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CASE REPORT

Two cases of fetal congenital oral masses

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ABSTRACT

Fetal oral masses rarely occur *in utero*. Their detection is of paramount importance because the mass may close the oral cavity, leading to polyhydramnios and eventually resulting in preterm labor and/or fetal loss due to postpartum asphyxia. We present two cases of fetal oral masses diagnosed with fetal congenital epulis and fetal thyroglossal duct cyst each, in which the masses were excised by *ex utero* intrapartum treatment during cesarean deliveries.

Keywords: Prenatal diagnosis, gingival neoplasms, oral masses.

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INTRODUCTION

Fetal oral masses rarely occur *in utero*. They may be benign or malignant; therefore, their differential diagnosis is of paramount importance. The size of the cyst and the amount of amniotic fluid should be monitored during pregnancy. The increase in the size of the mass may close the oral cavity, leading to polyhydramnios and eventually resulting in preterm labor. If fetal oral masses are unrecognized, sudden fetal losses may occur due to postpartum asphyxia. The differential diagnosis includes epignathus, epulis, thyroglossal duct cyst, and ranula. *Ex utero* intrapartum treatment (EXIT) is required for masses that close the oral cavity and compromise the airway.

We present two cases of fetal oral masses diagnosed with fetal congenital epulis and fetal thyroglossal duct cyst each.

CASE REPORTS

Case 1: Fetal Congenital Epulis

A 28-year-old woman with a singleton pregnancy was referred to the perinatology clinic at 36 weeks of gestation because of a mass in the fetal mouth. The first trimester scan performed at another center was unremarkable. The second trimester was not performed. At the current presentation, ultrasonography (USG) showed a solid mass in the fetal mouth at girl fetus, measuring 30 by 35 mm (Fig. 1). Doppler USG showed blood flow in the mass (Fig. 2). On USG, the fetal growth and the amniotic fluid index were unremarkable, with no evidence for any anomaly. Magnetic resonance imaging revealed a well-circumscribed solid mass anterior to the alveolar process of the maxillary bone, approximately 35 by 30 mm in diameter, extending to the extraoral area. Appearance of the mass was suggestive of a congenital epulis. At 39 weeks of gestation, a scheduled cesarean section delivery was performed, during which the mass was simultaneously removed (Fig. 3). The newborn was not intubated and was discharged on the 3rd day from the neonatal intensive care unit. Histopathological examination confirmed the diagnosis of congenital epulis.

Case 2: Fetal Thyroglossal Duct Cyst

A 20-year-old woman with a singleton pregnancy was referred to the perinatology clinic at 26 weeks of gestation for the evaluation of a mass lesion in the fetal mouth. The first- and second-trimester scans performed at 12 and 20 weeks of gestation, respectively, at another center had been normal. USG showed a cystic mass, 19 by 12 mm in size, arising from the tongue, with no solid component compromising the oral cavity (Fig. 4). Doppler USG showed no blood flow in the cyst. On USG, the fetal growth and the amniotic fluid index were normal, with no evidence for an anomaly. Fetal magnetic resonance imaging showed a cystic mass, 20 by 15 mm in diameter, originating from the tongue, with no solid component (Fig. 5). The localization and appearance of the mass were suggestive of a ranula or a thyroglossal duct cyst. Repeat USG scans obtained 2 weeks apart until delivery showed no change in the cyst size. The patient underwent a scheduled cesarean section delivery at 39 weeks of gestation, and the mass was simultaneously removed by an EXIT treatment by Eskişehir Osmangazi University of pediatric surgery team. Intubation was not required. The baby was discharged on the 3rd day after an uneventful neonatal care. On histopathological examination, a diagnosis of thyroglossal duct cyst was made.

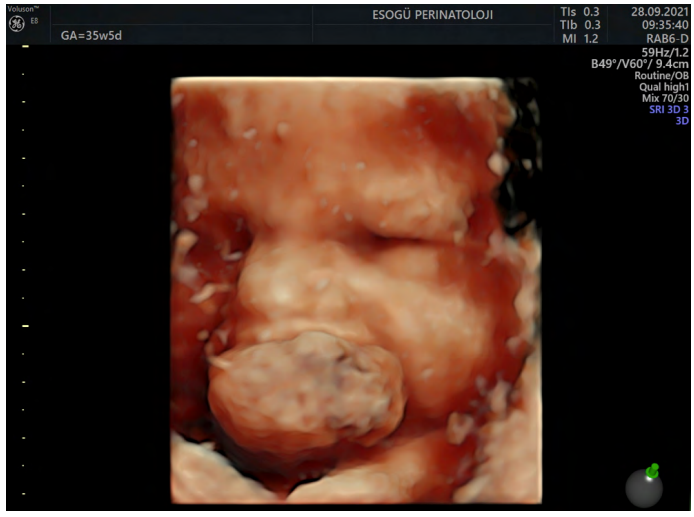

Figure 1: 3D ultrasonographic view of oral mass.

