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Case Report

Oral synechiae with cleft palate- A case report

Kirti Nagpal¹, Shweta Gogia^{1*}, Alok Agarwal¹

¹Dept of ENT, Ganga Ram Institute of Postgraduate Medical Education and Research, New Delhi, India.

Abstract

Oral synechiae is a rare clinical entity which may be seen in association with other congenital defects or part of a syndrome. The condition needs early recognition and addressal for the management of the respiratory and swallowing concerns that are inherent to the condition. This condition has been associated with persistent buccopharyngeal membrane. A 11 day old male child was referred with the complaints of inability to feed and respiratory distress. On examination, the infant was found to have bands connecting the hard palate with the floor of the mouth. The child was able to maintain the oxygenation through spontaneous respiration but was being fed through a nasogastric tube. It is prime that such congenital conditions are identified early and addressed to prevent subsequent sequela.

Keywords: Oral synechiae, Buccopharyngeal membrane, Fibrous band, Cleft palate

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1. Introduction

The incidence of congenital anomalies in neonates is typically reported approximately 2.5% globally.¹ Congenital oral synechiae is an uncommon clinical condition with very few documented cases in the literature. There is a strong association of this condition with cleft palate and it may be a part of a syndrome such as Vander Woude Syndrome, Oromandibular Limb Hypogenesis syndrome, Popliteal Pterygium Syndrome. The condition is usually sporadic in occurrence but familial cases have also been reported in literature. Lateral adhesions between the maxillary and mandibular regions are more commonly seen as compared to midline synechiae. The presentation is usually one of feeding issues but respiratory distress may also be seen in conjunction. Early recognition and treatment is prudent in these cases to prevent sequelae. The objective of treatment is thus to achieve an adequate oral cavity, thereby to enhance the patency of airway and also the feeding ability. This, in turn, facilitates normal oral and facial growth and enables the timely surgical repair of the cleft. In this context, the present study documents a rare case of intraoral synechiae with cleft palate.

*Corresponding author: Shweta Gogia
Email: Docnagpal28@gmail.com

The aim of this study is to facilitate the early identification of these congenital anomalies, ensuring timely intervention to prevent potential complications and longterm sequelae.

2. Case Report

An eleven day old male child was referred from a peripheral health centre to the neonatal care unit of our hospital with an unknown defect in the oral cavity due to which the child was not able to feed. The child was born at 37 weeks of gestation by normal vaginal delivery with no significant pre- natal history. The birth weight was 2200 gms, birth length was 46.5 gms, and head circumference was 32.5cms. The Apgar score was not available. The examination of the child revealed a diffuse mucosal band extending from the floor of the mouth to the palate with a small opening in the midline through which the oropharynx could be seen. The airway was assessed with flexible scopy to rule out other airway abnormalities which showed a cleft palate involving hard and soft palate. The nasopharynx with rest of the airway was normal with normal mobility of the vocal cords. There was retrognathia and the bands limited the mouth opening.

Bilateral fifth toe was hypoplastic with absence of nail on both toes. 2D echocardiogram was done which showed a large ventricular and atrial septal defect with bidirectional shunt with severe pulmonary artery hypertension with dilated right atrium and ventricle.

The patient was planned for the release of the bands under general anaesthesia. After pre-oxygenation, the bands and the oral airway was assessed and intubation was not attempted as the opening did not provide enough space for the same, so the bands were divided using radiofrequency from the floor of the mouth. Following this the child was intubated using C-Mac laryngoscope to facilitate the visualisation of the larynx which was difficult due to the tongue prolapse into the oropharynx. Following intubation, glossopey was performed to prevent the fall of the tongue. The child was kept on high flow oxygen following the procedure for three days and was then weaned off.



Figure 1: Congenital diffuse oral mucosal band (synechiae) with cleft palate. Tongue is behind the fibrous band.

