ISSN 2218-4333 (online)

World Journal of *Clinical Oncology*

World J Clin Oncol 2024 September 24; 15(9): 1117-1255

Published by Baishideng Publishing Group Inc

World Journal of Clinical Oncology

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Monthly Volume 15 Number 9 September 24, 2024

World Journal of Clinical Oncology

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ABOUT COVER

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RESPONSIBLE EDITORS FOR THIS ISSUE

Production Editor: *Yu-Qing Zhao.*; **Production Department Director:** *Xu Guo*; **Cover Editor:** *Xu Guo.*

NAME OF JOURNAL
World Journal of Clinical Oncology

ISSN
ISSN 2218-4333 (online)

LAUNCH DATE
November 10, 2010

FREQUENCY
Monthly

EDITORS-IN-CHIEF
Hiten RH Patel, Stephen Safe, Jian-Hua Mao, Ken H Young

EDITORIAL BOARD MEMBERS
https://www.wjgnet.com/2218-4333/editorialboard.htm

PUBLICATION DATE
September 24, 2024

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World J Clin Oncol 2024 September 24; 15(9): 1245-1250

DOI: 10.5306/wjco.v15.i9.1245

ISSN 2218-4333 (online)

CASE REPORT

Prenatal ultrasound diagnosis of fetal maxillofacial teratoma: Two case reports

Chuan-Fen Gao, Pei Zhou, Chen Zhang

Specialty type: Obstetrics and gynecology

Provenance and peer review: Unsolicited article; Externally peer reviewed.

Peer-review model: Single blind

Peer-review report's classification

Scientific Quality: Grade B

Novelty: Grade A

Creativity or Innovation: Grade B

Scientific Significance: Grade B

P-Reviewer: Mohey NM

Received: May 17, 2024

Revised: July 25, 2024

Accepted: August 7, 2024

Published online: September 24, 2024

Processing time: 103 Days and 23.3 Hours

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Abstract

BACKGROUND

Facial teratoma is a rare benign tumor that accounts for about 1.6% of all teratomas and can be diagnosed by prenatal ultrasound (US). The purpose of this report was to describe our experience with the diagnosis of fetal facial teratoma by prenatal US at second trimester to provide a reference for clinical diagnosis of fetal maxillofacial teratoma.

CASE SUMMARY

We present two cases of patients with abnormal fetal facial findings on US at second trimester of pregnancy in our department. Case 1 was a 31-year-old G3 P1 + 1 female, with US revealing a heterogeneous echogenicity of 32 mm × 20 mm × 31 mm on the fetal face, most of it located outside the oral cavity and filling the root of the oral cavity. Case 2 was a 29-year-old G1P0 female, with fetal head and neck US revealing a cystic-solid echo mass measuring 42 mm × 33 mm × 44 mm, the upper edge of the lesion reaching the palate and filling the oral cavity. The contours of the lesions were visualized using three-dimensional (3D) US imaging. Both patients decided to give up treatment. Biopsies of the lesions were performed after induction of labor, and diagnosed as maxillofacial teratoma.

CONCLUSION

Fetal maxillofacial teratomas can be diagnosed by US in early pregnancy, allowing parents to expedite treatment decisions.

Key Words: Fetal maxillofacial teratoma; Prenatal ultrasound; Diagnosis; Ultrasound; Case

report

Core Tip: Facial teratoma is a rare benign tumor that accounts for only about 1.6% of all teratomas and can be diagnosed by prenatal ultrasound (US). In this study, we presented two cases of pregnant women with prenatally diagnosed fetal maxillofacial teratomas by US. The study highlights the value of prenatal US in the identification of maxillofacial teratoma in early pregnancy and describes the ultrasonographic features of these tumors.

Citation: Gao CF, Zhou P, Zhang C. Prenatal ultrasound diagnosis of fetal maxillofacial teratoma: Two case reports. *World J Clin Oncol* 2024; 15(9): 1245-1250
URL: https://www.wjgnet.com/2218-4333/full/v15/i9/1245.htm
DOI: https://dx.doi.org/10.5306/wjco.v15.i9.1245

INTRODUCTION

Teratomas are rare fetal and neonatal tumors composed of elements derived from the three germ layers. They typically develop in midline structures, the most common forms being ovarian (37%-43%), sacrococcygeal (28%-30%), testicular (10%-15%), and mediastinal (5%-6%)[1]. Head and neck teratomas account for approximately 5% of all teratomas, with an incidence of between 1/20000 and 1/40000 births. In contrast, the incidence of neonatal facial teratomas is less than 1:200000[2,3]. Although fetal facial teratomas are most likely to be histologically benign, there have been reports of invasive recurrence and malignant transformation following incomplete resection[3]. Additionally, maxillofacial teratomas carry a significant risk of possible secondary polyhydramnios, pharyngeal obstruction, and intrauterine death[4,5]. Therefore, early and accurate identification of fetal teratomas is of great significance for physicians, to evaluate potential risks and provide treatment options, and for patients, to make decisions based on risk profiles and management options.

