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Report of Fetus in Fetu in A Four-Year-Old Child

Fetus in fetu is a rare condition in which a fetiform calcified mass is often present in the abdomen of its host, a newborn or infant; the mass is considered as a parasitic twin originated from a diamnion-monozygotic pregnancy. Herein, we report on a new case of a 4-year-old child who was admitted for left upper quadrant abdominal mass without any symptom.

Keywords: teratoma, imaging, fetus in fetu.

Introduction

The term "fetus in fetu" (FIF) was first coined by Meckel in 18th Century. The first case of FIF was reported by Young in 1809.[1,2] FIF is most commonly diagnosed in infancy, as a retroperitoneal mass, and may be totally asymptomatic.[3] They are usually one in number, although more than one FIF has also been reported.[4]

Case Report

A 4-year-old boy presented with left upper quadrant (LUQ) abdominal mass. The mass was firm, non-motile and crossed the midline without tenderness. He had a normal weight and height for his age. His appetite was also normal. He did not have diarrhea or constipation. His liver and spleen were normal in size. No abnormalities were found in his biochemical and hematologic tests. Clinical differential diagnosis included neuroblastoma or nephroblastoma. Abdominal roentgenogram showed a space-occupying mass in LUQ displacing bowel gas to the right side. The mass contained calcified densities and structures similar to long bones (Figure 1). Abdominal ultrasound (Hitachi EUB 525, Japan) revealed a calcified mass with mixed echo and in LUQ with measured 140×160 mm. Abdomino-pelvic multidetector computed tomography (MDCT) (4-slice GE Light Speed CT Scanner, USA) was performed. Axial images and additional multiplanar reformatted views (MPR) in coronal and sagittal planes showed a heterogenous mass containing soft tissue, fat and calcified densities with apparent small and long bones resembling of fetal limbs, vertebral column and flat bone (Figures 2a-d). According to imaging data, FIF and teratoma were added to our differential diagnosis list. However, serum α-fetoprotein and β-HCG were negative.

The patient was operated. A retroperitoneal mass, located in front of pancreas and kidney at the base of the transverse mesocolon was resected. The mass had a well-defined capsule facilitated complete resection, was composed of hair, sebaceous materials and bony structures in gross appearance (Figure 3).

In pathological examination, vertebral column, limbs, cartilage, nerve fibers, full-thickness dermal elements, epithelium of urogenital and respiratory systems (Figures. 4a-c), muscle fibers, endocrine glands, vessels, hair, and also foreign body granulation tissue were detected. No evidence of immature components

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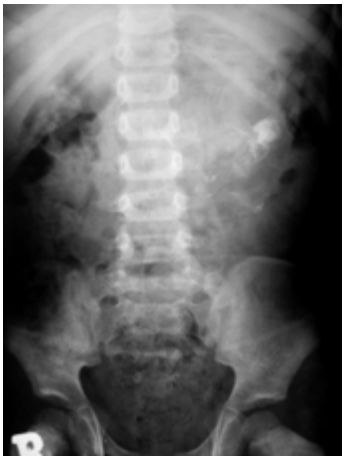

Fig 1. Plain abdominal x-ray shows a mass contain calcified and bony sructures and also fat densities in LUQ displacing bowel gas shadow.

in the resected mass, suggestive for immature teratoma, were seen.

Discussion

FIF with an incidence of one in 500,000 births, is a very rare entity.[1,5,6]

Many theories have been postulated to elucidate the exact pathegonesis of FIF. Though we cannot accurately mention the number of reported cases of FIF, the number should not be more than 100. It is postulated that FIF arises from inclusion of a diamnion-monozygotic twin within the bearer.[4,8]

Unequal division of totipotential cells of blastocyst may result in inclusion of the smaller cellular mass in the more mature embryo[1,4], where the parasitic twin is stopped to grow in the body of its partner.[9] Symptoms, if present, are related to mass effect and include abdominal distention, poor feeding, emesis, jaundice and dyspnea.[3]

