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Fetus-in-fetu: imaging and pathologic findings

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Abstract

A 3.5-month-old boy was hospitalized because of an abdominal mass found accidentally. On physical examination, a smooth, firm, nontender mass was present in the right upper quadrant. Abdominal ultrasonography revealed a large, hyperechoic, heterogenous mass with clear boundaries, and scarce blood flow. Abdominal CT scan showed a bulky right retroperitoneal mass. Three-dimensional CT imaging demonstrated spine, iliac bone, and long bones of limbs. The mass was excised successfully. After opening the sac it was noted to contain an incompletely developed fetus with grossly visible limbs, clearly discernible male genitalia, hairs, and a poorly formed head. The fetus was connected to the sac via an 8 cm cord-like structure. Microscopic examination of the mass revealed the presence of skin, cartilage, bone, intestine, and cysts with simple cuboidal epithelium. The use of CT scans enhanced the accuracy of pre-operative diagnosis. Identification of the vertebral column and the long bones of limbs are important indications for the diagnosis. Pathologically, fetus in fetu has many characteristics different from teratoma.

Key words: Computed tomography—Diagnosis—Fetus in fetu—Pathology—Abdominal mass

Fetus-in-fetu is a rare entity estimated to occur in 1:500,000 deliveries, with fewer than 100 cases reported worldwide [1]. Generally, fetus in fetu is a single parasitic twin, but there can be multiple fetuses-in-fetu sometimes [2]. It is predominantly retroperitoneal in 80% of cases, while reported uncommon sites include the oral cavity, sacrococcygeal region, and scrotum [3, 4]. We report a case of fetus-in-fetu in a 3.5-month-old boy with regard to its imaging and pathologic findings.

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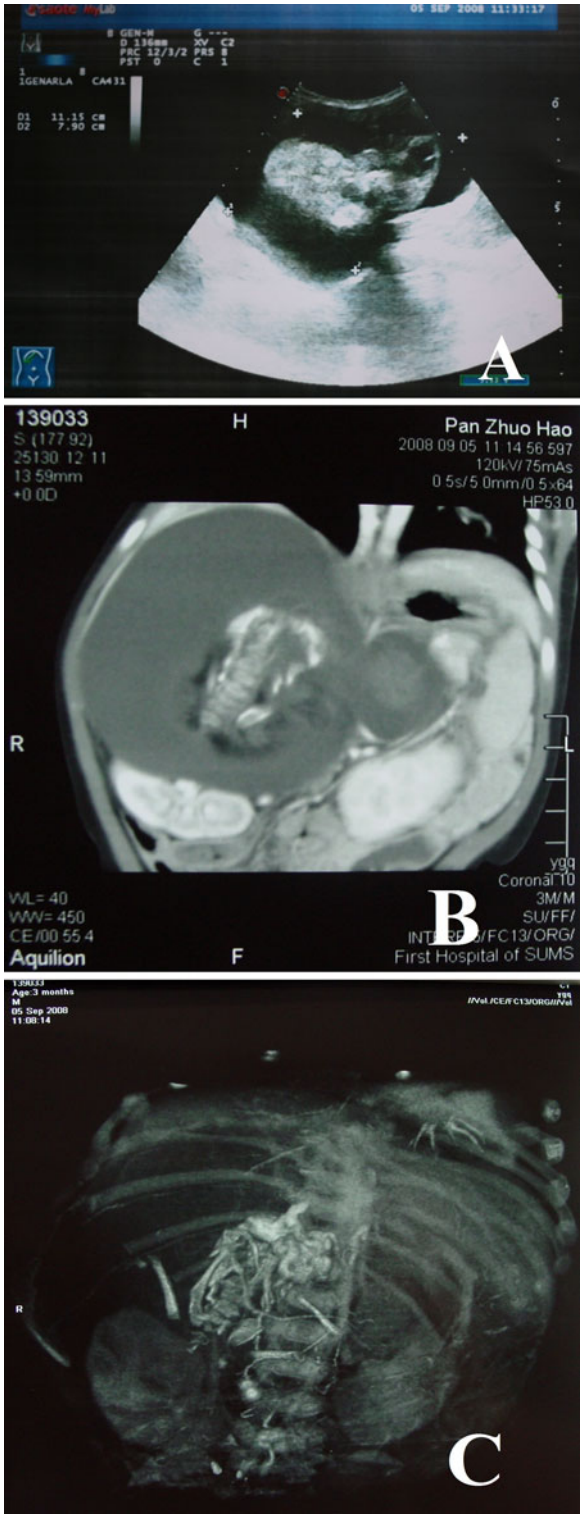
Case presentation