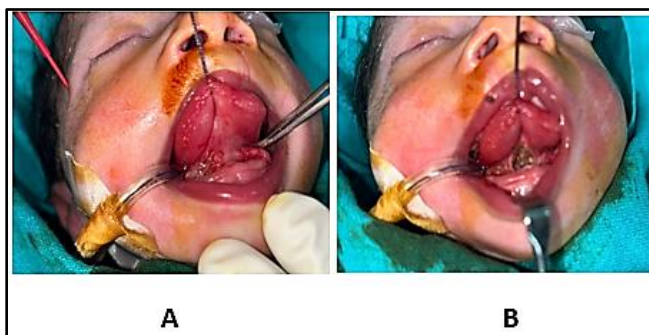


Figure 2: a; Release of fibrous band (synechiae) by radiofrequency. b; Tongue was pushed forward and Glossopey was performed.

3. Discussion

Synechiae is defined as fibrous bands which are composed of only soft tissue, and if there is bony involvement with soft tissue, it is termed as Synostosis. Syngnathia is defined as bands involving everything such as soft tissue, bone, muscle, and connective tissue. Oral synechiae are quite rare, only 60 cases have been identified worldwide.² Congenital fibrous

bands may occur in any part of the oral cavity. It may either be due to congenital fusion of mandible or maxilla, or may involve any part of the oral cavity such as between the palate and tongue. This may be seen isolated or may coexist with cleft palate. These fibrous bands may be associated with various syndromes such as Vander Woude or Popliteal pterygium syndrome, Pierre Robin, or may be non syndromic. The non syndromic with cleft palate is even rarer, with only 5 cases reported in the literature.³

The etiology behind the formation of fibrous bands along with cleft palate remains elusive. Various theories have been proposed for this. One of the widely accepted theories proposes that the buccopharyngeal membrane fails to regress during development, obstructing the tongue's forward and downward movement. This mechanical restriction subsequently hinders the palatal shelves from merging properly, leading to the formation of a cleft palate.⁴ The other theories include the presence of amniotic constriction bands affecting the developing branchial arches, exposure to environmental teratogens, and maternal intake of certain drugs, such as high-dose vitamin A during pregnancy.⁵⁻⁶

Ogino et al described synechiae into five different types.⁷

1. Adhesion of the alveolar mucosa to one or both sides of the upper and lower jaw (alveolar synechiae).
2. Membranous adhesion on the hard palate and floor of the mouth, excluding the rear of the tongue (lateral synechiae).
3. Bands partially involving the hard palate and tongue.
4. Bands involving the soft palate and tongue.
5. Bands involving a membranous adhesion between the hard palate and lower lip.

There is no classification which classifies synechiae with cleft palate into any type. Therefore there is need to devise a new classification which classifies the synechiae with other congenital defects.

Congenital fibrous bands can significantly compromise vital functions, most notably feeding and respiration. These anomalies may adversely affect the overall health, growth, and development of the child. They can also complicate procedures such as endotracheal intubation during anesthesia and increase the risk of life-threatening complications, including airway obstruction and aspiration pneumonitis.

Prompt intervention is therefore essential, with initial priority given to securing a patent airway. Subsequent management should focus on addressing feeding difficulties, since delay in treatment can predispose the neonate to asphyxia, recurrent aspiration, malnutrition, growth retardation, aberrant craniofacial development, and misalignment of erupting teeth. The choice of therapeutic approach, however, must be tailored to the severity, extent, and specific nature of the anomaly.

4. Conclusion

The occurrence of fibrous bands within the oral cavity in association with cleft palate is extremely uncommon. This case underscores the critical importance of early recognition, as such anomalies may significantly compromise feeding and respiration, increase the risk of aspiration, and complicate airway management, particularly in the event of acute deterioration. Prompt identification not only facilitates timely intervention but also emphasises the need for a multidisciplinary approach.

5. Source of Funding

None.

6. Conflict of Interest

None.

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