

Twin Fetus in Fetu in an Infant

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Abstract

Twin fetus in fetu is a very rare anomaly among fetus in fetus. Fetus in fetu is a rare developmental abnormality in which a mass of tissue resembling a fetus forms inside the body. We reviewed a case of 6 months old male baby who was admitted in the Department of Pediatric Surgery in Bangabandhu Sheikh Mujib Medical University (Dhaka, Bangladesh) due to gradual distension since birth. Our patient recovered well after surgery. He was discharged home on 8th Postoperative day. To rule out any recurrence the child was followed up through clinical examination, abdominal ultrasound and serum alpha feto protein. After two and half year, no recurrence of previous symptoms.

Keywords: *Twin Fetus in Fetu; Fetus in Fetus; Alpha Feto Protein*

Introduction

Fetus in fetu (FIF) is a rare condition with an estimated incidence of 1 in 500000 births [1]. To our knowledge fetu in fetus was originally described by Meckel in the 18th century. It is a rare variety of parasitic twin where a developmentally abnormal mono zygomatic monochromic diamniotic parasitic twin is completely encapsulated within the abdomen of the otherwise normally developed host twin [2]. Fetus in fetus is thought to result from unequal division of totipotent inner mass. The mass of the cells that are ancestral precursor to all cells in the body. The unequal division is thought to occur during the blastocyst, which can also result in parasitic and conjoined twins.

Twin fetus in fetus is a very rare anomaly among fetus in fetus [4]. Most cases that have been reported in the literature occur within the upper retroperitoneum or within the abdomen [5]. The fetus in fetus (FIF) complex is characteristically composed of fibrous membrane (Equivalent to the chorioamniotic complex) that contains some fluid (equivalent to the amniotic fluid) and the fetus suspended by cord or pedicle [6]. In uterus the growth of an FIF initially parallel that of its twin but stops abruptly because either the vascular dominance of the host twin or an inherent defect in the parasitic twin [7]. In the past the scientists speculated that fetus in fetus was the result of a highly developed teratoma rather than product of abnormal embryonic development. In order to classify abnormal growth as fetus in fetu rather than a teratoma several characteristics must be present. FIF is a benign growth of embryological origin and is located behind the abdomen lining of the host twin. To diagnose fetus in fetus there must be evidence of body plan organization including vertebrae, limbs buds and organ tissue, abdominal radiographs, CT scan and ultrasonography (USG) are helpful technologies to determine whether a mass is teratoma or fetus in fetus. The surgical removal of the twin fetus is the treatment of choice. In most cases of FIF only one fetus exists inside the baby. Only in extreme rare cases more than one fetus is found.

Case Presentation

A 6 months old male baby was admitted in the Department of Pediatric Surgery in BSMMU due to gradual distension since birth. On examination abdomen was distended, non-tender, smooth surface and firm. Conventional abdominal x-ray showed a large soft tissue

mass with vertebrae like column. An ultrasound of the baby's abdomen showed two encysted hyper echoic and calcified heterogeneous complex mass. CT scan of his abdomen revealed two (one large and one small) fetal skeletons are seen within upper peritoneal cavity. Evidence of anencephaly was seen in both fetuses. The structures are causing displacement of adjacent loops.

A detailed ultrasound of the abdomen showed a complex cystic mass within this single thin walled cyst, there were two heterogeneous solid mass, which contained a well-ossified spine and two ossified long bones at the caudal end resembling the configuration of fetal femur. Alfa fetoprotein and beta human chorionic gonadotropin level were normal for age. Routine hematological investigation was within normal limits.

Antenatal at 9 months pregnancy ultrasonography showed a fetus with a mass in the abdomen of the fetus. The full term born baby was born by lower uterine caesarian section and perinatal period was uneventful. The abdomen was found distended after birth also.

Elective laparotomy was performed. We found a large encysted retroperitoneal mass compressing right kidney and pushing the hepatic flexure of ascending colon anteriorly and medially. After mobilization of the right colon and hepatic flexure we found a well capsulated cystic retroperitoneal mass. The mass had a cord like structure, which contained its artery and vein. These were arising from abdominal aorta and vena cava. This feeding vessel pedicle was ligated and divided. The mass was removed easily. The sac contained two-amniotic fetus connected to each other by a cord like structure at the middle. The miniature fetuses had well-defined limbs, foot, skin with hairs, a convex and pliable skull bone.

On gross examination the entire mass was enclosed in a firm thin covering. On opening this covering there was small amount of fluid with sticky brownish coating the entire multi structure. The mass corresponded to an incompletely developed twin fetus.

Our patient recovered well after surgery. He was discharged home on 8th postoperative day. To rule out any recurrence the child was followed up through clinical examination, abdominal ultrasound and serum alpha fetoprotein. After two and half year, no recurrence of previous symptoms.

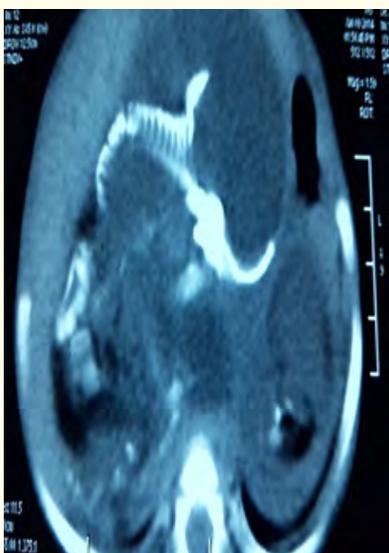


Figure 1: CT scan showing the spine of fetus.



Figure 2: Contrast MDCT showing two fetal skeletons within upper peritoneal cavity.

