Open Journal of Clinical & Medical Images

www.jclinmedimages.org

Short Report

Open Access, Volume 3

Fetus in fetu: A case report and literature review

Su Chen[1†]; Jie Weng[5†]; Zejun Chen[2]; Haiyang Wang[3]; Yi Ling[4]*

[1]Department of Fetal Medicine and Prenatal Diagnosis Center, the First Affiliated Hospital of Hainan Medical University, Haikou, Hainan, 570011, China.

[2]Department of Hepatic and Infectious Disease, the First Affiliated Hospital of Hainan Medical University, Haikou, Hainan, 570011, China.

[3]Department of Pediatric Surgery, the First Affiliated Hospital of Hainan Medical University, Haikou, Hainan, 570011, China.

[4]Department of Fetal Medicine and Prenatal Diagnosis Center, the First Affiliated Hospital of Hainan Medical University, Haikou, Hainan, 570011, China.

[5]Department of Hepatic Surgery, the First Affiliated Hospital of Hainan Medical University, Haikou, Hainan, 570011, China.

[†]Co-first authors

***Corresponding Author: Yi Ling**

Department of Fetal medicine and Prenatal Diagnosis Center, the First Affiliated Hospital of Hainan Medical University, Haikou, Hainan, 570011, China.

Email: dianalingyi@163.com

Received: Dec 13, 2022

Accepted: Jan 30, 2023

Published: Feb 06, 2023

Archived: www.jclinmedimages.org

Abstract

Fetus in fetu is a very rare condition, with a reported incidence of one in 500,000 live births. It most likely represents a monozygotic twin that implants itself and grows within the body of its normal karyotypically identical sibling. Fetus in fetu is often overlooked in the differential diagnosis of an abdominal mass. The increasing use of routine obstetric ultrasound has identified more cases during the antenatal period. This allows more time for both the neonatology team and the parents to develop a coordinated treatment plan for the anticipated child. We report a case of a fetus in fetu which was first seen as a complex cyst-solidary mass during antenatal ultrasound at 22 weeks' estimated gestational age and was confirmed by subsequent imaging and surgical treatment shortly after birth.

Keywords: Fetus in fetu; Prenatal diagnosis; Teratoma; Aberrant fetus.

Introduction

Fetus in fetu is an extremely rare condition of abnormal twinning during embryogenesis, with a reported incidence of one in 500,000 live births [1]. The increasing use of routine obstetric ultrasound has identified more cases during antenatal period. This will facilitate a planned birth of the anticipated child and immediate pediatric input. We report a case of a fetus in fetu which was first detected at 22 weeks' gestation and was confirmed by subsequent imaging and surgical treatment shortly after birth.

Case report

A 28-year-old pregnant woman, gravida 2 para 1 abortion 1, presented to our department with an abnormal ultrasound scan at 22 weeks' gestation that revealed a fetal abdominal mass. Initially this was thought to be a teratoma. Family history was negative for congenital malformations, and there was no history of medication and drug use during pregnancy. Upon detailed sonography scanning in our institution a 25 mm in diameter heterogeneous abdominal mass with an internal density mass was visualized. The mass was well circumscribed and power dop-

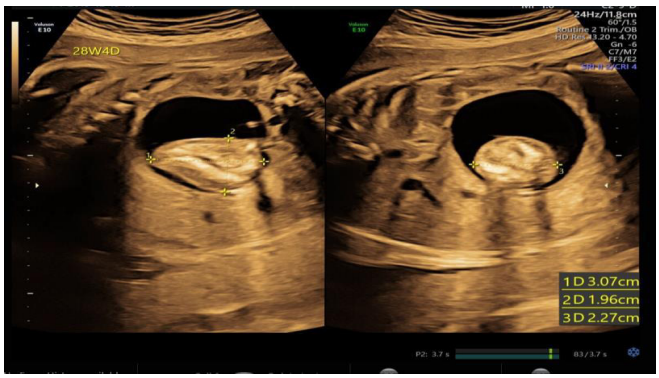

Figure 1: Prenatal ultrasound examination of the FIF at 26 weeks' gestation (left) and 22 weeks' gestation (right).

Figure 2: Specimen manifestations after FIF resection. The lower part of body with a limb and an umbilical cord could be seen.

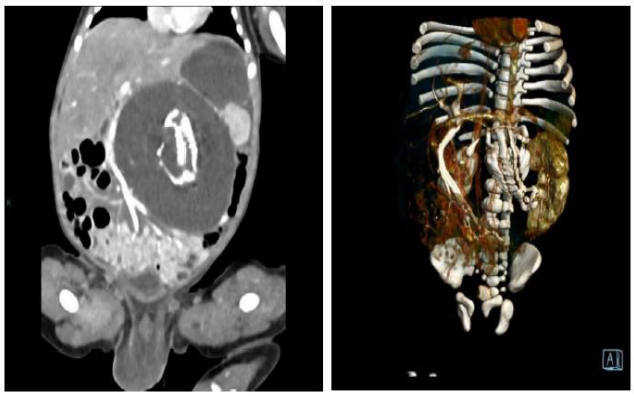

Figure 3: MRI images of newborn after birth. A vertebral axis can be seen in the cavum abdominis of the infant.

