A fetus with a mass in the oral cavity: a rare case of oral eruption cyst

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Case report

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Abstract

Background

An ultrasonographic examination is widely used for prenatal screening of abnormal findings. The approach to the oral cavity of the fetus requires special attention because many aspects are unique and peculiar to this period of life. Obstetricians and pediatricians should be aware of the characteristics of normal patterns and be prepared to make a correct diagnosis. Congenital eruption cysts(EC) are rarely observed in fetuses and newborns, because at this stage of life, tooth eruption is unusual.

Case presentation

We report a 26-year-old woman who had a sonographic examination at 27^{+5} weeks of pregnancy that showed a fetal oral cyst. During an ultrasound examination, we detected a hyperechogenic solid mass with a homogeneous texture. This mass was well circumscribed with a layer of anechoic area with no abnormal vessel. The cyst appeared to originate from the body of left mandible. Real-time assessment showed that the cyst was fixed and confined to inferior maxilla. Her neonate was finally diagnosed with EC.

Conclusion

Ultrasound can reveal the distinctive features of oral cysts lesion and can be useful for the differential diagnosis. The treatment was required mainly by the symptomatology of the newborn. Therefore, prenatal ultrasonography could be used to conjecture the position and size of the mass, which is considered to be useful for localized and qualitative diagnosis prior to afterbirth treatment. Our findings indicate that ultrasound is an appropriate diagnostic tool to identify antenatal EC.

Background

Malformations of the oral cavity may manifest at birth or later, after development of the involved disturbed structures. Congenital malformations of the oral cavity include cleft lip, palate, and alveolus defects, developmental anomalies of the tongue, vascular anomalies, developmental disturbances of the teeth and/or tooth structure, and congenital dysontogenetic tumors or tumorlike conditions[1]. Although congenital oral masses are rare, they are readily detectable during fetal ultrasound screening.

EC is a benign, developmental odontogenic cyst, which creats a pathological cavity having fluid, semifluid, or gaseous content, that accompanies an erupting primary tooth, forming shortly before the tooth's appearance in the oral cavity. In the past, they were classified as a dentigerous cyst but according to the World Health Organization’s classification, it is a form of dentigerous cyst occurring within the soft tissues overlying a tooth in eruption with no bone involvement[2, 3]. EC usually occurs in the first decade of life, and it could be in any forms in the jaw arch with preference in the maxillary arch. And the majority of eruption cysts are located in the incisal and molar areas, followed by canine and premolar areas[4].

A number of complications can arise are associated with congenital pathologies in the oral cavity: pain on sucking, refusal to feed, respiratory difficulty because of airway obstruction, aspiration of fluids or teeth when natal or neonatal teeth are present[5]. Because of this, oral cysts are also of great concern to parents who may be confused and misinformed about these pathologies, generating misconceptions and myths around the newborn child. Information to parents of the pathology affecting the child and its implications is of paramount importance.

We reported and discussed an oral mass which was diagnosed incidentally at mid-trimester exam and repaired successfully in the infancy. The present article is the first case of prenatal observation from China regarding EC.

Case Presentation

A 26-year-old, gravida 1, para 0 woman was admitted for further screening at 27^{+5} weeks of gestation in our department because congenital aortic valvar stenosis was suspected in a local hospital. During an ultrasound examination, we detected a hyperechogenic solid mass with a homogeneous texture. This mass was well circumscribed with a layer of anechoic area with no abnormal vessel architecture as shown by Doppler ultrasound (Fig. 1, Supplementary video). The cyst measured 0.6×0.5×0.5 cm and appeared to originate from the body of left mandible. Real-time assessment showed that the cyst was fixed and confined to inferior maxilla. In oblique coronal plane view, there was interruption of echogenicity on the palate with intact upper alveolar echogenicity (Fig. 2A). Cleft palate was suspected (Fig. 2B). Three-dimensional (3D) reconstruction of the face showed intact lips (Fig. 2C), but the 3D imaging effect of this small cyst was not ideal. Fetal echocardiography showed aortic valve stenosis, pulmonary valve stenosis, and tortuous ductus arteriosus, with no major hemodynamic abnormalities. There were no major changes during the rest of pregnancy.

