Annals of Case Reports & Reviews

Case Report

doi: 10.39127/2574-5747/ACRR:1000227
Mohamed R, et al. Annal Cas Rep Rev: ACRR-227

# Fetus-In-Fetu Diagnosed Ante-Natally Using Fetal 2D Ultrasound and Successfully Excised Post-Natally. A Case Report

***(Running title: Fetus-in-fetu diagnosis and excision)***

**Rehab Mohamed F Mahgoub[1], Amr H Farag[1*], Mohamed H Nasr El-din[1]**

[1]Department of Obstetrics and Gynecology, Ain Shams University

[*]**Corresponding author:** Amr Hassaan Farag, Assistant Professor of Obstetrics and Gynaecology, Ain Shams University Maternity Hospital, Abbasiya, Cairo, Egypt and Consultant Obstetrician and Gynaecologist, Walsall Healthcare NHS Trust, UK. Tel: 00447383985465; Email: amr.farag1@nhs.net

**Citation:** Mohamed R, Farag AH, El-deen MHN (2021) Fetus-In-Fetu Diagnosed Ante-Natally Using Fetal 2D Ultrasound and Successfully Excised Post-Natally. A Case Report. Annal Cas Rep Rev: ACRR-227.

**Received Date:** 09 April 2021; **Accepted Date:** 14 April 2021; **Published Date:** 20 April 2021

***Summary***

*Fetus-in-fetu is a rare congenital anomaly in which malformed fetus grows within the body of its twin. We present a case with a rare condition of fetus-in-fetu suspected by antenatal 2D ultrasound scan at 36 weeks gestation. The baby was delivered by cesarean section followed by elective laparotomy that revealed a well-encapsulated retroperitoneal mass which was successfully excised. We demonstrate that fetus-in-fetu should be differentiated from teratoma with complete excision being curative and confirmatory of the diagnosis.*

***Keywords:*** *Antenatal, Fetus-in-fetu.*

## Introduction

The rare fetus-in-fetu (FIF) condition is usually retroperitoneal or intraperitoneal but may also be present in scrotum or in cranial cavity [1]. Several theories have been postulated regarding the pathogenesis with the 'included twin theory' suggesting an aberration of monochorionic diamniotic monozygotic twinning in which unequal division of the totipotent inner cell mass of the developing blastocyst leads to the inclusion of smaller cell mass within a maturing sister embryo due to persistent anastomosis of vitelline circulation [2].

Another theory considers FIF as a highly differentiated mature teratoma [3], while some literature suggests it can be differentiated from teratoma by the presence of vertebral organization with limb buds and other organ systems. Also, FIF is a benign disorder whereas the teratoma bears malignant potential [4].

The present report is a rare case FIF diagnosed ante-natally using 2D ultrasound and Doppler at 36 weeks gestation followed by successful surgical excision.

## Case Presentation

A 26-year-old primigravid lady was referred to Fetal Medicine Unit, Ain Shams University Maternity Hospital, Egypt, at 36 weeks gestation for evaluation of fetal intraabdominal cystic mass. A 2D ultrasound scan with Doppler showed a viable intrauterine pregnancy with normal amniotic fluid volume and normal fetal movements with cephalic presentation. Within the fetal abdominal cavity, there was a retro-peritoneal complex heterogenous mass containing cystic areas and measuring 9.5 x 6 x 9 cm (Figure 1). The mass was sited below the diaphragm and related to the right lobe of the liver.