A 3.5-month-old boy was hospitalized because of an abdominal mass found by his parents accidentally. On physical examination, a smooth, firm, nontender mass was present in the right upper quadrant. Routine prenatal ultrasound examination failed to find the mass.

Abdominal ultrasonography revealed a large (11.5 × 8.5 × 7.9 cm), hyperechoic, heterogenous intra-abdominal mass with clear boundaries, and scarce blood flow. Patchy areas of calcifications were evident within the mass (Fig. 1A). The liver was compressed by the mass. Abdominal computed tomography (CT) scan showed a well-limited, 12.4 × 9.5 × 8.2 cm, apparently right retroperitoneal mass presenting bulky intra-abdominal growth, with the left part of the mass across the median line and the adjacent organs (e.g., liver, pancreas, and right kidney) compressed. The central mass was solid (containing fat and bony components), surrounded by liquid density (Fig. 1B). Three-dimensional CT imaging demonstrated spine, iliac bone, and long bones of limbs (Fig. 1C). Based on these imaging findings, a pre-operative diagnosis of retroperitoneal fetus-in-fetu was made.

Elective laparotomy was performed. A large retroperitoneal cystic mass was found over the upper pole of the right kidney (Fig. 2A, B). There were only some small vessels around the mass. The sac contained a clear fluid which was decompressed by accidental rupture. In toto excision of the mass was done. The post-operative course was uneventful and the patient was discharged on the 8th day postoperatively.

After opening the sac it was noted to contain an incompletely developed fetus with grossly visible limbs, clearly discernible male genitalia, hairs, and a poorly formed head. The fetus measured 10.0 × 8.5 × 7.0 cm, and was connected to the sac via 8 cm cord-like structure (Fig. 2C). Microscopic examination revealed the presence of fibrous connective tissues in the sac, and skin, cartilage, bone, intestine, and cysts coated with simple cuboidal epithelium in the fetus (Fig. 3).



◀**Fig. 1.** Preoperative studies. **A** Abdominal ultrasonography revealed a large mass with clear boundaries and scarce blood flow. Patchy areas of calcifications are seen in the internal mass. **B** Abdominal computed tomography (CT) scan showed a right retroperitoneal mass presenting bulky growth, the adjacent organs were compressed. The central mass was solid (containing spine), surrounded by liquid density. **C** Three-dimensional CT imaging demonstrated spine, iliac bone, and long bones of limbs.

After 1 year the patient has shown no recurrence or complications and is leading a normal healthy life.

Discussion

A fetus-in-fetu is a fetiform mass located within a basically normal fetus. There is controversy as to whether fetus-in-fetu is a distinct entity or represents a highly organized teratoma. Most investigators suggest that fetus-in-fetu is a pathologic entity that is distinct from teratoma [5]. A teratoma may have a slight potential for malignancy. However, malignant degeneration associated with fetus-in-fetu is extremely rare [6].

Classically the fetus is almost always anencephalic. The vertebral column and the limbs are present in almost all cases [5]. The lower limbs are more developed than the upper limbs. The presence of an axial skeleton is considered to be a distinctive feature to distinguish it from teratoma. However, in about 9% of cases of fetus-in-fetu, there is no vertebral column, particularly when the lesion is intracranial [7].

