DOI: 10.7860/IJARS/2022/53184.2772

Case Report

Radiology Section

# Radiologic Features of Foetus-in-foetu: A Case Report

MEENU AMAR[1], RENU YADAV[2]



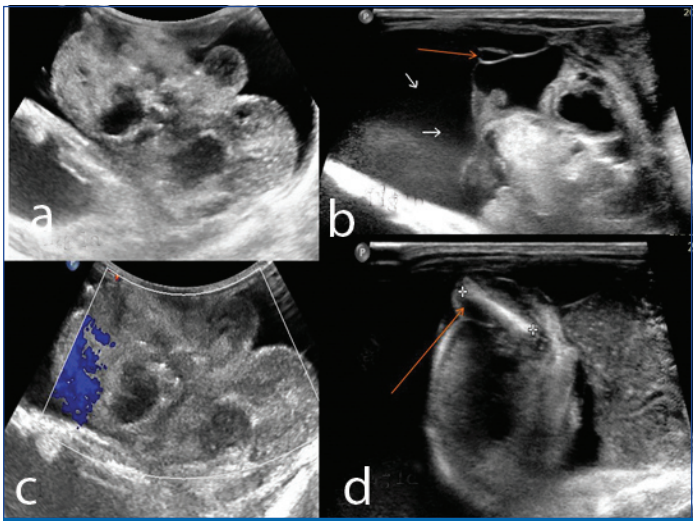

## ABSTRACT

Foetus-in-foetu is a rare pathology which occurs due to abnormal embryogenesis in a diamniotic monochorionic pregnancy. This unusual condition is characterised by development of foetal parts in a normally developing foetus. The authors present a case of two-day-old male neonate delivered successfully via normal vaginal delivery presented to Emergency Department with complaint of non tender abdominal distension from birth. There was no associated complaint like fever, vomiting or non passage of stools or urine. Antenatal scan showed large abdomino-pelvic mass occupying almost all the quadrants. On clinical examination, abdomen was soft and revealed a large, non tender, soft to firm large mass which required further evaluation. Bowel sounds were heard normally. Sonography and Computed Tomography (CT) was done for diagnostic evaluation which suggested possible diagnosis of Foetus-in-foetu confirmed on histopathological examination of the postoperative surgical specimen.


**Keywords:** Abnormal embryogenesis, Neonates, Teratoma

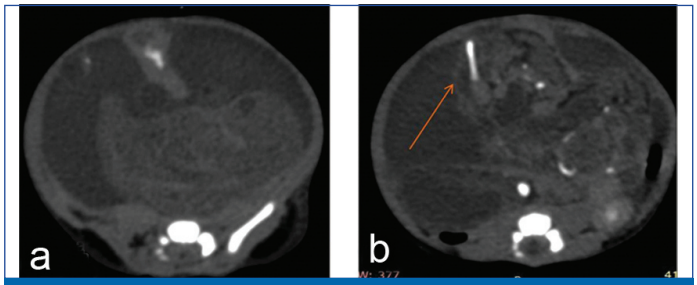
## CASE REPORT

A two-day-old male neonate was brought to the Emergency Department with complaint of non tender abdominal distension from birth. There was no associated fever, vomiting or non passage of stools or urine. Clinical examination revealed a large, non tender, soft to firm, abdomino-pelvic mass occupying almost all the quadrants. Bowel sounds were heard normally. After initial stabilisation, he was sent for an Ultrasonography (USG) of abdomen which revealed a large solid-cystic mass in the abdomino-pelvic cavity enclosed by a well-defined thin, echogenic membrane. The lesion was seen to displace and compress the right kidney posteriorly and small bowel loops were also seen displaced anteriorly and to the left of the abdomen. The solid component appeared as dysmorphic fetoid parts. A well-defined linear echogenic structure with distal acoustic shadowing resembling long bone (osseous element) was seen within the solid component. In addition, multiple ill-defined densely calcified structures were noted within the mass. Few globular echogenic areas were also seen within, without posterior acoustic shadowing, suggestive of possible fat component. No internal vascularity was elicited within the lesion on doppler evaluation. No free fluid was seen in peritoneal cavity [Table/Fig-1].