A male neonate was delivered by cesarean section at 37^{+4} weeks of gestation owing to premature rupture of the membranes and breech presentation. The operation was carried out successfully. The neonate had Apgar scores of 10, 10 at five and ten minutes. No cleft palate was found in the first neonatal examination. There was a mass on the inferior alveola, which measured approximately 0.5×1.0 cm, and was firm in consistency, pink, and had a smooth surface (Fig. 3A). The neonate was examined by a stomatologist. Surgical incision of the cyst was carried out and a tooth was exposed (Fig. 3B). The final histopathological diagnosis was an EC.

The patient was reevaluated at a follow-up visit 8 months after the surgery. The child’s recovery was uneventful and there was no aggravation of the heart condition.

Discussion

EC is a benign, developmental odontogenic cyst, which creates a pathological cavity with fluid, semifluid, or gaseous content. EC accompanies an erupting primary tooth and forms shortly before the tooth’s

appearance in the oral cavity. The prevalence of EC has not been well studied. An extensive literature review showed a low prevalence of EC [6, 7]. In some surveys, an increased incidence of EC has been observed in Caucasians [2, 7, 8]. Most reports of ECs are in the first decade of life, and only a few studies have shown this type of lesion in newborns [8, 9]. To the best of our knowledge, there are have been no prenatal reports of EC. We observed one case of EC in an Asian fetus during prenatal screening, which was confirmed after delivery. Ultrasound may provide some information for the diagnosis of EC, including the extent and contents of the mass, which may affect the mode of delivery.

The origin of an EC is still controversial. Aguiló et al. [10] studied 36 cases and found that trauma, caries, infection, and less space for eruption were possible factors that may cause EC in older children. Some authors have attributed the origin of EC to degenerative changes in reduced enamel epithelium after the end of amelogenesis or have suggested that this cyst develops from the remnants of the dental lamina that coats the erupting teeth [2, 6]. On the basis of prenatal discovery of EC, we speculate that the latter histological interpretation was likely in our case.

Standard ultrasound screening views or targeted views can help to detect the mobility, extent of involvement, and tissue characteristics of this lesion. Mandibular central incisors and permanent first molars are the most common sites for EC [9]. We found that an EC during the fetal period was detected as a lesion that comprised a solid mass and cystic fluid with low mobility. The homogeneous hyperechogenic mass of the lesion was finally determined to be a tooth. In the oblique coronal plane view, the signal was absent behind the tooth, which strongly absorbed ultrasonic waves, and acoustic shadowing of the tooth caused interruption of the echo on the palate [11]. Therefore the artifact of EC on the palate can easily be incorrectly diagnosed as orofacial clefts. 3D imaging allows visualization of structures in planes that are difficult to obtain with conventional 2D sonography. This visualization is achieved by collecting a multiplanar reconstruction view displaying a series of 2D slices in three perpendicular orthogonal planes. However, poor image acquisition and visualization using 3D imaging occur when fetal movement or position does not allow adequate imaging of the specific area. Additionally, 3D ultrasound imaging is subject to the same types of artifacts seen in 2D images [12]. Therefore, screening views should include multiple sections from different angles to avoid the artifacts.

Treatment options for EC include no treatment and follow-up or surgical extraction. EC may be associated with natal or neonatal teeth, such as in our case. When the cyst interferes with feeding, natal teeth have high mobility and/or are poorly developed, and they need to be removed.

In conclusion, ultrasound can reveal the distinctive features of fetal EC. The treatment required for EC is mainly determined by the symptomatology of the newborn. Prenatal ultrasonography can be used to identify the position and the size of the mass, which are considered to be useful for localized and qualitative diagnosis before birth.

Declarations

Ethics approval and consent to participate: Not applicable

Consent for publication: Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Availability of data and materials: Data sharing not applicable to this article as no datasets were generated or analysed during the current study.

Competing interests: The authors declare that they have no conflict of interest.

