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Case Report

Fetus *in fetu*: rare Indian case of infancy

Girraj Prasad Sharma[1], Jagriti Chauhan[2]*

[1]All India Institute of Medical Sciences, Bhopal, Madhya Pradesh, India
[2]All India Institute of Medical Sciences, Delhi, India

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***Correspondence:**
Jagriti Chauhan,
E-mail: jagritichauhan1004@gmail.com

ABSTRACT

Fetus in fetu (FIF) is an extremely rare condition in which malformed twin develop inside the body of living twin. It is most commonly found within the retroperitoneum. Currently, at least 200 cases have been reported across the globe, yet very few instances seen in India. We describe a case of five months old infant born via normal vaginal delivery. She had a firm palpable mass measuring 7×10×7 cm in the left hypochondriac epigastric region crossing midline. A preoperative diagnosis of FIF was made by using ultrasonography (USG) and computed tomography (CT). An exploratory laparotomy and excision of mass was performed successfully. The excised mass was proven to be FIF on the basis of gross and histopathological examination. Even though teratoma is a differential diagnosis, surgical removal is the most suitable treatment method.

Keywords: Fetus in fetu, Infant, Teratoma

INTRODUCTION

Fetus in fetu (FIF) is an extremely rare congenital condition in which malformed fetus develop inside the body of its twin. The incidence of FIF is about 1 in 5,00,000 live births.[1] It is most commonly found in retroperitoneum, although it can occur in other body parts like thorax, pelvis and sacral region.[2] Lack of a separate circulatory system accounts for the later growth retardation. Since it is still in the developmental stage of the primitive streak, it has a vertebral body and organs that are oriented around an axis. This sets it apart from fetiform teratoma, which lacks this kind of organization. The "included-twin" theory, which describes FIF as a diamniotic, monochorionic, monozygotic twin embodying within the body of the host twin following colligation of the vitelline circulation, has been used to characterise the embryo pathophysiology of FIF.[3] There is a male predominance, and the majority of cases are documented before the age of two.[4] Reviewing the literature, we aim to report this case since very few FIF reported in Indian female infants.

CASE REPORT

A five months old infant was brought by her parents to the outpatient department (OPD) complaining of abdominal distention that had been becoming worse over the last 1.5 months. The female baby was born via normal term vaginal delivery, had a healthy birth weight and no hospitalization to a neonatal intensive care unit, but she had a history of a fever episode at two-months age. There was no family history of teratoma or twin birth.

On abdominal examination, a globular hard mass was palpated in left hypochondriac and epigastric region crossing midline. On evaluation her alpha feto protein and routine blood investigations were within normal limits. For further diagnostic evaluation, ultrasonography (USG) was done. USG report showed a cystic lesion with well-defined wall contiguous with gastrointestinal tract measuring around 48×45 mm (Figure 1). Later, a contrast enhanced CT scan was done which showed a well-defined, oval-shaped, mixed-density solid cystic mass measuring 7.1×10.2×7.9 cm in the retroperitoneum that was perhaps a well-differentiated teratoma or FIF.

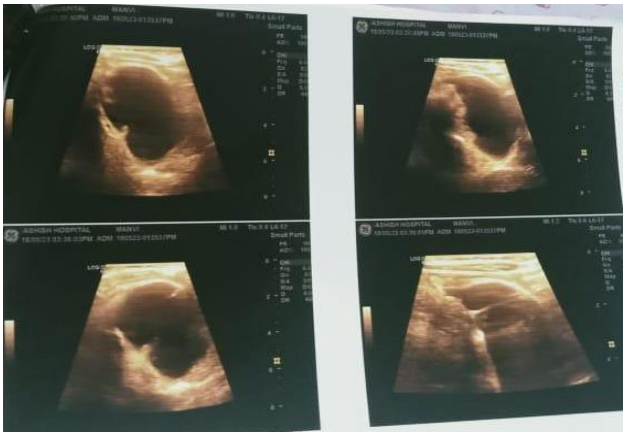

Figure 1: Ultrasonography showing gastrointestinal cyst.

A preoperative diagnosis of FIF was made and an exploratory laparotomy was planned. The excision of mass was performed via supra umbilical transverse incision. Approximately 7×10×7 cm in size, the mass mostly had a cystic appearance and contained water, soft tissue, and bone (Figure 2).