Upper retroperitoneum is the most common site affected, albeit, cranial cavity, pelvis, scrotal sac, sacrococcygeal region, mesentry, right iliac fossa and oral cavity are also rare locations affected.[2,4,6,10,11]

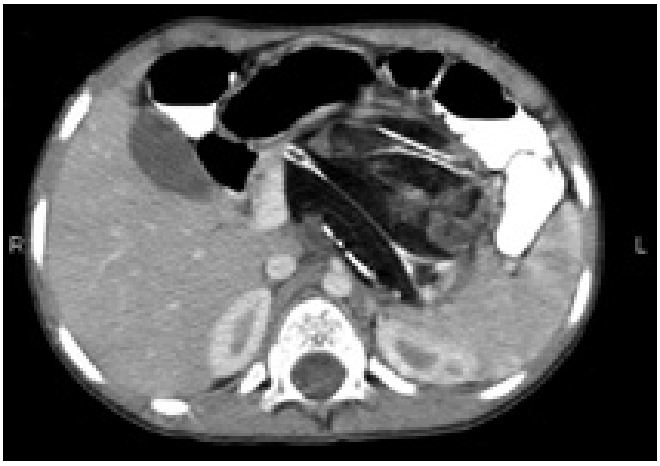

Fig 2a. Axial abdominal CT reveals a large Lt retroperitoneal mixed density mass anterior to Lt kidney. Fat and soft tissue densities and bony structures can be seen.

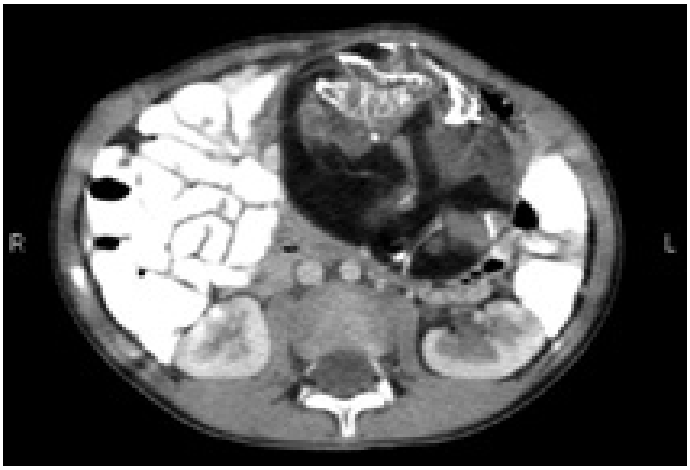

Fig 2b. Another section lower than 2a reveals bony elements of spinal column and also position of the mass anterior to the pancreas.

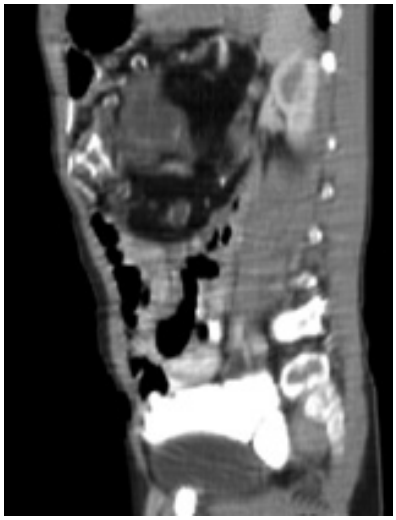

Fig 2c. Coronal reformatted view shows exact position and relation of the mass and also it is content.

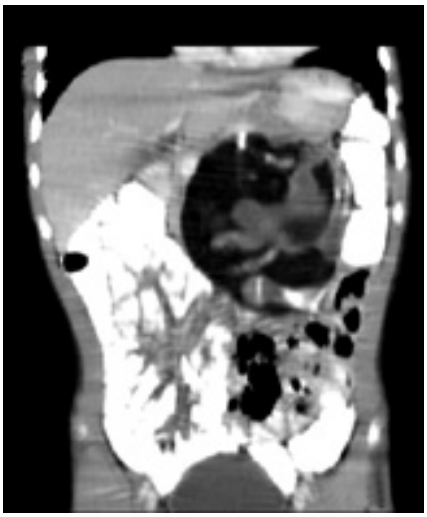

Fig 2d. Sagital reformatted view; fat and soft tissue densities and also bony elements are seen.

