CASE REPORT

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Fetus in Fetu: Rare and Intriguing Case Report

Erna SUPARMAN[a], John WANTANIA[a], Juneke Joice KAENG[a], Eddy SUPARMAN[a], Leonardo ALEXANDRA[a]

[a]Department of Obstetrics and Gynecology, Faculty of Medicine Sam Ratulangi University, North Sulawesi, Indonesia

ABSTRACT Fetus-in-fetu is a very rare case, happening in approximately 1/500,000 live births, and a rare phenomenon where parasitic deformed twin pregnancies happen in the abdominal cavity of the other twin. Most fetus-in-fetu cases can be diagnosed in the antenatal period with various imaging modalities. The recommended management is surgical excision with pathology examination to provide an exact diagnosis of fetus-in-fetu. Several laboratory markers are used to monitor the case after surgical excision. Monitoring is essential in this case because there is a potential for the case to become malignant. This case was diagnosed accidentally in the routine antenatal ultrasound, imaging study of the baby followed by surgical excision was done several days after the baby was born to minimize the impact of the fetus inside her. This case report is made to increase the awareness and acuity of routine antenatal examinations therefore planned management can be done to provide better outcomes.

Keywords: Fetus-in-fetu; fetiform twin; parasitic twin; yolk-sac tumor; twin pregnancy

Fetus-in-fetu is a very rare case, happening in approximately 1/500,000 live births, and a rare phenomenon where parasitic deformed twin pregnancies occur in the abdominal cavity of the other twin.[1]

There are at least two theories explaining fetus-in-fetu, firstly that fetus-in-fetu is a monochorionic monozygotic diamniotic that grows inside its host. This theory is supported by the fact that the genetic material of the host baby and fetiform mass are genetically identical. The proposed mechanism is due to the deviation of monozygotic diamniotic twins that asymmetrically division between totipotent cells from blastocyst that grows and causes inclusion from smaller cells of the sister embryo.[2-4]

The second theory is a very well-differentiated teratoma. This theory is supported by several arguments that the size of fetus-in-fetu and teratoma is very much similar. Teratoma also consists of pluripotent cells without the occurrence of organogenesis or vertebral segmentation.[5]

The majority of fetus-in-fetu is diagnosed at antenatal care or when the patient has the symptoms. With the advancement of diagnostic imaging, many modalities can be used to help diagnose fetus-in-fetu, such as ultrasound, computed tomography (CT)-scan, and magnetic resonance imaging (MRI).[6-8]

The diagnostic criteria of fetus-in-fetu are if one of these criteria are met, the criteria are: a mass that is covered by sac, partial or complete part of the mass is covered by skin, can be anatomically distinguished, and connected to the host by pedicle containing major blood vessels.

The recommended management in fetus-in-fetu is surgical excision. Surveillance after surgery should be done, considering the risk of recurrence, and routine follow-up should be done.

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Correspondence: Erna SUPARMAN
Department of Obstetrics and Gynecology, Faculty of Medicine Sam Ratulangi University, North Sulawesi, Indonesia
E-mail: ernasuparman@yahoo.com

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CASE REPORT

A 35-year-old multiparous female with term pregnancy was referred with a suspected fetus-in-fetu. The suspicions were made at a routine antenatal examination at the previous hospital. The history of this pregnancy was remarkable, with no known comorbidities in this patient.

A maternal-fetal specialist was attended to further evaluate the pregnancy, from ultrasound examination, fetus-in-fetu appeared inside the abdominal cavity of the twin fetus with CRL of 3.92 cm (Figure 1).

A planned cesarean section was done to manage the obstetric history, female baby was born with a birth weight of 3,300 grams, birth length of 50 cm, and Apgar score of 7-9.

Babygram was done the day after the baby was born and normal babygram was reported with no sign of fetal component found.

Abdominal multi-slice computed tomography (MSCT) was done further, and a heterogenous mass sized + 5.6x6x6 cm, consisting of cystic and solid with calcification shaped like vertebrae and extremities in the right upper abdomen of the baby, was found. The mass appears to be pressing the surrounding organs. These findings were following fetus-in-fetu appearance.

One week later, laparotomy to excise the tumor with ureterolysis, retroperitoneal lymphadenectomy, and blood vessel ligation was done. The tumor was a fetus shaped inside of an amniotic sac, the amniotic sac was opened and the fetal part was distinguished from the abdomen to the lower extremity. Three days after the surgery, the patient was discharged with a planned abdominal CT scan for re-evaluation. The patient has informed and agreed for this case to be published.

