Ultrasound diagnosis of a retroperitoneal fetus in fetu: A case report

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Abstract. Fetus in fetu (FIF) is a rare anomaly of diamniotic monochorionic twins, where a malformed fetus resides within the body of its twin. Most FIF occurs in the retroperitoneal region around the host spine and appears prenatally as a solid-cystic mass consisting of fetal-like structures. Imaging has an important role in the diagnosis of FIF. The present study reported a single case, a 45-year-old woman, with a teratoma in a third-trimester fetus diagnosed after prenatal ultrasonography (US), which showed a mass containing fetus-like echoes. FIF was considered after the US showed that the mixed solid-cystic retroperitoneal mass around the vertebral axis of the host fetus consisted of two separate masses, each containing distinct fetal visceral structures. One fetus was acardiac and the other parasitic fetus was visible with a weak heartbeat. Postpartum magnetic resonance imaging and ultrasonography (US) scans of the newborn showed a retroperitoneal cystic space-occupying mass with distinctive limbs and visceral structures. The pathological examination further confirmed the diagnosis of retroperitoneal FIF. Also, a prenatal US could detect FIF *in utero*. A cystic-solid mass containing long bones, vascular pedicles, or visceral structures around the vertebral axis of the host fetus in the US might suggest the possibility of a FIF.

Introduction

Fetus in fetu (FIF) is a very rare congenital anomaly where a malformed fetus is enclosed within the body of a twin fetus, with an incidence of 1 in 500,000 live births (1). FIF mainly occurs in the retroperitoneal region but has also been reported in other locations, such as the cranial cavity and the scrotum (2). The FIF has the same blood type, sex chromosome, protein polymorphism and DNA as the host fetus (3). The exact pathogenesis of FIF remains to be elucidated. One hypothesis suggests that FIF arises from an abnormal division of monochorionic monozygotic twins during early embryogenesis (3). Another hypothesis suggests that FIF and teratoma are two related congenital manifestations with the same pathogenesis (4).

Imaging modalities, including ultrasonography (US) and magnetic resonance imaging (MRI), have an important role in the early diagnosis and management of FIF. Since US is the most common choice/imaging in the prenatal examination, obstetricians must become familiar with the ultrasound findings suggestive of FIF. The present study reported a single case of a retroperitoneal FIF diagnosed by prenatal US.

Case presentation

A 45-year-old woman (gravida 2, para 1) was admitted at 37^{+4} gestational weeks to the Ultrasonography Department of Huidong People's Hospital, the People's Government of Liangshan Yi Autonomous Prefecture, Sichuan, with an obstetric US diagnosis of a teratoma. The initial US at 37^{+3} weeks of gestation showed a fetal-like echo in the thoracic cavity of the fetus. After admission, a repeat ultrasound examination revealed a live third-trimester singleton fetus in a cephalic presentation with a retroperitoneal mass of mixed echogenicity. One fetus had a weak heartbeat and the other was acardiac. The woman and her husband were both healthy. Her first child had no congenital abnormalities; the husband was not the father of her first child. There was no history of twin gestation or newborns with deformities.

Obstetric US revealed a mixed cystic-solid mass at the right retroperitoneal space of the host fetus. There were two parallel fetal-like signals in the upper and lower ends of the mass. The fetal-like echoes were parallel to the sagittal plane of the fetal spine (Fig. 1A and B) and closer to the ventral side of the host fetus. The fetal-like tissue, 4.7x2.1 cm in size, showed mixed echogenicity and intra-cyst septation that closely adhered to the cyst wall. Furthermore, multi-sectional

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Abbreviations: FIF, Fetus in fetu; US, ultrasonography

Key words: Fetus in fetu, fetal abnormalities, ultrasonography, prenatal, case report