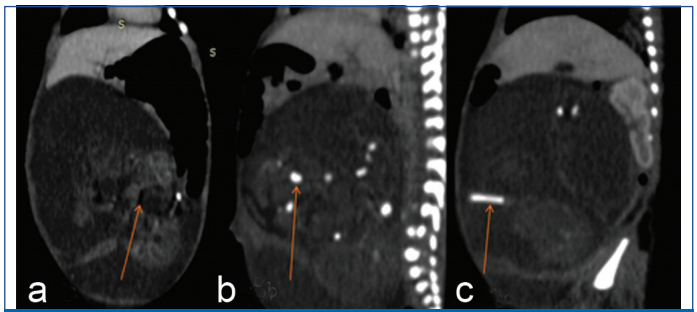



**[Table/Fig-1]:** a) Mass lesion with solid and cystic areas is most likely dysmorphic foetus; b) White arrows demonstrating the cystic component of the lesion and the long orange arrow showing the linear echogenic membranes within the lesion; c) No colour flow was demonstrated within the lesion following doppler examination; d) A linear echogenic structure with posterior acoustic shadowing, in keeping with the long bone (shown by orange arrow).

Based on the USG findings, Contrast Enhanced Computed Tomography (CECT) of abdomen was performed for better characterisation of the mass. Magnetic Resonance Imaging (MRI) could not be done due to logistic issues. The CECT examination revealed a large complex heterogeneous solid and cystic mass occupying almost the entire abdomino-pelvic cavity which appeared to be intraperitoneal. This mass lesion was seen anterior to the ascending and descending colon and was seen to displace the small bowel loops superolaterally to the left side and there was mass effect exerted on to the right kidney and it was compressed and displaced posteriorly. The solid component showed heterogeneous enhancement with interspersed areas of fat attenuation and multiple areas of coarse calcification. Partially formed vertebral elements and relatively well-formed long bones were identified within the mass [Table/Fig-2,3].




**[Table/Fig-2]:**a) Non contrast axial of the abdomen shows large complex mass lesion with solid and cystic areas within and solid areas resembling foetoid morphology; b) Linear hyperdense structure (HU 970) resembling that of a long bone denoted by arrow.




**[Table/Fig-3]:** a) Contrast enhanced coronal and sagittal reformatted images shows a large complex mixed density mass lesion with solid and cystic areas within. Non enhancing hypodense areas of fat attenuation within the lesion denoted by arrow. b) Multiple small hyperdensities of bone attenuation are also visualised within the lesion, may be ill-defined vertebral bones; c) Sagittal reformations denoting the same linear bony structure resembling that of long bone.

Based on these imaging findings, a provisional diagnosis of 'foetus-in-foetu' was made and the child was taken up for surgery. Intraoperatively the lesion was found to be in the retroperitoneum. On gross inspection, the mass lesion was well encapsulated covered by a pink membrane and was bluish in appearance [Table/Fig-4]. The mass was removed without any intraoperative complications.

**[Table/Fig-4]:** Gross specimen shows enclosed sac containing fat, solid soft tissue and malformed bony elements.

On cut section, serosanguineous fluid was obtained. Areas of fat, cartilage, glial tissue, primitive neuroepithelium, choroid plexus, skin and partially formed vertebral bones and long bones were seen on histopathology. Postoperative course went uneventful. The patient was discharged on day 10 of surgery and was suggested regular monthly follow-up with consultation from the surgical team. Regular tumour markers level was done as a part of follow-up.