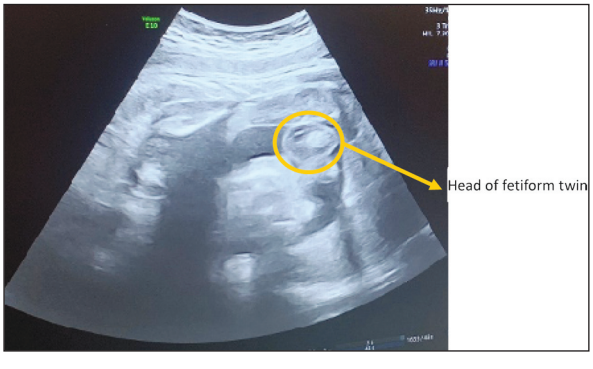

FIGURE 1: Ultrasound finding of fetus-in-fetu (courtesy of author's documentation)

DISCUSSION

Fetus-in-fetu is a rare phenomenon estimated to occur in 1/500,000 live births. In the 18th century, Meeker explained fetus-in-fetu as a twin deformity that is parasitic in the abdominal cavity inside the healthy twin.[1]

Several studies found that male babies are twice as prevalent as female babies.[9] However, no difference in the prevalence of male and female babies, as in this case, a female baby with a fetus-in-fetus was born.

The majority of fetus-in-fetu is diagnosed at antenatal care or when the patient has the symptoms. With the advancement of diagnostic imaging, many modalities can be used to help diagnose fetus-in-fetu. The guiding image in the fetus-in-fetu is the axial bone. Plain X-ray can be used to help diagnose fetus-in-fetu but have low resolution therefore, in cases where the mass is situated at the retroperitoneal space, a clear image was rarely found.[6-8] In this case, a plain babygram photo was taken with the conclusion of normal findings (Figure 2).

Due to inconclusive findings in the babygram, an abdominal MSCT scan was done (Figure 3) to help diagnose fetus-in-fetu, from the imaging study found a heterogenous mass with mixed of cystic and solid parts with calcification shaped like vertebrae and extremity in the upper right abdomen, The mass appears to press surrounding organs. In another reported case, a CT scan study found mixed density mass with irregular shape and well-circumscribed mass, the finding showed a whole axial bone.[6-8] The mass finding location was suggested in the retroperitoneal space, according to literature most fetus-in-fetu located in the retroperitoneal space, at the midline, even though there were reports where fetus-in-fetu situated at other places like the cerebral ventricle, liver, and pelvic cavity, scrotum, and mediastinum.[10,11]

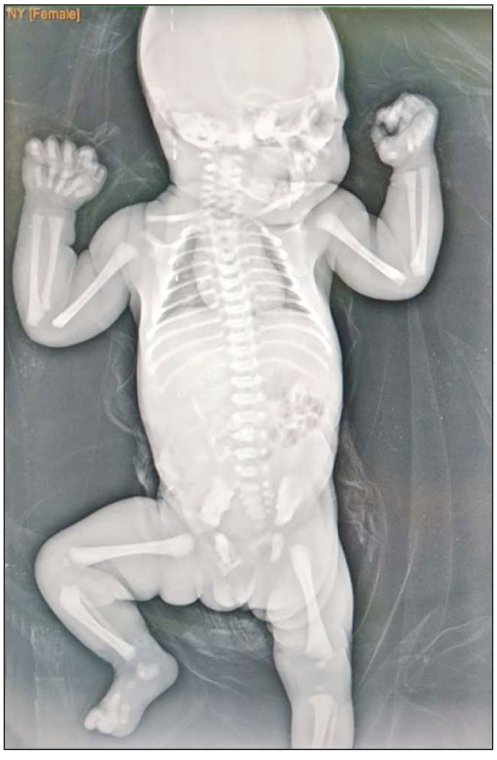

FIGURE 2: Babygram (courtesy of author's documentation).

Other modalities to detect fetus-in-fetu in antenatal care is by using ultrasound (Figure 1) which is safer, easier, more affordable, and more feasible with pathognomonic signs of axial bone. This finding might mimic teratoma, therefore the false positive rate is increased. In this case, the first suspected fetus-in-fetu diagnosis was when the mother went for a routine antenatal ultrasound. For a better resolution, MRI can be used to diagnose fetus-in-fetu with superior resolution and safe for pregnancy.[6-8]

In this case, the patient is yet to be symptomatic, but from the MSCT finding (Figure 3), the mass appears to press the surrounding organs. These findings if left untreated, might become symptoms, as Ji et al. reported that most of the cases' symptoms are due to pressure of the mass, such are abdominal distention, feeding difficulties, emesis, icteric, and up-to-pressure to the renal and respiratory system.[12] Wu et al. even reported a case where fetus-in-fetu caused cardiac arrest due to pressure in the mediastinum.[13]

The recommended management in fetus-in-fetu is surgical excision, with attention to excised the whole fetiform mass to prevent recurrence. Due to the similarity of fetus-in-fetu and teratoma, several diagnosis criteria were made to distinguish the two. In our cases, the surgical finding (Figure 4) was following literature with distinguished anatomy of the head, thorax, abdomen, and extremity easily distinguished.

