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Prenatal diagnosis of well-developed fetus in fetu with spontaneous movement in a resource-limited setting: A case report

Bella Stevanny [a,*], Putri Mirani [b,**], Elsye Ruth Frida Thene [c], Cindy Kesty [a]

[a] Department of Obstetrics and Gynecology, Dr. Mohammad Hoesin General Hospital/Faculty of Medicine, Universitas Sriwijaya, Palembang, Indonesia
[b] Division of Maternal-Fetal Medicine, Department of Obstetrics and Gynecology, Dr. Mohammad Hoesin General Hospital/Faculty of Medicine Universitas Sriwijaya, Palembang, Indonesia
[c] Department of Radiology, Prof. Dr. W.Z. Johannes Hospital, Kupang, Indonesia

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ABSTRACT

Fetus in fetu (FIF) is a rare congenital anomaly characterized by the presence of a parasitic monozygotic twin encased within the body of its host twin. Because FIF is asymptomatic throughout pregnancy, it is mainly diagnosed in children with an abdominal mass after birth. In the case reported here, at 38–39 weeks of gestation, a 33-year-old woman (gravida 4, para 3) was referred for routine obstetric ultrasonography. Fluid accumulation was identified along with calcification resembling two well-developed legs and trunk with undifferentiated organs inside. Slight spontaneous movement of the legs was observed. The fetus was delivered based on the presumed diagnosis of FIF. Postnatal sonography and computed tomography (CT) supported the diagnosis. The neonate underwent surgical excision of the tumor and was discharged on the eighth postoperative day. Ultrasound can be used to provide accurate prenatal diagnosis of FIF. Early diagnosis is important to improve outcomes.

1. Introduction

Fetus in fetu (FIF), also known as cryptodidymus, is a very rare clinical entity, with fewer than 200 cases reported worldwide. The term, meaning fetus within fetus, was first used in the late 18th century, by Johann Friedrich Meckel [1]. Because FIF is asymptomatic throughout pregnancy, it is mainly diagnosed in children with an abdominal mass after birth. By that time, the tumor impact on nearby organs has already resulted in considerable morbidity to the host twin [2]. Early diagnosis is crucial as a significant rise in negative health outcomes, such as failure to thrive and organ dysfunction, has been observed among those who receive a diagnosis at a more advanced stage of the disease[3]. Prenatal diagnosis will allow the delivery to take place in a hospital with the necessary resources to manage any complications that may arise after birth, as well as the ability to perform surgery to remove the tumor shortly after birth [4].

Ultrasonography can provide a preliminary diagnosis of FIF, but magnetic resonance imaging (MRI) is the preferred imaging method for a more accurate diagnosis, treatment plans, and prognosis. Unfortunately, MRI is frequently unavailable in regions with limited resources [5]. Only a limited number of cases (7%) of FIF have been reported to be diagnosed prenatally [6]. In this report, we describe a case of well-developed intraabdominal FIF identified by prenatal ultrasonography in a community hospital where MRI is not available. This case has been reported in line with the CARE 2013 guidelines [7]. An abstract describing this case was presented at the 31st World Congress on Controversies in Obstetrics, Gynecology & Infertility (COGI) as an e-poster.

2. Case Presentation

A 33-year-old woman (gravida 4, para 3) and a history of caesarean section was referred for an obstetric ultrasonography scan at 38–39 weeks of gestation. There was no family history to suggest congenital abnormalities, and there was no reported history of infectious diseases, radiation exposure, prenatal drug usage, or consanguineous marriages. The antenatal sonography showed a fetus corresponding to 38 weeks of gestation with a heart rate of 131 bpm and an estimated weight of 3940

Abbreviations: CT, computed tomography; FIF, fetus in fetu; MRI, magnetic resonance imaging.

* Corresponding authors at: Dokter Muhammad Ali, Sekip Jaya, Palembang, South Sumatra 30114, Indonesia.
** Corresponding authors.
E-mail addresses: bellastevanny@student.unsri.ac.id (B. Stevanny), putrimirani@fk.unsri.ac.id (P. Mirani).

