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Fetus in Fetu at Prof. Dr. R. D. Kandou Hospital: A Rare Case Report

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Abstract: Fetus in fetu (FIF) is a rare congenital disorder in which a malformed fetus-like structure is inside the actual body of the fetus. We reported a 3-day old female baby, born through sectio caesaria with a lump on her right abdomen. On physical examination, a rounded mass of 8x8 cm was palpable in the right hypochondrium. CT-scan of the abdomen with contrast revealed a heterogeneous mass of mixed cystic and solid accompanied by calcification in the form of vertebrae and extremities in the upper right abdomen (size ±5,6 x 6 x 6 cm). The mass appeared to be abutting with the liver, gallbladder, pancreas, and right kidney by pressing the intestinal loops to the left. A laparotomy was performed with extensive tumor excision, and a malformed fetus-shaped mass with good differentiation characterized by a hand-like structure, fairly complete toes and skin on its surface was found. The tumor was diagnosed as a FIF since the benchmark for diagnosing FIF was the shape of a fetus-like mass, a mature bone structure with components such as cranium, vertebrae, pelvis, extremities and even fingers as found in this patient. The patient was carried out a 5-day treatment after surgery, and then was referred to the Pediatric Surgery polyclinic for a follow-up on the histopathological result to confirm the diagnosis but the patient did not come for further treatment.
Keywords: fetus in fetu; fetal tumor; aberrant monozygotic twinning; CT-scan abdomen

INTRODUCTION

Fetus in fetu (FIF) is a rare congenital disorder in which the form of abnormalities is in the form of a malformed fetus-like structure inside the actual body of the fetus.[1] More than 80 cases have been reported in the literature with varying locations varying from retroperitoneal and intraperitoneal most commonly to intracranial, intraoral, and intrascrotal.[2] Another benchmark is the finding of more mature organs such as intestines, adrenal glands, esophagus, trachea, lungs, fingers, skin, hair, smooth muscles, striated muscles, and others.[1] Embryologically, the cause of FIF is controversial, but the pathogenesis of the disorder is aberrant monozygotic twinning.[3] The FIF is most commonly found in males with a ratio of 2:1. Clinical most often it is an intra-abdominal mass in the first year of life. The number of FIFs is usually single but several cases of multiple FIFs have been reported. The disorder appears in 1:500.000 births.[4]

CASE REPORT

A female baby, 3-day old, was born through sectio caesaria with a birth weight of 3300 grams and a body length of 47 cm. There is a lump on the right abdomen. There was no history of maternal illness, radiation exposure, or drug consumption during pregnancy. There was no family history of multiple pregnancies. On physical examination palpable large, hard, round mass, size 8x8 cm in the right hypochondrium with good vital signs. CT-scan of the abdomen with contrast revealed a heterogeneous mass of mixed cystic and solid accompanied by calcification in the form of vertebrae and extremities in the upper right abdomen (size 5.6 x 6 x 6 cm). After giving the contrast, there was a buffer on the solid part. The mass appeared to be abutting with the liver, gallbladder, pancreas, and right kidney by pressing the intestinal loops to the left (Figure 1).

The patient was then performed laparatomy with extensive tumor excision. In intraoperative exploration, duodenum coiled above the tumor, colon tranversum also coiled above the tumor, adhesiolysis was done from colon tranversum and duodenum to tumor. Identification of the kidneys and ureterolysis of the ureters to identify the ureters, then extensive excision of the tumor was decided to protect the vena cava and portal vein. Furthermore, exploration of the common bile duct (CBD) to ascertain the direction and structure of the CBD, and retroperitoneal lymphadenectomy was performed. Control bleeding was performed by doing ligation of the veins branching from the right hepatic artery that bled around the CBD. A mass encapsulated with white membrane and yellow liquid was removed. After uncovering the membrane, there appeared a malformed fetus-shaped mass with good differentiation characterized by a hand-like structure with fairly complete toes and skin on its surface (Figure 2). The patient was carried out a 5-day treatment after surgery. On physical examination, it appeared that the surgical wound was well treated, no acute signs of abdomen, and vital signs were found within normal limits (Figure 3). Therefore, the patient was referred to the Pediatric Surgery polyclinic for a follow-up on the pathology result but the patient did not come for further treatment.