Presence of vertebral column which develops from the primitive streak and appropriate arrangement of other organs and limbs relative to the vertebral column are the diagnostic features of FIF and help to differentiated it from the highly differentiated teratomas.[7,8]

However, this entity is also differentiated from teratoma by its fetiform shape and metameric segmentation of the spinal axis.[4]

In a comprehensive review articles of 87 cases, 76 (83%) aged under 18 months, 47 (54.02 %) of patients were male, 35 (40.23%) were female and 5 (5.74%) had undetermined sex.[4]

The organs which could be identified in FIF included vertebral column in 91%, limbs (one to four) in 82.5%, central nervous system in 55.8%, gastrointestinal structures in 45%, vessels in 40%, and genitourinary structures in 26.5% of the studied FIF. The fetuses were always anencephalic.The fetuses varied from 4 cm to 24.5 cm in size and from1.2 g to 1,800 g in weight. In about 9% of cases of FIF, there was no vertebral column, even on pathologic examination.[4,12,13]

The mass is often enclosed within a membrane resembling amnion. A vascular anastomosis with the host vessels is also identifiable. Chromosomal study showed normal chromosomes, identical to the host, and the same blood group as their bearer. The serum α-fetoprotein may be elevated or normal in the host.[3,11]

FIF has characteristic imaging features: An abdominal plain x-ray shows a vertebral column and or bony structures within a soft tissue mass. Computed tomography is diagnostic and a mass comprising of fat collection around a central bony structures are the typical finding in FIF.[2]

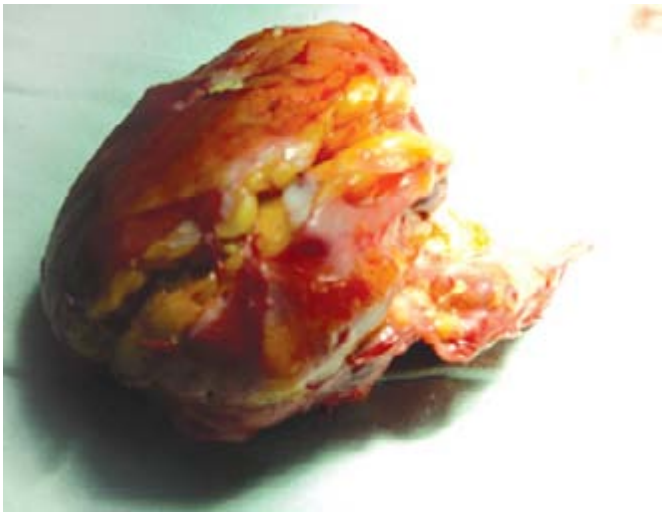

Fig 3. Surgical sample which shows membrane envelop the mass and also spinal column.

Evidence of axial skeleton formation in FIF can distinct it from teratoma. Differential diagnosis of FIF includes teratoma, which can contain any type of normal human tissue, the commonest of which is the ectoderm and its derivatives. In our case, the arrangement of the long bones of the fetal limbs around the axial bones and also other histologic findings compatible with FIF were nicely shown.

In multiple (twice the normal) slices which were taken from the resected mass, there was no evidence of malignancy or immature teratoma. The location of the mass was similar to that of other cases. However, in our case, the serum α-fetoprotein was negative; chromosomal study was not done. Contrast enhanced computed tomography can well-elucidate the extent and relation of the mass with its adjacent structures, which is necessary in surgery—the treatment of choice for FIF.[2]

After six months of surgery, no recurrence is noticed. The patient is in good condition.

