

Case Report

A fetus with a mass in the oral cavity: a rare case of oral immature teratoma

Yong Huang¹, Hongxia Pan²

¹Department of Pathology, 251 Hospital of PLA, Zhangjiakou 075000, PR China; ²Department of Diagnostic Ultrasonic and Diagnostic Imaging, 251 Hospital of PLA, Zhangjiakou 075000, PR China

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Abstract: Teratomas are congenital germ cell tumors composed of diverse tissues of ectodermal, mesodermal, and endodermal origin with variable levels of maturity. Immature teratoma is an uncommon prenatal finding, which was rarely reported in the oral region. We herein report a unique case of a male neonate with a mass arising in his oral cavity. Ultrasonic studies diagnosed it as fetus-in-fetus or teratoma and pathological studies of the resected mass provided supportive evidence for the case of an oral immature teratoma. To our knowledge, there are few cases of oral immature teratoma arising in the fetus reviewing the literature.

Keywords: Immature teratoma, fetus, oral

Introduction

Teratomas of head and neck are rarely encountered and account for less than 5% of reported cases [1-5]. The most common sites of occurrence are the nasopharynx and cervical region. Teratomas of the palate are rare. We describe histopathologic features of an oral immature teratoma arising in the palate of a fetus and review the literatures.

Case report

A 28-year-old woman, gesta 1, para 0, was referred for prenatal care because of fetal abnormality. The spouses were unrelated. She denied a prenatal exposure to teratogenic drugs, ovulation-inducing agents. The patient's medical and family histories were unremarkable. Obstetric sonography at 18 weeks' gestation found a single fetus of appropriate size for gestational age with normal amount of amniotic fluid and placental characteristics. Two-dimensional ultrasound revealed a 4.7×3.2×2.6 cm, cystic-solid mass at the right parotid and oral part of the fetus (**Figure 1A**). Three-dimensional ultrasonography revealed an irregular mass at the orofacial region (**Figure 1B**). The appearance of mass was irregular, with

limb-like structure. Based on these findings, teratoma or fetus in fetus was presumed to be the most likely diagnosis. An induction delivery was performed.

Pathological findings

Macroscopically, the mass was measured 5.5×4.1×2.8 cm, regularly, located beneath the palate, with irregular and segmented appearance (**Figure 1C, 1D**). Microscopically, on low power field, mature bone and immature cartilage were seen. Beneath the mature bone tissue, a striking feature was the proliferation of neural tissues to form nodular mass (**Figure 2A**). The immature neuroepithelium showed neural tube formation and rosettes (**Figure 2B**). The microscopic observation of other focuses, mature embryonic tissue containing elements of the 3 germinative layers, such as striated muscle, intestinal epithelium, keratotic epithelium, etc. were found. The final pathologic diagnosis was oral immature teratoma.

Discussion

Teratomas of the head and neck are rare congenital lesions. Mature teratomas are composed of tissues derived from the three germ