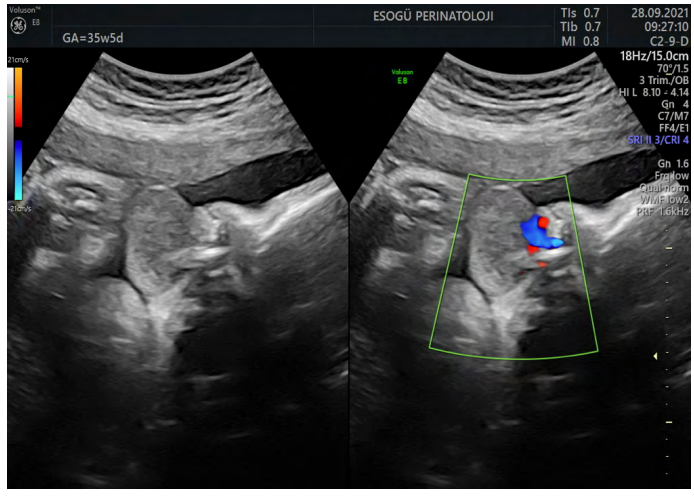

Figure 2: Doppler flow at oral mass.

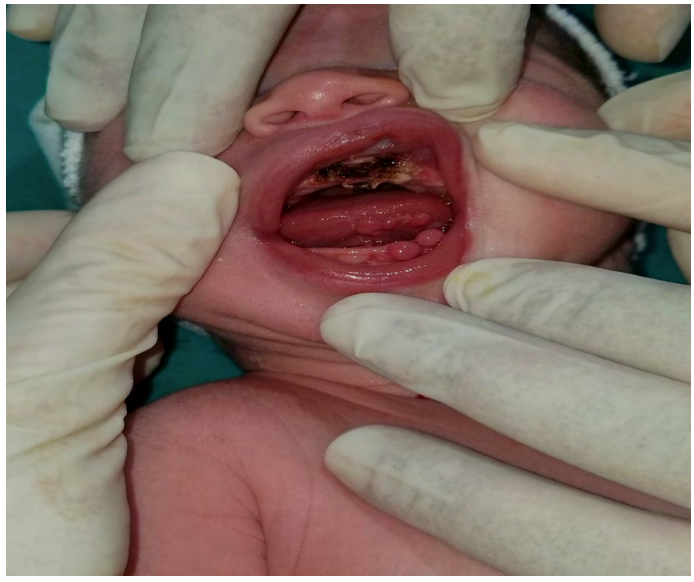

Figure 3: Operation view.

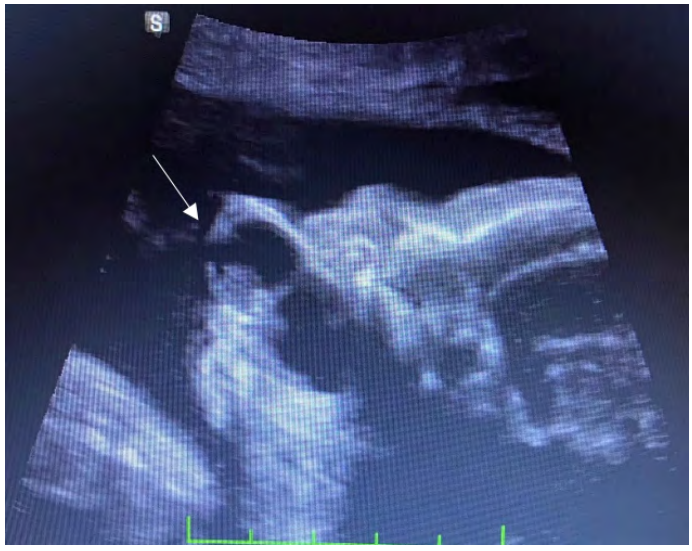

Figure 4: Ultrasonographic view of oral mass (white arrow).