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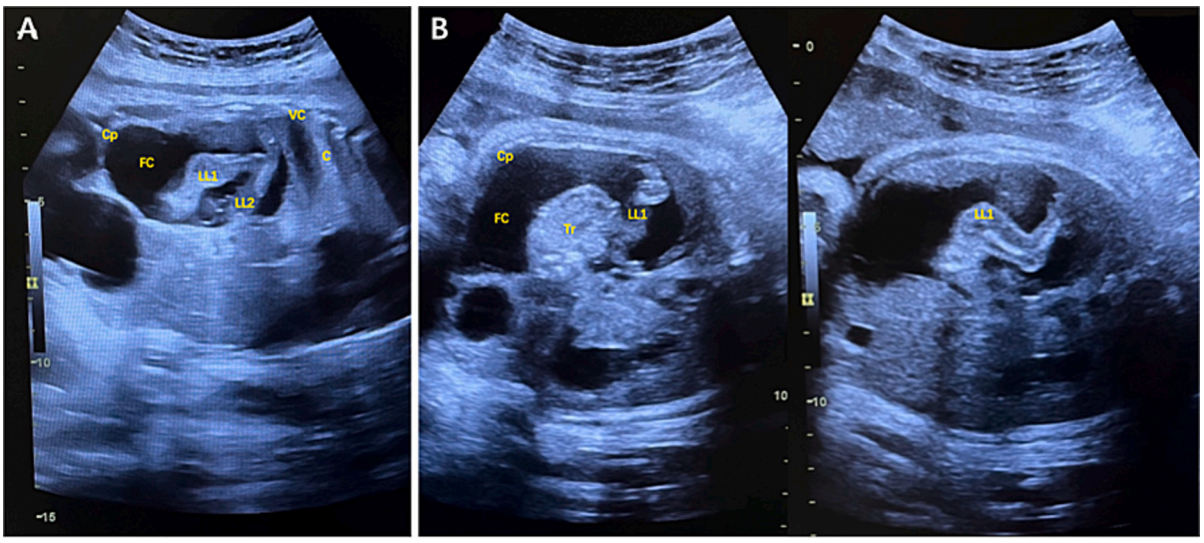

Fig. 1. Prenatal ultrasonography showed an intraabdominal encapsulated heterogenous mass suspected of FIF. (A) Sagittal view showing solid mass and calcification resembling fetal lower limbs (LL1 and LL2). (B) Transverse view showing the trunk (Tr) and lower limb of the parasite fetus. Cp: capsule; FC: fluid collection; VC: vertebral collumns of host fetus; C: costae of host fetus.

