CASE REPORT

Coexisting Congenital Subglosso-palatal Membrane and Tongue Dermoid in a Neonate

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Background: Neonatal respiratory distress due to coexisting subglosso-palatal membrane and tongue dermoid has not been reported yet. **Case characteristics:** A newborn with respiratory distress having a membrane in the oral cavity. Excision of membrane revealed a tongue mass with cleft palate, obstructing the nasopharynx completely. Elective ventilation was followed by excision of mass. **Outcome:** The child was cured with uneventful course at follow-up of six months. **Message:** Co-existing congenital anomalies causing airway obstruction may be missed in presence of subglosso-palatal membrane.

Keywords: *Infant, Respiratory distress, Stridor*.

Congenital anomalies affecting the oral cavity are rare. Often these can cause respiratory distress, aspiration, and bleeding [1]. It is unusual to have various congenital pathologies of the oral cavity in same patient, resulting in complexity in presentation and management. A congenital mass in the oral cavity can be a cause of respiratory distress [1]. Similarly, the subglosso-palatal membrane which is thought to be a remnant of the buccopharyngeal membrane is also known to be associated with respiratory distress [2]. We share our experience and challenges faced during management of a neonate with severe respiratory distress, who had a co-existence of these two birth defects.

CASE REPORT

A full-term male baby weighing 2.8 kg was delivered by spontaneous vaginal delivery to an unbooked primigravida mother at a peripheral center. The child presented to us with severe respiratory distress with a heart rate of 180/min, a respiratory rate of 60/min along with inspiratory stridor and subcostal retraction. The air entry was diminished but equal on both the sides. The oxygen saturation on high flow oxygen, *via* high flow nasal cannula, at the rate of 10L/min, was 90-95%. On oral examination, there was a membrane extending from the floor of the mouth to the junction of the soft and hard palates with its lateral extension up to the molar trigone bilaterally. Nasal suction was performed to clear the secretions and to rule out associated choanal atresia.

The child deteriorated rapidly and progressed to respiratory failure. The respiratory rate fell to 20-30/min and saturation dropped to 75-80% on high flow oxygen. A diagnosis of the sub-glossopalatal membrane with severe respiratory distress and respiratory failure due to upper airway obstruction was made. The bag and mask ventilation was started. The distended stomach was decompressed with a infant feeding tube, though there was difficulty in negotiating it through the nasopharynx.

As the bag and mask ventilation was ineffective, an emergency excision of the membrane was performed. On excision of the membrane, a mass was seen to be arising from the dorsum of the tongue. The lesion was nearly filling the oropharynx and the nasopharynx (***Fig.* 1**). The tongue was gently pulled out of the mouth, and the child was intubated to secure the airway. The child was shifted on the ventilator. After 24 hours, the baby improved and was extubated after which he was nursed in prone position.

A computed tomography scan showed a cystic mass arising from the dorsum of the tongue extending into oropharynx and nasopharynx, and associated with cleft palate. Echocardiography and abdominal ultrasound were normal. An elective excision of the mass was planned. Per-operative, the mass was tense, cystic and lobulated with extension into the substance of tongue. The upper portion of the mass was filling the space of palatal cleft while the lower and posterior portions were occupying the nasopharynx and oropharynx. Primary closure of dorsum of the tongue was achieved after the complete excision of the mass. The postoperative course was uneventful. The child is awaiting cleft palate repair. Histopathology of the mass showed features suggestive of a dermoid cyst.

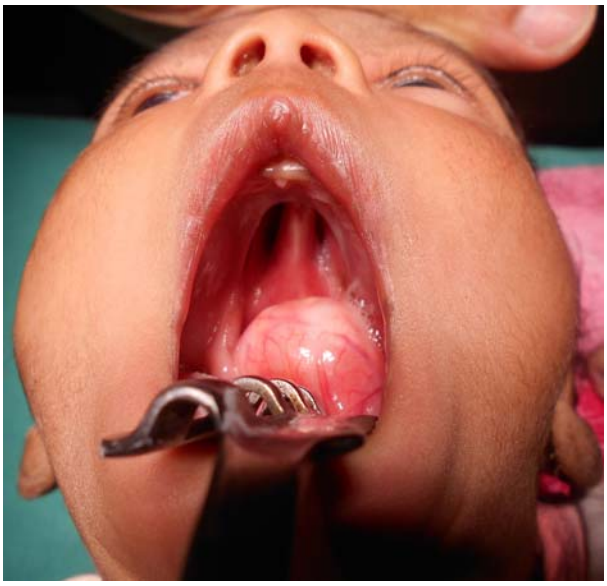

FIG. 1 *The mass arising from the dorsum of tongue with palatal cleft.*

DISCUSSION

Congenital anomalies of the oral cavity are rare. As these result from abnormal development at a very early stage, they can be associated with multiple complications. Further, it is rare to find multiple congenital lesions affecting the oral cavity in one patient. A congenital lesion in the oral cavity may cause upper airway obstruction and lead to respiratory distress at the time of birth [2]. Similarly, the sub-glossopalatal membrane can compromise the upper airway patency and present with respiratory distress [3]. Both lesions presenting simultaneously in one patient is extremely unusual.

In this case, the child had respiratory distress due to upper airway obstruction caused by congenital anomalies. Upper airway in neonates may be blocked due to anomalies like choanal atresia, tumors such as glioma, encephalocele, teratoma or dermoid, vocal cord paralysis, and subglottic stenosis [1,5]. Many of these cases may require emergency surgical intervention for restoration of a secure airway [1]. Subglossopalatal membrane is a remnant of the buccopharyngeal membrane [3]. As neonates are obligate nasal breathers, the subglosso-palatal membrane may not cause respiratory distress in all cases [5]. This created a diagnostic dilemma in our case, and we looked for associated choanal atresia, as this association has previously been reported [6].

Congenital tongue mass can cause upper airway obstruction and child may have respiratory distress at birth [2]. The development of tongue is one of the earliest events in fetal life (4th–5th week) and can affect the development of palate and other maxillofacial structures [7]. Thus, a mass on the dorsum of the tongue can protrude into the defect between the palatal shelves (palate develops between 6th and 7th week) and prevent their fusion, and therefore, can cause palatal cleft [8]. Further, as the musculature of the tongue is formed there can be fusion, entrapment, and proliferation of epithelial debris, which can lead to the development of lingual dermoid cyst [9]. This can explain the infiltration of dermoid into the substance of tongue in our case [10]. The development of tongue and glossopharyngeal membrane and other maxillofacial structure overlap closely with each other. Thus, one anomaly may lead to the development of others.

Through this case, we documenting rare co-existence of subglosso-palatal membrane, cleft palate and dorsal tongue dermoid. Subglosso-palatal membrane excision should be performed in an emergency if the child is not improving on high flow oxygen because some other associated anomaly can be missed due to blocked view.

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