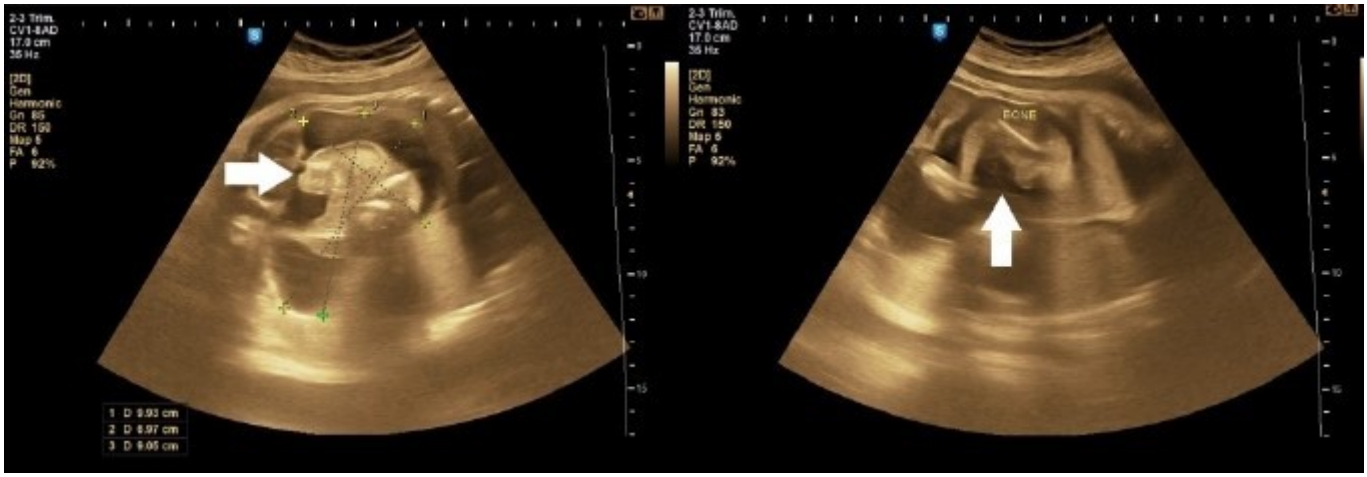


**Figure 1:** 2D sonography images of fetal abdomen at 36 weeks gestation showing a fetal retroperitoneal abdominal complex mass showing heterogeneity with presence of cystic areas below the diaphragm and related to the right lobe of the liver.

A local multidisciplinary team (MDT) consisting of a consultant obstetrician, a consultant radiologist, a consultant neonatologist and a consultant paediatric surgeon have made the diagnosis of a retro-peritoneal FIF and were all involved in planning the woman's care and delivery. The recommendation was delivery in a tertiary center with a setup for urgent neonatal surgery and neonatal intensive care unit (NNICU) admission. The woman and her husband were fully informed and counselled and an agreement has been achieved.

At 39 weeks gestation the patient was delivered by elective Caesarean section and the baby had a nasogastric tube inserted for feeding. The neonate showed clinical stability and the parents were counselled for surgery. Elective laparotomy was performed at 10 days following delivery, where a large retro-peritoneal cystic mass was found displacing the urinary bladder anteriorly and the gut laterally. Dilated ureters were present on postero-lateral aspect of the mass. The urinary bladder was dissected free from the mass and the cystic component of the mass was decompressed and the mass composing the parasitic fetus with its membrane was removed after ligation and sectioning of its umbilical cord (Figure 2).

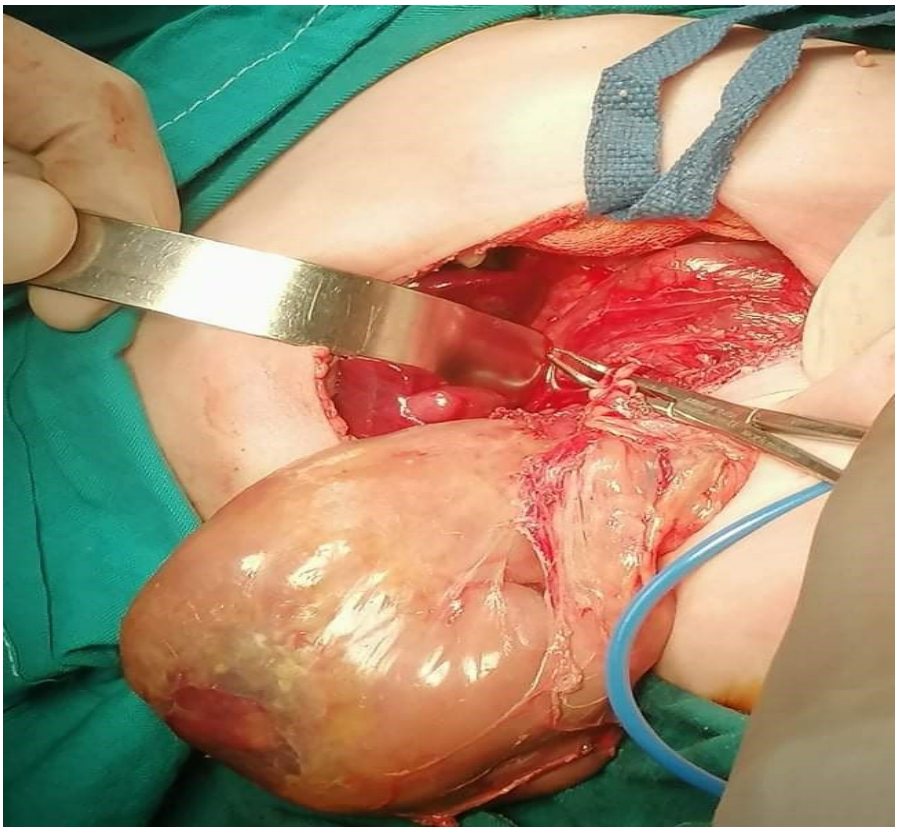

**Figure 2:** A medical photograph at laparotomy showing cystic component of the mass surrounded by membrane covering the parasitic fetus.