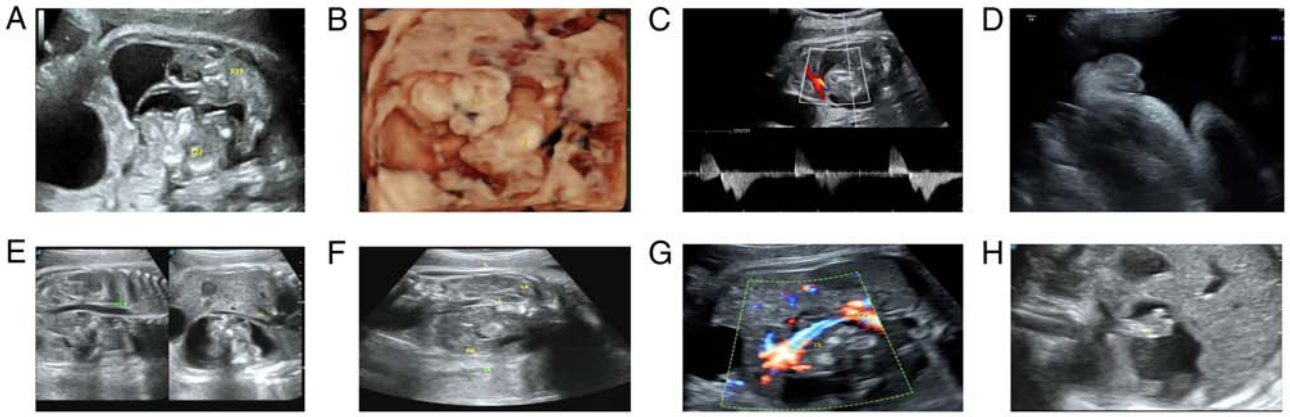

Figure 1. US of FIF. (A) US of the twin FIF in the abdominal cavity of the host fetus. (B) Surface image of the FIF in the abdominal cavity of the host fetus. (C) The spectrum of the heartbeat of the FIF in the abdominal cavity of the host fetus. (D) Limb image of the FIF in the abdominal cavity of the host fetus. (E) The abdominal aorta and inferior vena cava of host fetus were compressed and shifted inferiorly. (F) The position of the right kidney of the host fetus was relatively low and closely adjacent to the bladder. (G) The right renal artery originates from the abdominal aorta of the host fetus. (H) Ultrasound scan of the fetal genitals. US, ultrasonography; FIF, Fetus in fetu.

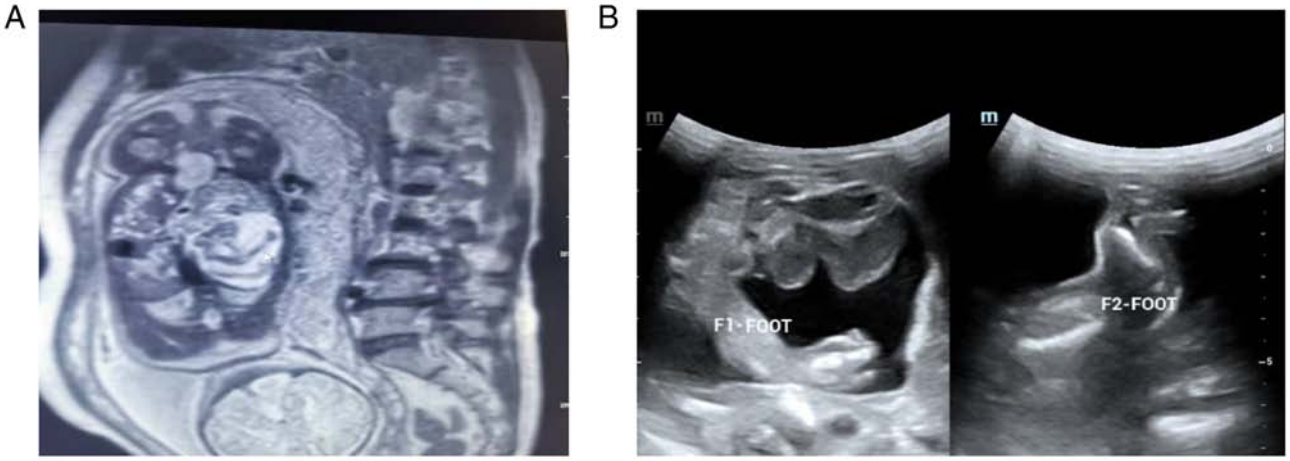

Figure 2. MRI of FIF. (A) MRI image of the fetus showed signals of a long bone in a retroperitoneal cystic mass of the host fetus. (B) After the delivery of the fetus, US showed retroperitoneal twin FIF in the host fetus. MRI, magnetic resonance imaging; US, ultrasonography; FIF, Fetus in fetu.

