Fetus-in-fetu

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ABSTRACT

Introduction: Fetus-in-fetu (FIF) is a pathological condition in which the malformed foetus is found in the body of its twin. We report this rare case of a 3-month-old female baby whose diagnostic work-up suggested a mesenteric cyst but the mass at surgery was a FIF. **Discussion:** Although current imaging modalities like computerized Tomography and magnetic resonance imaging have a higher Sensitivity for accurate delineation of tissues, cheaper imaging alternatives like plain abdomen and ultrasonography still have a place in the preoperative work-up of FIF.

Key words: Fetus-in-fetu, intraabdominal mass, operative diagnosis, teratoma, ultrasonography

INTRODUCTION

Fetus-in-fetu (FIF) is a pathological condition in which the malformed foetus is found in the body of its twin. It is a form of monozygotic diamniotic twin with an unequal division of the totipotent inner cell mass of the developing blastocyst, which results in inclusion of a small cell mass in the more mature embryo. It has a 2:1 male predominance [1,2] with most patients presenting with an abdominal mass in infancy.[1] It remains controversial whether FIF is a distinct entity or represents a highly organized teratoma.

We report this rare case of a 3-month-old female baby whose diagnostic work-up suggested a mesenteric cyst but the content at surgery was an FIF.

CASE REPORT

A 3-month-old female presented with progressively increasing abdominal swelling and poor weight gain noticed 2 weeks before presentation. Delivery was at term via the vagina, with no complications. Pregnancy and delivery were uneventful; there was no history of abdominal mass in her two siblings.

On examination, the abdomen was asymmetrically distended and a small umbilical hernia was noted. A firm mass measuring 12x12 cm was palpated in the left lumbar region. It had a well-defined lower margin but the upper margin was ill defined. The mass was irregular, fixed and nontender. The right kidney was palpable while the left was obscured by the mass.

Plain radiograph showed a large soft tissue mass in the left upper and middle abdominal quadrants, displacing the bowel loops peripherally and the stomach to the right side. Marginal linear calcification was noted in the superior left margin of the mass [Figure 1].

Ultrasound scan of the mass revealed a noncalcified complex multiloculated cyst with solid internal content [Figure 2].

Intravenous urography showed normal kidneys on both sides, but the left kidney was displaced downwards into the pelvis by the mass.

A preoperative impression of a complex retroperitoneal mass was made to rule out a mesenteric cyst.

At an elective laparatomy, there was a well-encapsulated cystic retroperitoneal mass displacing the spleen and the pancreas cephalad and the left kidney laterally and caudally. The mass had blood supply from the left renal vessels. Fifty millilitres of clear fluid was aspirated from the mass. The mass was completely excised. The soft tissue contained a malformed dead foetus with a well-defined foot and skin with hairs as well as some recognizable brain tissue and a convex pliable piece of skull bone among other undifferentiated tissues [Figure 3].

Other associated findings include malrotated midgut and Ladd's band. X-ray of the mass did not reveal any calcified vertebral tissue. Treatment was complete surgical resection of the mass. The patient did well after surgery and was discharged.

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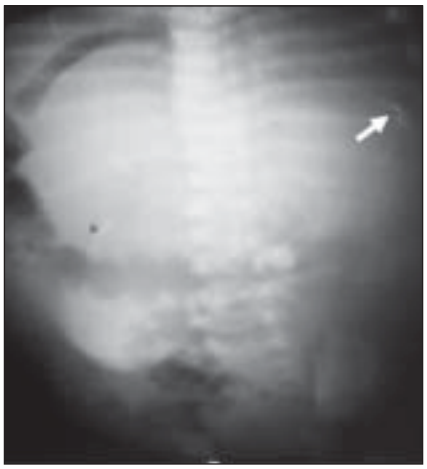


Figure 1: Plain radiograph of the abdomen showing displaced bowel loops by the mass. Note the curvilinear calcifi cation [arrow]



Figure 2: Ultrasonography of the mass showing a well-circumscribed complex mass, which is mainly cystic

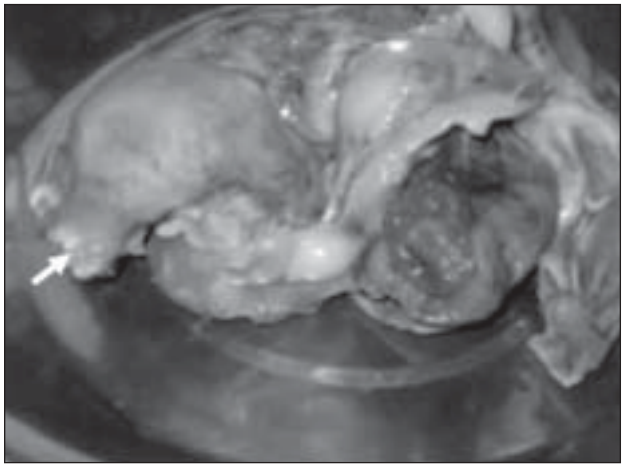


Figure 3: Photograph of the dissected foetoid mass demonstrating the right lower limb with well-formed digits [arrow]

DISCUSSION

FIF is a rare condition. It was first reported by Young in 1809,[3] but the term "Fetus-in-fetu" was first coined by Merkel in the 18th century.[4]

FIF is discovered most commonly in infancy as a retroperitoneal mass.[5] Other more unusual sites, including the cranial cavity, oral cavity, sacrococcygeal region and scrotum, have been reported.[2]

Usually, only one foetus is present, but multiple foetuses have been reported.[1] The foetus itself is incomplete, containing a variable number of identifiable organs. They are always anencephalic and acardiac.[6,7]The lower limbs are more developed than the upper limbs.

Symptoms may be absent or are due to mass effect and include abdominal distension, feeding difficulties, emesis, jaundice and dyspnoea.[1,7]

The preoperative diagnosis of FIF depends on radiological findings. Plain abdominal X-ray may be helpful, and up to half of the cases show the presence of a vertebral column and axial skeleton.[6,8] The vertebral column was not identified in this case, neither was any calcification seen in the excised mass on X-ray. However, Hoeffel *et al*. [8] explained that nonvisualization of the vertebral axis on radiography or computerized tomography (CT) does not exclude the diagnosis because this may be due to an underdeveloped and markedly dysplastic spinal column that prevented its identification at imaging.

Plain abdominal radiograph in our case showed a soft tissue mass with rim calcification displacing the bowel loops peripherally. This led to the suggestion of a mesenteric cyst. Another possibility was meconium pseudocyst. The sonographic findings were those of a complex cystic mass with an ill-defined solid internal component. This made us suggest a mature cystic teratoma.

Imaging played an important role in the ability to diagnose FIF. CT and magnetic resonance imaging (MRI) have proven very helpful in suggesting a preoperative diagnosis;[7] however; these were not carried out in our case due to their high cost, which parents cannot afford.

The foetus is typically suspended by a pedicle within a complete sac containing fluid or sebaceous material. There is no placenta or chorionic villi at the point of attachment to the host.[6]

In conclusion, the diagnosis of FIF should be considered in a child with progressively increasing abdominal swelling. Where current imaging modalities like CT and MRI services are unavailable, plain X-rays and

ultrasonography are still useful in the work-up before surgery.

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Source of Support: Nil, **Conflict of Interest:** None declared.