FOETUS-IN-FETU, A PAEDIATRIC RARITY IN AN INFANT AT IRRUA SPECIALIST TEACHING HOSPITAL, IRRUA, NIGERIA

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Submission Date: 16th Aug., 2023 Date of Acceptance: 30th Dec., 2023 Publication Date: 30th Jan., 2024

ABSTRACT

Introduction: Fetus in fetu is a paediatric rarity. It involves the presence of a mass resembling a fetus inside the body of a child or an adult. It is described as a twin growing inside the body of the other. It can be located in different parts of the body but commonly the retroperitoneum. It is usually benign. Case presentation: The patient was a 4 month old male infant who presented to the hospital with complaints of abdominal distention. The distention was noticed two months prior to presenting to our hospital. The distension was generalized and has been progressively increasing until presentation. There was no associated abdominal pain and no other abdominal symptoms.

Conclusion: Treatment is by complete excision for histological examination.

Keywords: Fetus-in-fetu (FIF), Twin, Retroperitoneum

INTRODUCTION

Fetus-in-fetu (or foetus in fetus) is a rare developmental abnormality in which a mass of tissue resembling a fetus forms inside the body of its twin. 1 Fetus-in-fetu has been defined as the presence of one of the twins in the body of the other. It is most frequently located in retroperitoneal area; however, it has been reported in other locations as well.² An early example of the phenomenon was described in 1808 by George William Young.3 This rare congenital anomaly, has been reported around 100 times since its first definition in the nineteenth century.4 Clinical manifestations vary. Detection is most often in infancy, the oldest reported age being 47 years.⁵ Foetus in fetu is differentiated from the teratoma by the presence of vertebral column often with an appropriate arrangement of other organs or limbs around it. Unlike teratomas, fetus-in-fetu is a benign disorder.6 Despite the requirement of the presence of a vertebral column for diagnosis; there are reports of cases without a vertebral column.⁶ Fetus in fetu most frequently (80%) inhabits the retroperitoneal region. However, there have been few reports of FIF location in the head, sacrum, scrotum and the mouth. Different organs can be seen in foetus in fetu, including vertebral column (91%), limbs (82.5%), central nervous system (55.8%), gastrointestinal tract (45%), vessels (40%), and genitourinary tract (26.5%).6

In order to qualify as a foetus-in-fetu, one of the following characteristics must be present: a mass enclosed within a distinct sac, partially or be completed covered by skin, grossly recognizable anatomic features and attached to the host by a pedicle containing a few

We report a case of foetus-in-fetu in a 4 month old male infant who presented with abdominal distension. Preoperative diagnosis of foetus in fetu in this case was made on Magnetic Resonance Imaging, and the patient underwent exploratory laparotomy with complete excision of mass. The excised mass in a sac was proven to be foetus in fetu on the basis of gross and histopathological examination.

CASE PRESENTATION

The patient was a 4 month old male infant who presented to the hospital with complaints of abdominal distention. The distention was noticed two months prior to presenting to our hospital. The distension was generalized and has been progressively increasing until presentation (Figure 1). There was no associated abdominal pain and no other abdominal symptoms.

Patient is the fourth child in a monogamous setting of four children. There is no history of twining in the family or in parents family. Prenatal period was essentially uneventful with two prenatal ultrasound



Figure 1: 4 month old male distended abdomen

scans which were normal. He was delivered through vagina delivery and birth weight was 4.6kg.

On examination, the abdomen was grossly distended. An abdominal ultrasound scan was done which revealed a huge heterogeneous predominantly cystic multiloculated intra-abdominal mass measuring about 26cm by 22cm containing areas of calcifications which appear to be arising from the upper right side of the abdomen extending inferiorly towards the pelvis and crossing the midline. The ultrasound scan suggested a teratoma. The electrolytes, urea and creatinine levels were normal. The full blood count and liver function test were also normal. The level of alpha –fetoprotein (AFP) and beta –HCG were normal.

