Fetus-In-Fetu or fetaform teratoma: Clinical dilemma

Adnan Amin Alsulaimani, Alaaeldin Mohammed Elsayed EI-Zeky

ABSTRACT
Fetus is fetu (FIF) is an uncommon anomaly in Pediatric age group. The literature cites around 100 cases worldwide of twin fetus in fetu. Hypothesis vary about whether FIF represents an enclosure or failed monozygotic twin or a well-differentiated variant of mature teratomas. The presence of a vertebral column has been considered to be mandatory for the diagnosis of fetus in fetu. It should be differentiated from a teratoma because of the teratomas malignant potential. Our case is another example of lesion that fits in this spectrum of interesting dilemma and highlight on the magnitude of imaging studies to differentiate between them.

Keywords: Fetus in fetu, Fetaform teratoma

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INTRODUCTION
Fetuses in fetus and retroperinatal teratoma have been considered as two distinct entities [1]; Recent literature question the necessity of presence of axial skeleton to be mandatory for the diagnosis of fetus-in–fetu [2]. This entity is differentiated from teratoma by its embryological origin, its unusual location in the retroperitoneal space, and the presence of vertebral organization with limb buds and well developed organ system. In most cases of FIF, only one fetus exists inside the baby, only on extremely rare cases are multiple fetuses found [3]. The surgical removal of the twin fetus is the treatment of choice.

CASE REPORT
A two days old, full term male newborn born to a Saudi mother G^5P^5 without any pregnancy or familial significant risk factors. The baby delivered by cesarian section due to breech presentation. Upon examination of the baby abdomen was discovered that a firm, irregular and non-tender mass was present in the left hypochondrium extending to the umbilical region, genitalia were normal apart from undescended bilateral testis. Conventional X-ray of the abdomen showed multiple dense radio-opacities in the left upper quadrant of the abdomen (Figure 1). An ultrasound of the baby abdomen showed a large encysted, hyperechoic heterogeneous complex mass. MRI lumbar spine and CT scan of the baby's abdomen revealed (Figures 2 & 3) an encapsulated cystic peritoneal mass contain multiple bony elements of axial skeleton with pelvic girdle, lower extremity buds, ribs and no cranial vault can be appreciated. On laparotomy, a left upper quadrant retroperineal mass in 8x6 cm in size, covered with a sac containing a partially developed fetus with partial differentiation of the limbs, skull, spine and vertebrae (Figure 4). Both testes were felt retroperitonally. Baby did well and discharged home after few days in good general condition. The chromosome analysis confirm a male chromosome complement without structural anomalies. The tumor marker, Carcinoembryonic antigen (CEA) was normal.
DISCUSSION

Fetus in fetu was generally considered a highly differential teratom till 1935. Willis et al [1] defined FIF as a separate entity containing some organs, limbs and vertebral column. The phenomenon of FIF has been described at least since the 17th century [4], but the credit with first description of it go to Meckel in 1800 [5]. In review of the literature, about nine percent of cases of FIF, no vertebral skeleton could be identified an imaging or histopathologically. Moreover, more cases reported with hands, phalanges and nail were found but no vertebral column were exists [3, 6]. Most investigators believe that the presence of a developed vertebral column is not a prerequisite for diagnosis [7, 8]. Spencer [9] has suggested that an FIF must have one or more of the following condition:

- Be enclosed within a distinct sac.
- Be partially or completely covered by normal skin.

Figure 1: X-ray of abdomen showed heterogenous and amorphous large area of calcification, mostly peritoneal calcification seen in the left hypochondrial area. A small calcified area in right suprarenal region.

Figure 2: MRI Lumbar spine showed encapsulated cystic peritoneal mass contain multiple bony elements of axial skeleton with pelvic girdle.

Figure 3: CT scan of the baby's abdomen showed encapsulated cystic peritoneal mass contain multiple bony elements of axial skeleton with pelvic girdle.

Figure 4: A mass sized 8x6 cm covered with a sac containing partially developed fetus.
• Have grossly recognizable anatomic parts.
• Be attached to the host by only a few relatively large blood vessels.
• Either be located immediately adjacent to one of the site of attachment of conjoined twins or be associated with the neutral tube or the gastrointestinal system.

A teratoma, on the other hand, shows lesser organization of microscopically identifiable tissues. Teratoma have a broader attachment site with multiple small blood vessels. The differential diagnosis of a complex mass with calcified components in a fetus or neonate include FIF, teratoma, meconium pseudocyst and neuroblastoma [2]. Our case include most of spencer characteristics of FIF. The normal value of CEA marker, the coexistence of vertebral column and the finding of the testis in the retroperitoneum, all confirm the diagnosis of FIF in this case. Although the prognosis for FIF is more favorable than for cystic teratoma, the presence of immature elements nevertheless indicates the need for close clinical radiological, serological and tumor marker work-up.

CONCLUSION

We conclude Fetus in fetu is an extremely rare condition. Currently it is considered as a benign condition; however, controversy continues regarding its future malignant potential or association. When mass compression becomes significant, as in our case, the child or adult can suffer from poor growth or development, infections and even lack of organ function. With surgical treatment, normal anatomy and physiology can be restored and the malignant potential excised. Definitive diagnosis is best made using CT and MRI techniques serology test and tumor marker surveillance and in addition to genetic testing and pathologic reviews, physicians must consider the potential for a minimum of 18 months follow up to avoid a missed malignancy. Surgical outcomes are reportedly good following excision, but further data collection is required for long term results.

REFERENCES


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Alaaelddin Mohammed Elsayed El-Zeky – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

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Written informed consent was obtained from the patient for publication of this case report.

Conflict of Interest
Authors declare no conflict of interest.

Data Availability
All relevant data are within the paper and its Supporting Information files.

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