

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 12×11×10cm 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:

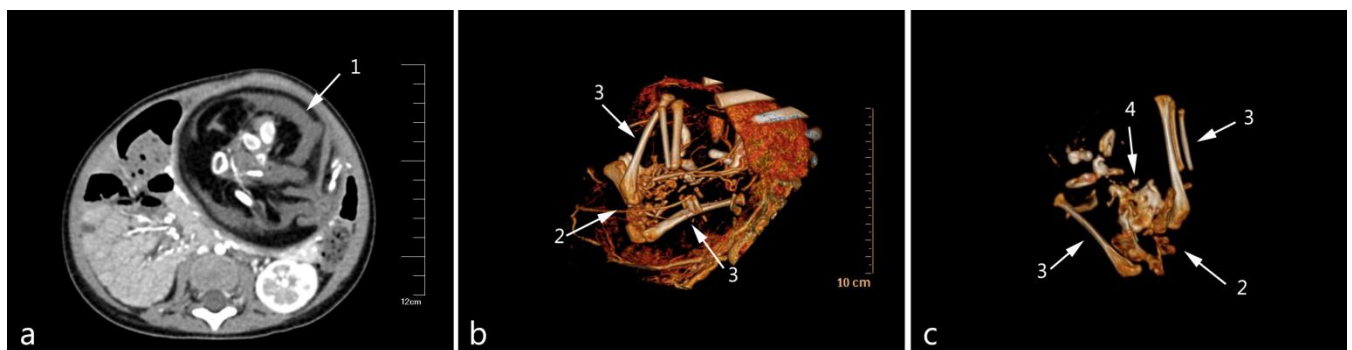


Fig 1. a: Three-dimensional CT imaging showed pelvic bones, vertebral bones, femur and other long bones (arrow 1). b,c: Three-dimensional CT showing long bones like pelvis (arrow 2), thighbone (arrow 3), vertebra (arrow 4) and in the mass.

Figure 2:



- 1 **Fig 2.** a. b: Excised encapsulated mass before and after opening the capsule.
- 2 c: After removing the capsule: we can see Fetus-in-fetu with hairy head (arrow 1) ; limb
- 3 buds (arrow 2); umbilical cord(arrow 3).

4 **Table 1:**

Case No.	Gender	Age	Vertebral column on radiology	Long bones	Pathologic
1.[16]	Female	2-months	Not seen	Lower limbs with toes, upper limbs with fingers (CT,US,X-Ray Gross pathologic)	Umbilical cord, brain tissue ,gut-like structure, fingers, limbs
2.[17]	Male	30-months	Seen (CT)	Vertebral,2 lower limbs,1 foot with 9 toes,1 foot with 4 toes(CT ,Gross pathologic)	Skin, vertebral column, muscle, lymphoid tissue, adipose tissue, male genitalia, neural and bone tissue
3.[17]	Male	Neonate	Seen (US,CT)	Long bones, axial skeleton vertebral column,2 upper extremities,1 hand with 5 fingers,1hand with 4 fingers, thoracic cage(CT ,US, Gross pathologic)	Skin, fat, skeletal , muscles, intestines, ganglion, nerve
4.[18]	Female	5-days	Not seen (X-Ray ,US ,CT,)	Finger, coccyx(X-Ray ,US ,CT, Gross pathologic)	Skin, fat tissue, liver, pancreas, well-developed gastrointestinal tract,
5.[19]	--	18-days	Seen (MRI, X-Ray ,US ,CT)	Limbs, 4 fingers with nails, spinal column (MRI, X-Ray ,US ,CT Gross pathologic)	Brain tissue, gastrointestinal, genitalia , cartilage, vessels, myxoid materia
6.[20]	Female	4.5-years	Not seen	3 vestigial arm	---

			(X-Ray ,US)	(Gross pathologic)	
7.[21]	Male	3.5-months	Seen (CT , US)	Long bones of limbs, Vertebral column, spine(CT, Gross pathologic)	Skin, hairs, cartilage, bone, intestine, epithelium, vessel, genitalia,
8.[22]	Female	6-months	Seen (MRI, X-Ray)	Long Bones, vertebral column, 5 limbs, hands, feet (X-Ray, MRI, Gross pathologic)	Neural tissue, epithelial, choroid plexus, intestine, pancreatic tissue, adipose tissue
9.[23]	--	6-weeks	Seen (CT)	Vertebral column with rib cage(X-Ray, CT, Gross pathologic)	Hair, vertebral column, brain, stomach, intestine, liver, kidney
10.[24]	Male	2-years	Seen (CT)	Verbebral axis,limbs (CT, Gross pathologic)	Vertebral axis, hair, placenta, limbs
11.[25]	Male	4-months	Seen (CT)	Vertebrae, femur, tibia, fibula, limbs, hand (CT, X-Ray, Gross pathologic)	Umbilical cord, limbs, hands, hair, scrotum, calcification
12.[26]	Female	19-days	Seen (CT,US,X-Ray)	Well-formed spine, limb (CT,US,X-Ray)	Umbilicus cord, well-developed trunk, skin, hair, well-develop arms and forearms
13. [27]	Male	7-days	Seen (CT,US,X-Ray)	Spine, femur, scapula (CT, US, X-Ray Gross pathologic)	limbs with skin and hair, mature brain tissue, epithelium, cartilage, gut-like structure, fat

14.[28]		2-weeks	Seen (X-Ray)	Ossify(US), vertebral axis bones(X-Ray)	Skin
15.[29]		17-months	Seen (CT,US)	Extremity bone, vertebral (CT, US, Gross pathologic)	Vertebral, extremity bone, fat
16.[30]		3-months	Not seen	Limb buds, long bones(US, X-Ray)	Limbs, rudimentary pulmonary
17.[31]	Male	4-days	Seen (MRI,US)	Spine, Extremity, thorax(MRI,US, Gross pathologic)	Mature bone tissue, muscular tissue, adipose tissue, bowel tissue, pancreatic tissue, myeloid tissue, umbilical cord
18.[32]	Male	18-months	Seen (CT,US)	Pelvis , vertebral with rib cage , upper limbs with fingers, lower limbs with feet (US,CT)	Neurological tissue, vertebral, external genitalia
19.[32]	Female	9-years	Seen (X-Ray, US)	Long bones, vertebrae, upper and lower limbs (X-Ray, Gross pathologic)	Hair, genitalia, vertebrae
20.[33]	Female	Neonate	Seen (US,CT)	2 fetuses-in-fetus with spine, pelvic, femurs, limbs, long bone, rib cage (CT,US, Gross pathologic)	Skin, intestines, brain tissue, spine, umbilical cord, cartilage, muscles