Prenatal ultrasound (US) screening has been reported to have extremely high sensitivity (100%) and a low false-positive rate (3.3%) in identification of fetal teratomas[6]. Due to the rare nature of fetal maxillofacial teratomas, there are few published data on US screening of these lesions. Thus, in this study we reported the ultrasonographic features of two prenatally diagnosed maxillofacial teratomas and highlighted the importance of prenatal US screening for early identification of these tumors. This study was approved by the Ethics Review Board of the First Affiliated Hospital of Anhui Medical University.

CASE PRESENTATION

Chief complaints

Case 1: A 31-year-old female, gravida 3, para 1, miscarriage 1, presented to the US department at 16 weeks of gestation. US revealed a heterogeneous echogenicity on the fetal face.

Case 2: A 29-year-old female, gravida 1, para 0, presented to the US department at 16 weeks and 6 days of gestation. US revealed a cystic-solid echogenicity with irregular shape and uneven echo of the solid part.

History of present illness

Case 1: The patient had no complaints of fever, bleeding, or vaginal discharge.

Case 2: The patient had no subjective symptoms.

History of past illness

Case 1: The patient had no special medical history.

Case 2: The patient had no known past medical history.

Personal and family history

Case 1: The patient had a miscarriage of a previous pregnancy, had no history of morphological abnormalities during delivery, and had no family history.

Case 2: The patient had no relevant personal or family history.

Physical examination

Case 1: The vital signs of the patient were stable. Cardiovascular, respiratory, and neurological examinations were normal, and abdominal examination revealed no specific abnormalities.

Case 2: The vital signs of the patient were stable and physical examination revealed no abnormal findings.

Laboratory examinations

Case 1: Laboratory tests including total blood count, renal function, liver function, and other basic biochemical tests were normal.

Case 2: No obviously abnormal laboratory markers were observed.

Imaging examinations

Case 1: US revealed a 32 mm × 20 mm × 31 mm heterogeneous echogenic mass on the fetus's orofacial surface. Most of it was located outside the oral cavity, with roots filling the oral cavity and the fetus in a persistent open-mouth state (Figure 1). The mass was irregular in morphology, with heterogeneous and uneven internal echoes, predominantly solid ones; anechoic and strong echoes, but no obvious intracranial space-occupying lesions, were also seen. Microvascular flow imaging (MVFI) showed abundant blood flow signals, with blood supply originating from the deep part of the oral cavity. Three-dimensional US (3DUS) crystal imaging showed that the mass protruded from the face and was irregular in shape.

Case 2: US scanning of the fetal face and neck revealed a cystic-solid echogenic mass of 42 mm × 33 mm × 44 mm, mainly solid with irregular shape, with the solid part showing uneven low echo (Figure 2). MVFI showed rich blood flow signals within the mass. The upper edge of the mass reached the level of the palate and filled the oral cavity, while the lower edge reached the base of the neck. The lower, but not the upper, alveolus was visible, the mandible was deformed and flared, there was significant elevation of the bilateral facets, and the mass did not break through the skin. The nose and eye sockets were visible. 3DUS crystal imaging revealed the contours of the lesion in the coronal plane, markedly protruding up to the palate, down to the base of the neck, and on both sides of the cheeks.

FINAL DIAGNOSIS

Case 1

The final diagnosis was maxillofacial teratoma, oral origin.

Case 2

The final diagnosis was maxillofacial immature teratoma.

TREATMENT

Case 1

The patient decided to give up treatment and underwent labor induction after counseling.

Case 2

The patient decided to give up treatment and underwent labor induction after counseling.

OUTCOME AND FOLLOW-UP

Case 1

After induction of labor, the fetus showed a protuberant mass in the oral cavity that presented a translucent surface interconnecting the posterior palate and had a disorganized tissue structure (Figure 3). A biopsy of the lesion tissue was performed, and pathology revealed multiple tissue sources. Based on all the examinations, a diagnosis of maxillofacial teratoma (Pathological No. GX2205411) was confirmed.

