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Case report

Fetus in fetu in an adult woman

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SUMMARY

Fetus in fetu (FIF) is an extremely rare condition in which malformed fetus is found most commonly in the retroperitoneum of living twin. It occurs in about 1 in 500 000 live births and less than 200 cases have been reported in medical literature. We report FIF in a 17-year-old woman unlike other cases which were usually detected in early age and in male gender. This is the eighth case of adult FIF and the first case of FIF in adult woman worldwide. Preoperative diagnosis of FIF on CT was made and planned for exploratory laparotomy. The excised malformed fetus in a sac was proven as FIF on the basis of histopathological examination. In view to avoid such late presentation, early detection of FIF with radiological imaging in clinically suspected case is recommended. Surgical excision is the ideal treatment even teratoma is the differential diagnosis.

BACKGROUND

Fetus in fetu (FIF) is a rare developmental abnormality in which a malformed fetus is found within the body of other twin. It was first described by Johann Friedrich Meckel in the late 18th century. Majority of cases have been described in neonates and children and only seven cases have been reported after the age of 15 years (table 1). The first reported case of FIF in an adult man, whose age was 47 years, is by Dagradi *et al*¹ All reported cases of adult FIF were in male gender. We aim to report this case since to the best of our knowledge this case is the first case of FIF in a female gender of 17 years age and reviewing the literature.

CASE PRESENTATION

A 17-year-old woman presented with abdominal lump for 5 years, which was gradually increasing in size. It was associated with pain in abdomen which was on and off in nature. She was also complaining of early satiety but there was no history of significant weight loss. There was no history of altered bowel habits and urinary symptoms. Her menstrual history was within the normal limits. There was no history of twin birth or teratoma in the family. Her general, physical and systemic examinations were within the normal limits. On abdominal examination, there was a mass involving the whole of the abdomen (figure 1). This abdominal lump was firm to hard in consistency, its surface was irregular, margins were ill-defined and it was not moving with respiration. With these clinical information except to say abdominal tumour, it was not possible to conclude a final diagnosis.

INVESTIGATIONS

On evaluation, beta-human chorionic gonadotropin (β -HCG), alpha fetoprotein and routine blood investigations were found within the normal limits. An abdominal contrast-enhanced computed tomography (CECT) scan showed a well-defined mass that measured approximately $25 \times 23 \times 15$ cm, extending from epigastrium to upper pelvis (figure 2). It was showing fat density areas, soft tissue and multiple calcified density components of various sizes and shapes resembling the shape of vertebrae, ribs and long bones. This mass was causing displacement and compression of adjacent abdominal viscera.

DIFFERENTIAL DIAGNOSIS

On the basis of clinical findings (slow growing abdominal lump with firm to hard consistency, irregular surface and ill-defined margins) and CECT findings (well-defined mass with fat density areas and multiple calcified density resembling shape of vertebrae, ribs and long bones), a clinical diagnosis of FIF was made preoperatively.

TREATMENT

On exploration, there was a large retroperitoneal encapsulated mass, extending from the under surface of the liver reaching up to the pelvic rim (figure 3). It was grossly displacing intra-abdominal viscera however no invasion was noted. The contents of the tumour consisted of hairs, mature bones and other body parts (figure 4). The contents were removed in toto and a part of cyst wall which was densely adherent to the mesenteric vessels was left behind in order to prevent devascularization of gastrointestinal tract. The remaining cyst wall was cauterised. On macroscopic examination, the mass measured $30 \times 16 \times 10$ cm and was composed of hairy cheesy material, multiple teeth and structures

Table 1 Fetus in fetu in adult cases published in between 1999 and 2016

Adult FIF cases reported by (authors)	Year of reporting	Age of patient at the time of diagnosis	Sex	Location
Shrivastava	1999	27 Years	Male	Retroperitoneum (RP)
Patankar	2000	16 Years	Male	RP
Awasthi	2001	30 Years	Male	RP
Masad	2001	27 Years	Male	RP
Lee	2005	39 Years	Male	Intraperitoneal
Sharma	2007	36 Years	Male	RP
Daga	2009	20 Years	Male	RP



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Figure 1 Showing abdominal lump with ill-defined margin.

resembling limb buds. On cutting section, it showed fatty, cartilaginous and bony areas along with another cystic area of size 8×7 cm. Microscopically, there was an admixture of different elements including neural, intestinal, cartilaginous and bone along with skin adnexal structures and adipose tissue. However, no immature elements were seen. A final diagnosis of FIF was given on the basis of histopathological findings.