Surveillance after surgery should be done, considering the risk of recurrence, and routine follow-up should include beta-human chorionic gonadotropin, alpha feto protein serum, and carcinoembryonic antigen.[12]

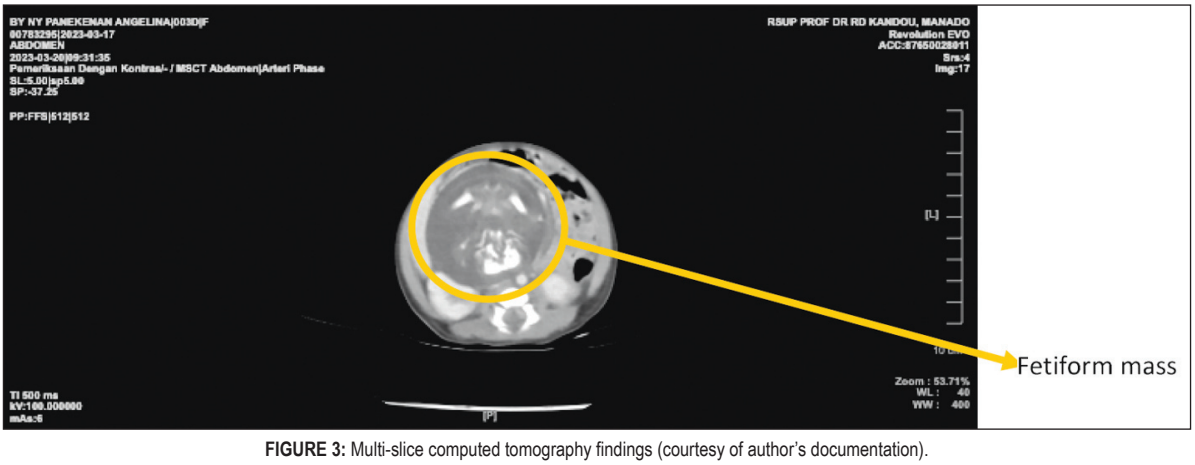

FIGURE 3: Multi-slice computed tomography findings (courtesy of author's documentation).

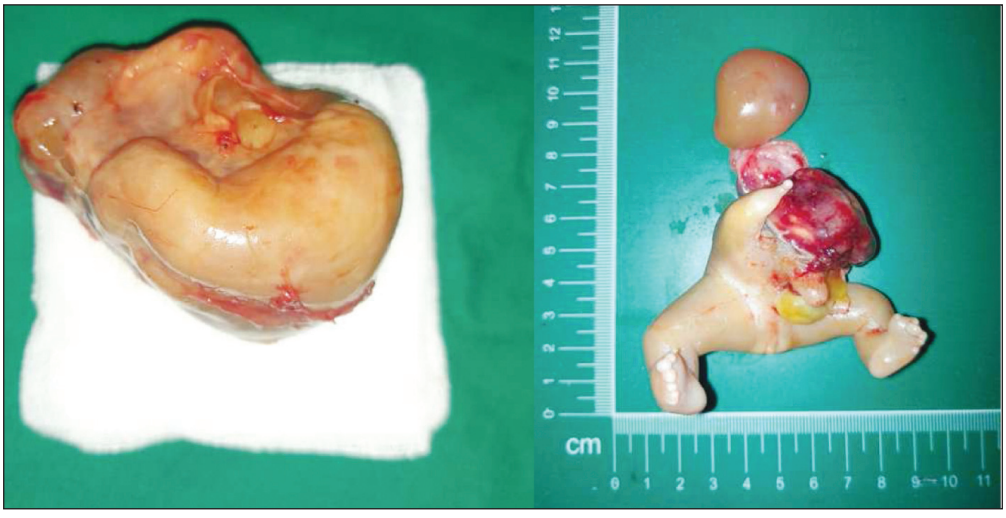

FIGURE 4: Surgical finding (courtesy of author's documentation).

Fetus-in-fetu is a very rare case, and thorough evaluation needs to be done because fetus-in-fetu might become a problem for the born baby, like symptoms of space-occupying lesion, such as feeding difficulty, icteric, hepatic disorder, and others. The differential diagnosis of fetus-in-fetu is teratoma, which carries a 10% risk of malignancy.

Thorough evaluation might diagnose this case early and earlier treatment can be done with a better prognosis for the baby born. In this case, the treatment is following the literature and longer follow-ups still need to be done to evaluate further risk of malignancy.

Source of Finance

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Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

All authors contributed equally while this study preparing.

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