Prenatal Diagnosis of Oral Teratoma by Ultrasound

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Abstract

A pregnant woman had a normal second-trimester anatomic survey at 22 weeks gestation. She was revealed to have a fetal oral mass with polyhydramnios and invisible stomach bubble by ultrasound at 28 weeks. A 50 mm × 36 mm × 42 mm, solid mass was found in the fetal mouth, filling the entire oral cavity. Fetal magnetic resonance imaging showed a homogeneous solid mass in the oral cavity compressing the hypopharynx. At 33 weeks, preterm labor occurred because of the continuation of increased amniotic fluid volume, and a female infant was vaginally delivered. The infant died shortly after tracheal intubation attempt failed. Autopsy confirmed the prenatal sonographic finding. The final pathologic diagnosis was oral immature teratoma. Our study indicates that although oral teratomas are rare, they are readily apparent at prenatal sonographic examinations. Respiratory compromise is the frequent complication of oral teratomas, which is associated with high perinatal mortality.

Keywords: Airway obstruction, fetal teratoma, oral mass, polyhydramnios, prenatal diagnosis

Introduction

Mature teratomas are most often composed of derivatives of two or three germ cell layers. Immature teratomas, however, contain immature elements in addition to the mature components, most often consisting of immature neural tissue. The common sites of occurrence are the sacrococcyx, anterior mediastinum, testicle, ovary, and the retroperitoneum. Fetal teratomas are the most common tumors diagnosed prenatally.[1] The majority of these cases are cured by complete resection of the tumors during the neonatal period. Prenatal identification of teratomas can be benefitted in perinatal management, which differs dramatically depending on their size and location.[2] Teratomas are exceedingly rare in the orofacial region, while those arising from the oral cavity and diagnosed *in utero* are rarer still.[3] In this study, we report the prenatal diagnosis of a case of oral teratoma arising in the palate with sonographic and histopathologic features.

Case Report

A 33-year-old pregnant woman, G2P1, was referred to our unit at the third trimester of gestation because of polyhydramnios. She had a healthy boy. The woman denied any exposure to teratogenic agents during pregnancy. She had a normal nuchal translucency scan at 12 weeks and a normal anatomic scan at 22 weeks, respectively. At 28 weeks, the woman complained that she felt abdominal distention. An in-detail sonographic evaluation revealed a female fetus of appropriate size for gestational age but an increased amniotic fluid index (AFI) of 271 mm. A 50 mm × 36 mm × 42 mm, solid mass was found in the fetal mouth, filling the entire oral cavity [Figure 1a and b]. Fetal lips could not be closed, but the mass did not protrude out of the mouth. The fetal stomach bubble was invisible in a 30-min period on ultrasound. No other structural anomalies were observed. Fetal echocardiography was normal. Fetal magnetic resonance imaging (MRI) showed a homogeneous solid mass in the oral cavity compressing the hypopharynx [Figure 1c]. Based on these findings, an oral tumor was presumed to be the most likely diagnosis. At 33 weeks, the AFI rose to 310 mm. The fetal prognosis was explained to the parents, and the mother was advised to come back for hospitalization in 2 weeks. Unfortunately, premature rupture of membranes (PROMs) occurred on the same day of

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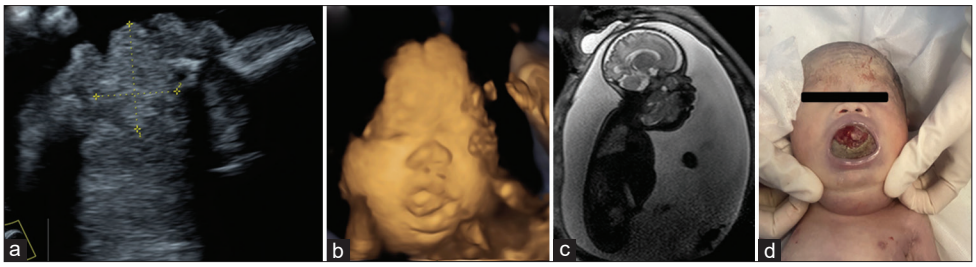

Figure 1: A fetal case of an oral immature teratoma identified by prenatal ultrasound. (a and b) Two- and three-dimensional ultrasounds revealed an oral mass. (c) Magnetic resonance imaging of an oral mass. (d) A mass filling the mouth of the fetus

ultrasound examination. Labor rapidly progressed, and there was no time to arrange *ex utero* intrapartum treatment (EXIT). A female infant was vaginally delivered and died shortly after a tracheal intubation attempt failed.

Autopsy confirmed the prenatal sonographic finding [Figure 1d]. The mass was measured 62 mm × 46 mm × 51 mm in diameter and originated from the anterior hard palate near the midline. Microscopically, immature elements originating from all three germ layers were seen [Supplementary Figure 1]. The immature elements originating from neuroectoderm showed neural tube formation and rosettes. Immature glial tissue, cartilage, fat, fibrous connective tissue, choroidal tissue, ependymal tissue, and squamous epithelium were also observed. The final pathologic diagnosis was oral immature teratoma. Chromosomal microarray analysis of the fetal tissue was normal.

