



Fetus in Fetu as a Retroperitoneal Mass in Infant: A Rare Case Report

TB Odih Rhomdani Wahid*; Muhammad Ilham Herzoni; Alfath-hu Rahmat

¹Department of Pediatric Surgery, Arifin Achmad General Hospital, Faculty of Medicine, Riau University, Indonesia.

²Department of Pediatric Surgery, Arifin Achmad General Hospital, Faculty of Medicine, Riau University, Indonesia.

³Surgical Resident of General Surgery, Arifin Achmad General Hospital, Faculty of Medicine, Riau University, Indonesia.

*Corresponding Author(s): TB Odih Rhomdani Wahid

Department of Pediatric Surgery, Arifin Achmad General Hospital, Faculty of Medicine, Riau University, Indonesia.

Tel: +628117574599;

Email: tubagus7606@lecturer.unri.ac.id

Received: Aug 10 2024

Accepted: Sep 28, 2024

Published Online: Oct 04, 2024

Journal: Annals of Pediatrics

Publisher: MedDocs Publishers LLC

Online edition: <http://meddocsonline.org/>

Copyright: © Rhomdani Wahid OTB (2024).

This Article is distributed under the terms of Creative Commons Attribution 4.0 International License

Keywords: Fetus in fetu; Mass; Retroperitoneal.

Abstract

Background: Fetus In Fetu (FIF) is identified by the appearance of a partially formed fetus inside a twin which is an extremely rare congenital pathology. This rare disease has less than 200 reported cases globally. FIF can arise from a deviation in the twinning process. It usually occurs within the abdomen, often in the retroperitoneal region, and typically presents during infancy as an abdominal mass. FIF rarity remains a diagnostic challenge and diverse clinical symptom. The recommended treatment is complete surgical removal of the mass. Early intervention provides a good prognosis.

Clinical Description: A female infant was taken to the emergency department by her parents due to her enlarged abdomen. Physical examination discovered a cystic mass in the left abdominal cavity.

Management & Outcome: Surgery was performed with an exploration of the abdomen. In the retroperitoneum, we found a well-encapsulated mass containing the left kidney. To preserve vital structures, cautious dissection was performed and removed the mass successfully without rupture. The patient recovered uneventfully after surgery.

Conclusion: FIF is diagnostically challenging and often requires laparotomy exploration to release the mass. For favorable result, surgical excision is crucial especially to avoid probable complications.

Introduction

Fetus in Fetu (FIF) in children manifests as an abdominal mass. It is a rare congenital anomaly, mostly during infancy. It was described first in the past 18th century by Johann Friedrich Meckel. Less than 200 cases of FIF have been reported globally, with a frequency of 1 in 500,000 births [1-5]. The abnormality arises in the retroperitoneal of the normal fetus's body

in about 80% of cases and other areas such as the sacral, pelvis, and thorax region [3,6]. The cause of FIF is still debated. One theory suggests that FIF occurs when within its twin trapped a malformed fetus, relying on the twin's blood supply in the case of monozygotic monochorionic diamniotic twin pregnancies [2,4-7].



Cite this article: Zvekic M, Herbert M, Morales A, Softic S. Growth Hormone Treatment Normalized Liver Enzymes in an Adolescent with Obesity and Short Statute. *Ann Pediatr.* 2024; 7(2): 1146.

FIF typically impacts nearby structures due to mass effect and appears as localized swelling. Imaging studies can be made to diagnose the FIF and confirmed with exploratory laparotomy, during which the entire mass is removed surgically [3,7]. Highly differentiated teratoma can be considered as FIF [8]. Discerning a FIF from teratomas is based on the presence of the vertebral axis, which plays a crucial role in differentiation [3]. Early diagnosis as early intervention results in a better prognosis [9]. While preoperative diagnosis relies on radiological discovery, a definitive diagnosis is established post-operative [10]. We report an infant with a rare mass that was managed successfully.

Clinical description

A 1-month-old baby girl was brought by her parents to the emergency department due to a 3-week history of abdominal enlargement. The child had experienced five episodes of non-bilious vomiting. The baby was born at full term to a 32-year-old primipara via cesarean section delivery due to cephalopelvic disproportion. Her birth weight was 2800 grams. During the neonatal period, no significant complications were found. The mother did not have any known medical conditions before or during her pregnancy. She was a non-smoker and was only taking prescribed prenatal supplements.

