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Case Report

# Two Cases of a Rare Congenital Anomaly: Fetus in Fetu

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## ABSTRACT

Fetus in fetu is a rare anomaly seen in 1 in 500,000 births. It presents as a retroperitoneal mass which may remain unnoticed till later age. We present two interesting cases of fetus in fetu (FIF), its diagnosis, treatment, and histological findings. The first case was ten-month-old male baby who presented with symptoms and was diagnosed pre-operatively as FIF, later confirmed by postoperative radiograph and histology. The second case was six-month-old female child diagnosed as having a mass suggestive of teratoma in retroperitoneum and the diagnosis of FIF was made intra and postoperatively. We conclude that FIF is a benign retroperitoneal mass having varied presentation and may be confused with teratoma. Advances in diagnostic modalities have made it possible to diagnose this condition prenatally. Surgical excision is curative with good outcome. Histology can confirm the diagnosis as it is important to differentiate FIF from teratoma which has malignant potential.

**KEYWORDS:** Fetus In Fetu, Teratoma, Congenital Anomaly, Retroperitoneal Mass, Histology.

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## INTRODUCTION

Fetus in fetu (FIF) is a rare anomaly seen in 1 in 500,000 births. The entity was first described by Meckel in 18th century. Though the exact pathogenesis is unclear, inclusion of diamniotic monochorionic twin within the host is said to be a probable theory as also the unequal division of a totipotent cell of inner cell mass [1,2]. It presents as a retroperitoneal mass which may remain unnoticed till later age [1,3]. It may also be confused with mature teratomas [4]. Here we present two cases of FIF and discuss its diagnosis, treatment, and histological findings.

**Case History:**

**Case 1:** A ten-month-old male patient presented with constipation, straining, and crying during defecation for last 3 months. There was no significant birth history or past history. The child was on breast feeds and top feeds. There was no distension of abdomen, palpation revealed a single mass in hypogastric and umbilical regions measuring 12 x 12 cm. The edges were well defined, and the mass had a smooth surface and solid consistency with dull note on percussion. On per-rectal

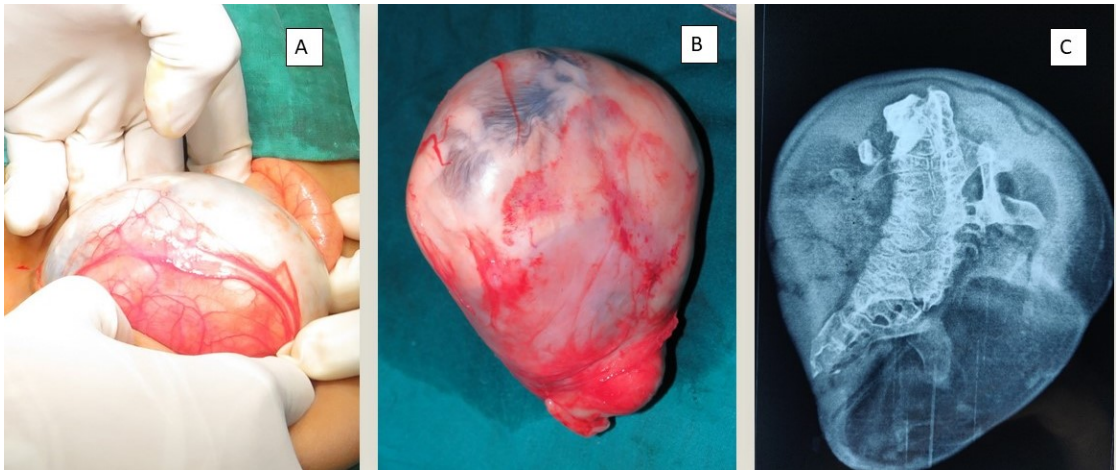



**Fig. 1:** Case 1- A. Intraoperative well encapsulated mass, B. Excised mass – Showing hair at one end. C. Plain Radiograph showing the vertebral axis.