Oral immature teratoma

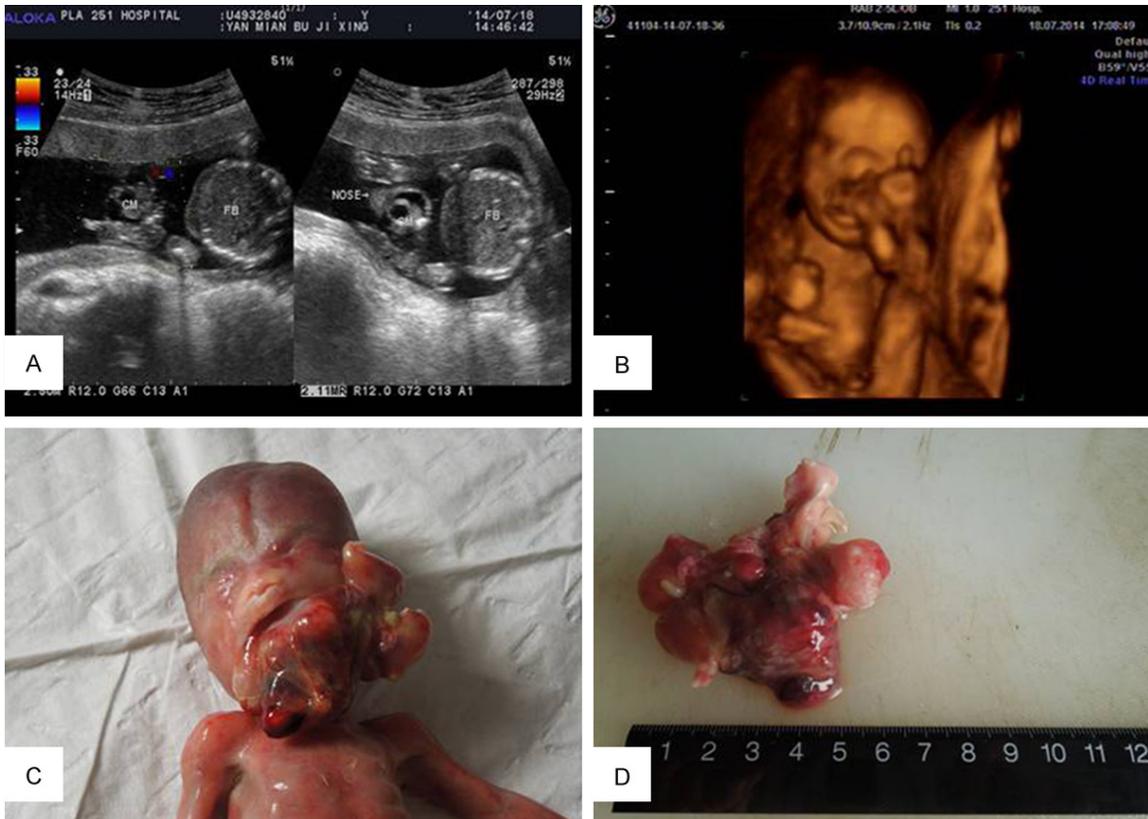


Figure 1. A. Two-dimensional ultrasound revealed a cystic-solid mass at the right parotid and oral part of the fetus. B. Three-dimensional ultrasonography revealed an irregular mass at the orofacial region. C, D. The mass, measured 5.5×4.1×2.8 cm, was located beneath the palate, with irregular and segmented appearance.

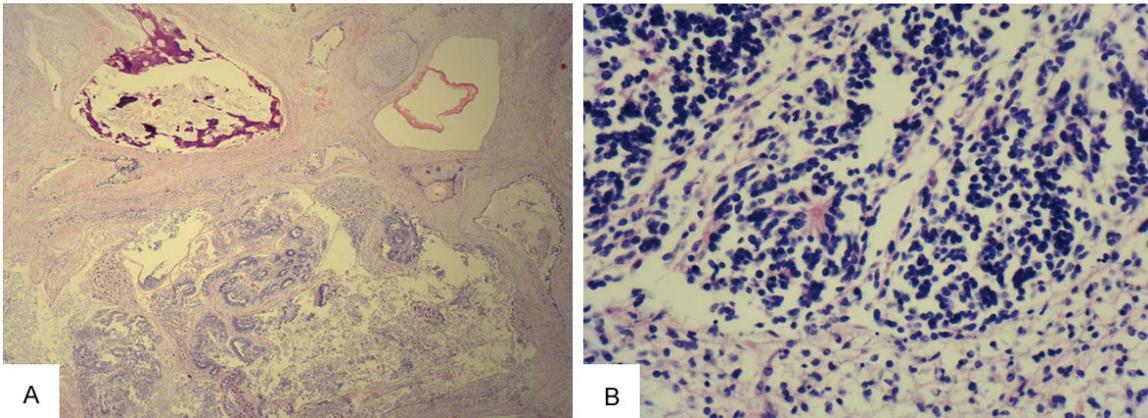


Figure 2. A. On low power field, mature bone and immature cartilage were seen. Beneath the mature bone tissue, a striking feature was the proliferation of neural tissues to form nodular mass. B. The immature elements originating from neuroectoderm show neural tube formation and rosettes.

layers. Immature teratomas are different to mature teratoma, which contain elements of all three germ layers with a prominent neuroectodermal component. Occasionally, the tumor may be composed of a small number of tissues.

The occurrence of a congenital oral teratoma (epignathus) is 2-9% of all teratomas [6, 7]. The aetiology of epignathus remains unclear. The most common theory supposes that an epignathus derives from pluripotential cells in Rathke's

pouch that grow in a disorganized manner [8]. Reviewing the literature, Cleft palate is most frequently involved. It is thought to be caused by the migration of embryonic tissue into the nasopharynx at a very early stage of fetal life, before the normal union of the bilateral palatal shelves [9]. Oral teratoma typically emerges during the neonatal period and is associated with airway obstruction and high infant mortality rate.

In the immature teratomas, tissues might arrange from embryonic to mature, which are scattered haphazardly throughout the tumor, which differ from the orderly organoid arrangement seen in a mature teratoma. Sometimes, the tumor is composed mainly of mature tissues, differentiation from mature teratoma may be difficult, careful examination and thorough sampling of the tumor is strongly recommended. Immature teratoma might be combined with other neoplastic germ cell elements, especially arising in ovary, testis and pineal body. The grade of immature teratoma has been tailored to estimate the amount of immature tissue in the teratoma. Assessments of grade were based on the overall extent of immature neuroepithelium.

The primary differential diagnosis of teratoma is fetus in fetus. Fetus in fetus can be distinguished from teratoma by the presence of a vertebra. What's more, teratomas are most commonly seen in the sacrococcygeal or head and neck region, whereas fetus in fetus cases is typically located in the retroperitoneal position. Moreover, enclosure of the malformed fetus within the amniotic membrane helps distinguish it from teratoma [10].

Disclosure of conflict of interest

None.

Address correspondence to: Dr. Hongxia Pan, Department of Diagnostic Ultrasonic and Diagnostic Imaging, 251 Hospital of PLA, 13 Jianguo Road, Zhang Jiakou 075000, Hebei, PR China. Tel: +86-313-8785267; Fax: +86-313-8785267; E-mail: zjk-phx@163.com

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