The patient had an MRI done which shows an extremely huge well circumscribed heterogeneous solid-cystic mass with varying signal intensities including fat located in the abdomen to the right possibly within the retroperitoneum. The cystic components are multilobular and do not show marginal enhancement following contrast administration. It measures 18.2cm by 15.8cm by 15.9cm. The MRI findings was that of features in keeping with a mature cystic teratoma of the retroperitonuem.

The patient had an exploratory laparotomy. The patient was put under general anaesthesia and endotracheal intubation and through a transverse skin crease supraumbilical incision the peritoneal cavity was entered.

At laparotomy there was a huge cystic retroperitoneal mass covered by a transparent membrane (Figure 2). This was displacing the bowels to the left (Figure 2).



Figure 2: Cystic mass displacing bowel to the left

The sac was opened revealing a huge mass with multiple fetal parts like the head with hair, back, poorly developed limbs and trunk. (Figure 3,4,5)

The mass was displacing the right kidney inferiorly and the bowels to the right. The mass crossed the midline



Figure 3: Content showing hair, limbs ear lobe



Figure 4: Mass with fetal parts

and was observed to be supplied by a vessel from a branch of the ascending aorta.



Figure 5: Completely excised mass

The mass was completely removed (Figure 5) and sent for histological examination.

Histological examination revealed on macroscopy; an irregularly shaped solid to cystic mass weighing 650g and measured 16.0 by 12.0 by 7.0cm. The mass had hair attached to the superior part of the mass, surface appendages which look like poorly developed extremities and other poorly defined fetal structures. The cut section revealed solid to cystic structures. Three tubular tissues were identified. The solid components contained bony structures in the midline appearing like vertebra column.

The microscopy showed sections of retroperitoneal mass showing benign lesions and composed of well formed organs and structures from three germ layers. The structures from the ectoderm included the skin and appendages, neural tissue, ectocervical epithelium, meninges with psammoma bodies and brain tissues.



Figure 6: Patient 3weeks after surgery

Parts of the tissues from the mesoderm include chondroid and spongy bone, ectocervical stroma and myometrium. Parts of the endodermal tissues were intestinal glands.

The patient recovered smoothly and was discharged on the 12th day post operation with good healing at first clinic attendance. (Figure 6)

DISCUSSION

Fetus-in-fetu is a rare variety of parasitic twins, where the developmentally abnormal parasitic twin is completely encapsulated within the body of the otherwise normally developed host twin. In the late eighteenth century, German anatomist Johann Friedrich Meckel was the first to described fetus in fetu, which translates to fetus within fetus. Fetus-in-fetu is thought to result from the unequal division of the totipotent inner cell mass, the mass of cells that is the ancestral precursor to all cells in the body. The unequal division is thought to occur during the formation of the blastocyst, which can also result in parasitic and conjoined twins. Fetus-in-fetu represents a developmental anomaly that has prompted developmental biologists to further examine the mechanisms for how twins arise.

Preceded by Willis in 1935, in 1954, Lord claimed the presence of a vertebral column and extremities and organs located at appropriate places around it as the basic diagnostic feature for Foetus-in-fetu. These criteria are still, to a wide extent, valid today.^{8,9,10,11}. These criteria for diagnosis were present in this patient.

This condition is usually benign as most of the patient appears relatively healthy with obvious progressively enlarging abdomen if it is retroperitoneal as seen in this case.

The treatment of fetus-in-fetu is complete excision. This mass after excision was sent for histological examination which revealed other findings like the vertebra column needed to establish a diagnosis.

Fetus-in-fetu is considered a paediatric rarity with about 100 cases reported since the nineteenth century⁴, with yet no specific figures for Africa and Blacks.

There is no evidence to suggest any untoward sequalae in any patient with fetus-in-fetu post-operatively. This patient recovered smoothly from surgery and has been seen twice after discharge.

CONCLUSION

Foetus- in- fetu is a rare malformation presenting as an abdominal mass resembling a foetus with rudimentary fetal parts growing inside a child. The presence of a vertebral column on histology is usually used to confirm the condition. The condition is not usually life-threatening and complete excision is the modality of treatment.

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