Fetus-in-fetu in an 8-month-old girl

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**Abstract:** Fetus in fetu (FIF) is a very rare congenital malformation with an incidence of 1/500,000 live births. These children are often hospitalized because of vomiting, abdominal distension or abdominal mass found by their parents. The most common position for the parasitic fetus is the retroperitoneum but other position such as the scrotal sac, cranial cavity, sacrococcygeal region, back, oral cavity has been reported. FIF is often misdiagnosed as a mature teratoma. Ultrasound, CT, MRI are of great significance in distinguishing between FIF and teratoma. The postoperative pathological examination may reveal axial skeleton which can make a definitive diagnosis of FIF. In this report, we present a case of FIF in an 8-month-old girl.

**Keywords:** Fetus-in-fetu(FIF), differential diagnosis, pathologic histology

An 8-month-old girl was found with an abdominal mass which was detected on ultrasound on the 4th antenatal examination. The ultrasound scan at that time found a mass in the upper left quadrant of the fetus, which was suspected to be a retroperitoneal teratoma. At the age of 8-months, she admitted to our hospital with vomiting and abdominal distension. Her vital signs at the time of admission were normal. The laboratory results including her blood routine, urinalysis, liver function, renal function, alpha-fetoprotein [AFP], carcinoembryonic antigen [CEA]) were all within normal range. Ultrasound examination revealed a retroperitoneal mass which was suspected to be a mature teratoma and the mass is about 10.0x9.5x9.0cm in size. An abdominal computed
tomography (CT) scan showed of a 12×11×10 cm mass with a clearly boundary. There is a cystic structure containing a large amount of calcification in the mass.

Three-dimensional CT imaging showed pelvic bones, vertebral bones, femur and other long bones (Figure 1). A diagnosis of retroperitoneal fetus-in-fetu was made.

A laparotomy was performed. During the surgery, a retroperitoneal cystic mass was found. After opening the mass, we can see an incompletely developed fetus with grossly visible limbs, hair, and a poorly formed head. The fetus measured 11.5×10×9 cm. (Figure 2) After operation, histopathology showed the presence of cartilage in the mass, as well as skin, bones, and adipose tissue in the fetus. The postoperative course was uneventful and the patient was discharged on postoperative day 7. The patient was asymptomatic at the 6-month follow-up.

Discussion:

Fetus in fetu (FIF) was first described by Meckel in the 19th century. It was defined as a mass containing a vertebral axis often associated with other organs or limbs around this central axis [1]. Its incidence is estimated to be approximately 1/500,000 [2,3]. In most of the cases, FIF is located in the retroperitoneal, while uncommon sites include the sacrococcygeal region, lung, oral cavity, cranial cavity, mediastinum and scrotum [4-10]. The major clinical manifestation of FIF is a palpable abdominal mass. The other symptoms are secondary to the mass such as vomiting, jaundice, intestinal obstruction, respiratory distress, meconium peritonitis, and hydronephrosis [11-13]. Imaging
examinations plays a important role in diagnosing FIF. Abdominal X-ray plain shows the presence of a vertebral column and axial skeleton about in half of the reported cases [14]. CT and MRI can give a more accurate diagnosis and can define the relationship of FIF with the other abdominal organs. The presence of a vertebral column in FIF is an significant feature that distinction FIF with teratoma.[14-15] In this report, we summarized the imaging performance and postoperative pathologic histology inspection of retroperitoneal FIF which have been published in English from 2010 to 2015.[16-33] (Table 1). The main differential diagnosis is teratoma. To distinguish FIF from teratoma, Gonzalez-Crussi defined it as any structure in which the fetal form exhibits very advanced development of organogenesis and has a vertebral axis[34]. FIF should also be differentiated from other cystic masses in fetus, such as meconium pseudocyst, intestinal duplication, hematoma, and fetal infection[35].

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**Figure 1:**

![Three-dimensional CT imaging showed pelvic bones, vertebral bones, femur and other long bones (arrow 1). b,c: Three-dimensional CT showing long bones like pelvis(arrow 2)、thighbone (arrow 3)、vertebra (arrow 4) and in the mass.]

**Figure 2:**

![Images of the cystic form of meconium peritonitis]
Fig 2. a. b: Excised encapsulated mass before and after opening the capsule.

c: After removing the capsule: we can see Fetus-in-fetu with hairy head (arrow 1); limb buds (arrow 2); umbilical cord(arrow 3).

Table 1:

