# Fetus-in Fetu in an Infant – A Ccase Report.

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**How to cite this article:**
Singh S, Vaishnavi M, Koshyari H, Naaz A. Fetus-in fetu in an Infant – A Case Report. Iranian Journal of Pediatric Surgery 2025; 11(2): 285 – 293.

**DOI:** https://doi.org/10.22037/irjps.v11i2.47949

## Abstract

Fetus in fetu (FIF) is a rare pathological condition, mostly presenting as a fetiform, calcified abdominal mass in its living host which is usually a fetus or a newborn. Due to its rarity with an incidence of 1:500000, FIF is often missed in the differential diagnosis of an intra-abdominal mass in a newborn.

**received:** 5 April 2025
**accepted:** 19 July 2025
**Published online:** Nov 2025

## Abstract

FIF should also be differentiated from a teratoma because of the malignant potential of the latter. The presence of vertebral segments and an encapsulating cyst can reliably distinguish FIF from teratoma. Condition can be diagnosed in-utero as well as preoperatively in a new born by imaging. Surgical excision is curative. Recurrence is extremely unusual.

An 8-month-old, full-term, male infant was brought with gradually increasing abdominal distension. Examination revealed a distended abdomen with a non-tender, irregular, firm mass in the right flank. Initial US imaging revealed the presence of a large foetoid tumor - like mass which was later confirmed as FIF on CT scan. There was evidence of a vertebral column along with bony appendages and formed organs. A laparotomy was done where sac was found to be loosely attached to the posterior abdominal wall. The entire mass was successfully removed. Baby made good recovery . At the end of six months of follow-up, he is doing well with no new symptoms.

## Keywords

- Parasitic twin
- Teratoma
- Newborn

## Introduction

Fetus-in-fetu (FIF) is an extremely rare developmental abnormality where a malformed, vertebrate fetus-like mass develops within the body of the other twin, the host. It was first described by Johann Friedrich Meckel in the late 18th century. Majority of cases have been described in neonates and children and only seven cases have been reported after the age of 15 years.

Two main hypotheses on the pathogenesis of FIF have been described. The "included-twin" or "parasitic-twin" theory suggests that FIF arises from an anomalous monozygotic monochorionic diamniotic twin pregnancy. The malformed fetus becomes entrapped within its twin while dependent on its blood supply via persistent anastomosis of vitelline circulation.[1-2] Alternative theory, the "teratoma-

spectrum" theory, suggests that FIF arises as a highly differentiated mature fetiform teratoma. Teratoma is a type of germ cell neoplasm consisting of at least two of three germ layers, i.e., endoderm, mesoderm, and ectoderm. FIF usually presents as a localized swelling that may compress adjacent structures due to the growing mass effect.

Due to its rarity, FIF is often missed in the differential diagnosis of an intra-abdominal mass in a new born or infant. It mainly occurs in an intra-abdominal location although there are reports of FIF in other locations as well. There are embryo-pathogenetic debates over whether it belongs to the spectrum of highly differentiated teratomas or is a parasitic twinning from a monozygotic, monochorionic, diamniotic pregnancy. The presence of vertebral segments and an encapsulating cyst can reliably distinguish FIF from teratoma. Condition can be diagnosed in-utero as well as preoperatively in a new born by imaging. Surgical excision is curative. Recurrence is extremely unusual.

**Case presentation**

An 8-month-old male baby presented with gradually increasing abdominal distension noticed by the mother. Baby was the product of a non-consanguineous marriage, delivered at 39-weeks by lower-segment caesarean section. Apgar score at 1- and 5-minutes was 8 and 9 respectively. Antenatal history was not significant. There was no history of any addiction, any drug intake other than the prescribed medications, or radiation exposure. His birth weight was 2800 grams. Post-natal period was uneventful with no history of any NICU stay. The complete neonatal examination was remarkable for a distended abdomen with a large, palpable, irregular, midline abdominal mass. Rest of the examination was unremarkable. Laboratory investigation revealed serum α-fetoprotein levels of 8.2 ng/ml which was normal for the age. Routine haematological investigations were within normal limits.

Plain x-ray abdomen **(Figure 1)** showed well-formed long limb bones in a soft tissue density which was displacing the bowel loops supero-laterally. Initial US imaging revealed the presence of a large foetoid tumor - like mass which was later confirmed as FIF on CT scan.


**Figure 1:** showing axial skeleton within a soft-tissue density displacing the bowel superiorly.

There was evidence of a vertebral column along with appendicular bones and formed organs. An elective laparotomy was planned. There was a complete sac with the fetoid mass within. It showed a malformed head and an ill-defined trunk with aborted limbs **(Figure 2)**.

