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Case Report

Intra-abdominal fetus in fetu presenting at 31 weeks gestational age

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ABSTRACT

Fetus in fetu (FIF) is a rare anomaly of diamniotic monochorionic twins, in which a malformed fetus resides in its twin's body. This report shows a case with the prenatal diagnosis of FIF at Tu Du hospital. A 23-year-old woman, first-time pregnant, presented at the hospital with an abdominal mass in the fetus at 31 weeks and 4 days of gestation. The ultrasound showed an abnormal mass with the images of calcified features located in the left hypochondriac region and the kidney's upper pole. These images had shapes of skull, femur, spine and became more apparent as the fetus grew. Then, the patient was monitored and delivered at our hospital. The infant was moved to the Children's Hospital 1 and diagnosed with FIF by ultrasound. The surgical resection was performed at 12 days of birth.

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Background

Fetus in fetu (FIF) is a rare anomaly with less than 190 reported cases and mostly diagnosed after birth with the rate of 1/500,000 infants [1]. Ultrasound is the first-line tool in diagnosis FIF. In most cases, the parasitic fetus has neither head nor heart while the spine and limbs still present. FIF is frequently located in retroperitoneum (80%), and due to the similar ultrasound features, it is differentially diagnosed with teratoma. After delivery, computerized tomography (CT) scan and magnetic resonance imaging (MRI) 's indication is usually to minimize the differential diagnosis and to evaluate the patients'

condition before the operation, whereas histopathology helps confirm the diagnosis. Surgical resection is the treatment recommendation for FIF. This defect is often forgotten and misdiagnosed with other congenital anomalies. Hence, we would like to share our experience by doing this case report.

Case report

A 23-year-old patient, G0 P 0-0-0-0, presented at our faculty for 31 weeks and 4 days fetus with an abdominal mass. The ultrasound detected a heterogeneous echogenicity mass, of which size increased with the fetus's age, located in the left hypochondriac region and the kidney's upper pole. This mass figure was 45 × 50 × 42 mm at 31-week and 4-day, 48 × 58 × 50

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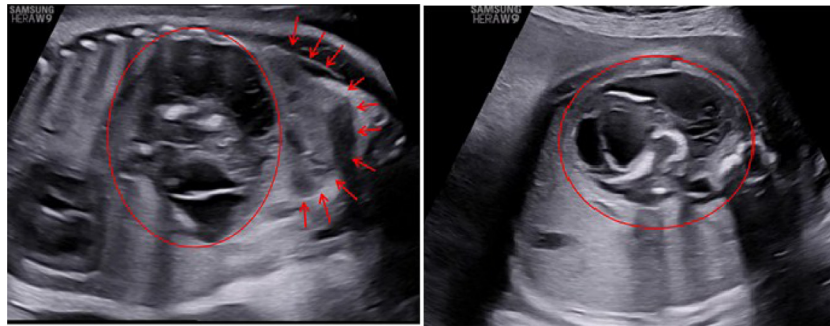


Fig. 1 – At 31-week and 4-day of gestation, on the transverse and sagittal plane, the image indicated a heterogeneous mass located in the left hypochondriac region, below the diaphragm (circle), and pushing the left kidney downwards (arrowhead).

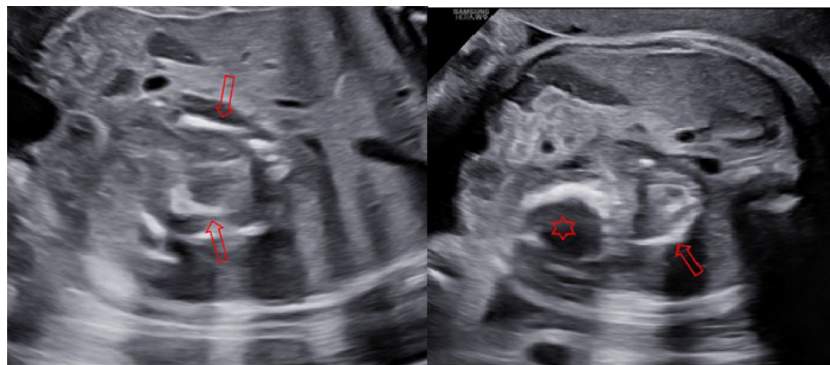


Fig. 2 – At 34-week and 2-day of gestation, on the transverse and sagittal plane, structures with shapes of a skull (asterisk), spine, and femur (arrowhead) were shown.