Macroscopic Examination of the excised mass revealed a soft tissue resembling a fetus with anencephaly and covered entirely with intact skin. Vertebral column was palpable with the presence of bilateral, asymmetrical and well-developed lower limbs with structures resembling toes. Rudimentary upper limbs were also present (Figure 3).

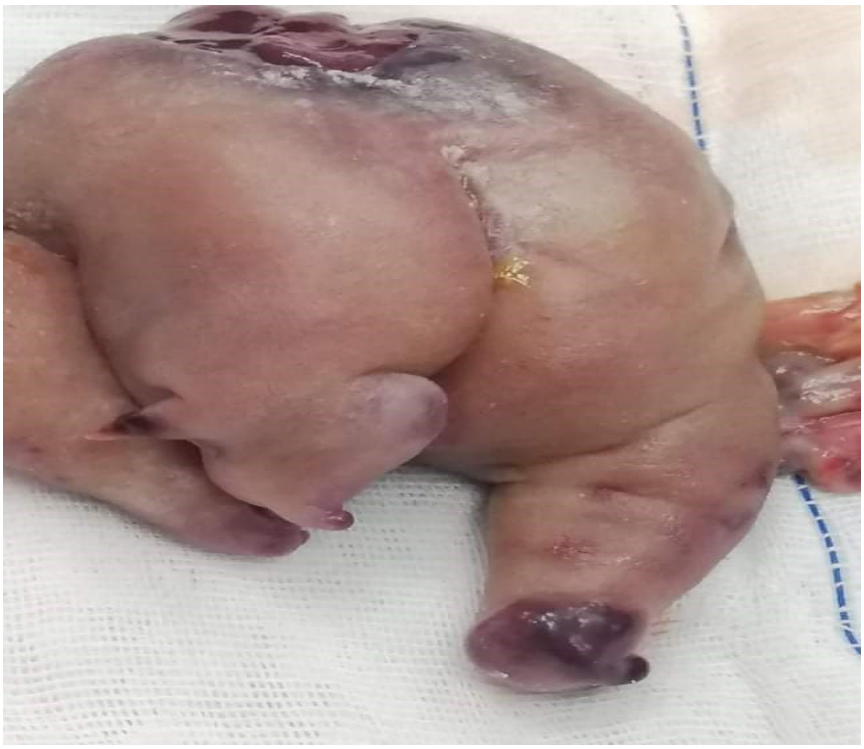

**Figure 3:** A medical photograph of the excised parasitic fetus showing well-developed lower limbs with structures resembling toes.

## Discussion

Fetus-in-fetu is a rare condition occurring once in 500,000 deliveries [5] and less than 200 cases reported with the first case reported by George William Young in 1808 [6]. FIF usually presents as a lump in the abdomen (70%) and the retro-peritoneal space is the commonest site (80%). Other rare sites include sacro-coccygeal region, intracranial, thorax, pelvis and the scrotum [7]. Thakral et al. [8] reported equal male and female predisposition but Federici et al. [9] noted 2:1 male predominance.

Different organs can be seen in FIF including vertebral column (91%), limbs (82.5%), CNS (55.8%), GIT (45%), vessels (40%), and genitourinary (26.5%) [10]. Visualization of the vertebral column and limbs on imaging, gross, and histopathological examination confirms the diagnosis. This fulfills the 'Willis criteria' which stresses much emphasis on the development of axial skeleton and vertebral axis [11].

According to Brand et al., diagnosing FIF, requires at least one of the following characteristics to be present; a mass enclosed within a distinct sac, partially or completed covered by skin, grossly recognizable anatomic features and attached to the host by a pedicle containing a few relatively large blood vessels [12]. In most cases, there is a single parasitic twin but rarely, more than one parasitic twin is observed in the host body [13].

Infants with FIF commonly present with abdominal mass. However, symptoms mainly relate to its mass effect and include abdominal distension, feeding difficulty, emesis, jaundice, pressure effects on renal system and dyspnea [8,10]. Pre-operative diagnosis is possible by pre- or post-natal ultrasonography. Plain abdominal X-ray may be helpful in diagnosis. In half of the cases, X-ray shows the vertebral column and axial skeleton [14], and the presence of a bony vertebral axis with appropriate limb arrangement on gross examination is an important diagnostic feature which was observed in the studied case, thus confirming the diagnosis of FIF [12]. Other differentiating features include normal levels of alfa feto-protein and molecular analysis using an informative genetic marker, for uni-parental isodisomy of chromosomes 14 and 15 that must show no genetic difference between the host and the fetiform mass to confirm the diagnosis of FIF [9].