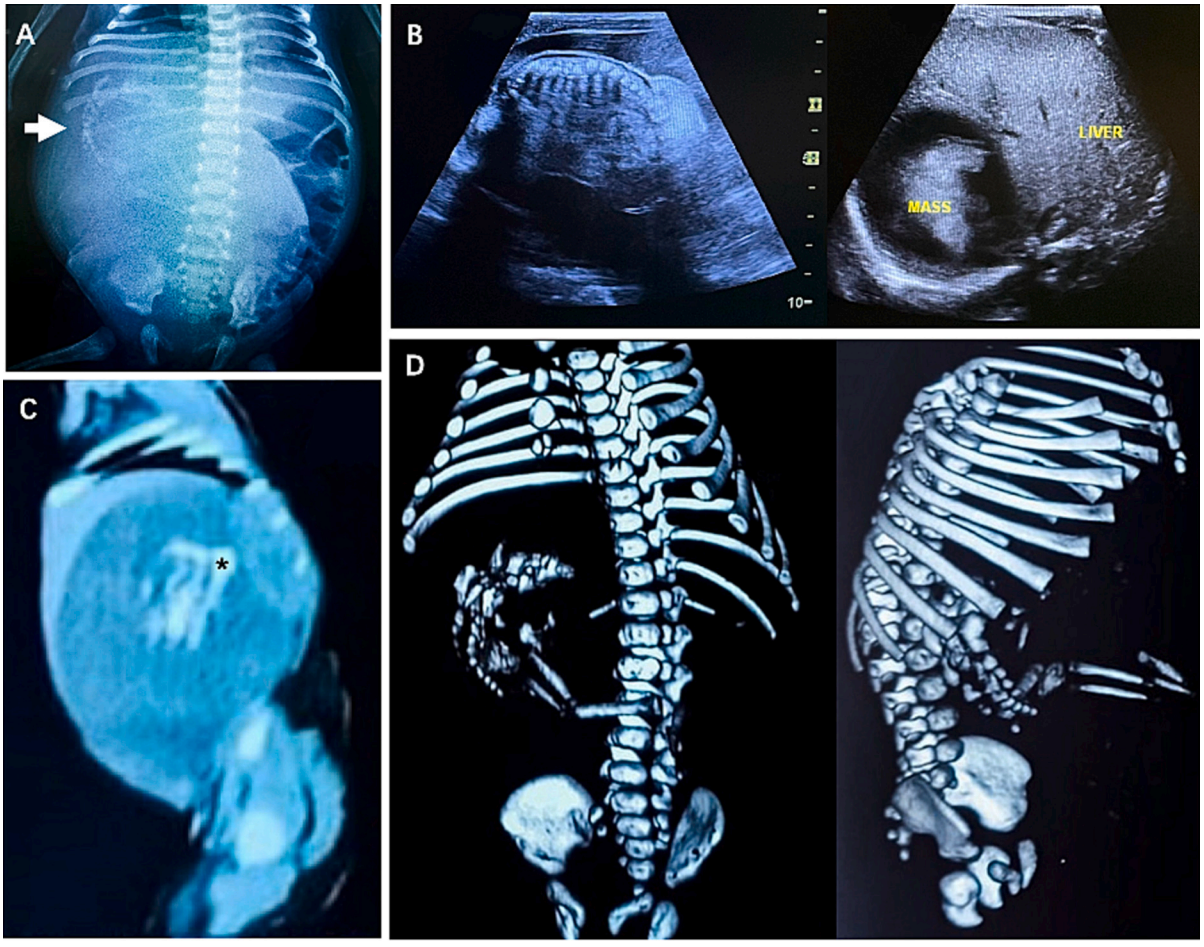

Fig. 2. The radiographic, ultrasonographic, and computed tomographic (CT) image of the abdominal region revealed the presence of a complex soft-tissue mass with bony components. (A) Abdominal X-ray showed calcified mass in the right hemiabdomen which is pressing on the intestines. (B) Postnatal abdominal ultrasonography showed a retroperitoneal cystic mass in the right hemiabdomen containing an incomplete fetus, which is pressing on the liver. (C) Sagittal CT scan. In front of the right kidney, there is a soft tissue mass with calcified opacities corresponding to fetal costae. (D) Three-dimensional reconstruction from the abdominal CT scan demonstrated the spatial relationship between autosite's lumbar spine and pelvis with visualized portion of the fetus in fetu's skeleton including vertebral columns, costae, pelvic bones, and bones of the lower extremity.

± 576 g. Hydrocele in the left scrotum was also identified. In the host fetus's right hemiabdominal cavity, there was a 10.2 × 8 × 8.2 cm cystic mass consisting of fluid collection as well as a 9–10 cm solid mass with calcification resembling two well-developed legs and trunk with undifferentiated organs inside (Fig. 1). Slight spontaneous movement of thelegs of the FIF was detected during the examination. The placenta was anterior with grade 3 maturation. The results of the amniotic fluid and fetal velocimetries were normal.

Five days later, the host twin was delivered by caesarean section with a weight of 3000 g, body length of 48 cm, and abdominal bulging. The baby was subjected to a babygram, abdominal ultrasound, and abdominal computed tomography (CT) scan, which confirmed the diagnosis of FIF (Fig. 2).

Surgical resection of the tumor was done by an experienced pediatric surgeon. Subsequent pathological evaluation of the abdominal mass confirmed the presence of an acardiac and anencephalic fetus covered by skin surrounded by a semitransparent bag-like capsule, with malformed trunk, two lower limbs, upper limb buds, and male genitalia, supporting the diagnosis of FIF. A feeding vessel was identified and connected to the abdominal wall plexus. The histopathological analysis of the excised membrane revealed findings that were indicative of a fetal sac, resembling the amniotic membrane. Pathologic analysis of the fetiform mass revealed bone, cartilage, dermal tissue, adipose tissue, muscle fibers, fibrous tissue, and dilated vessels, with no evidence of immature elements or malignancy. The differential diagnosis of teratoma was therefore excluded.

The host twin experienced a smooth recuperation and was discharged from the hospital on the eighth day following the surgical resection. The baby was in good health at two-month follow-up.

3. Discussion

Fetus in fetu is described as a parasitic monozygotic twin growing within and supplied by its host twin of the same sex [8]. In FIF, the most recognizable structures were the vertebral column and limbs. Lower-limb growth is usually faster than upper-limb development [9]. In our case, the vertebral column was not clearly visualized on prenatal ultrasound, but the FIF had two well-developed legs with slight spontaneous movement. To the best of our knowledge, only one case of a well-developed FIF with spontaneous movement has been previously reported. The movement was observed using prenatal three-dimensional sonographic imaging [4]. In a scenario with limited resources, we successfully diagnosed this condition solely by two-dimensional ultrasonography. Ultrasound can typically identify fetal body movements at early stages of gestation. Spontaneous movements of the limbs can be observed between the ninth and twelfth weeks of gestation. These movements are ascribed to an intrinsic autonomic mechanism [10]. The spontaneous movement suggested the presence of a functional nervous system in this fetus in fetu.