**Figure 2:** showing malformed head, trunk and aborted limb

This was loosely attached to the posterior abdominal wall with flimsy adhesions. There were no well-defined feeding vessels. The entire mass was removed in-toto. Baby made a good recovery. Feeding was started on post-operative day 2 and was completely resumed by day 4. There was no morbidity observed. He was discharged on 5$^{th}$ post-operative day. Histopathological examination of the excised mass confirmed it to be a malformed fetus. At six-month follow-up, he is asymptomatic and thriving well.

## Discussion

FIF is a rare developmental anomaly with an incidence of less than one in 500,000 births with a slight male preponderance (2:1).[3] Despite the two pathogenetic controversies on whether FIF represents a distinct pathogenetic entity (the parasitic-twin theory) or is a part of the fetiform teratoma spectrum (the teratoma-spectrum theory), distinguishing FIF from teratoma is extremely important. That is because even mature cystic teratoma carries a risk of malignancy from 3.5% to as high as 6.67%, while immature teratoma should be regarded as malignant.[4-5] In contrast, there are only two known cases of malignant recurrences in the case of FIF.[6-7] The fetiform mass in FIF is genetically identical to its host body and is usually single, although the case of multiple FIF masses in a single host has also been reported.[8]

Two main hypotheses on the pathogenesis of FIF have been described. The "included-twin" or "parasitic-twin" theory suggests that FIF arises from an anomalous monozygotic monochorionic diamniotic twin pregnancy. The malformed fetus becomes entrapped within its twin while dependent on its blood supply via persistent anastomosis of vitelline circulation.[1-2] Alternative theory, the "teratoma-spectrum" theory, suggests that FIF arises as a highly differentiated mature fetiform teratoma. Teratoma is a type of germ cell neoplasm consisting of at least two of three germ layers, i.e., endoderm, mesoderm, and ectoderm. FIF usually presents as a localized swelling that may compress adjacent structures due to the growing mass effect.

Most cases of FIF occur retroperitoneally in the abdominal region (80%). This may be explained by the fact that vitelline circulation embryologically develops into the superior mesenteric artery that is located retroperitoneally. However, its occurrence in other areas has also been reported, including intracranial[9], intrahepatic[10], intrathoracic[11], oral cavity[12], sacrococcygeal[13], and even in an undescended testis.[14]

The diagnostic findings favouring FIF over teratoma include the presence of an axial skeleton and limb buds on imaging modalities. Willis et al. regarded the presence of a vertebral column as one of the important diagnostic criteria of FIF.[15] The presence of vertebrae indicates an organized embryological development that has developed notochord, the precursor of vertebral bones, during the primitive streak

stage. In contrast, teratoma develops as a result of disorganized and uncontrolled pluripotent cell replication, thus vertebral segmentation and organogenesis are not usually found.[1,16] In the neonate, other differential diagnoses should include meconium pseudocyst, which is the result of meconium peritonitis caused by prenatal bowel perforation. In females of reproductive age, ectopic pregnancy should also be considered as a potential differential diagnosis of the fetiform mass.[17-18]

Spencer et al. have proposed that the diagnosis of FIF requires at least one of the following conditions met[2]: (a) it is enclosed within a distinct cyst; (b) it is partially or entirely covered in normal skin; (c) it has anatomical structures that are grossly recognizable; (d) it is connected to the host by a small number of relatively large blood vessels; and (e) it is either positioned immediately next to one of the sites where conjoined twins attached, or be connected to the neural tube or the gastrointestinal system.

## Conclusion

In conclusion, FIF should be considered as a differential diagnosis in a new born or infant who presents with an encysted fetiform mass[18]. Early diagnosis is made more frequently nowadays due to routine antenatal imaging. In doubtful cases, further imaging modalities such as CT or MRI can reliably differentiate FIF from teratoma by the presence of the encapsulating cyst, vertebral organization, and limb bones in the fetiform mass. The condition should also be differentiated from ectopic pregnancy in adult females and meconium pseudocyst in neonates. Despite its benign nature, post-operative follow-up with serum AFP and imaging modalities may be necessary.

Prognosis after complete excision is excellent.

## Ethical Consideration

This issue has been raised and approved by the research committee of Himalayan Institute of Medical Sciences, Dehradun, India.

(Ref.No:SRHU/HIMS/RC/2025/162)

## Acknowledgment

Not applicable

## Funding/Support

Not applicable

## Conflict of interests

There is no conflict of interest

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