During the physical examination, a cystic mass was palpable in the left abdomen region, leading to abdominal distention [see Figure 1A]. The routine blood tests came back normal. A plain babygram radiograph revealed a heterogeneous mass in the abdomen at the left quadrant, pushing the intestine to the contralateral side [see Figure 1B].

Management and Outcome

During the elective laparotomy exploration, a transverse incision was made to identify a large encapsulated mass in the retroperitoneal region containing the left kidney [see Figure 2A]. To preserve vital structures careful dissection was performed. The mass was removed in toto without rupture [see Figure 2B&C]. As the left kidney was located in the pelvic region, a nephropexy of the left kidney was performed against the psoas muscle.

We examined the mass after being resected. Macroscopically, it was embryonic with a sac-like membrane attached and weighed 800 grams. Sections showed precursors of a solid organ, a spine, and legs [see Figure 3]. Based on the findings of laparotomy exploration a diagnosis of FIF was made. Histopathological examination revealed skin, fibrous bone, adipose tissue, skeletal muscle, vascular tissue, and primitive neuroepithelial elements. In the neonatal intensive care unit, the patient's postoperative course was uneventful and discharged in stable condition.

Discussion

Fetus in Fetu (FIF) incidence is a slight male majority with a 2:1 ratio male-to-female [6,7]. The existence of a vertebral axis (cranium, spine, sternum, ribs) and the presentation of limbs or solid organs forcibly suggest the FIF presence [1,9]. This indicates that the fetus after gastrulation has proceeded through a primary phase, involving tube formation, metamerization, and symmetrical neural development around this axis [6]. FIF in our female infant revealed precursors of limbs and solid organs.

A FIF cannot survive independently and remains a living tissue within the host, receiving nutrition from the host. Our patient showed clinical signs and symptoms mainly due to pres-

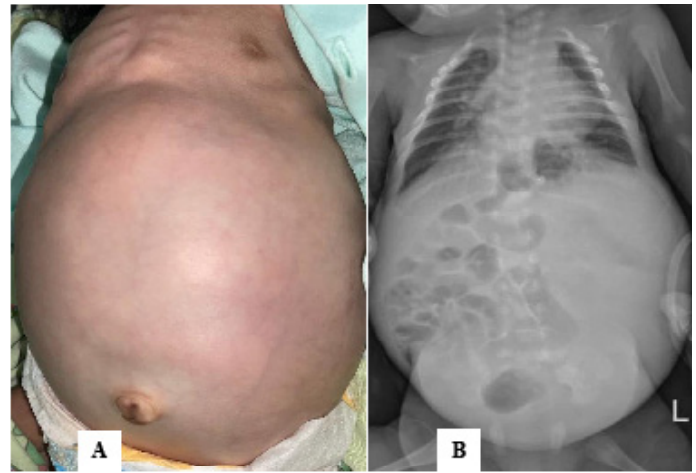


Table 1: (A) Abdominal distension. (B) Babygram X-Ray.

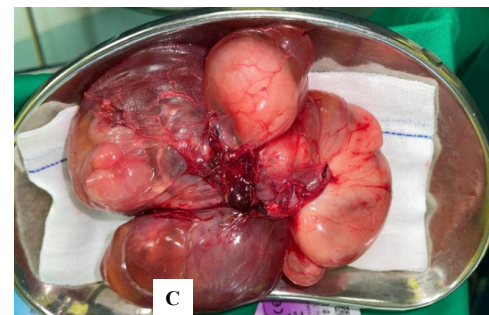
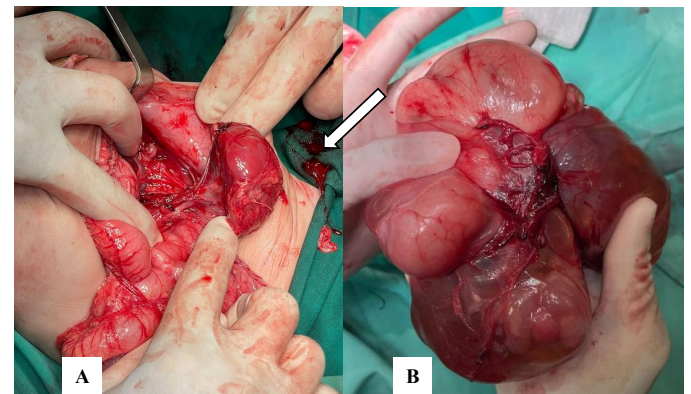


Table 2: (A) Mass contains the left kidney (arrow). (B&C) Encapsulated mass.