It is important to differentiate between teratoma and FIF. Teratoma is defined as disorganized array of pluripotent cells with 10% malignancy rate and representing all three germ layers that, unlike FIF, does not develop past the primitive streak stage (days 12 to 15) and demonstrates no organogenesis or vertebral segmentation. In contrast, FIF is always benign and can be clinically differentiated from teratoma by the presence of vertebral bodies and limbs [15].

Fetus-in-fetu derives its blood supply from the rich vascular plexus around the cyst wall. Vascular pedicle is rare and is usually observed in large growing masses with delayed

presentation [9] and surgical excision is the treatment of choice in both fetiform teratoma and FIF where complete excision of the mass with its surrounding membrane gives complete cure [8]. Surgical expertise and careful dissection is crucial to avoid injury to surrounding structures. In our case, the mass was carefully and fully resected with no subsequent complications to the best of our knowledge. Post-operatively the patient was followed up with tumor markers including alpha-fetoprotein and beta-hCG that returned back to normal levels.

Fetus-in-fetu is a rare interesting condition that usually presents during childhood and commonly presenting with intraabdominal mass and rarely seen ante-natally as in this case. It should be differentiated from teratoma with complete excision being curative and confirmatory of the diagnosis.

## Declarations

**Funding:** None.

**Conflicts of interest/Competing interests:** The authors declare that there is no conflict of interest regarding the publication of this article.

**Ethics approval:** Ethical approval was waived at Ain Shams University as it is not an institutional requirement for publishing an anonymous case report.

**Consent to participate:** Not applicable.

**Consent for publication:** A written informed consent to publish this case was obtained from the patient.

**Availability of data and material (data transparency):** Data material is available on request.

**Code availability:** Not applicable.

**Authors' contributions:**

**R M F Mahgoub:** Data collection, performed imaging and reviewed images.
**A H Farag:** Manuscript editing and correspondence
**M H Nasr El-din:** Has managed the case obstetrically and planned care and delivery.

## References

1. Suttiwan P, Sutthiwan I, Tree-traken T. Fetus-in-fetu. J Pediatr Surg. 1983;8(3):290-292.
2. Spencer R. Parasitic conjoined twins: external, internal (Fetuses in fetus-in-fetu and teratomas), and detached (acardiacs). Clin Anat. 2001;14(6):428-444.
3. Gangopadhyay AN, Srivastava A, Srivastava P, et al. Twin fetus-in-fetu in a child: a case report and review of the literature. J Med Case Reps. 2010; 4:96.
4. Hopkins KL, Dickson PK, Ball TI, O'Shea PA, Abramowsky CR. Fetus-in-fetu with malignant recurrence. J Pediatr Surg. 1997; 32:1476-1479.
5. Grant P, Pearn JH. Foetus in foetu, Med J. Aust. 1969;1:1016-1020.
6. Young GW. Case of fetus found in the abdomen of a boy. Med Chir Trans. 1809; 1:234.
7. Nagar A, Raut AA, Narlawar RS. Foetus in fetu. J Postgrad Med. 2002; 48:133-134.
8. Thakral CL, Maji DC, Sajwani MJ. Fetus-in-fetu: a case report and review of the literature. J Pediatr Surg. 1998;33(9):1432-1434.
9. Federici S, Ceccarelli PL, Ferrari M, et al. Fetus-in-fetu: report of three cases and review of literature. Pediatr Surg Int. 1991; 6:60-65.
10. Hoeffel CC, Nguyen KQ, Phan HT. Fetus-in-fetu: a case report and literature review. Pediatrics. 2000;105(6):1335-1344.
11. Willis RA. The borderland of embryology and pathology 2nd ed. Washington DC: Butterworth, 1962; 442-462.
12. Brand A, Alves MC, Saraiva C, et al. Fetus-in-fetu-diagnostic criteria and differential diagnosis–a case report and literature review. J Pediatr Surg. 2004;39(4):616-618.
13. Kimmel DL, Moyer EK, Peale AR, Winborne LW, Gotwals JE. A cerebral tumor containing five human fetuses. A case of fetus-in-fetus-in-fetu. Anat Rec. 1950;106(2):141-158.
14. Khalifa NM, Maximous DW, ABD-Elsayed AA. Fetus-in-fetu: a case report. J Med Case Reports. 2008;10(2):2.
15. Nocera RM, Davis M, Hayden CK, Schwartz M, Swischuk LE. Fetus-in-fetu. Am J Roentgenol. 1982;138(4):762-764.