<table>
<thead>
<tr>
<th>Case No.</th>
<th>Gender</th>
<th>Age</th>
<th>Vertebral column on radiology</th>
<th>Long bones</th>
<th>Pathologic</th>
</tr>
</thead>
<tbody>
<tr>
<td>1.[16]</td>
<td>Female</td>
<td>2-months</td>
<td>Not seen</td>
<td>Lower limbs with toes, upper limbs with fingers (CT,US,X-Ray Gross pathologic)</td>
<td>Umbilical cord, brain tissue ,gut-like structure, fingers, limbs</td>
</tr>
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<td>2.[17]</td>
<td>Male</td>
<td>30-months</td>
<td>Seen (CT)</td>
<td>Vertebral, 2 lower limbs, 1 foot with 9 toes, 1 foot with 4 toes(CT ,Gross pathologic)</td>
<td>Skin, vertebral column, muscle, lymphoid tissue, adipose tissue, male genitalia, neural and bone tissue</td>
</tr>
<tr>
<td>3.[17]</td>
<td>Male</td>
<td>Neonate</td>
<td>Seen (US,CT)</td>
<td>Long bones, axial skeleton vertebral column, 2 upper extremities, 1 hand with 5 fingers, 1 hand with 4 fingers, thoracic cage(CT ,US, Gross pathologic)</td>
<td>Skin, fat, skeletal , muscles, intestines, ganglion, nerve</td>
</tr>
<tr>
<td>4.[18]</td>
<td>Female</td>
<td>5-days</td>
<td>Not seen (X-Ray ,US ,CT, )</td>
<td>Finger, coccyx(X-Ray ,US ,CT, Gross pathologic)</td>
<td>Skin, fat tissue, liver, pancreas, well-developed gastrointestinal tract,</td>
</tr>
<tr>
<td>5.[19]</td>
<td>--</td>
<td>18-days</td>
<td>Seen (MRI, X-Ray ,US ,CT)</td>
<td>Limbs, 4 fingers with nails, spinal column (MRI, X-Ray ,US ,CT Gross pathologic)</td>
<td>Brain tissue, gastrointestinal, genitalia , cartilage, vessels, myxoid materia</td>
</tr>
<tr>
<td>6.[20]</td>
<td>Female</td>
<td>4.5-years</td>
<td>Not seen</td>
<td>3 vestigial arm</td>
<td>---</td>
</tr>
<tr>
<td></td>
<td>Gender</td>
<td>Age</td>
<td>Observations</td>
<td>Imaging Techniques</td>
<td>Gross Pathologic</td>
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<tr>
<td>7.</td>
<td>Male</td>
<td>3.5-months</td>
<td>Seen (CT, US)</td>
<td>Long bones of limbs, Vertebral column, spine (CT, Gross pathologic)</td>
<td>Skin, hairs, cartilage, bone, intestine, epithelium, vessel, genitalia,</td>
</tr>
<tr>
<td>8.</td>
<td>Female</td>
<td>6-months</td>
<td>Seen (MRI, X-Ray)</td>
<td>Long Bones, vertebral column, 5 limbs, hands, feet (X-Ray, MRI, Gross pathologic)</td>
<td>Neural tissue, epithelial, choroid plexus, intestine, pancreatic tissue, adipose tissue</td>
</tr>
<tr>
<td>9.</td>
<td>--</td>
<td>6-weeks</td>
<td>Seen (CT)</td>
<td>Vertebral column with rib cage (X-Ray, CT, Gross pathologic)</td>
<td>Hair, vertebral column, brain, stomach, intestine, liver, kidney</td>
</tr>
<tr>
<td>10.</td>
<td>Male</td>
<td>2-years</td>
<td>Seen (CT)</td>
<td>Vertebral axis, limbs (CT, Gross pathologic)</td>
<td>Vertebral axis, hair, placenta, limbs</td>
</tr>
<tr>
<td>11.</td>
<td>Male</td>
<td>4-months</td>
<td>Seen (CT)</td>
<td>Vertebrae, femur, tibia, fibula, limbs, hand (CT, X-Ray, Gross pathologic)</td>
<td>Umbilical cord, limbs, hands, hair, scrotum, calcification</td>
</tr>
<tr>
<td>12.</td>
<td>Female</td>
<td>19-days</td>
<td>Seen (CT, US, X-Ray)</td>
<td>Well-formed spine, limb (CT, US, X-Ray)</td>
<td>Umbilicus cord, well-developed trunk, skin, hair, well-developed arms and forearms</td>
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<tr>
<td>Case No.</td>
<td>Age</td>
<td>Status</td>
<td>Findings</td>
<td>Tissue Types</td>
<td></td>
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<tr>
<td>14.[28]</td>
<td>2-weeks</td>
<td>Seen (X-Ray)</td>
<td>Ossify (US), vertebral axis bones (X-Ray)</td>
<td>Skin</td>
<td></td>
</tr>
<tr>
<td>15.[29]</td>
<td>17-months</td>
<td>Seen (CT,US)</td>
<td>Extremity bone, vertebral (CT, US, Gross pathologic)</td>
<td>Vertebral, extremity bone, fat</td>
<td></td>
</tr>
<tr>
<td>16.[30]</td>
<td>3-months</td>
<td>Not seen</td>
<td>Limb buds, long bones (US, X-Ray)</td>
<td>Limbs, rudimentary pulmonary</td>
<td></td>
</tr>
<tr>
<td>17.[31]</td>
<td>Male</td>
<td>4-days</td>
<td>Seen (MRI,US)</td>
<td>Mature bone tissue, muscular tissue, adipose tissue, bowel tissue, pancreatic tissue, myeloid tissue, umbilical cord</td>
<td></td>
</tr>
<tr>
<td>18.[32]</td>
<td>Male</td>
<td>18-months</td>
<td>Seen (CT,US)</td>
<td>Pelvis, vertebral with rib cage, upper limbs with fingers, lower limbs with feet (US,CT)</td>
<td>Neurological tissue, vertebral, external genitalia</td>
</tr>
<tr>
<td>20.[33]</td>
<td>Female</td>
<td>Neonate</td>
<td>Seen (US,CT)</td>
<td>2 fetuses-in-fetus with spine, pelvic, femurs, limbs, long bone, rib cage (CT,US, Gross pathologic)</td>
<td>Skin, intestines, brain tissue, spine, umbilical cord, cartilage, muscles</td>
</tr>
</tbody>
</table>