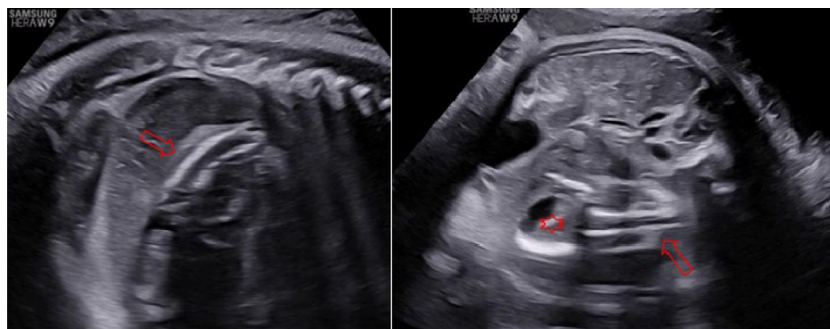


Fig. 3 – At 39 weeks of gestation, on the transverse and sagittal plane, the skull-shaped (asterisk) and spine-shaped (arrowhead) structures became more apparent.

mm at 34-week and 3-day, and $80 \times 74 \times 66$ mm at 39-week. The ultrasound image of calcified features had shapes of skull, femur, spine, and became more apparent as the fetus grew. The color Doppler showed a branch of the fetus's abdominal aorta with the umbilical arterial waveform, which supplied blood to the mass. Prenatal diagnosis: FIF. Because skull-shape and spine-shape images were the typical features of FIF, we did not give other differential diagnoses for this case (Figs. 1–5). At 39 weeks and 2 days of gestation, the patient got cesarean delivery due to pelvic disproportion. After delivery, the infant weighted 3500gr with APGAR: 8-9, and

then it was transferred to Children's hospital 1. The infant's ultrasound image showed a heterogeneous echogenic area ($53 \times 81 \times 75$ mm) with features of the skull, spine, long bones, and adipose tissue, located on the left side of the retroperitoneum (Fig. 6). These structures pushed the left kidney into the pelvic fossa. Preoperative diagnosis: FIF. The resection was performed at 12 days of birth. The mass's gross description was similar to the prenatal and preoperative diagnosis: an incomplete fetus with head, spine, limbs, and ambiguous genitalia.

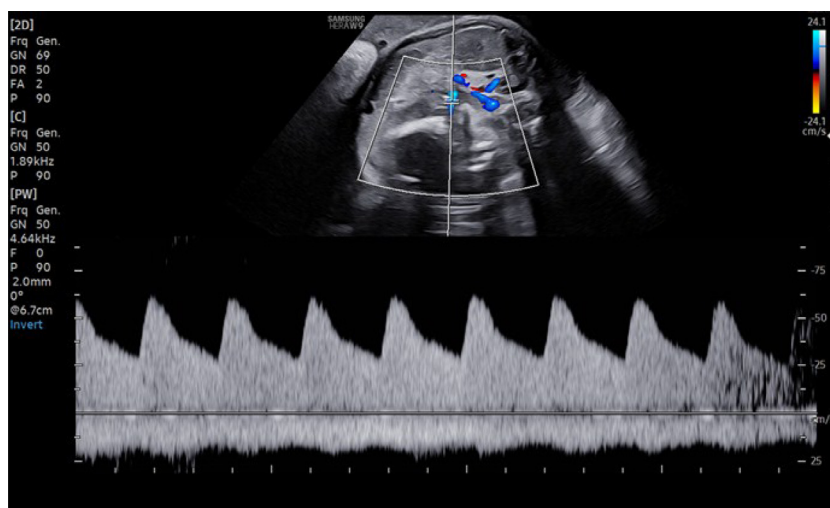


Fig. 4 – The color Doppler showed a vessel branch, originated from the fetus's abdominal aorta, supplied blood to the mass. On pulse-Doppler, it had umbilical arterial waveform.