Before making a diagnosis of a fetus-in-fetu, one or more of the following characteristics should be considered: (a) enclosed within a distinct sac, (b) partially or completely covered by normal skin, (c) with grossly recognizable anatomic parts, and (d) attached to the autosite by a pedicle containing a few relatively large blood vessels [5]. This case had all of the above characteristics.

Ultrasonography is usually the first step for an abdominal mass. The presence of diffuse calcifications within a cystic mass should raise the suspicion of fetus-in-fetu [8]. As in this case, ultrasonography is difficult to differentiate between fetus-in-fetu and teratoma. The use of CT scans has since enhanced the accuracy of preoperative diagnosis [9]. Identification of the vertebral column, the long bones of limbs, are important indications for the diagnosis, which was the situation in the reported case. In particular, the three-dimensional CT imaging displayed the shape of fetus-in-fetu in a more intuitive way. Although radiologists think that three-dimensional CT may lose a lot of details, the surgeons prefer to use it to guide the surgery.

Complete excision of the mass is curative. Pathologically, the intra-abdominal fetus-in-fetu is usually contained in a complete sac, without any major vascular connections to the host [3, 5]. Besides the vertebral column, commonly found in fetus-in-fetu are dermal structures, limbs, gastrointestinal tract, and the central nervous system. Less commonly found are the gonads, adrenal glands, heart, the primitive respiratory unit, pancreas, and spleen. There were some cysts coated with simple cuboidal epithelium in the fetal pelvic cavity in