OUTCOME AND FOLLOW-UP

Her postoperative period was uneventful. She has now been on our follow-up for the last 24 months with normal level of β-HCG and is doing well.

DISCUSSION

FIF is a condition, commonly occurs in retroperitoneal space (80%).² Others rare sites like sacrococcygeal region, intracranial, thorax, pelvis, scrotum, back and oral cavity have also been described in the literature.^{3–5} There are two theories for the development of FIF. One is parasitic twin theory in which parasitic malformed fetus formed inside the body of its host twin and share common blood supply. The parasitic twin is anencephalic with lack of many internal organs and it usually dies before birth.⁶ The other theory is that FIF is a highly differentiated form of teratoma.⁷ However, Willis (1953) emphasised the definition of FIF as a mass containing a vertebral axis along

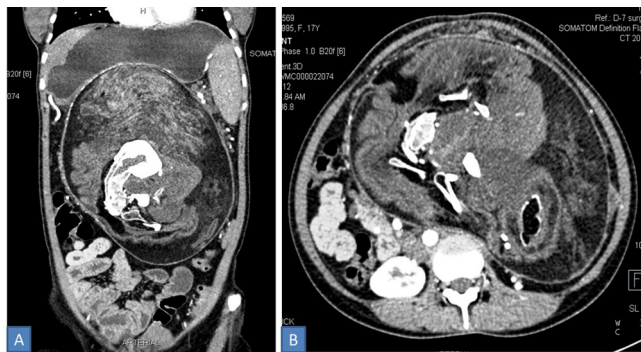


Figure 2 Abdominal contrast-enhanced computed tomography showing 25×23×15 cm mass with multiple calcified densities compressing the adjacent abdominal viscera.

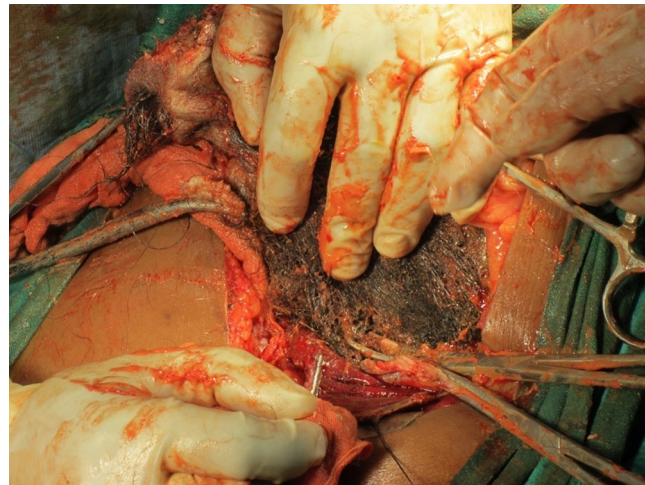


Figure 3 Showing encapsulated mass extending from the under surface of the liver reaching up to the pelvic rim.

with other organs or limbs.⁸ Recently, many cases have reported as FIF without the presence of vertebral column but features of advanced stage of organogenesis.⁹

Majority of the reported cases of FIF are found in infancy and neonatal age group unlike our case.^{10–14} Excluding the present case, only seven adult cases of FIF have been reported in the literature.¹⁵ Among adult cases of FIF, the oldest and the youngest case was 47 years and 20 years old respectively, while the age of present case was only 17 years.¹⁶ In older age, although diagnosis is more in favour of retroperitoneal teratoma the possibility of FIF cannot be ruled out. Moreover it is in fact very difficult to differentiate these two conditions. Willis in 1953 set the criteria to differentiate these two conditions that FIF contains a vertebral axis with organs and limbs arranged around it, whereas teratoma is an accumulation of pluripotent cells in which there is neither organogenesis nor vertebral segmentation.¹⁷ The preoperative diagnosis of FIF can be made on radiological findings.¹⁸ Plain abdominal X-rays may be helpful in diagnosis of a vertebral column and axial skeleton. A CECT would easily pick the presence of axial skeleton and fatty tissue around it as in our case. Complete excision of FIF with its all surrounding membrane is the treatment of choice because of the possibility of malignant transformation.¹⁹ The possibility of malignant transformation is more found if the



Figure 4 Specimen consists of hairs, mature bones and other body parts.

tissue of FIF remains left after surgery.²⁰ Although the prognosis for FIF is more favourable than for cystic teratoma, the presence of immature elements mandates close clinical, radiological and serological follow-up to detect recurrence. This case will be follow-up at every year in view to detect malignancy as some adherent tissue was left during surgery.