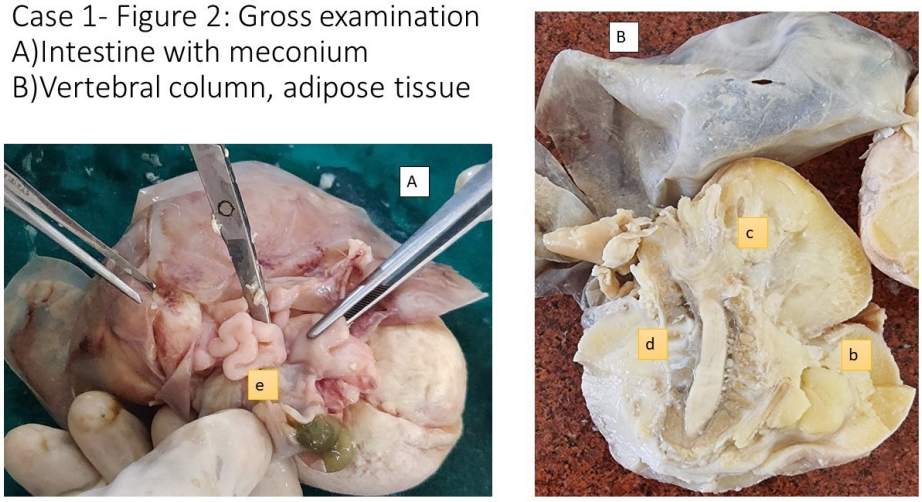



**Fig. 2:** Case 1- Gross examination A. Intestine with meconium B. Vertebral column, adipose tissue.

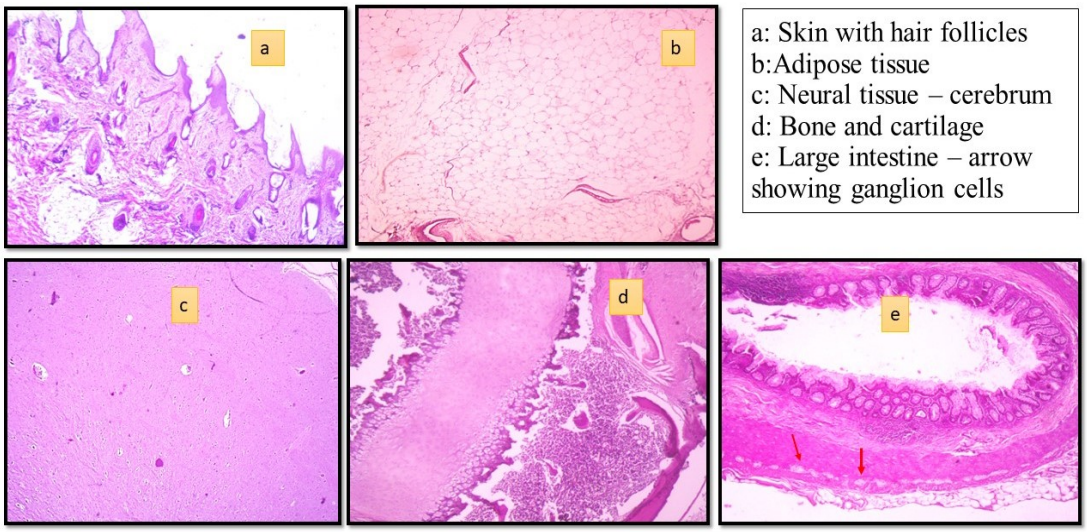



**Fig. 3:** Case 1- Microscopic view of the fetal parts.

examination a solid mass was palpable one finger deep on anterior wall of rectum and was fixed. Rectum was loaded with hard stools.

X-Ray showed a soft tissue mass in abdomen, displacing bowel loops. The mass showed foetal skeletal structure. Ultrasound showed heteroechoic lesion 9x8.5 cm in the pelvis with dense calcification. CT abdomen showed large pelvi-abdominal well encapsulated mass with mixed fat, soft tissue and bone densities suggestive of teratoma or foetus in fetu. Laparotomy was done and the retroperitoneal

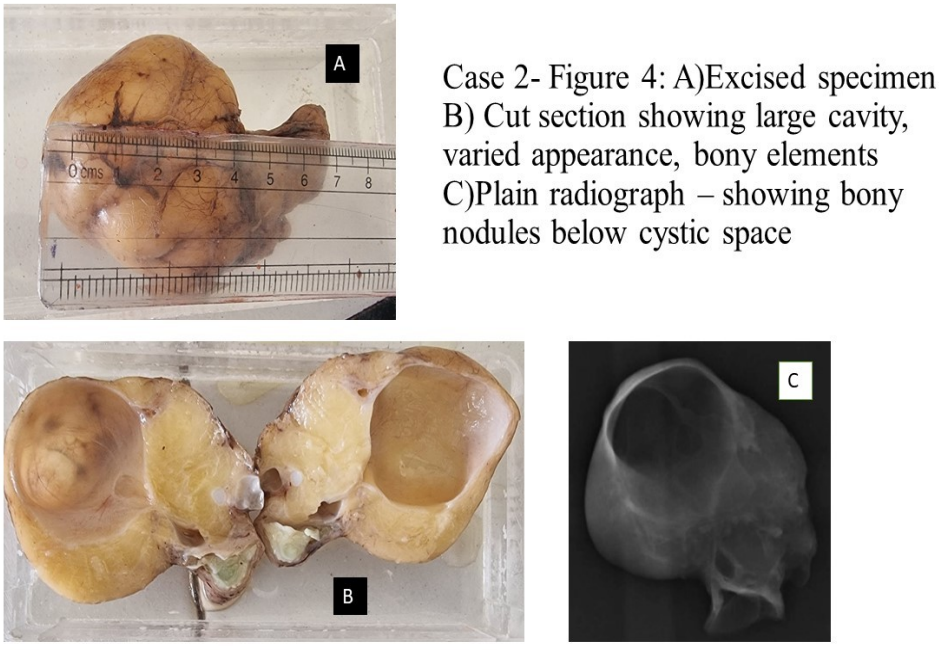
mass was excised in toto. (Figure 1)