and multimodal scanning of the fetal-like tissue showed an irregular morphology and outline, with no fetal head, spine, upper limb, or heart echoes. The abdominal and thoracic cavities were not distinguishable. However, continuous sagittal and transverse scanning showed that one limb was continuous. The limb was covered with skin and had a long bone, a distal footpad and toe-like echoes, with a stiff morphology (Fig. 1C and D). In addition, an umbilical cord-like vascular echo was also found connected to the fetal-like mass, but no blood flow signal was found by color Doppler scan. At its rear side, another fetal sonogram with a size of about 3.9x2.1 cm could be seen. Multi-sectional and multimodal scanning showed the following: Irregular shape and contour, no fetal head, spine, or upper limb; the thorax and abdominal cavity could not be distinguished. However, continuous sagittal and transverse sections revealed the buttock-like contour at the posterior part of the body; the fetal limb was seen at the far end, similar to femoral echo; plantar echo was seen at the far end; long bone echo was seen in the proximal limb and the limb was covered with skin. A faint fetal heartbeat was seen in the chest cavity. Color doppler flow image showed blood flow signal. Pulsed wave Doppler showed a positive and negative two-way arterial spectrum. The fetal movement was continuously observed. The FIF compressed the host fetus's inferior vena cava and abdominal aorta, which showed arch-shaped shifting. The intestine of the host fetus was also compressed and shifted to the left; the right kidney was compressed and displaced into the pelvic cavity. The blood flow to the two kidneys of the FIF was from the abdominal aorta of the host fetus (Fig. 1E-H). Finally, an ultrasound diagnosis of a retroperitoneal FIF was made (Video S1 and S2).

An abdominopelvic MRI at 37^{+5} weeks of gestation showed a cystic space-occupying lesion at the right epigastric region of the fetus, suggesting a FIF with developmental deformity (Fig. 2A). The US performed on the newborn suggested a retroperitoneal twin FIF (Fig. 2B). A computed tomography examination in the newborn showed a gigantic space-occupying lesion at the right retroperitoneal region containing adipose tissue, long bone and axial skeleton-like structures, which wrapped the right renal artery and vein and compressed the pancreas, liver, intestine and surrounding blood vessels. The findings highly suggested FIF.

At 20 days after the child's birth, the patient was admitted to the Department of critical care medicine of West China Hospital of Sichuan University. He was diagnosed with FIF and underwent surgery on December 21, 2021. During the operation, a 15x13x14 cm mass was resected from the retroperitoneum and the final diagnosis was of a right retroperitoneal giant tumor (teratoma) following the surgery.

Pathological examination revealed a mass with a bunch of hand- and foot-shaped teratoma-like masses on gross inspection, with an approximate overall size of 13.5x7.8x4.6 cm; microscopy showed immature teratoma-like characteristics.

At the time of writing, the child is 1 year and 8 months old, 80 cm tall and 10 kg in weight and in good health.

Discussion

The present study presented a case of retroperitoneal FIF in a live intrauterine fetus diagnosed by US at 37 weeks of gestation. One fetus had weak cardiac activity, while the other had no heartbeat. The FIF fed on the host blood supply and compressed adjacent organs. This case represents a prenatal diagnosis of FIF, demonstrating the critical role of the US in FIF diagnosis.

FIF can be single or multiple (5) and while most FIFs reported in the literature showed no cardiac activity, the present case had FIF in which one was acardiac and the other demonstrated weak cardiac activity and visible fetal motion. The FIF had limbs, digital pads and skin. Doppler scan showed that renal blood flow of the live FIF originated from the host's abdominal aorta. A recent study also reported a viable FIF with recognizable cardiac chambers using a prenatal Doppler ultrasound (6).