## DISCUSSION

Foetus-in-foetu is a rare condition with reported incidence of 1 in 500,000 births [1]. Majority of cases present in infancy. It is an unusual condition which develops due to abnormal embryogenesis in a normally developing foetus which serves as the host for this parasitic twin. It usually presents as a malformed parasitic twin included in the body of the host twin due to unequal division of totipotent cells of a blastocyst, resulting in the inclusion of a small cellular mass in a more mature embryo, and thus forming a monozygotic, diamniotic twin pregnancy [2]. Another theory is that during ventral folding of trilaminar embryonic cyst (2nd and 3rd weeks of development), the diamniotic monochorionic twin is included within its host. This inclusion in the sister embryo is speculated to be due to persistent anastomosis of the vitelline circulation during development [3]. The most common site is retroperitoneum of the normal foetus. Other reported sites are cranial cavity, mediastinum, liver, oropharynnx and scrotum [4]. Preoperative diagnosis is usually made on imaging. It needs to be differentiated from its closest imaging differential diagnosis teratoma. The imaging modalities available are plain radiography, USG and cross-sectional imaging.

The diagnosis is usually made preoperatively on imaging. The first imaging investigation usually is a plain radiograph which is less sensitive and specific as it mostly reveals a large, partly calcified mass which may raise the suspicion of teratoma, especially in neonatal age-group. However, if vertebrae or any other mature skeletal structure is identified, it usually indicates the diagnosis of foetus-in-foetu. Advances in sonography have added accuracy in the diagnosis of foetus-in-foetu. Sonography shows large complex solid and cystic mass with foci of calcification. Further evaluation can be done by CECT which has higher sensitivity and specificity to identify the axial and appendicular skeletal elements. In addition, it is important to comment on the vascular supply to the parasitic twin which is usually derived from mesenteric vessels [5].

Usually, the parasitic twin tends to be acardiac and anencephalic. Various other structures reportedly identified in them, are axial and appendicular skeleton, intestine, neural, pulmonary, gonadal, pancreatic, and adrenal tissue [3]. Single malformed foetus-in-foetu was seen in retroperitoneum in our study. Single foetus is the most common presentation (88.4%) was seen in cases and most common location being the retroperitoneum (72%). However, multiple foetus have also been reported [4,5]. Majority of the patients present in neonatal age-group and infancy, although cases have been reported in adult population as well [6-8]. According to Wills criteria, if there is well visualisation of vertebral column or limbs, the diagnosis is definitely foetus-in-foetu. Visualisation of the vertebral column and limbs on imaging, gross specimen, and histopathological examination confirms the diagnosis [8,9].

As in this case, on imaging basis well-defined osseous linear structure resembling long bone and ill-defined vertebral bones were identified suggesting the strong possibility of foetus-in-foetu rather than the possibility of teratoma. Mature teratoma is a strong differential diagnosis, however absence of axial or appendicular skeleton rules out its possibility. Although Wills criteria helps in differentiating foetus-in-foetu from the mature teratoma, sometimes it is difficult to differentiate both of these, therefore as per the previous literature the management of these follow the same guidelines that is complete surgical resection and follow-up as the teratoma carry 10% malignancy rate [3,9].

Malignant transformation is rare in foetus-in-foetu [10,11]. Hopkin KL et al., stated that five-year-old boy had a retroperitoneal Foetus-in-foetu and who later developed a right abdominal mass which proved to be a teratoma with malignant components necessitating chemotherapy [11]. Associated teratomas and rarely a coincidental yolk sac carcinoma has been reported along with foetus and fetu [3,11,12] Therefore, follow-up every two years with the help of imaging and serological markers i.e., alpha fetoproteins or beta-human chorionic gonadotrophin levels is recommended in the patients diagnosed with foetus-in-foetu. Tumour marker surveillance and imaging should be done for these patients to avoid any malignancy in the follow-up protocol [12].

