheal tube was then placed and oral cavity was thoroughly examined. The infant had a high arched palate with cleft of the soft palate. No complications were

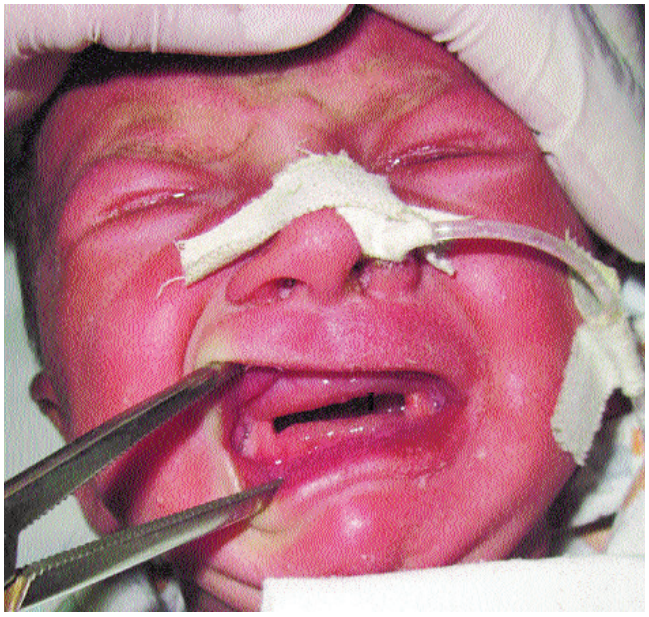

Figure-1: Bilateral alveolar bands with open bite.

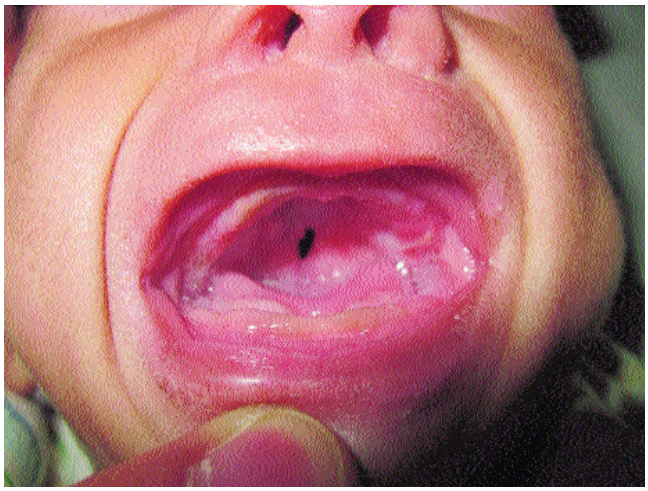

Figure-2: Three months follow up with no residual scars and adequate mouth opening.

encountered in the postoperative period. Patient had a good recovery without residual scars or adhesions and achieved 25mm mouth opening (Figure-2). Subsequently palate repair was also done with uneventful recovery at 9 month of age.

Discussion

Congenital alveolar synechiae is an uncommon condition. It can occur in any part of oral cavity and usually contain membranes or bands of epithelium, fibrous tissue and supportive tissue like muscle or bone. It can be associated with cleft palate or syndromes like Vander de woude, Pierre robin sequence[1] and popliteal pterygium syndrome.[6] In this case there was no syndromic association.

Etiology of Alveolar synechiae is still unknown although several hypotheses have been proposed. Postulated factors include disturbance of the interactions of growth factors,[2] teratogenic drugs like meclizine,[7] trauma late in pregnancy and local ischaemia and amniotic bands causing pressure on the first branchial arch.[8] The maternal history did not reveal any such cause in this case.

Management of alveolar synechiae starts with assessment of general condition of patient, presence of other anomalies and syndromic associations, need for airway and feeding tubes if mouth opening is restricted. Radiographs should be obtained to rule out TMJ ankylosis and bony union. The main goal of surgery is early division of the adhesions to release trismus, achieve normal feeding, avoid upper airway obstruction and to allow for normal mandibular growth and function. In this case there were fibrous bands with no bony union at maloar regions of upper and lower alveolar ridges.

It must be remembered that although surgery is simple anaesthesia is sometimes difficult. Fiber optic bronchoscope, nasal endotracheal intubation and even tracheostomy is required.[8] Another important fact to be occasionally mentioned is that normal-range mouth opening often cannot be obtained in the early postoperative period but it usually regains within 1-2 weeks postoperatively so forceful opening of mouth should not be attempted.[9] Use of silastic bite blocks, sheets, rubber and gauze can be used as a splint for mouth opening and prevent secondary adhesions of raw areas.[9,10] In this case transoral approach were used and after division of bands endotracheal tube was inserted. Good mouth opening was achieved 10 days after surgery without manual manipulation.

Conclusion

Congenital alveolar synechiae frequently have syndromic associations although few cases occur in isolation. Fibrous alveolar synechiae once excised properly have excellent outcomes because there is no associated bony or muscular abnormality. Bony fusion must be ruled out radiologically in every case. To treat the patient effectively, it is vital to secure the airways, ensure adequate nutrition and achieve enough mouth opening without any complication or recurrence.

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