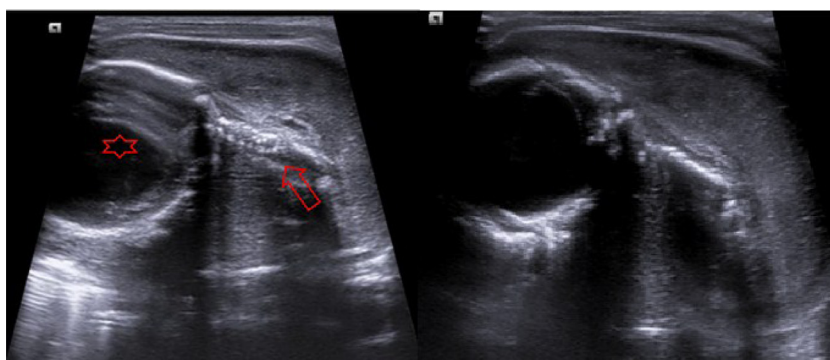


Fig. 5 – On prenatal ultrasound, a mass with skull-shape (asterisk) and spine-shape (arrowhead), located in retroperitoneum and upper the kidney, is shown.

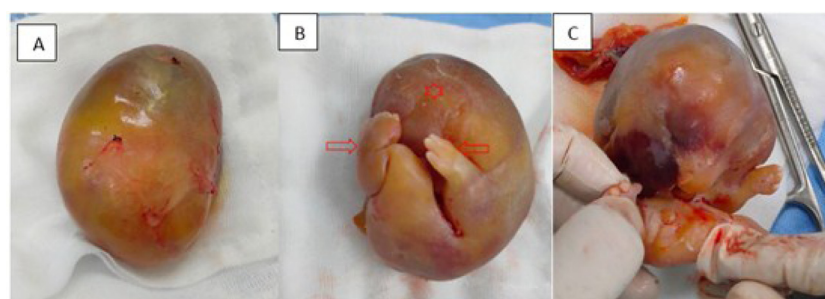


Fig. 6 – The gross description of operative remains at 12 days of birth. (A) An abnormal mass is about 85 x 65 mm in size and covered by an amniotic-like membrane. (B) After dissecting the membrane, there was an immature fetus with a head (asterisk) and limbs (arrowheads). (C) Between the lower limbs, the external genitalia is undefined.

Discussion

FIF, named by Willis and first described by Johann Friedrich Meckel in the 18th century [2], is a rare anomaly of monozygotic twins in which a malformed fetus resides in the body of its partner. The rate among infants is 1/500.000 and mostly diagnosed prenatally [1].

Pathogenesis

There are two hypotheses of the pathogenesis mechanism [3]. The teratoma hypothesis suggests that FIF is a highly differentiated teratoma, and the diamniotic monozygotic twins' hypothesis demonstrates that FIF is a parasitic fetus growing in the body of its twin. Following the latter idea, FIF occurs at a very early stage, during the embryonic folding at the second

and third week of gestation. Due to the unequal division of germ cells in the blastocyst stage, some germ cells remain in the normal embryo. The parasitic fetus is usually covered by a membrane and supplied blood from the host fetus. Though most scientists advocate the theory of monochorionic diamniotic twins, this can not demonstrate odd cases in which multiple fetuses parasitize in 1 host (Multiple fetuses in fetu) or the FIF disease combines with teratoma [4].

Imaging features

Ultrasound is the first-line tool in diagnosing FIF with many merits: safe, easy to perform, and feasible to repeat multiple times. The advancement of ultrasound has enhanced the image quality and accuracy of diagnosis in a shorter time. The prenatal image of FIF shows a fluid-filled mass with floating solids inside. In most cases, the parasitic fetus has neither brain nor heart, while the spine and limbs still present (91% and 82.5%, respectively). Lower limbs are usually more mature than the upper ones. The external genitalia is often found. According to a study by Lindsay among 95 FIF cases, there were 50 male patients (63%), 34 female patients (37%), and 3 patients having ambiguous genitalia. Single parasitic fetus counted 89% of all cases, whereas a unique patient had 11 parasitic fetuses. The most common location of FIF was retroperitoneum (72%), which was significantly more frequent than other positions, such as coccyx, intracranium, chest, mouth, and neck [5].