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Figures

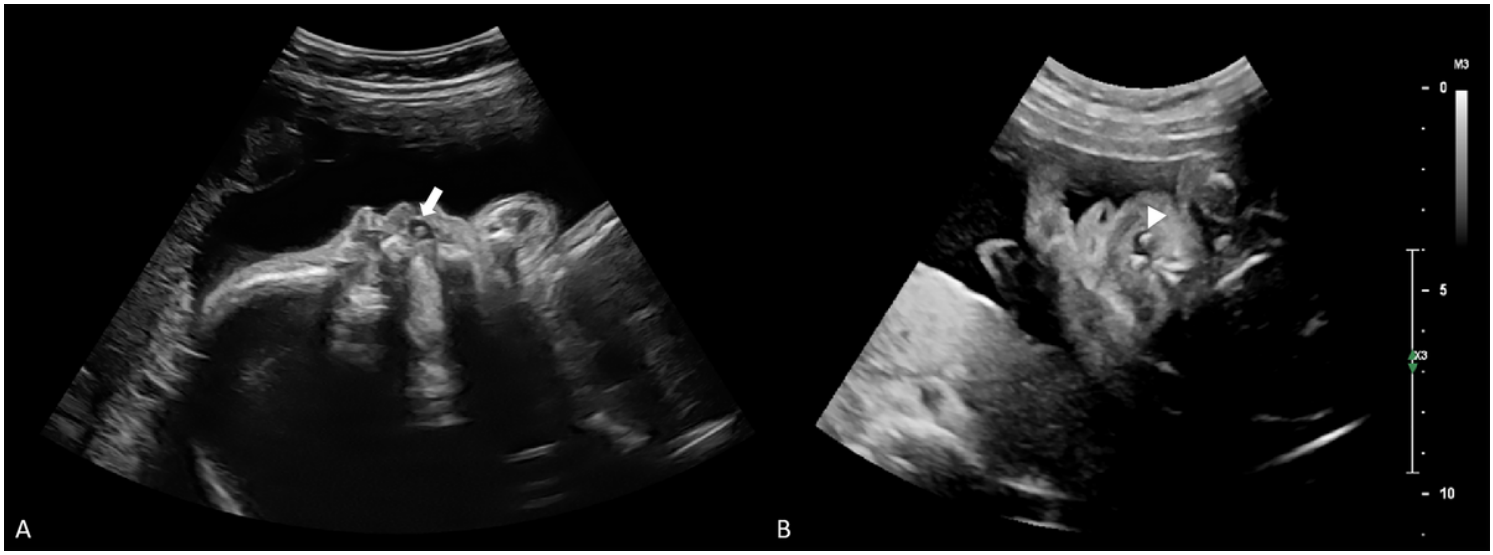

Figure 1

Two-dimensional ultrasound imaging of the cyst. A. Prenatal sonography shows a cystic mass (arrow), which includes a homogeneous hyperechogenic solid mass and a layer of anechoic fluid space. B. The mass originates from the left inferior maxilla region (arrowhead).