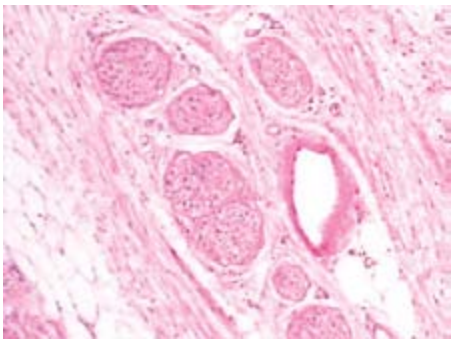

Fig 4a. Nerve bundles.

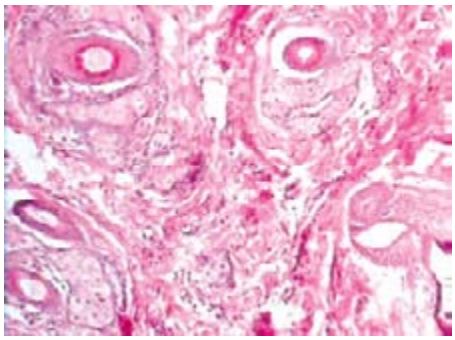

Fig 4b. Full thickness dermis and dermal elements especially hair follicles and epiderm.

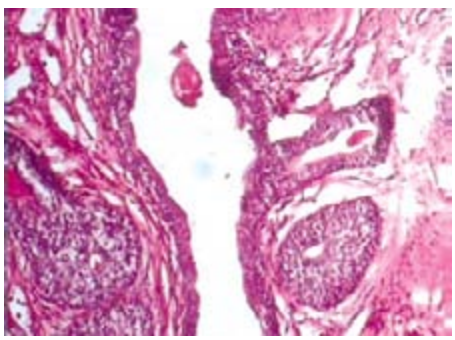

Fig 4c. Respiratory epithelium.

References

1. Borges E, Lim-Dunham JE, Vade A. Fetus in fetu appearing as a prenateal neck mass. J Ultrasound Med 2005; 24(9): 1313-1316.
2. Nagar A, Raut AA, Narlawar RS. Foetus in foetu. J Postgrad Med 2002; 48:133-134.
3. Hopkin KL, Dickson PK, Ball TF, Ricketts RR, O'shea PA. Abramovosky CR. Fetus in fetu with malignant recurrence. J Pediatr Surg 1997; 32: 1476-1479.
4. Hoeffel CC, Nguyen KQ, Phan H, Thruong NH, Nguyen TS, Thanhtran T, et al. Fetus in fetu: a case report and literature review. Pediatrics 2000; 105(6): 1335-1344.
5. Grant P, Pearn JH. Foetus in foetu. Med J Aust 1969; 1: 1016-1020.
6. Micheal J, Varanelli MD, Syed A, Jamal Bokhari MD, Fredely M, Kotai MB, et al. Case 55: fetus in fetu. Radiology 2003; 226: 517-520.
7. Willis RA. The borderland of embryology and pathology. 2nd ed. London: botterworths; 1962: 442.
8. Kajbafzadeh A, Baharnoori M. Fetus in fetu: Br J Ultrasound 2004; Supp 2:73.
9. Galatius-Jensen F, Rah DH, UHM IK. Foetus in foetu. Br J radiol 1965; 38: 305.
10. Taori KB, Khurana SD, Dhomne SP, Rathi V. Fetus in fetu: arare case. Ind J radiol imag 2003; 13(1): 85-87.
11. Eng HL, Chaung JH, Lee Ty, Chen WJ. Fetus in fetu: Case report and review of the literature. J pediatr surg 1989; 24: 296-299.
12. Luzzatto C, Talenti E , Tregnaghi A, Fabris S, Scapinello A, Gugliemi M. Double fetus in fetu: diagnostic imaging. Pediatr Radiol 1994; 24: 602-603.
13. Lee Ey. Foetus in foetu. Arch Dis child 1965; 40: 689-693.