## CONCLUSION(S)

Foetus-in-foetu is a rare benign condition. There is always a clinical and imaging dilemma on the differentiation between foetus-in-foetu and teratomas as the latter carries malignant potential. Early surgical exploration is needed to avoid maturation and growth of developing immature organs. Hence, prompt and correct diagnosis coupled with close follow-up after every two years should be done in these cases.

## REFERENCES

[1] Hoeffel CC, Nguyen KQ, Phan HT, Truong NH, Nguyen TS, Tran TT, et al. Fetus in fetu: a case report and literature review. Pediatrics. 2000;105:1335-44.
[2] Gurses N, Bernay F. Twin fetuses-in-fetu and a review of the literature. Z Kinderchir. 1990;45:319-322.
[3] Prescher LM, Butler WJ, Vachon TA, Henry MC, Latendresse T, Ignacio RC. Fetus in fetu: Review of the literature over the past 15 years. J Pediatr Surg Case Rep. 2015;12:554-62.
[4] Sharma S, Gupta P K, Regmi B, Gupta A, Lamichhane U. Fetus in fetu in an adult female and brief review of literature. Case Reports in Radiology. 2021;2021:6660277.
[5] Gunaydin M, Celik FC, Tander B, Bozkurter AT, Sullu Y, Baris S, et al. Two cases of fetus in fetu. J Pediatr Surg. 2011;46:e09-12.
[6] Spencer R. Parasitic conjoint twins: external, internal (fetus in fetu and tertaoma), and detached (acardiac). Clinical Anatomy. 2001;14:428-44.
[7] Aoki K, Matsumoto Y, Hamazaki M, Sano M, Fukumoto K, Fukaya T, et al. MRI reveals fetus in fetu in the mediastinum. Pediatr Radiol. 2004;34:1017-19.
[8] Willis RA. The Borderland of Embryology and Pathology. Butterworths, Washington DC 1962.
[9] Harigovind D, Babu SP H, Nair SV, Sangram N. Fetus in fetu-a rare developmental anamoly. Radiol Case Rep. 2018;14:333-36.

[10] Phatak SV, Kolwadkar PK, Phatak MS. Fetus in fetu: A case report. Indian J Radiol Imaging. 2003;13:93-94.
[11] Hopkins KL, Dickson PK, Ball TI, Ricketts RR, O'Shea PA, Abramovosky CR. Fetus in fetu with malignant recurrence. J Pediatr Syrg. 1997;32(10):1476-79.
[12] Gilbert-Barness E, Opitz JM, Debich-Spicer D, Mueller T, Arnold SR, Quintero R. Fetus-in-fetu form of monozygotic twinning with retroperitoneal teratoma. Am J Med Genet A. 2003;120A:406.

**PARTICULARS OF CONTRIBUTORS:**
1. Radiologist, Department of Radiodiagnosis, Vardhman Mahavir Medical College and Safdarjung Hospital, South West Delhi, Delhi, India.
2. Senior Resident, Department of Radiodiagnosis, Vardhman Mahavir Medical College and Safdarjung Hospital, South West Delhi, Delhi, India.

**NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:**
Renu Yadav,
RZ-6C, Gali No. 30, Indira Park, Palam Village, Dwarka, South West Delhi,
Palam, Delhi-110045, India.
E-mail: DOC.RENU0803@GMAIL.COM

**PLAGIARISM CHECKING METHODS:** [Jain H et al.]
- Plagiarism X-checker: Nov 10, 2021
- Manual Googling: Jan 01, 2022
- iThenticate Software: Mar 05, 2022 (12%)

**ETYMOLOGY:** Author Origin

**AUTHOR DECLARATION:**
- Financial or Other Competing Interests: None
- Was informed consent obtained from the subjects involved in the study? Yes
- For any images presented appropriate consent has been obtained from the subjects. Yes

Date of Submission: **Nov 07, 2021**
Date of Peer Review: **Dec 29, 2021**
Date of Acceptance: **Jan 01, 2022**
Date of Publishing: **Apr 01, 2022**