Table 3: Precursors of a solid organ, an intestine, and legs.

sure from the mass, including abdominal distension, painless mass, and vomiting. Upon physical examination, the abdomen was swollen and supple, and there was a mass palpable in the abdomen. The mass had a medium texture, clear borders, a smooth surface, and poor mobility [8].

At least one of the following criteria is required to diagnose the FIF: A visible cyst; normal skin covered partially or completely; anatomical structures grossly recognizable; a small number of relatively large blood vessels connection to the host; and positioning next to where conjoined twins connected or attached to the neural tube or the gastrointestinal tract [2,6,7]. Diagnosis of FIF has been reported in 16.7% of cases preoperative. In our case, most of the criteria for diagnosis were met and the diagnosis was made intraoperatively [4].

Visible of the vertebral column and long limb bones from a plain radiograph can be used to diagnose FIF [4]. However, plain radiographs have low resolution, and only space-occupying lesions cast a shadow of soft tissue denseness and mass if the FIF is buckled in the body. The parasitized fetal bone may not be completely calcified due to retroperitoneal organs, intestines, fat, etc. imaging overlapping. Therefore, to distinguish the spinal images it is difficult at this time. In our case, only space-occupying lesions were shown from radiographic examinations [8].

The encapsulated mass is surgically removed from the retroperitoneum area to prevent further compression. From the isolation and removal process, it is important to preserve the unity of the mass and its covering [1]. The vitelline circulation originates in the retroperitoneum and the superior mesenteric artery developed, which is embryonically located in the retroperitoneum [3,6,7]. Therefore, in our case, we completely removed the mass located in the retroperitoneal region with an exploratory laparotomy.

The mass size relative to the blood supply can range from 13 to 2000 grams. In this patient, the weight of the FIF was 800 grams [4]. A thin fibrous capsule enclosed by a FIF enfolded by a single layer of epithelium or squamous epithelium, making the tumor evident around the organs and tissues [6]. In general, the lower limbs development is often superior to that of the upper limbs, so in this patient we can observe the primordial leg from the cut section [8].

Conclusion

The initial diagnosis of our case presented some challenges. It is crucial to perform prompt surgical excision to prevent potential complications. Although reports have shown positive surgical outcomes after excision, more data is needed to determine long-term results. Early recognition of FIF is challenging, and to establish reliable preoperative diagnostic methods needs further research.

References

1. Agarwal A, Agrawal M, Ahuja S. Fetus in fetu in a 1-year-old female: A case report. *J Pediatr Surg Case Rep.* 2024; 106: 1-16.
2. Malwade S, Mane S, Sneha N, Agarkhedkar S. Fetus in fetu. *Medical Journal of Dr DY Patil Vidyapeeth.* 2023; 16(5): 807-10.
3. Ravishankar N, Sheeladevi CS, Pazhayattil J. Fetus in Fetu as a suprarenal mass in a neonate – a rare and perplexing entity. *Autops Case Rep.* 2022; 12: 1-5.
4. Hasan B, Ebrahim M. Fetus in Fetu: A Case of Vanishing Triplet Phenomena. *Cureus.* 2022; 14(10): 1-7.
5. Kazia A, Mathews S, Vasu P, Krishnadas S. Fetus in fetu: A rare differential diagnosis for an antenatally identified ultrasonographic intra-abdominal mass. *Indian Pediatrics Case Reports.* 2022; 2(1): 29-31.
6. Cantarero MC, Osejo Cantarero AL, Mendieta LR. The First Case of Fetus in Fetu in Nicaragua: The Management Experience of the Pediatric Neurosurgery Team. *Cureus.* 2023; 15(1): 1-5.
7. Jihwaprani MC, Mousa AA, Mohamed AA, Alkouz Y, Bahlawan IH. Fetus-in-Fetu: A Differential Diagnosis of Neonatal Fetiform Encysted Abdominal Mass. *Cureus.* 2023; 15(1): 1-7.
8. Xiaowen M, Lingxi C, Song L, Shengbao P, Xiaohong Y, et al. Rare Fetus-in-Fetu: Experience from a Large Tertiary Pediatric Referral Center. *Front Pediatr.* 2021; 9: 1-8.
9. Puranik RU, Joshi P, Jahanvi V, Handu AT, Puli KR. Antenatally detected fetus in fetu case report. *BJR|case reports.* 2023; 9(3): 1-4.
10. Lu T, Ma J, Yang X. A rare case of fetus in fetu in the sacrococcygeal region: CT and MRI findings. *BMC Pediatr.* 2021; 21(575): 1-6.