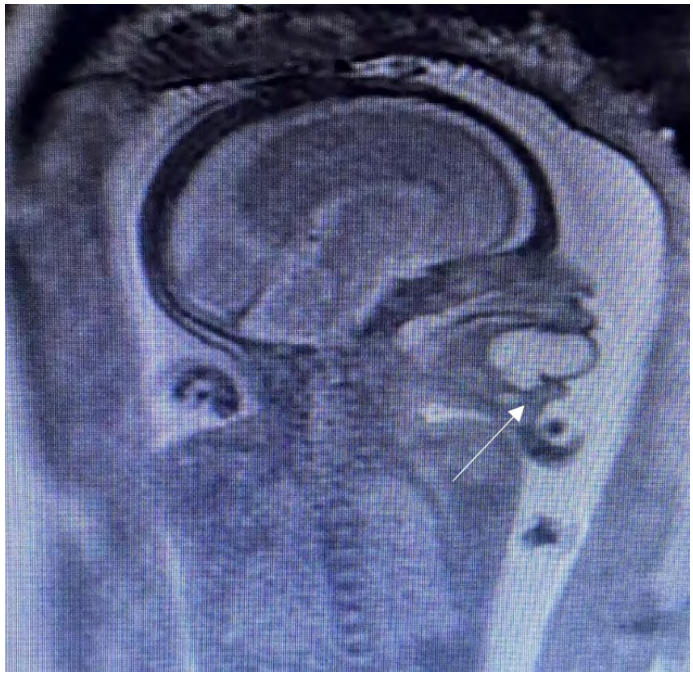

Figure 5: Fetal magnetic resonance imaging of oral mass (white arrow).

DISCUSSION

Congenital fetal oral masses are rare. Although most congenital oral masses are benign, they may result in mechanical airway obstruction, leading to poor neonatal outcomes at delivery.

The differential diagnosis of fetal oral masses includes macroglossia, epignathus, epulis, thyroglossal duct cyst, teratoma, hemangioma, and ranula.

Congenital epulis, also known as congenital gingival granular cell tumor, is a benign soft-tissue tumor arising from the alveolar ridge.[1] The etiology of congenital epulis is unknown. A hormonal etiology has been proposed, because it is 10 times more common in female fetuses than in male fetuses.[2] The size of an epulis may vary from a few millimeters to several centimeters. It is usually solitary and, unlike epignathus with its irregular, heterogeneous, and mixed echogenic shape,[3] it has homogeneous echogenicity. Epulis usually occurs in the third trimester, which may explain its absence on the first two trimester ultrasound examinations.[4]

The blood flow pattern may help distinguish congenital epulis from the other oral masses. Unlike hemangioma with a disorganized high-flow pattern, it has a single feeding vessel.[5]

Thyroglossal duct anomalies are the most common malformation in the neck, occurring anywhere in the area of migration of the thyroid tissue.[6] In approximately 2% of cases, they originate from the foramen cecum at the base of the tongue and extend to the pyramidal lobe of the thyroid gland.[7,8] Due to the similarities between a ranula and a thyroglossal duct cyst with respect to their avascular and anechoic nature,[9] ultrasonography was not helpful in distinguishing between a ranula and a thyroglossal duct cyst. The definite diagnosis was made by histopathological examination.

When a fetal oral mass is detected, it should be evaluated for obstruction of the oral cavity. The absence of the normal fluid-filled stomach and the presence of polyhydramnios are signs of the absence of fetal swallowing. The size of the mass and the amount of the amniotic fluid should be monitored during pregnancy.[10] If there is any sign of obstruction, cesarean delivery should be preferred. If the mass closes the oral cavity, asphyxia can be prevented by EXIT, in which the mass is excised without interrupting the fetomaternal circulation.[11]

Statement

Informed Consent: Written informed consent was obtained from patients who participated in this study.

Peer-review: Externally peer-reviewed.

Author Contributions: Concept – ZB, GŞ; Design – KS, AK; Supervision – HMT, MV; Resource – BÖÖ, SU; Materials – ZB, KS; Data Collection and/or Processing – GŞ, AK; Analysis and/or Interpretation – HMT, BÖÖ; Literature Search – MV, SU; Writing – ZB, GŞ; Critical Reviews – MV, HMT.

Conflict of Interest: The authors have no conflict of interest to declare.

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