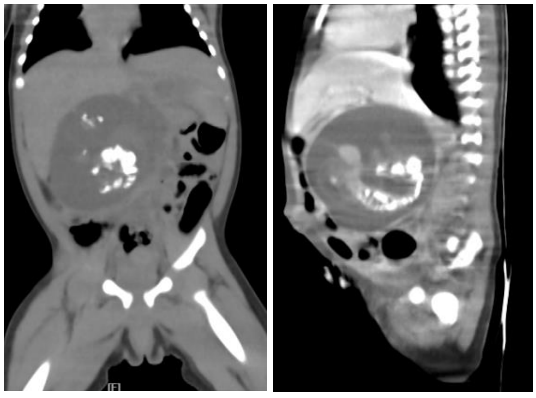

Figure 1. CT-scan of the abdomen with contrast revealed a heterogeneous mass of a mixture of cystic and solid accompanied by calcification in the form of vertebrae and extremities in the upper right abdomen (size ± 5.6x6x6cm)

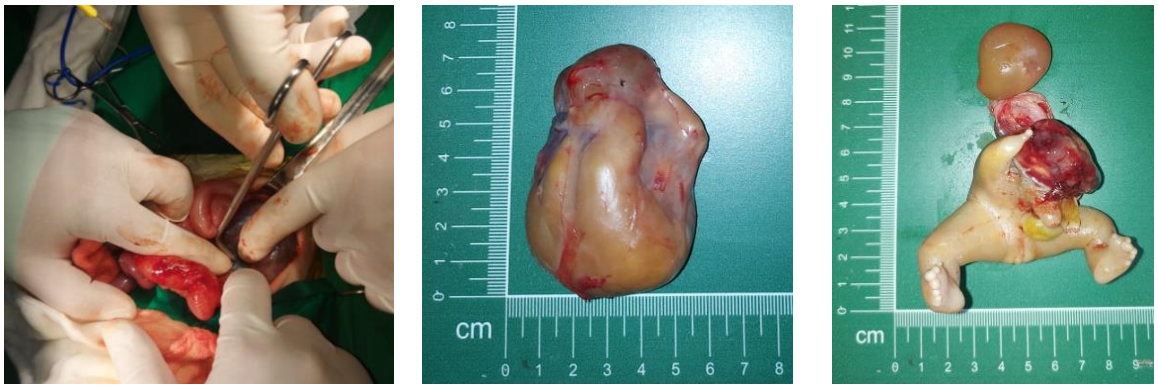

Figure 2. The membrane of the mass wall was white with yellow liquid. After the tumor mass and the membrane were removed, there appeared a malformed fetus-shaped mass with good differentiation characterized by a hand-like structure with fairly complete toes and skin on its surface

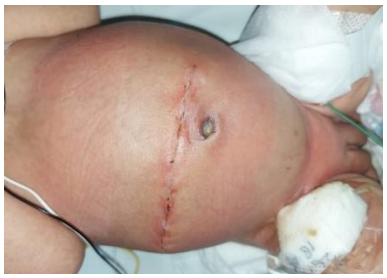

Figure 3. After a laparotomic surgery, the surgical wound was well maintained and no acute signs of abdomen were found

DISCUSSION

Fetus in fetu (FIF) is most commonly found in males with a ratio of 2:1.[4] It can occur anywhere in the body, ranging from the oral cavity, retroperitoneal, intraperitoneal, intra-cranial, neck, scrotum, and others. The benchmark for diagnosing FIF is first the form of a fetus-like mass, a mature bone structure with components such as the cranium, vertebrae, pelvis, extremities and even fingers. Another benchmark is the finding of organs that are more mature such as the intestines, adrenal glands, esophagus, trachea, lungs, fingers, skin, hair, smooth muscles, striated muscles, and others.[5] Radiological modalities that can be used as a comparison to diagnose FIF based on its location are ultrasound, plain photos, CT-scan, and MRI. Post-natal patients without history of FIF usually come to the hospital with complaints of masses that continue to grow and are painless. A plain photo is the simplest check that can be done. The image found is usually a soft tissue mass with a solid structure/calcified bone that gives the effect of mass pushing organs around. The mass effect causes disruption of the function of the organ if treatment is not carried out as soon as possible.[6]

The closest differential diagnosis of FIF is mature teratoma.[7] Although each fetal tumor has its own characteristics, a common sign of all fetal tumors is polyhydramnios, which appear in one-third of pregnant women and can be the first clinical sign of increased fundal height on obstetric examination. Other congenital abnormalities can also be found in 20% of cases, especially in teratomas. Hydrops fetalis is also found in 17% of cases. Accurate prenatal diagnosis is essential for prenatal and postnatal management planning.[5]