MRI is required in cases that ultrasound imaging is unclear. Its application minimizes the number of differential diagnoses. This imaging technique is safe for fetuses and allows physicians to accurately assess the mass's location inside the host and its structure, especially in patients with obesity. Besides, MRI's disadvantages are expensive, long waiting, and sometimes unavailable in local hospitals or clinics. The use of MRI is also limited if the fetus moves regularly, and in women who have difficulty in lying down for a long time or fear of closed spaces. CT scan gives more information about the relation between the FIF mass and the surrounded structures, improving the rate of accurate diagnosis and planning for a proper treatment. Due to the radiation exposure, CT scan is commonly indicated antenatally and required the parents' consent [4,5].

Differential diagnosis

A heterogeneous abdominal tumor in the fetus with internal calcification is suspected of teratoma, neuroblastoma, meconium pseudocysts, or FIF. Teratoma usually arises in the lower abdomen, ovaries, sacrococcygeal region, and rarely in retroperitoneum [5]. Fetiform teratoma is different from the FIF by the absence of spine. However, teratoma is much more common than FIF. Pathologically, FIF is made up of well-differentiated vertebra tissue, whereas teratoma is composed of germ cells that yet develop into organs. In general, FIF and mature teratoma have many similar features excepting that mature teratoma has a high risk of malignancy [3].

Congenital neuroblastoma, the most common neoplasm among infants, can be detected at the fetal period, in which

the typical features are tissue mass and microcalcification with or without hemorrhagic area.

In fetal peritonitis and meconium pseudocyst, the primary aetiology is the intestinal perforation during the fetal period, in which the peritoneal mass is calcified and then covered by the omentum and intestine. Overall, the final diagnosis of FIF requires pathologic confirmation.

Treatment and prognosis

Although FIF is mostly a benign disorder, its potential for later malignancy is still controversial. The development of the parasitic fetus can slow the growth of the host, increase the risk of infection, and reduce organs' function. Surgical treatment can restore the normal anatomy and physiology of the host fetus as well as decrease cancerous potential. After the operation, though most reports show positive results, patients should be monitored by imaging techniques and biomarkers, such as Alpha-fetoprotein and Human Chorionic Gonadotropin. A minimum follow-up period of 2 years is necessary to avoid omission of malignancy.

Conclusion

FIF is a rare anomaly of diamniotic monochorionic twins, of which very few cases have been reported, and the majority are diagnosed postnatally. Prenatal ultrasound shows a fluid-filled mass with solid pieces and bone-shaped calcification inside. Image of the spine is the typical sign suggesting FIF. Teratoma is the principal differential diagnosis, which is noticeably more popular than FIF.

Patient consent statement

Written informed consent was obtained from the patient for their anonymized information to be published in this article.

REFERENCES

- [1] Hoeffel CC, Nguyen KQ, Phan HT, Truong NH, Nguyen TS, Tran TT, et al. Fetus in fetu: a case report and literature review. *Pediatrics* 2000;105(6):1335–44.
- [2] Willis RA. *The borderland of embryology and pathology*. Washinton DC: Butterworths; 1962. p. 442–62.
- [3] Narayanasamy JN, Nallusamy MA, Baharuddin ND. Fetus-in-fetu: a pediatric rarity. *J Surg Case Rep* 2014;2014(2):rju001.
- [4] Gilbert-Barness E, Opitz JM, Debich-Spicer D, Mueller T, Arnold SR, Quintero R. Fetus-in-fetu form of monozygotic twinning with retroperitoneal teratoma. *Am J Med Genet A* 2003;120:406–12.
- [5] Prescher LM, Butler WJ, Vachon TA, Henry M, Latendresse T, Ignacio R. Fetus in fetu: Review of the literature over the past 15 years. *J Pediatr Surg Case Rep* 2015;3(12):554–62.