Case 2

The postpartum gross appearance was darkly swollen from the maxillofacial region to the neck (Figure 4). A biopsy of the lesion was performed, and pathological results revealed a teratoma (Pathological No. 2214830).

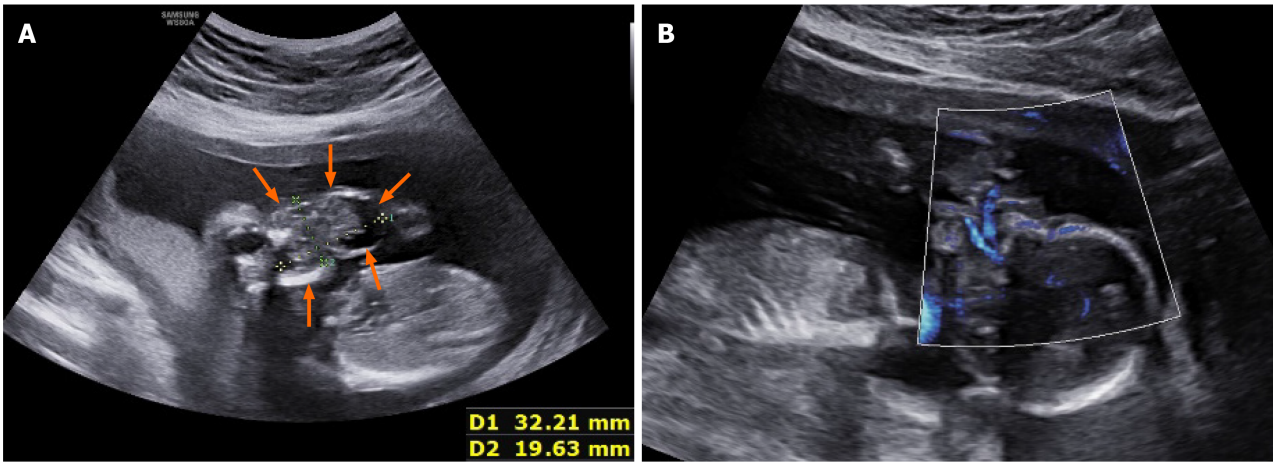

Figure 1 Prenatal ultrasound of a teratoma at 16 weeks of gestation (case 1). A: Cystic-solid echo of the fetal orofacial exophytic process (orange arrows); B: Microvascular flow imaging showed that blood flow originates from the deep oral cavity.

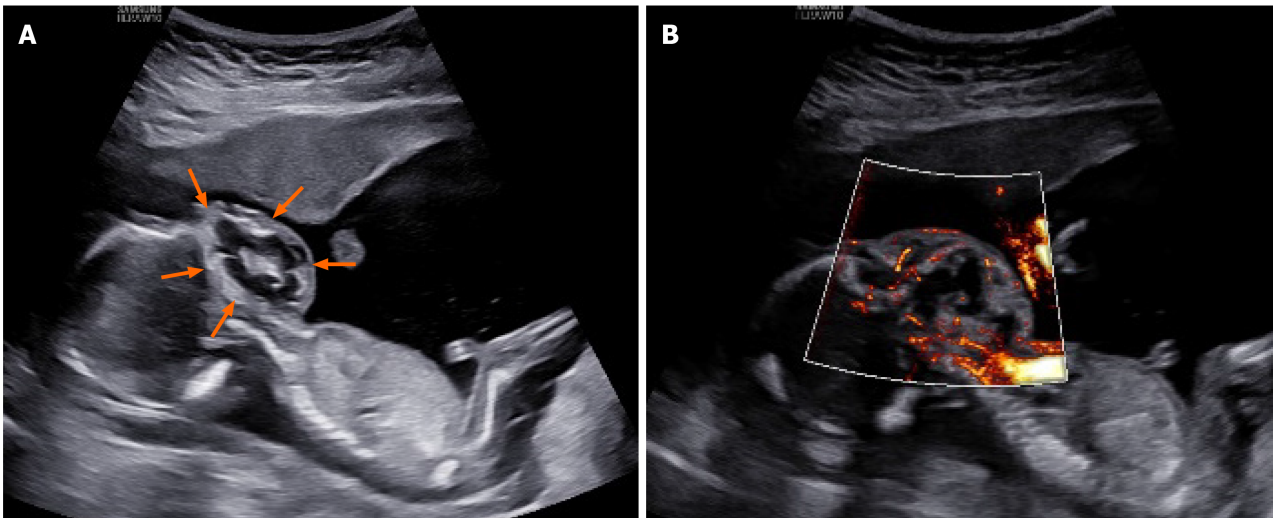

Figure 2 Prenatal ultrasound of an orofacial teratoma at 16 weeks of gestation (case 2). A: Cystic-solid echo of the fetal face (orange arrow); B: Microvascular flow imaging superimposed on section A, showing slightly abundant blood flow signals in the interior.