Discussion

Teratomas are the most common solid tumors in fetuses. They can be seen in any parts of fetal body along the midline from the coccyx to the pineal gland. Embryologically, teratomas are thought to arise from the primitive germ cells during their migration from yolk sac to genital ridge.[4] Compared to adult teratomas, ectodermal components dominate in fetal teratomas, and immature components are more common. In earlier studies, oral teratomas have been found to be associated with chromosomal abnormalities.[5-7] However, we believe that, as evidenced by our case, there is currently only a very small chance of this association in case of teratomas because of the routine implementation of first trimester aneuploidy screening in clinical practice.

As the 18–22-week anatomic sonography has become one part of prenatal care, oral teratomas can be detected *in utero*. The standard anatomy protocol includes a detailed evaluation of the head and neck, including a sagittal view of the facial profile and a coronal view of the face for visualizing nose and lip. With these views, an oral mass or macroglossia mimicking an oral mass can be identified. On sonography, the tumors appear as cystic or solid masses in the oral cavity that displace the tongue. They fill the mouth and can protrude outside when the tumors grow larger. Persistent opened fetal mouth is also a clue to the diagnosis.[8] The large size of the mass usually precludes definition of its origin or its relationship to intracranial structures. Most often, the tumors arise from the hard or soft palate. However, they can also be lingual teratomas, expand directly into the cranium, or coexist with an intracranial tumor without communication.[9,10] In these cases, fetal MRI is indicated as it is able to determine airway obstruction by the expansion of oral masses and assess the possible intracranial abnormalities. It should be emphasized that late-onset teratomas may be missed by the traditional structural scan. In a study, a fetal oral mass was not present on two prior detailed anatomic surveys at 15 and 22 weeks, and it was revealed as a solitary solid mass, measuring 2 cm × 1.6 cm, filling the oral cavity at 32 weeks.[11] Another study reported prenatal diagnosis of oral masses in six cases; five were diagnosed at a gestational age of >33 weeks and only one diagnosed at 24 weeks.[12] The present case had a normal anatomic sonogram and was not diagnosed until at 28 weeks when the rapidly growing tumor resulted in the patient's clinical symptoms. In this case, the mass inhibited fetal swallowing, which manifested at sonogram as an absent stomach and polyhydramnios, placing the pregnancy at risk for PROM.

For teratomas found prenatally in the head or neck, high perinatal mortality usually occurs, mainly as the result of upper airway obstruction due to local mass effect.[13,14] The size of oral teratomas is an important prognostic factor because larger tumors often cause both feeding difficulties and mechanical airway obstruction. For those identified in the second or third trimester, repeated sonographic evaluations should be undertaken, focusing on the growth of the mass and assessing fetal breathing and swallowing. Tracheal occlusion should be anticipated before birth. Such potential impairments may necessitate an immediate intervention to secure the neonate's airway following delivery, and severe cases may require an EXIT procedure.[15]

Conclusion

We presented a case of oral immature teratoma identified by a third-trimester ultrasound. The pregnancy was interrupted unexpectedly with PROM due to severe polyhydramnios. The infant passed away because of an unplanned delivery with inadequate preparation of rescue measures. Our report indicates that for pregnancies with polyhydramnios, a detailed ultrasound is warranted, including a survey of fetal oral cavity.

Respiratory compromise is the frequent complication of oral teratomas, which is associated with high perinatal mortality. Our report also indicates that for a pregnancy with a fetal oral mass and signs of possible impairments in breathing functions such as severe polyhydramnios, elective delivery should be considered at a proper gestational week before PROM occurs, and pediatric team should be notified in advanced and prepared to rapidly secure the neonatal airway.

Declaration of patient consent

The authors certify that they have obtained appropriate patient's consent form. In the form, the patient has given the consent for the prenatal images and other clinical information to be reported in the journal. The patient understands that the name and initial will not be published and due efforts will be made to conceal the identity, but anonymity cannot be guaranteed.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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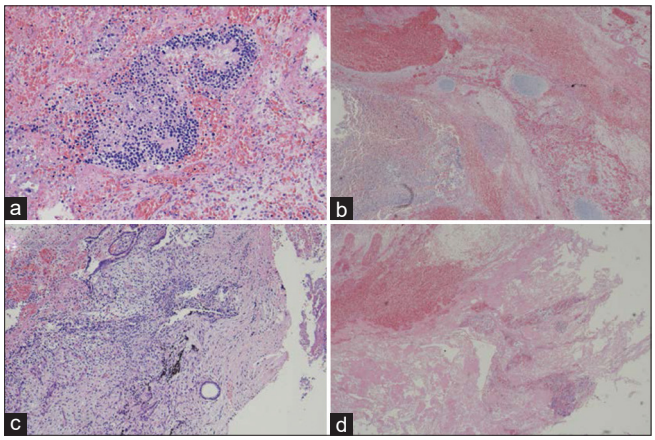
Supplementary Material



Supplementary Figure 1: Histological results of the oral tumor. (a) Neuroectoderm showing neural tube formation (H and E, ×200). (b) Glial tissue, cartilage, fat, fibrous connective tissue (H and E, ×100). (c) Choroidal tissue (H and E, ×100). (d) Squamous epithelium (H and E, ×100)