Patient's perspective

I was much worried about my abdominal lump, after operation I am feeling very well and my abdomen is now flat and my parents are also very happy. Thanks to all operating doctors.

Learning points

- ▶ Fetus in fetu (FIF) is a rare condition usually occurs in infancy and child age group.
- ▶ FIF in a 17-year-old woman is an extremely rare condition.
- ▶ This case is the eighth case of adult FIF and the first case of FIF in adult woman worldwide.
- ▶ Among all reported cases of adult FIF until, this case represents the largest size of FIF measured 36×16×10 cm.

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REFERENCES

- 1 Dagradi AD, Mangiante GL, Serio GE, *et al.* Fetus in fetu removal in a 47-year-old man. *Surgery* 1992;112:598–602.
- 2 Gupta SK, Singhal P, Arya N. Fetus-in-fetu: A Rare Congenital Anomaly. *J Surg Tech Case Rep* 2010;2:77–80.
- 3 Abdurraheem NT, Nasir AA, Abdur-Rahman LO, *et al.* Oral fetus-in-fetu: A case report. *J Pediatr Surg Case Rep* 2015;3:171–3.
- 4 Peng B LD. Fetus in fetu in the back. *Case Reports* 2012;2012:bcr0320126150.
- 5 Trairisilp K, Srisupundit K, Suwansirikul S, *et al.* Intracranial fetus-in-fetu with numerous fully developed organs. *J Clin Ultrasound* 2018;46:487–93.
- 6 Chua JHY, Chui CH, Prasad TRS, *et al.* Fetus-in-fetu in the pelvis. *Ann Acad Med Singap* 2005;34:646–9.
- 7 Basu A, Jagdish S, Iyengar KR, *et al.* Fetus in fetu or differentiated teratomas? *Indian J Pathol Microbiol* 2006;49:563–5.
- 8 Willis RA. The structure of teratoma. *J Pathol Bacteriol* 1935;40:1–36.
- 9 Kumar B, Sinha Akumar, Kumar T, *et al.* Is definition of “fetus in fetu” needs revision?: A case report and review of literature. *J Pediatr Surg Case Rep* 2016;12:24–6.
- 10 Karaman I, Erdoğan D, Ozalevli S, *et al.* Fetus in fetu: A report of two cases. *J Indian Assoc Pediatr Surg* 2008;13:30–2.
- 11 Hoeffel CC, Nguyen KQ, Phan HT, *et al.* Fetus in fetu: a case report and literature review. *Pediatrics* 2000;105:1335–44.
- 12 Tofigh AM, Kavyani A, Abdollahi SM, *et al.* Fetus in fetu: report of a case and literature review. *Int J Surg* 2008;6:e94–e96.
- 13 Sitharama SA, Jindal B, Vuriti MK, *et al.* Fetus in fetu: case report and brief review of literature on embryologic origin, clinical presentation, imaging and differential diagnosis. *Pol J Radiol* 2017;82:46–9.
- 14 Aldosari SH, Alhudaif J, Alsasi O, *et al.* Fetus in fetu with skeletal dysplasia and hyperparathyroidism. *J Pediatr Surg Case Rep* 2019;44:101185.
- 15 Prescher LM, Butler WJ, Vachon TA, *et al.* Fetus in fetu: Review of the literature over the past 15 years. *J Pediatr Surg Case Rep* 2015;3:554–62.
- 16 Daga BV, Chaudhary VA, Ingle AS, *et al.* Double fetus-in-fetu: CT scan diagnosis in an adult. *Indian J Radiol Imaging* 2009;19:216–8.
- 17 Kim OH, Shinn KS. Postnatal growth of fetus-in-fetu. *Pediatr Radiol* 1993;23:411–2.
- 18 Gangopadhyay AN, Srivastava A, Srivastava P, *et al.* Twin fetus in fetu in a child: a case report and review of the literature. *J Med Case Rep* 2010;4:96.
- 19 Hopkins KL, Dickson PK, Ball TI, *et al.* Fetus-in-fetu with malignant recurrence. *J Pediatr Surg* 1997;32:1476–9.
- 20 Yaacob R, Zainal Mokhtar A, Abang Jamari DZH, *et al.* The entrapped twin: a case of fetus-in-fetu. *BMJ Case Rep* 2017;2017:bcr-2017-220801.

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