DISCUSSION

US is the main diagnostic method for oropharyngeal and maxillofacial teratomas. The two cases herein reported were diagnosed by US at 16 weeks of gestation, which is earlier than the time of diagnosis reported in most cases[7,8]. Our study demonstrated the significance of US in the diagnosis of fetal maxillofacial teratoma, which is generally consistent with previous findings[4,9].

US imaging of case 1 showed mainly a cystic-solid echo with strong echogenic calcification visible in the solid part. MVFI showed abundant blood flow signals in the mass, which originated from the deep part of the oral cavity, and a strong, uninterrupted echo of the upper palate. Both conventional US and 3DUS showed that most of the mass protruded outside of the mouth. This finding was confirmed at postpartum gross examination, which showed a mass completely protruding from the upper palate, without cleft palate. The US images of case 2 also showed cystic-solid echo densities but without typical calcification signs. Blood flow signal was visible in the solid part, and the mass was in a deep position, located mostly in the mouth and jaw. The skin surface was raised, but the mass did not break through the skin, and the deep contours of the coronal surface could be clearly displayed by 3D crystal imaging. The US features of the above two cases had the common characteristics of most fetal teratomas, namely mixed cystic echo and rich blood flow. The diagnosis of teratoma is more accurate if there is evidence of calcification. In this study, assessment of teratoma vascularization was performed by MVFI, which displays low-speed blood flow and is superior to conventional color Doppler and power Doppler[10]. Although the study confirmed the clinical value of US in diagnosing fetal maxillofacial teratomas, small oral and pharyngeal masses of the fetus are likely to be missed during prenatal US examination. Therefore, the development and combined application of new imaging techniques is expected to reduce the rate of missed diagnoses [11].

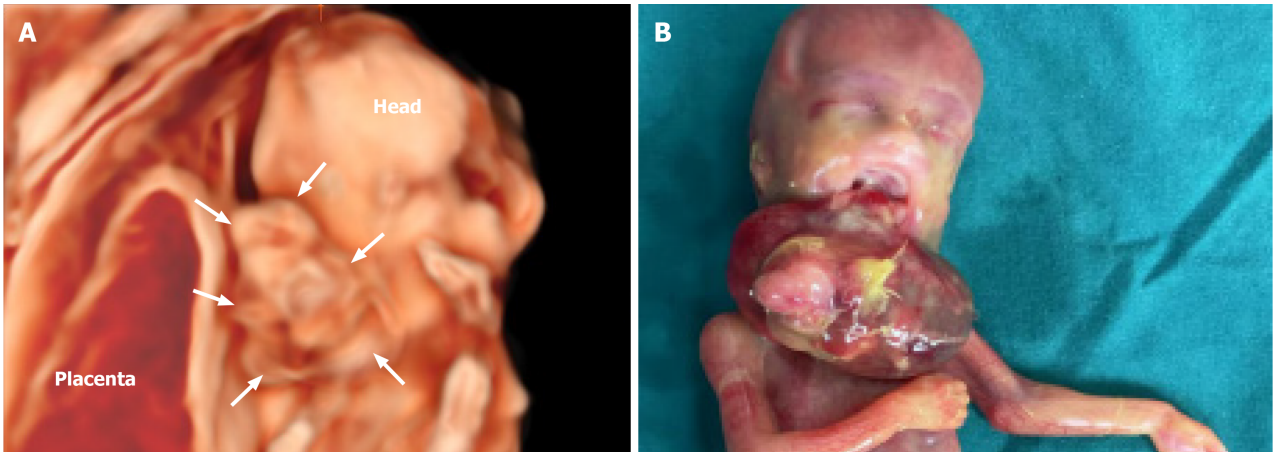

Figure 3 Gross specimen after induction of labor (case 1). A: Three-dimensional ultrasound imaging of the fetal face shows the outline of the mass (white arrows); B: The gross specimen after induction showed an extraoral mass with a disorderly structure, originating from the palate.

