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A Rare Case of Prenatal Diagnosis and Management of Fetal Oral Cystic Mass

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Abstract

Congenital fetal oral mass is a very rare entity. A correct ultrasound-based approach on the anatomic location, consistency, and vascularity will help us to narrow down the differentials antenatally but a confirmed diagnosis can be made by histopathological examination postnatally. We describe a case of a fetal oral cyst diagnosed antenatally at 20 weeks of gestation. On follow-up, fetal growth and amniotic fluid volume were normal with no changes in the size or the position of the cyst. The cyst was excised on the first neonatal day. Histopathology was consistent with a mucocele.

Keywords

- fetal oral mass mucocele
- ranula
- oral mucocele
- thyroglossal duct cyst
- foregut duplication cyst

Introduction

Congenital fetal oral masses are very rare and the exact epidemiology and natural history of these remain largely unknown.[1] These may be detected in second- or third-trimester scans.[2] Although accurate antenatal diagnosis and distinguishing the different oral masses are challenge, they are important for appropriate antenatal counseling, prognostication, deciding the mode of delivery, and for postnatal management.[3] The size and exact location of the oral mass are important prognostic factors because larger masses and those involving the nasopharynx can cause feeding difficulties and mechanical airway obstruction. Such potential impairments may necessitate a rapid intervention to secure the airway of the neonate after delivery, with severe cases requiring an EXIT procedure.[2] We present a case in which a detailed sonographic evaluation guided antenatal counseling as well as prognostication about the likelihood of neonatal breathing impairment.

Case Report

A 21 year old nonconsanguineous pregnant patient was booked at Fernandez Hospitals, Hyderabad, India, at 20 weeks of gestation for an anomaly scan. An $18 \times 13 \times 12$ mm avascular mass was noted in the fetal oral cavity. The mass was anechoic, homogeneous, localized, partly protruding outside the oral cavity with distinct margins (►**Fig. 1A, B**). The mass was arising from the ventral surface of the tongue and was moving along with tongue movements. Serial scans were performed every 4 weeks. Fetal swallowing movements, movement of mass along with the tongue (►**Video 1**), nasal breathing movements (using color Doppler) (►**Fig. 2**, ►**Video 2**), and jaw movements were assessed on real-time scan at each visit. Follow-up scans showed appropriate fetal growth and normal amniotic fluid index with no further change in the size or the position of the cyst. The mouth of the fetus was persistently open during all examinations because of the oral mass protruding partly outside the oral cavity. There was no polyhydramnios. The fetus had good

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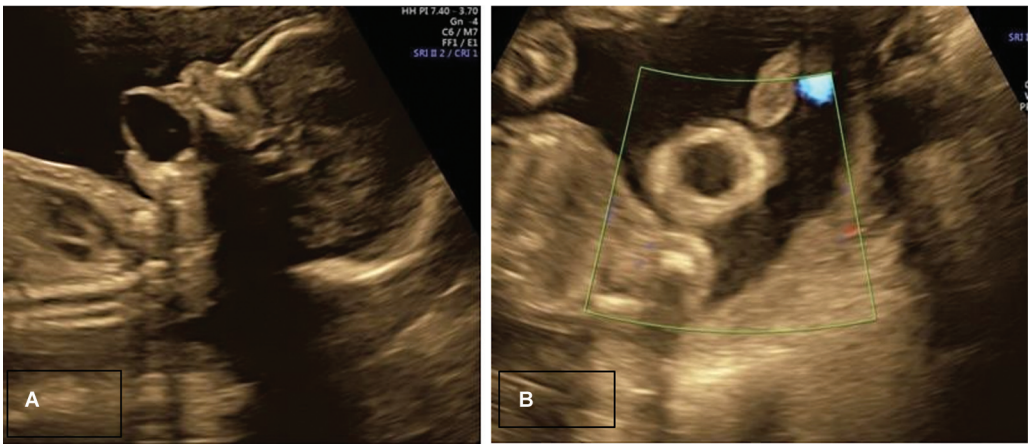

Fig. 1 (**A**) Fetal profile: Prenatal ultrasound examination demonstrates the presence of an echoic mass in the fetal mouth with no vascularity at 20 weeks. (**B**) Coronal image of the fetal face with persistently open fetal mouth.

sucking movements, which suggested intact swallowing and breathing movements. Three-dimensional (3D) rendering of the cyst was done to assess the anatomy of the mass (►**Fig. 3**). Magnetic resonance imaging (MRI) was planned at 24 weeks of gestation, and it confirmed our scan findings (►**Fig. 4A, B**). As there were no features of tracheoesophageal compression in the antenatal scan and MRI, the option of vaginal delivery was discussed with the couple.

