Case Report

Cleft palate lateral synechia syndrome

Deborah Sybil, Alok Sagtani

ABSTRACT

Cleft lip and palate are the most common congenital craniofacial anomaly in humans. The presence of oral synechia along with cleft palate is a rare syndrome. We encountered one case that had a cleft palate accompanied by congenital oral synechia due to a membranous adhesion between the floor of the mouth and the free margin of the cleft palate.

Key words: Cleft palate, synechia, syndrome

INTRODUCTION

Cleft lip and palate are the most common congenital craniofacial anomalies in humans. The presence of oral synechia along with cleft palate is a rare syndrome. We encountered one case of cleft palate accompanied by congenital lateral oral synechia.

CASE REPORT

The case involved a male infant 1 day old at the time of presentation. He was the first child of healthy parents. The infant was delivered normally at full term with a low birth weight of 1,750 g. There were no particularly notable points in the family history, medical history, or reproductive history.

On examination, the patient had a restricted mouth opening resulting from congenital oral synechia due to membranous adhesion between the free margin of the cleft palate and the floor of the mouth, lateral to the tongue on the left side. The cleft was of secondary palate with intact premaxilla. There were no other abnormalities found in the patient. No obvious auricular or nasal deformities were appreciated. Digits and extremities appeared normal, as did genitalia. Examination of the heart revealed no abnormalities.

The congenital oral synechia appeared as a thin membrane with a broad attachment at the floor of the mouth measuring approximately 2 cm in anteroposterior width. At the palatal margin the attachment narrowed to about 0.5 cm [Figure 1]. The membranous adhesion was pale pink in color and appeared to be avascular.

The infant faced feeding problems due to the restricted mouth opening, and therefore an immediate surgical excision of the synechia was decided on. Feeding was done using an infant feeding tube till anesthetic clearance was obtained for the procedure. At 1 week after birth, the congenital synechia was excised under sedation uneventfully restoring adequate mouth opening and allowing normal feeding [Figure 2]. A surgical closure of the cleft palate is planned for a later date.

DISCUSSION

The first report of oral synechia was by Illera in 1875,[1] but the first documented case of a lateral synechia between the floor of the mouth and free margin of cleft palate was by Hayward and Every in 1957.[2] Over the years, almost 60 cases of oral synechia have been reported out of which 52 are lateral synechia and 8 were of the median variety.[3]

Oral synchia can be of various degrees adhering at various locations. These can be classified into five types[3]: Synchia by cord-like adhesion of the alveolar mucosa on one or both sides of the upper and lower
jaw (alveolar synechia); synechia by a membranous adhesion on the hard palate and floor of the mouth, excluding the rear of the tongue (lateral synechia); synechia in which the hard palate and tongue are partially involved; synechia in which the soft palate and tongue are widely involved, such that continuity is interrupted between the oral cavity and the pharynx; and synechia by a membranous adhesion between the hard palate and lower lip.

Fuhrmann, et al.,[4] reported that five family members had cleft palates and synechia, one having a cleft palate without synechia, and one transmitted the gene but did not express it.

The etiology of intra‑oral bands or synechia of epithelial tissue has been debated, but many theories have been proposed. During the 7th to 8th week of embryological development, the alveolar ridges, tongue, and palatal shelves are in contact with each other. The ensuing palatal closure depends on downward contraction of the tongue. When the tongue protrudes from the mouth as a result of medial movements of the oral cavity walls, it prevents the alveolar ridges from fusing. Genetic, teratogenic, or mechanical insults during this critical stage may lead to periods of close, quiescent contact between oral structures, and this predisposes to abnormal fusion.[9]

Longacre asserts that oral synechia is due to the persistence of the buccopharyngeal membrane, and is for that reason associated with micrognathia and cleft palate.[6] Kruger speculates that the mechanical effect of the tongue may contribute to cases in which the periphery of the cleft palate adheres to mucous membranes on the floor of the mouth, and that adhesion in cases of cleft palate may occur as a result of obstruction by the tongue.[3] According to Mathis, when adhesion of the palatal shelf occurs during developmental stages, adhering epithelial rudiments, for some reason, lead to synechia.[8]

The general consensus on the treatment of cleft palate lateral synechia syndrome is excision of the synechia and palatal closure. Dalal et al.,[9] have reported a case of intra alveolar synechia in two siblings, one of whom had a spontaneous resolution of the adhesion. Donepudi, et al.,[10] have documented that the synechia provided additional tissue for surgical closure with less tension on the palatal flaps. We believe that such a use of the synechia has been possible, because the membranous adhesion in this case had fibromuscular bands in it unlike our situation where the synechia consisted of thin membranous tissue of unequal width.

Obtaining a safe airway for the release of the bands may be difficult. The conventional oro‑tracheal intubation or the use of a laryngeal mask may not be possible due to the presence of the synechia. Fiberoptic nasotracheal intubation may not be feasible due to technical difficulties of finding a bronchoscope small enough for a week old infant. Performing a procedure on the highly vascular floor of the mouth under local anesthesia in an infant would involve the risk of bleeding complications and aspiration. In cases where immediate excision of the synechia is deemed necessary either due to breathing or feeding problems, sedation can be used under constant anesthetic monitoring for the surgical procedure as was done in our case.

**References**

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