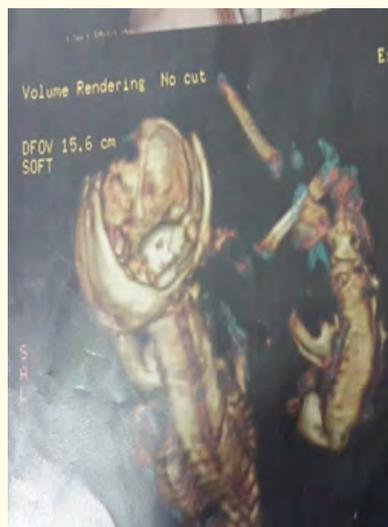


Figure 3: MDCT showing fetal skeleton within evidence of anencephaly in both fetuses.

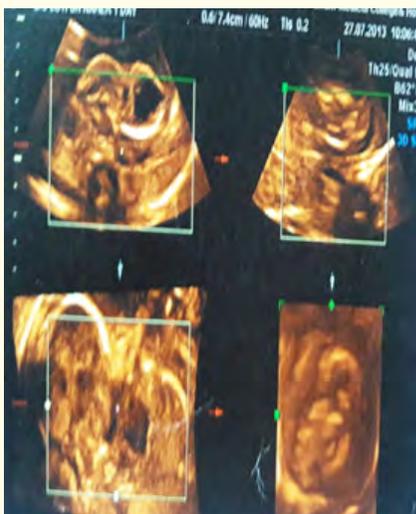


Figure 4: USG of whole abdomen showing incomplete spine along the right abdomen, lobulated clear cystic area is seen in left hypochondrium, fetal hearts could not be seen, bowel loops and intestinal loops are compressed posteriorly.



Figure 5: Under general anaesthesia, huge distension of abdomen is seen.



Figure 6: Scar mark at the time of discharge to home. There is no distension of abdomen.



Figure 7: Evisceration of gut along with the fetuses.



Figure 8: Ligation of blood vessel pedicle.

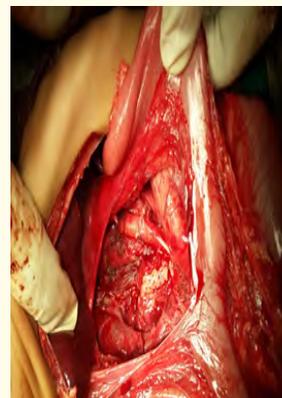


Figure 9: After expulsion of fetuses.

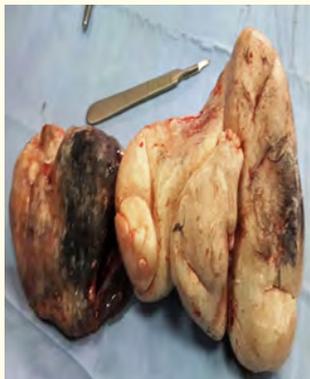


Figure 10: Specimen of the fetuses.

Discussion

Fetus in fetu is a rare developmental abnormality in which a mass of tissue resembling a fetus forms inside the body [1]. There are two schools of thoughts for the occurrence of FIF. One theory is that the mass begins as a normal fetus but becomes enveloped inside its twin; the other theory is that the mass is highly developed teratoma. And Twin fetus in fetu is a very rare anomaly among fetus in fetu [4].

This rare entity occurs only once every 500,000 births [1]. On global scenario less than 100 cases have been reported [7]. Most cases have been reported in the literature occur within the upper retroperitoneum or within abdomen. unusual location for fetus in fetu have included the scrotal sac, pelvis, iliac mesentery and in the cranial cavity. There is no predilection of sex for FIF i.e. occurs equally in male and female. In 70% of the reported cases, most of them present with an abdominal mass which is mostly retroperitoneum. Mass effect symptoms like feeding difficulties, emesis, and dyspnea are also predominate [8,9].

Most of the cases are diagnosed antenatal or immediately after birth. The differential diagnosis that one should consider are teratoma, wilm's tumor, hydronephrosis, and neuroblastoma. The differentiating point for teratomas are there are absence of vertebral segmentation, craniocaudal and lateral differentiation, body coelom or systemic organogenesis. Thus the presence of a mass with spinal organization and surrounded by fluid suggests the correct diagnosis as it was in our case. Neonatal evaluation with USG, CT and MRI scan should be done once the antenatal sonographic detection of fetal mass resembling a skeletal or other fetal parts have been made.

Sonographic findings are usually those of a complex cystic mass with ill-defined solid internal components⁹. Plain abdominal x-ray could be supplementary before operation as it can reveal the presence of vertebral column and axial skeleton. Further confirmation can be made by CT and MRI scans. In our case plain x-ray, Ultrasonography of abdomen and CT scan helped in evaluation of anatomy of FIF. Surgical extirpation is the treatment of choice for FIF. In our case we found a well-encapsulated cystic retroperitoneal mass. A cord like structure at the middle connected the two miniature fetuses to each other.

Studies have shown that the prognosis of FIF is better than Cystic teratoma. However the presence of immature elements should be taken seriously [3]. This need close monitoring clinically, radiologically, and serologically on follow up. Despite the AFP level normal with age preoperatively and postoperatively, a possible recurrence of a malignant teratoma after FIF resection must be kept in mind. That is why we continued to monitor with tumor marker on our follow up at 3 months, 6 months, 12 months and 18 months. No recurrences have been detected yet.

Conclusion

FIF is a rare entity. However it should be kept as differential diagnosis in a child presented with progressive growing abdominal mass. Different investigations help to diagnosis FIF preoperatively i.e. X-ray abdomen, USG, CT scan, MRI. further alpha fetoprotein (AFP) should be done preoperatively to exclude teratomas and serial tumor markers should be done postoperatively to monitor recurrence.

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