The majority of FIFs (80%) are found in the retroperitoneal region [11]. In our case, the FIF was in the right retroperitoneal region, pressing on the host fetus's liver, kidney, and intestines to the left. Although an FIF lacks the ability to live independently, it is nonetheless a living tissue in the host. It grows in tandem with the host since its nutrient supply is received from the host [9]. Because the FIF is connected to the abdominal wall, its blood supply usually originates from the abdominal wall plexus, as in the present case [12]. Timely diagnosis is essential to prevent the growing mass causing significant morbidity to the host twin.

With the advancement of acoustics technology, ultrasound can be used to detect intrauterine FIF. The majority of FIF cases were discovered in the second and third trimesters [2]. The presumptive diagnosis in the present case was based on prenatal ultrasound findings at 38 weeks of pregnancy. The woman had regular prenatal visits, but this was her first ultrasound scan because ultrasound was not available at her primary care facility, and the hospital was about 2 h away. Due to the highly developed characteristic of the FIF, the diagnosis could have been discovered sooner if the patient had undergone ultrasound screening in the second trimester. Prenatal diagnosis allows for adequate time for prenatal counseling to consider treatment options. It is controversial in some countries, including Indonesia, to terminate a viable fetus with serious congenital defects. Nonetheless, early prenatal diagnosis is critical for early management in order to avoid eventual morbidity [3].

Abdominal X-ray may detect the presence of an indistinct mass of soft tissue in the upper abdomen, along with many calcifications, the vertebral column, and limb buds [5], as in the present case. The density resolution of ordinary X-ray film, on the other hand, is low, and the FIF is buckled in the body [9]. A CT scan provides precise information about the specific location, blood supply, and the impact on surrounding tissues produced by the FIF. Three-dimensional reconstruction can pinpoint the precise anatomical location of FIF [5]. However, radiation exposure is a drawback of both plain radiographs and CT examinations [9]. Not only is ultrasound examination a relatively safe prenatal examination, but it can also be used to diagnose FIF prenatally and postnatally. Ultrasonography can detect calcific formations, but it can also lead to a misdiagnosis. MRI offers a broader scope of observation and superior spatial precision with no interdependence between the operator and the interpreter [8]. Unfortunately, MRI is not accessible in areas with limited resources. FIF can be successfully diagnosed solely by two-dimensional ultrasonography, as in the present case. Ultrasonography has been the investigation of choice for diagnosing fetal anomalies and growth problems due to its wide availability, low cost, and real-time imaging [5].

Despite the above-mentioned examinations, determining a definitive diagnosis prior to surgery is frequently difficult. To confirm the diagnosis, postoperative pathology exams should be performed. As in the present case, the mass is often enveloped by a delicate fibrous sac that contains fluid of a straw-like appearance, consisting of an anencephalic fetus (100%) with developing limb buds (83%), and a spinal column [5]. Because of the minor risk of malignancy associated with retroperitoneal teratomas, it is critical to distinguish between FIF and teratoma [13]. The diagnosis of FIF requires cytological and histological evaluation to rule out a teratoma [12]. The histological examination in the present case showed no immature elements or malignancy. The main approach for treating FIF is surgical removal of the tumor. Surgical treatment can restore normal anatomy and physiology, and also eliminate the malignant potential of the mass [3]. The prognosis varies greatly with location and rate of growth but is generally favorable due to its typical benign features [3]. The host twin in the present case experienced a smooth recuperation after surgery and was doing well at two-month follow-up.

4. Conclusion

The case presented therefore meets the diagnostic criteria for fetus in fetu. The diagnosis was based on prenatal ultrasonography that showed the presence of two well-developed legs with slight spontaneous movement. Accurate prenatal diagnosis of fetus in fetu is important to improve outcomes.

Contributors

Bella Stevanny contributed to patient care, conception of the case report, acquiring and interpreting the data, drafting the manuscript, undertaking the literature review, and revising the article critically for important intellectual content.

Putri Mirani contributed to drafting the manuscript, undertaking the literature review, and revising the article critically for important intellectual content.

Elsye Ruth Frida Thene contributed to patient care, acquiring and interpreting the data, and revising the article critically for important intellectual content.

Cindy Kesty contributed to drafting the manuscript, undertaking the

literature review, and revising the article critically for important intellectual content.

All authors approved the final submitted manuscript

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Patient consent

Written informed consent was obtained from the patient for publication of the case report and accompanying images.

Provenance and peer review

This article was not commissioned and was peer reviewed.

Conflict of interest statement

The authors declare that they have no conflict of interest regarding the publication of this case report.

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