Complex fetal tumor cases require multidisciplinary cooperation consisting of radiologists, perinatologists, neonatologists, pediatric surgeons, genetic counselors and social workers. It is important to understand which conditions can be corrected in utero and that diagnostic skills often exceed those in patient care, although sometimes in some cases the best option that needs to be taken is supportive care.[8]

Fetus in fetu mass removal is usually easy because the mass has a capsule with firm

boundaries, so separation of the mass from surrounding tissue becomes easier to do. Complications in the fetus can occur due to the effect of mass pushing the organs around, so the size and location of the mass in the body is important to determine the severity of complications that can occur. If the tumor in the fetus is not found before birth, there will be difficulties during childbirth or dystocia, with a high risk of tumor rupture and exsanguinations.[8]

Post-surgical follow-up imaging is the recommendation to rule out recurrence. Post-treatment complications are the same as any mass surgery in children, namely it is necessary to pay attention to bleeding, vital signs, mass effects on surrounding organs that can no longer return completely, healing of surgical wounds and the possibility of iatrogenic disorders of other organs post-operatively.[9,10]

The actual diagnosis can be made by physical examination and radiological examination. Even though the mass looks like a fetus, a histopathological examination has to be carried out to ensure that the type of mass tissue removed is malignant or not. In our case, the patient did not have a histopathological result since the tissue samples were given to the patient's family for the further examination but the patient was loss for further follow-up. Discussions and debates about FIF and teratomas still occur, but in fact these two types of tumor are different because the pathogenesis processes of FIF and teratoma abnormalities are different. To note, FIF comes from the patient's twin who is not well developed and is in the patient's body whereas teratomas are derived from primordial germ cells (PGCs) that do not migrate to where they should be and do not degenerate afterwards. In addition, the predilection of the place where teratoma occurs is also different from FIF. Genetic testing also states that patients with FIF are twins. Moreover, in a case report, it was stated that on ultrasound examination, involuntary movements of the FIF extremities were found.[6]

Although each fetal tumor has its own characteristics, a common sign in all fetal tumors is known, namely polyhydramnios which appears in one third of pregnant women and can be the first clinical sign accompanied by an increase in fundal height not in accordance with the gestational age on obstetric examination. Accurate prenatal diagnosis is essential for planning the prenatal and postnatal management. The diagnosis of FIF needs to be considered starting from the fetus on ultrasound, CT-Scan or MRI examinations, therefore, the diagnosis can be faster confirmed. Finding of cystic components with firm borders and dense complex tissue that can be accompanied by bone-shaped calcifications in it and begin to shape like a fetus even though it is not complete is crucial in diagnosing the FIF. The location and size of the FIF are major factors for assessing a patient's prognosis. If the FIF mass causes organ damage or even life-threatening by blocking the airway, the prognosis is poor.[5] Laboratory investigations typically reveal an elevated AFP (≥3000 nanograms/milliliter), further supporting the diagnosis of FIF. It is important to understand which conditions can be corrected in utero and that the ability to diagnose often exceeds the ability to treat the patient, although sometimes in some cases the best option to take is supportive management. In certain cases, a FIF mass can interfere with the airway, so that, the operation must be prepared well from prenatal, at birth, and postnatal. For masses that block the airway, an exit procedure can be performed which can quickly save the airway, so that subsequent operations to remove the mass are more controlled.[10]

CONCLUSION

We reported a 3-month female baby with a lump on the right abdomen since birth (birth weight of 3300 grams and body length of 47 cm). There was no history of maternal illness, radiation exposure, drug consumption during pregnancy, and multiple pregnancies. On physical examination a palpable large, hard, round mass, size 8x8 cm in the right hypochondrium was found. CT-scan of the abdomen with contrast revealed a heterogeneous mass of mixed cystic and solid accompanied by calcification in the form of vertebrae and extremities in the upper right abdomen (5.6 x 6 x 6 cm). After giving the contrast, there was a buffer on the solid part. The mass appeared to be abutting with the liver, gallbladder, pancreas, and right kidney by pressing the intestinal loops to the left. A laparotomy was performed with extensive tumor excision. The tumor

mass was encapsulated with white membrane. After removing the membrane, a malformed fetus-shaped mass with good differentiation characterized by a hand-like structure, fairly complete toes and skin on its surface was found which was diagnosed as FIF. The patient was carried out a 5-day treatment after surgery, and then was referred to the Pediatric Surgery polyclinic for a follow-up on the histopathology result to confirm the diagnosis, however, the patient did not come for further treatment.

Conflict of Interest

The author declares there is no conflict of interest regarding publication of current report.

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