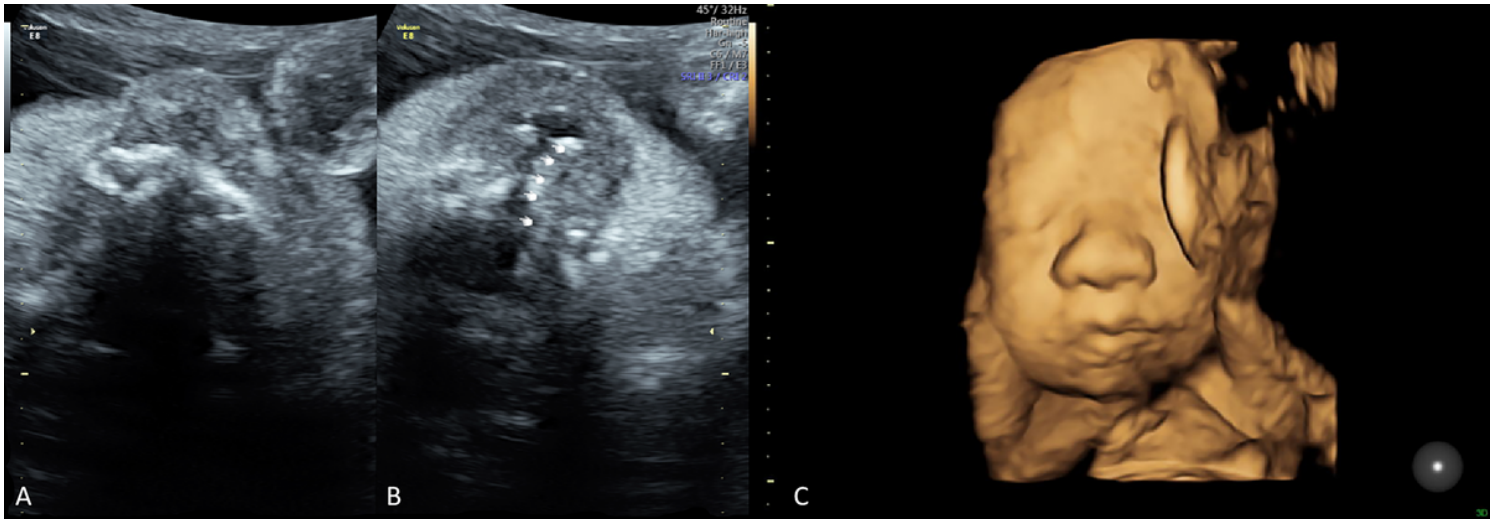

Figure 2

Two-dimensional ultrasound imaging in the axial transverse view of the fetal palate and three-dimensional volume reconstruction of the fetal face. A. Alveolar arch of the maxilla. B. Interruption of the echo on the palate. C. Three-dimensional reconstruction of the face showing intact lips.

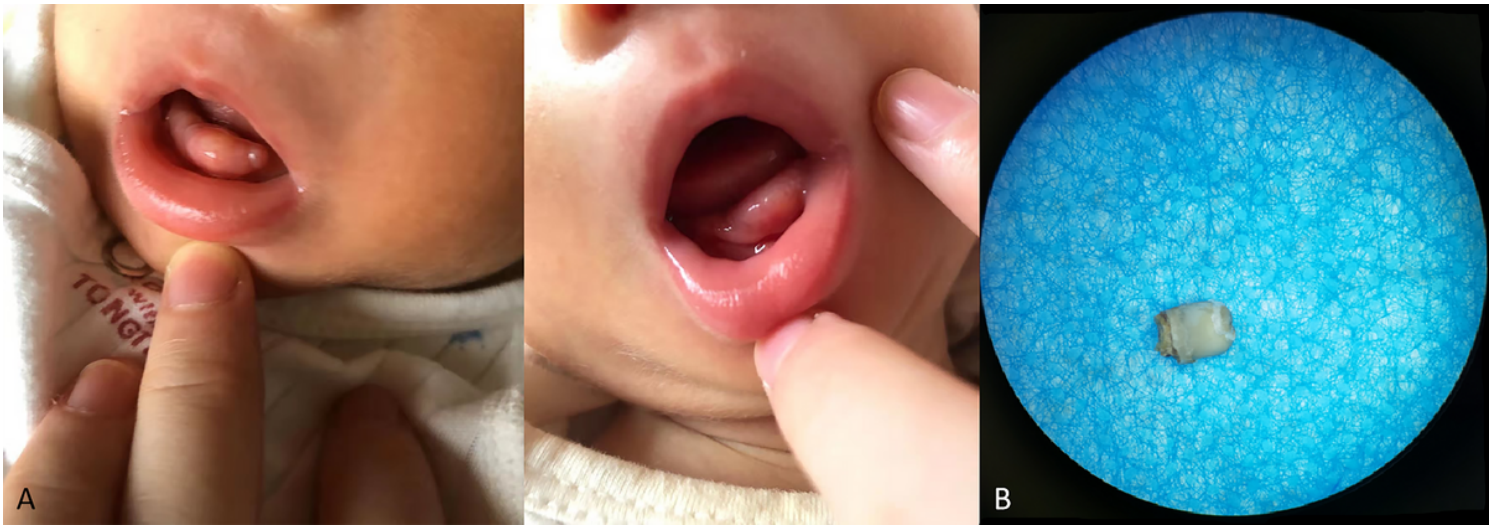

Figure 3

The first neonatal examination of the cyst. A. Gingival cyst in the anterior mandible, which is equivalent to where central incisors present. B. A tooth was exposed after surgical incision of the cyst.

Supplementary Files

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- Supplementaryvideo.mp4