Video 1 Anatomic location of the Oral mass

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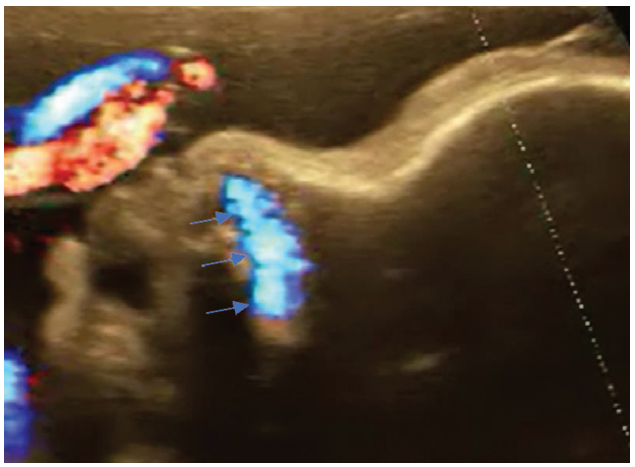

Fig. 2 Color Doppler assessment of the nasal passage (arrows) showing free flow of amniotic fluid through the nasal passages, confirming uninterrupted fetal nasal breathing.

Video 2 Demonstrating Color Doppler assessment of the nasal passage showing free flow of amniotic fluid through the nasal passages, confirming uninterrupted fetal nasal breathing

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A male baby weighing 3 kg was delivered at 38 weeks by vaginal delivery with normal Apgar scores. Pediatric and ENT surgeons were on standby for emergency airway management, but this was not required. Postnatal findings were consistent with antenatal scan findings (►**Fig. 5**). After birth, the baby was kept on nasal oxygen support and at 24 hours after birth, the cyst was excised (►**Fig. 6**) and sent for histopathologic evaluation (HPE). On postoperative day 2, the baby tolerated orogastric tube feeding and was gradually shifted to breast-feeding. Histopathology of the mass revealed a cyst wall lined by low columnar to mucinous columnar epithelial cells with subepithelial fibrocollagenous tissue suggestive of a mucocele. Currently, the infant is 6 months old and is doing well with no complications and no recurrence.

Discussion

Congenital fetal oral mass is a very rare entity.[1] A correct ultrasound (US) approach based on the anatomic location (►**Fig. 7**), size, extension, consistency (solid or cystic, presence or absence of calcification), echo texture (homogenous or heterogenous, unilocular or multilocular), relationship with surrounding structures, mobility, and vascularity will help us to narrow down the differentials antenatally, but the confirmed diagnosis is usually made by HPE postnatally. The differential diagnosis of antenatal oral mass includes mucoceles, ranula,

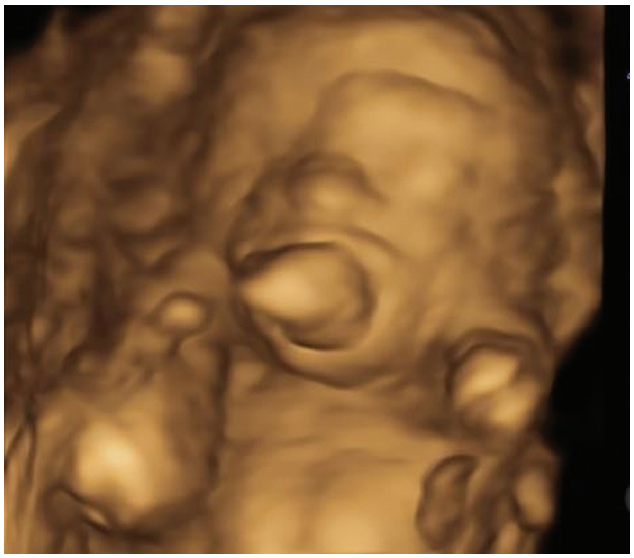

Fig. 3 Three-dimensional surface rendering showing the fetal face and protruding oral mass at 20 weeks.