US has an important role in diagnosing FIF, particularly during prenatal life. The key to diagnosing FIF *in utero* is detecting fetal structure formation in the host fetus. Moreover, Spencer *et al* (3) suggest looking for certain characteristics in the mass of the host fetus to increase suspicion of FIF. Identifying these characteristics in the prenatal US would assist in early diagnosis and informed decision-making. Consequently, the case in the present report demonstrated ultrasound characteristics similar to previous reports (6-8), including a retroperitoneal cystic-solid mass with a thin wall (representing the amniotic membrane) and clear boundaries; the solid components enlarged with gestational age, gradually showing fetal morphology surrounded by echo-free region (representing the amniotic fluid). Additionally, the mass contained an umbilical cord-like vascular pedicle in which a Doppler scan revealed a parallel artery and vein connected to the host fetus's artery and vein, respectively. Finally, the mass compressed the surrounding tissues of the host fetus without infiltration.

Some important clinical entities need to be distinguished from FIF. Teratoma is the most common differential diagnosis of FIF (9). However, teratoma is sporadic bone mass or calcification without vertebral bones, limbs, or viscera. Neuroblastoma is another congenital anomaly that may resemble FIF but is primarily characterized by a solid mass with no bones or other viscera. Last, meconium peritonitis can manifest as an abdominal mass devoid of fetal-like structures.

In conclusion, when cystic and solid masses are found in ultrasound examination during pregnancy, especially in the middle axis of the fetus, the diagnosis of an endoparasitic fetus should be considered and it should be carefully observed whether the mass contains the spinal axis, long bone, or organ structure. In addition, prenatal ultrasound diagnosis based on the spinal axis, long bone structure, skin and vascular pedicle has a high coincidence rate. Otherwise, teratoma should be considered.

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Availability of data and materials

All data generated or analyzed during this study are included in this published article.

Authors' contributions

The conception of the present study was by WTP, who was also responsible for methodology. WTP and SQZ were responsible for validation. WTP and MD preformed formal analysis. WTP and MD were responsible for resources. WTP and SQZ were responsible for data curation. WTP and MD wrote the original draft of the manuscript and WTP and SQZ reviewed and edited the manuscript. WTP supervised the present study. WTP was also responsible for project administration. WTP and SQZ confirm the authenticity of all the raw data. All authors read and approved the final manuscript.

Ethics approval and consent to participate

The present study was a simple single case with no patient-identifiable information and therefore did not require ethical approval.

Patient consent for publication

At the time of writing, as the patient is a minor, the present study obtained the informed consent of the parents of the patient.

Competing interests

The authors declare that they have no competing interests.

References

1. Hoeffel CC, Nguyen KQ, Phan HT, Truong NH, Nguyen TS, Tran TT and Fornes P: Fetus in fetu: A case report and literature review. Pediatrics 105: 1335-1344, 2000.
2. Lu T, Ma J and Yang X: A rare case of fetus in fetu in the sacrococcygeal region: CT and MRI findings. BMC Pediatr 21: 575, 2021.
3. Spencer R: Parasitic conjoined twins: External, internal (fetuses in fetu and teratomas) and detached (acardiacs). Clin Anat 14: 428-444, 2001.

4. Basu A, Jagdish S, Iyengar KR and Basu D: Fetus in fetu or differentiated teratomas? Indian J Pathol Microbiol 49: 563-565, 2006.
5. Gerber RE, Kamaya A, Miller SS, Madan A, Cronin DM, Dwyer B, Chueh J, Conner KE and Barth RA: Fetus in fetu: 11 fetoid forms in a single fetus: Review of the literature and imaging. J Ultrasound Med 27: 1381-1387, 2008.
6. Wang L, Long B, Zhou Q and Zeng S: Prenatal diagnosis of a 'living' oropharyngeal fetus in fetu: A case report. BMC Pregnancy Childbirth 19: 453, 2019.
7. Xia B, Li DD, Wei HX, Zhang XX, Li RM and Chen J: Retroperitoneal parasitic fetus: A case report. World J Clin Cases 9: 11482-11486, 2021.
8. Ruffo G, Di Meglio L, Di Meglio L, Sica C, Resta A and Cicatiello R: Fetus-in-fetu: Two case reports. J Matern Fetal Neonatal Med 32: 2812-2819, 2019.
9. Mohta A and Khurana N: Fetus-in-fetu or well-differentiated teratoma- a continued controversy. Indian J Surg 73: 372-374, 2011.