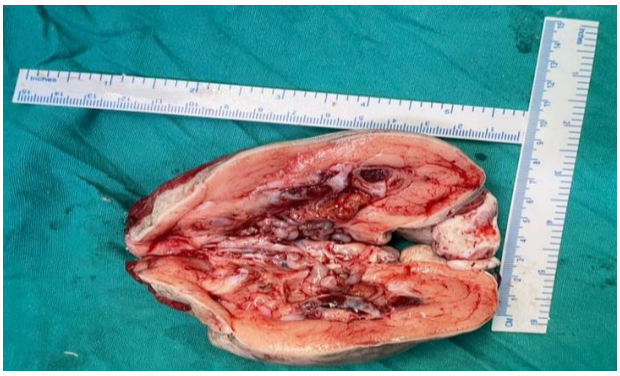

Figure 2: Cut section of mass showing bone, soft tissue and cystic water content.

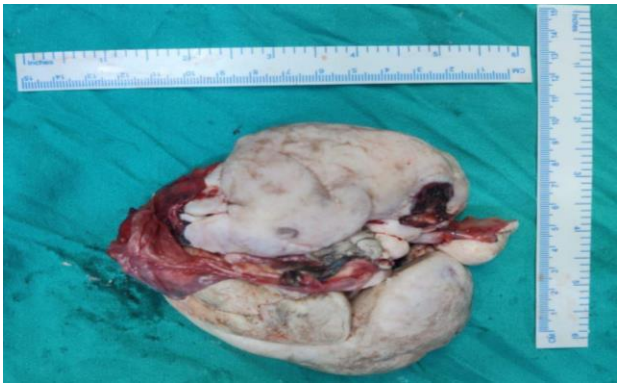

Figure 3: Gross section of excised mass.

The mass was sent for histopathological examination. According to histopathology, the removed mass was 11×8×6.5 cm in size and weighed 270 grams. Grossly four limbs were seen, cranial vault showed a crop of black hair and the cranium - caudal length was around 14 cm. The mass was covered with skin and lanugo hair. A 2.5–3 cm long, c-shaped spine and vertebral column were seen, but microscopic examination revealed no clear signs of the heart, liver, kidney, or spleen. Based on histological results, FIF was properly diagnosed.

After completion of surgery, infant was shifted to the pediatric surgical intensive care unit and later on to the surgical ward. Her post-operative period was uneventful with appropriate nursing care. She was discharged a week later and yearly follow-up was advised.

DISCUSSION

The term FIF was first described by Meckel in the late 18th century as rare condition where a malformed fetus resides inside the body of its living twin.[5]

The majority of the time, there is just one fetus there, however there have been cases where two or more coexist.[6] Symptoms like abdominal distention is due to the mass effect. The organs in these fetuses belong to several organ systems. The limbs and spinal column are two often observed organs. However, other organs like ribs, central nervous systems, gastrointestinal tract, blood vessels, and at times thymic tissues can also be visible. Being linked to the abdominal wall, the FIF receives its blood supply most frequently from the plexus of the abdominal wall. Depending on the blood supply, FIF varies in size and weight. However, fetal development retardation could nearly always be explained by the absence of a separate circulatory system. In half of the instances, intestines and brain tissue were found. Thyroid, parathyroid, pancreas, spleen, kidney, adrenal, testicles, ovaries, urinary bladder, and other rare organs have also been reported.[7] In the presented case, no clear signs of the heart, liver, kidney, or spleen were found.

The differential diagnosis of FIF includes conditions including meconium peritonitis, neuroblastoma, adrenal hemorrhage, and a retroperitoneal teratoma as causes of a newborn intra-abdominal tumor with calcification. Radiological analysis is the initial step in the study. When compared to solid tumors, meconium peritonitis, adrenal hemorrhage, and other conditions, ultrasonography reveals a complicated cystic mass with ill-defined solid internal components. By displaying the axial skeleton, more advanced techniques like contrast enhanced CT and magnetic resonance imaging (MRI) may be required to rule out a teratoma. Without histology, it may occasionally be impossible to tell the difference between FIF and a teratoma.[8]

Once FIF has been detected, surgical intervention is necessary, and the prognosis is good after total resection.[9] If the FIF tissue is left over after surgery, the risk of malignant transformation is increased.[10] Usually FIF cases are benign, but one malignant case have been reported.[11]

Although FIF has a better prognosis than cystic teratoma, thorough clinical, radiological and serological follow-up is required to look for recurrence because of the presence of immature components.[12]

CONCLUSION

The case we discuss in this report satisfies all the standards for an abdominal FIF. Preoperative FIF diagnosis is based on imaging modalities that show the vertebral column or limbs inside a mass. Complete resection with follow-up in certain cases is the preferred treatment for FIF. Also, additional research is required to figure out any connections between highly differentiated teratoma and FIF.

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