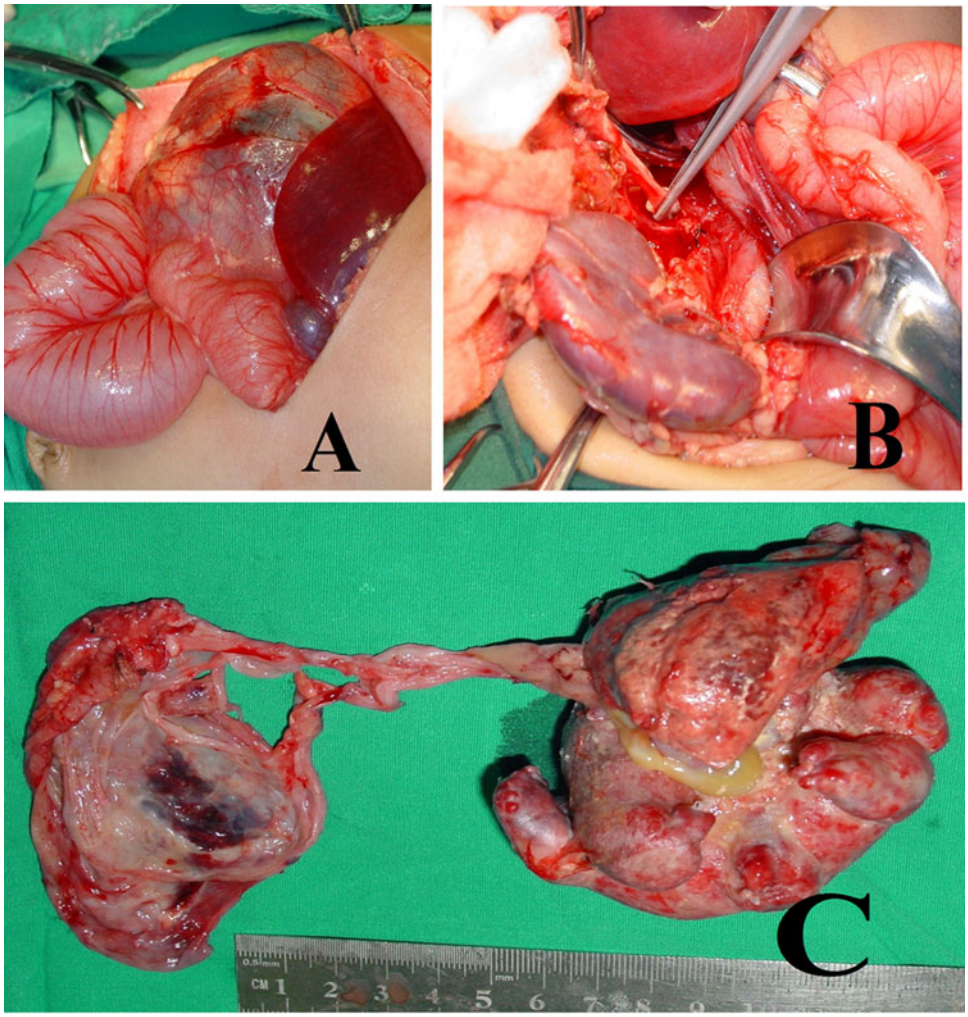


Fig. 2. Intraoperative pictures. **A** A large retroperitoneal cystic mass was found over the upper pole of the right kidney. **B** After the mass is removed, it can be clearly seen that the original mass was located between the liver, kidney, duodenum, hepatic flexure of colon, and the inferior vena cava. **C** The opened sac and an incompletely developed fetus were connected via a cord-like structure. The fetus shows grossly visible limbs, clearly discernible male genitalia, and a poorly formed head.

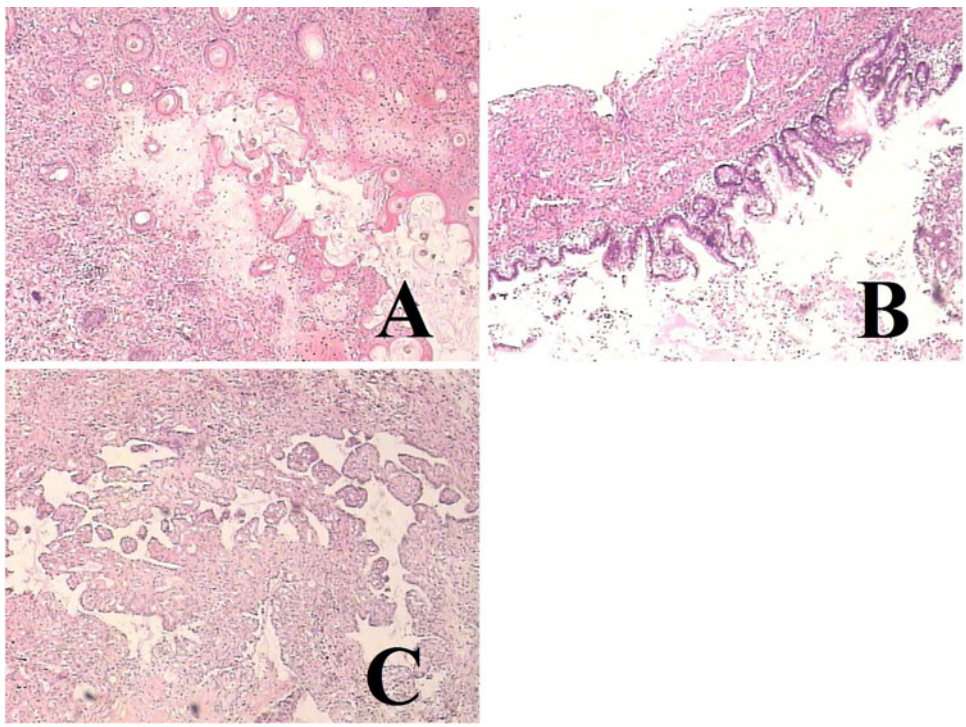


Fig. 3. Pathologic findings (HE × 40). **A** The skin with stratified squamous epithelium, hair follicles, and glands. **B** The intestine with all four layers. **C** Some cysts coated with simple cuboidal epithelium in the fetal pelvic cavity.

this case, and we suspect that may be a dysplastic bladder.

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