Post- operative radiograph of the mass showed a well-defined vertebral axis as shown in Figure 1. On gross examination the excised mass measured 9x8x4 cm, was encapsulated, ovoid and showed hair at the broader end. (Figure 1) On sectioning the mass there were cystic spaces, vertebral column, bone and adipose tissue along with loops of intestines clearly differentiated, which showed meconium inside the lumen. (Figure 2) The Haematoxylin and eosin-stained slides confirmed presence of skin with hair follicles, adipose tissue, nervous tissue showing histological appearance of cerebrum with pyramidal cells, cartilage, bone with osteoid formation and large intestine showing simple columnar lining epithelium with abundant goblet cells and intermuscular mature ganglion cell clusters. (Figure 3)

**Case 2:** A six-month-old female patient was incidentally diagnosed by the paediatrician with a vague lump in abdomen when the child had visited the paediatrician for diarrhoea. The abdomen was not distended and on palpation a vague lump was palpable in the periumbilical region. The borders were not clearly defined, and the mass was soft.

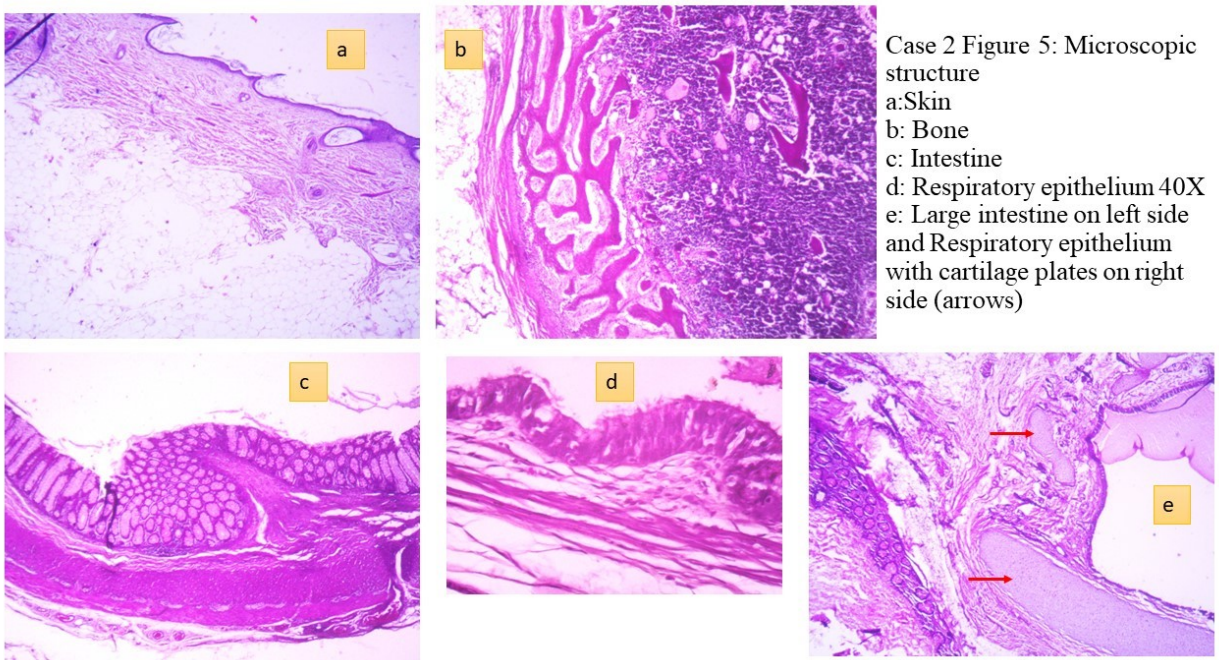
On ultrasonography a clear cystic lesion measuring 4x3.1 cm was seen in the lumbar region. The diagnosis of retroperitoneal teratoma was done. Laparotomy was done and the retroperitoneal mass having a well-defined vascular pedicle was dissected and excised in toto. On gross examination the mass was 7x5.5x3.5 cm. It was ovoid with a tapering end which was partially covered by skin with hair and showed bony elements. Cut section of the mass showed a large cystic space measuring 3x3 cm and containing serous fluid. (Figure 4) The radiograph of the mass showed multiple bony shadows suggestive of a central axis just below the large cystic cavity as shown in figure 4. The Haematoxylin and eosin-stained slides showed presence of skin, adipose tissue, cartilage, bone, large intestine with lymphoid follicles. The large cavity showed a lining of respiratory epithelium (pseudostratified columnar ciliated epithelium) with plates of hyaline cartilage in the wall. In one section the intestine is seen on one side and hyaline cartilage with respiratory lining seen on the other side. (Figure 5)

Considering the intraoperative findings, radiographic appearance suggestive of central axis and histology showing mature elements suggesting high development in organogenesis both the cases were diagnosed as fetus in fetu.