epulis, epignathus, hamartoma, dermoid cyst, epidermoid cyst, foregut duplication cyst (FDC), and thyroglossal duct cyst (TDC). Our case was a cystic avascular mass arising from the anterior part of the oral cavity so that we could rule out all solid and vascular oral masses like epulis, epignathus, hamartoma, hemangioma, and arteriovenous malformations. The mass was seen arising from the ventral surface of the tongue and was moving with the tongue, which further narrowed down our differentials to a FDC,[4] mucocele,[5] or a rare anterior presentation of TDC.[6] This approach-based examination helped us to rule out ranula, a common cause of fetal oral mass that arises from the floor of the mouth and does not move along with the tongue movements.[4,7]

Serial antenatal follow-up scans were done using 2D, 3D, real-time US, and color Doppler to look for change in size of the mass, check for secondary findings that indicate an obstruction to the airways or gastrointestinal tract (presence or absence of polyhydramnios, visualization of stomach bubble, fetal swallowing, breathing movement), and to monitor the fetal growth. On follow-up, the fetal growth and amniotic fluid volume were normal with no changes in the

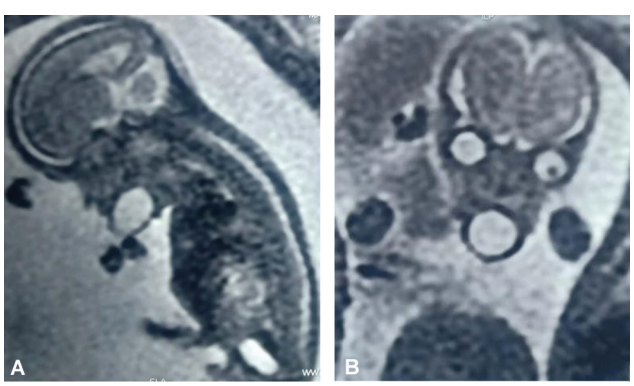

Fig. 4 Magnetic resonance imaging sagittal (**A**) and coronal image (**B**) revealing homogenous signal pattern (T2-hyperintense lesion) confined to the sublingual region with no extension.

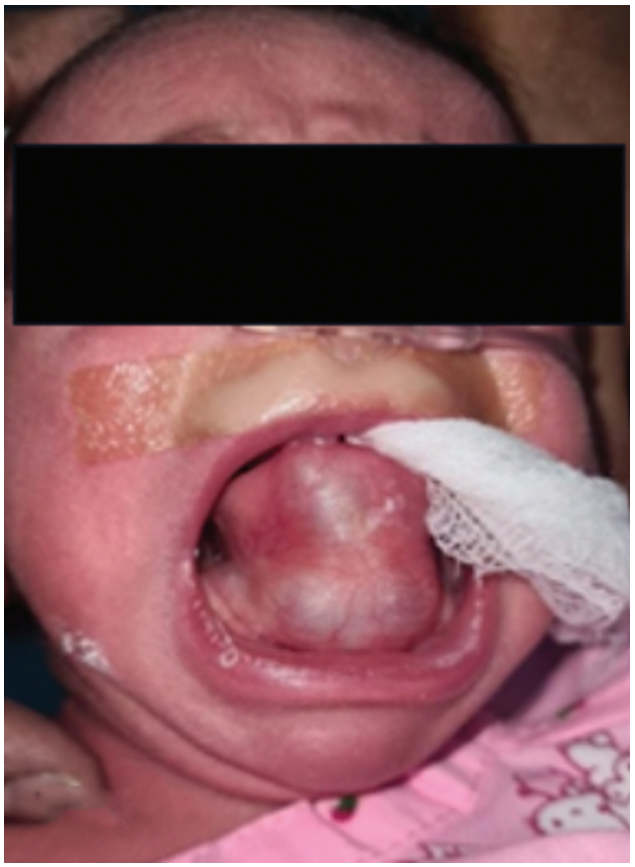

Fig. 5 Postnatal findings consistent with the prenatal diagnosis. A cystic mass is seen arising from the undersurface of the tongue with persistently elevated tongue.

size or the position of the cyst. Considering the anterior presentation of TDC being very rare, we were antenatally left with two differentials: an FDC and a mucocele. Antenatally it was very difficult and challenging to further differentiate