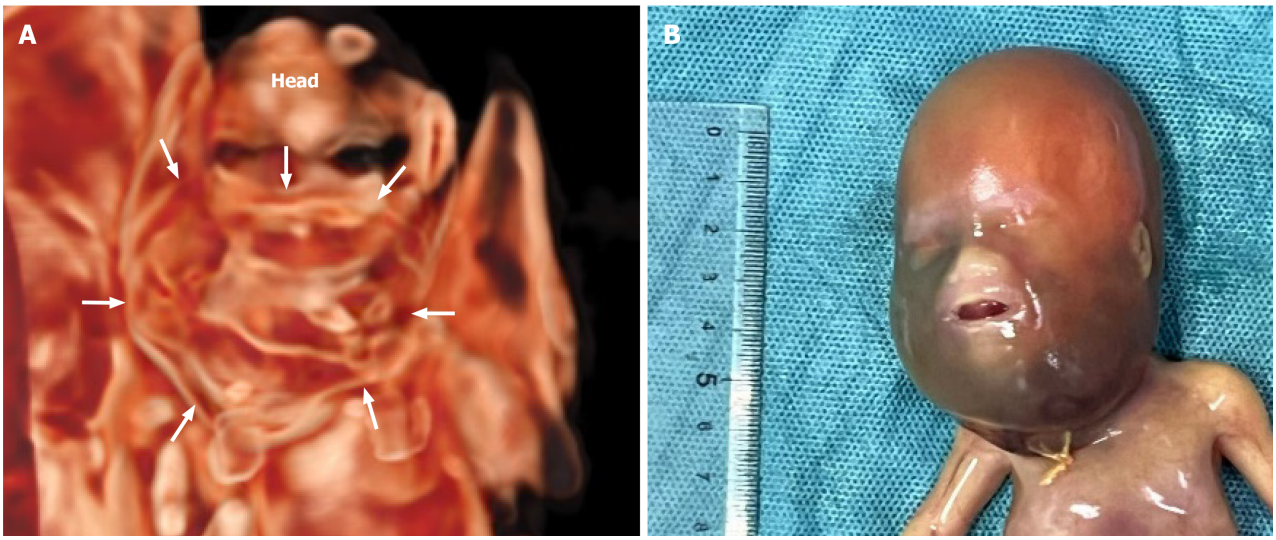

Figure 4 Gross specimen after induction of labor (case 2). A: Three-dimensional crystal imaging of the fetal face shows the internal outline of the mass in the coronal plane (white arrows); B: After induction of labor, the gross specimen showed swelling of the neck and dark color.

After consultation, both mothers and their families finally chose to induce labor due to poor prognosis. However, since they did not agree to autopsy, the exact origin of the teratomas could not be determined. Management of fetal oral and maxillofacial teratomas is challenging. Perinatal management includes ex-utero intrapartum treatment initiated by intubation during delivery or surgery on placental support to remove the tumor during cesarean section and before cutting the umbilical cord[12,13]. It has been reported that oropharyngeal teratomas have been successfully removed under fetoscopy, and that early surgery can avoid facial structure distortion and airway obstruction caused by further expansion of the mass[14]. However, fetal maxillofacial teratomas are also associated with congenital heart disease, cleft lip and palate, hypoplasia of the mandible, and other congenital malformations[15,16]. Therefore, in consideration of future development of the fetuses, the patients decided to give up treatment.

CONCLUSION

Fetal maxillofacial teratomas can be diagnosed by US in early pregnancy, allowing parents to expedite treatment decisions.

FOOTNOTES

Author contributions: Gao CF contributed to study concept and design, data collection, drafting, reviewing, and editing of the manuscript; Zhou P contributed to consultation and specimen photo acquisition; Zhang C contributed to data analysis; Both authors

have read and approved the final manuscript.

Supported by the Research Fund of the Anhui Medical University, No. 2022xkj145.

Informed consent statement: Informed written consent was obtained from the patients for publication of this report and any accompanying images.

Conflict-of-interest statement: All authors declare that they have no conflicts of interest to disclose.

CARE Checklist (2016) statement: The authors have read the CARE Checklist (2016), and the manuscript was prepared and revised according to the CARE Checklist (2016).

Country of origin: China

ORCID number: Chuan-Fen Gao 0009-0007-7387-3864.

S-Editor: Liu JH
L-Editor: A
P-Editor: Zhao YQ

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