Case 2- Figure 4: A)Excised specimen B) Cut section showing large cavity, varied appearance, bony elements C)Plain radiograph – showing bony nodules below cystic space

**Fig. 4:** Case 2- Gross examination and plain radiography.




Case 2 Figure 5: Microscopic structure
a:Skin
b: Bone
c: Intestine
d: Respiratory epithelium 40X
e: Large intestine on left side and Respiratory epithelium with cartilage plates on right side (arrows)

**Fig. 5:** Case 2- Microscopic view.

## DISCUSSION

Fetus in fetu is an extremely rare congenital anomaly. Literature review showed that some authors found an equal distribution in both sexes while some depicted a higher incidence of the anomaly in males, the male to female ratio being 2:1 [1,2]. Most of the cases (75%) are diagnosed within 2 years of life while some are detected at an early age [1], and there may be some which may remain undetected till 36 years of age [3]. We have encountered one male and one female case diagnosed at ten month and six months of age respectively. The presentation may be varied from abdominal distension, emesis, jaundice, poor feeding, dyspnoea etc [1,5].

It presents as a mass in abdomen, mostly retroperitoneal in 80 %, hence may remain undiagnosed for a longer period or present with symptoms of compression of surrounding organs like feeding difficulties, respiratory, renal, or hepatic insufficiency [1,6].

In the presented cases, the first one came for complaints of constipation while the second one was an incidental diagnosis. There was a palpable lump which during operation was retroperitoneal in both the patients. Ultrasonography, CT scan and MRI can diagnose the cases of FIF wherein presence of vertebral column or axis can be diagnostic. Radiographs can also help to achieve the diagnosis of FIF.

In our cases, imaging played an important role in the diagnosis FIF for the first case, however in the second case only ultrasound was done on which the diagnosis was given as teratoma, and the patient was operated. Advances in imaging techniques and availability of these at peripheral centres has enabled early diagnosis of cases of FIF during infancy or in the prenatal period [1, 7-9].

There are certain criteria to be fulfilled to label a case as FIF – like a mass covered by a sac, mass covered completely or partially by skin, showing anatomically identifiable tissue elements, vertebral axis and attached by a vascular pedicle [6]. However, recently FIF was defined by Gonzalez- Crussi as 'any structure in which the fetal form is in a very high development of organogenesis' and linked it 'to the presence of a vertebral axis' [8].

In our study, intraoperatively both the cases showed a mass which was well encapsulated, partially covered by skin with hair and was attached to the host by a single vascular pedicle. The first case was diagnosed as FIF pre-operatively and was confirmed after radiograph of the mass and histological examination. In the second case, the pre-operative diagnosis was teratoma, but considering the intraoperative findings and the gross examination, plain radiograph of the mass was done.

The radiograph showed multiple bony shadows in the central axis and histology confirmed FIF in the second case. The histological appearance may pose a dilemma between mature teratoma and FIF. The presence of vertebral axis, limb buds and highly differentiated organs like intestine, stomach, lymphoid tissue confirm a FIF. Teratomas on the other hand are true tumours or neoplasms that are not well encapsulated and do not have a single pedicle. They usually show exophytic growth with derivatives of multiple germ layers, but they are not foetal parts. Vertebral axis is never seen in a teratoma. The surgical excision of teratoma needs to be done in toto with a critical post operative workup for any metastasis, as it has a malignant potential. FIF is a benign condition and does not show any malignant potential [4]. Some authors consider that FIF and teratomas are not two different entities but two aspects of the same pathology which are at different stages of maturation [1].

Considering the rarity of FIF and the necessity to differentiate it from teratomas, there should be adequate awareness about the condition. A combination of imaging modalities can be very useful in confirming the diagnosis of FIF. Though the treatment for FIF and teratoma is same in case there is a doubt or suspicion of teratoma over FIF, close follow up and monitoring of alpha-fetoprotein levels will help in early detection of recurrence.

## Author Contributions

**Priya Patil:** Conceptualization, Methodology, Data curation, Writing- Original draft preparation.

**Santosh Patil**: Conceptualization, Methodology, Data curation, Writing- editing and reviewing.

**Nitin Mudiraj:** Supervision, Review of literature, Writing-Editing

**Sudhakar Jadhav**: Supervision, Writing- Reviewing

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## Conflicts of Interests: None

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