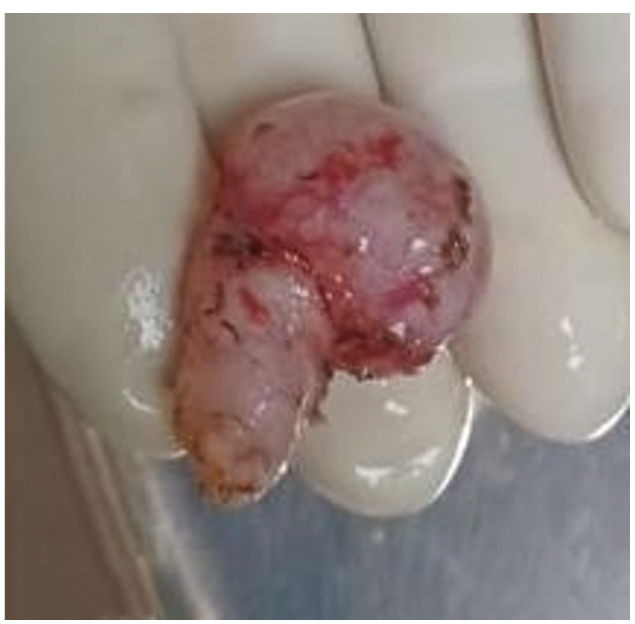

Fig. 6 Macroscopic appearance of the excised cyst.

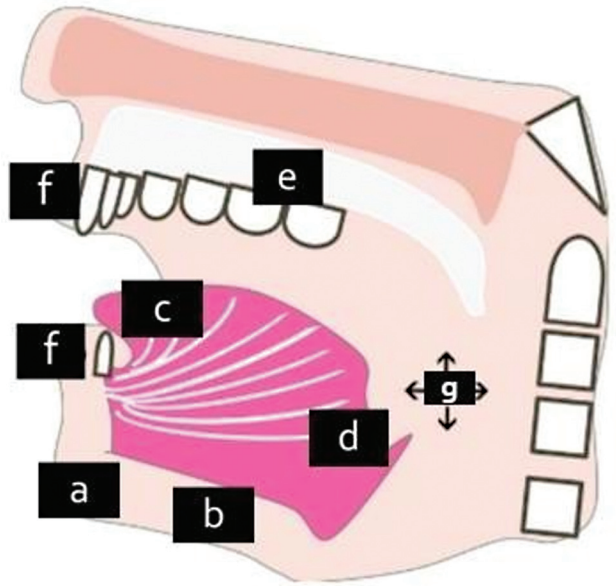

Fig. 7 Differential diagnosis for a congenital oral mass according to anatomic location in the oral cavity. A mass arising from the floor of the mouth in midline (**a**) raises suspicion for a dermoid cyst and off midline (**b**) points to a ranula/epidermoid cyst. Lesion in the anterior two-third of the tongue (**c**) can be an oral foregut duplication cyst, mucocele/ anterior presentation of thyroglossal duct cyst and that for a posterior one-third of the tongue (**d**) includes a congenital thyroid anomaly or lingual hamartoma. A mass arising from the midline palate (**e**) can be an epignathus and from the soft tissues of the maxilla or mandible (**f**) are suggestive of epulis. Lesions that span multiple neck spaces (**g**) are consistent with vascular anomalies.

between FDC and a mucocele. The final diagnosis could only be reached after delivery of the baby with histopathology consistent with a mucocele.

Ranula and mucoceles are the most common disorders of the salivary gland presenting as an oral cyst. A ranula is a mucocele usually situated at the floor of the mouth, but in our case, the mass was located on the ventral surface of the tongue that is one of the uncommon[8] presentations of a mucocele. In the literature search, it was noted to be one of the rare locations of the mucocele arising from the glands of Blandin and Nuhn.[5] They are submucous glands located within the ventral part of the muscles of the tongue. This rare location is consistent with our case.[9]

Around 70% of mucoceles arise from the minor salivary glands of the lower lip, and only 2.5% of mucoceles arise from the tongue.[10] There are two etiologies suggested for a mucocele. The "extravasation mucocele" is formed by leakage of mucus into connective tissues. The "retention mucocele" is formed by an obstruction of a salivary gland duct with resultant expansion, forming an epithelial lined cystic structure.[11] The treatment of choice is surgical excision of the lesion. Other options like corticosteroid injection and cryotherapy have also been reported. Recurrence of this lesion is uncommon.[12]

Conclusion

Practicing a systematic US approach (2D, 3D, real-time US, color Doppler) based on the anatomic location, size, consistency, echo texture, relationship with surrounding structures, mobility, and vascularity, we can narrow down the diagnosis antenatally that helps in prognosticating, counselling, and planning treatment options. Confirmation of the type of oral mass is usually done by postnatal histopathology.

Conflict of Interests
None declared.

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