pler investigation revealed normal blood flow. Monthly repeat studies showed the mass steadily enlarging. A formal follow-up ultrasound at 26 weeks gestation demonstrated that the mass was 38 mm in diameter with rudimentary spine, ribs, ilium and a limb. A subsequent magnetic resonance imaging verified the heterogenous mass with multiple bones and vertebral body. At this stage an endoparasitic FIF was diagnosed. Amniocentesis that performed at 23 weeks' gestation demonstrated a normal karyotype and testing of maternal alpha-fetoprotein levels was not performed. The boy was born full term by normal vaginal delivery with a birth weight of 3080 g. He had Apgar scores of 10 and 10 at 1 and 5 min, respectively. At laparotomy shortly after birth an encapsulated retroperitoneal mass was resected. Gross anatomy and pathological examination demonstrated a fetus in fetu, as well as DNA fingerprinting techniques confirming identical genetic material shared by the child and the mass. From the gross anatomy the lower part of body with a limb and an umbilical cord could be seen. The baby did well postoperatively.

Discussion

Fetus in fetu is a rare entity that describes a diamniotic, monochorionic, monozygotic twin which becomes internalized due to the anastamosis of vitillene circulation [2]. In the 19th century, Meckel first described FIF as a rare phenomenon in which a fetus of deformed twins was parasitized in the abdominal cavity of its healthy sibling [3]. The number of FIFs is usually single but multiples have been reported. In 1935 Willis distinguished a FIF from a teratoma by the presence of an axial skeleton with limbs and organs arranged about this axis [4]. More recently Gonzalez-Crussi further defined FIF as, 'any structure in which fetal form is in a very high development of organogenesis' and linked it 'to the presence of a vertebral axis' [5]. This reflects Kim's reverse definition of a teratoma as 'an accumulation of pluripotential cells in which there is neither organogenesis nor vertebral segmentation' [6]. As reported in literature most of FIF are benign and however 10% of teratoma are malignant. Mao X et al reported seven infants with FIF diagnosed in their hospital that none had diseases recurrence[7]. There is still the possibility of malignant transformation of FIF. Hopkins et al reported that malignant transformation occurred after FIF resection. The pathological diagnosis was recurrence of an endodermal sinus tumor after FIF [8]. FIF most tethered to the retroperitoneum as in our case. A review of 87 reported cases of FIF concluded that 91% of cases had a vertebral column, 82.5% had limbs, 55% had a central nervous system, 45% had a gastrointestinal tract and the lower limbs were more developed than the upper limbs [9]. The most controversial aspect of FIF is the etiology. It remains unclear what triggers the actual arrest in growth Tand further differentiation. Some theories focus on host factors or nutrient supply being size limitations. Some believe the parasitic twin will continue to grow if enough vascularization is present. Others concentrate on innate factors or even regression of development.

Conclusion

In conclusion, with advances in antenatal sonography the diagnosis of FIF can be made more confidently in the antenatal period. This will facilitate a planned birth of the child and immediate pediatric input. Following confirmation of the diagnosis in the neonate, complete surgical excision should be performed and the excised FIF sent for detailed pathologic investigation including genetic testing. Although most of FIFs are benign, the possibility of malignant recurrence should be kept in mind. That means long term follow up monitoring should be need.

Acknowledgements: This work was supported by Epigenetic regulation of placental growth factor gene and regulation of HIF-1α in the molecular mechanism of the pathogenesis of fetal growth restriction, Natural Science Foundation of Hainan Province 2022 (Project No. 822RC825) and Expression and significance of different transcripts of placental growth factor gene in selective fetal growth restriction, Health scientific Research Project of Hainan Province 2021 (Project No. 21A200063).

References

1. Grant P, Pearn J. Foetus-in-foetu. The Medical journal of Australia. 1969; 1: 1016-1019.
2. Chi J G, Lee Y S, Park Y S, et al. Fetus-in-fetu: report of a case. Am J Clin Pathol. 1984; 82: 115-119.
3. Lee E. Fetus in Fetu. Arch Dis Child. 1965; 689-693.
4. Willis R. The structure of teratoma. J Pathol Bacteriol. 1935; 40: 1-36.
5. Gonzalez-Crussi F. Extragonadal teratomas. Atlas of Tumor PathologyWashington DC: Armed Forces Institute of Pathology, 1. 1982.
6. Kim O H, Shinn K S. Postnatal growth of fetus-in-fetu. Pediatr Radiol. 1993; 23: 411-2.
7. Mao X, Cheng L, Lin S, et al. Rare Fetus-in-Fetu: Experience From a Large Tertiary Pediatric Referral Center. Front Pediatr. 2021; 9: 678479.
8. Hopkins KL, Dickson PK, Ball TI, et al. Fetus-in-fetu with malignant recurrence. J Pediatr Surg. 1997; 32: 1476-1479.
9. Hoeffel CC, Nguyen KQ, Phan HT, et al. Fetus in fetu: a case report and literature review. Pediatrics. 2000; 105: 1335-1344.