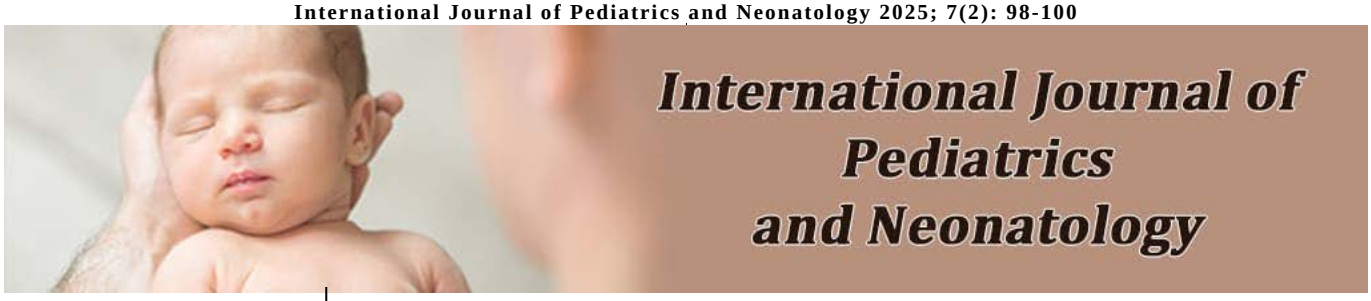

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Atypical location of an abdominal teratoma revealing a fetus in fetu in a 6-month-old infant at the chu of conakry

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Abstract
Introduction: Fetus in fetu (FIF) is an anomaly in embryonic development where a fetus is enclosed within the body of its twin. This anomaly is estimated to occur in 1 out of 500,000 live births. Between 1999 and 2013, 95 cases have been reported in the literature. The objective is to present this case of an atypical abdominal teratoma and demonstrate the importance of considering the diagnosis when encountering any abdominal mass in children.
Patient and Observation: A 6-month-old female infant, weighing 6.8 kg, was admitted to the pediatric surgery department at Donka National Hospital for an abdominal mass displacing the right kidney, spleen, pancreas, mesentery, and digestive loops. The mass, which contained bone structures, pelvis, hip, limbs, and teeth, was diagnosed via computed tomography. The mother had attended all prenatal consultations, and the pregnancy proceeded normally to term without any prenatal diagnosis, and the delivery was uneventful. After clinical, biological, and radiological investigations, the diagnosis of a mature abdominal teratoma was considered. During surgery, a large right retroperitoneal fetal mass was discovered. A surgical treatment with complete open resection was performed. The postoperative course was uneventful, and the patient was discharged on the sixth day.
Conclusion: Fetus in fetu is considered a benign condition, whereas the potentially malignant characteristics of the teratoma are the basis for discussion. This may constitute a difference in follow-up and treatment. Although there is a consensus on the benign nature of FIF, it is necessary to completely remove the mass, and periodic hormonal and ultrasound monitoring in the postoperative period constitutes an appropriate approach.

Keywords: Abdominal teratoma, fetus in fetu, total resection, monitoring

Introduction

Fetus in fetu (FIF) is an anomaly in embryonic development where a fetus is enclosed within the body of its twin [1]. The anomaly was first defined in the early 19th century by Meckel [2]. Despite its prevalence in infants and children, cases have been reported where the anomaly remained asymptomatic until an advanced age in the child [1]. This congenital anomaly is considered benign, while the potentially malignant nature of the teratoma raises debate regarding monitoring and treatment. Some researchers believe that FIF represents a spectrum of malignant teratomas [2]. The incidence of FIF is 1 in 500,000 births, with fewer than 100 cases reported worldwide. According to Willis, the presence of an axial skeleton distinguishes a teratoma from an FIF [3]. The preoperative diagnosis of FIF depends on radiological findings. Simple abdominal radiographs can be useful for diagnosis, in nearly half of the cases showing the presence of a spine and axial skeleton [4]. 3D ultrasound, computed tomography, and MRI have now further improved the accuracy of preoperative diagnosis before surgery. Complete resection, which requires careful dissection, is curative and allows for confirmation of the diagnosis [2]. Thus, this embryonic malformation, considered benign by some, should it be differentiated from potentially malignant teratomas? The objective is to present this case of an atypical abdominal teratoma and demonstrate the importance of considering the diagnosis when encountering any abdominal mass in children. This is to ensure appropriate management, taking into account the potentially malignant nature of the teratoma despite the presumed benign nature of FIF.

Patient and Observation

A 6-month-old female infant, weighing 6 kg, was referred by the pediatric oncology unit at Donka National Hospital for an abdominal mass. The infant was the fifth child, born to a 32-year-old mother, and the pregnancy had proceeded normally without prenatal diagnosis. After an uneventful delivery, the mother noticed a small abdominal mass in the right flank that gradually increased in size, causing concern among the parents. An initial ultrasound performed in the countryside suggested a cyst of the right kidney. Clinical examination revealed a large abdominal mass predominant in the right hemiabdomen with a protrusion in the homolateral hypochondrium. This mass was firm, smooth but irregular on the surface, fixed to the deep plane, and filled the right lumbar fossa. It measured 15 cm in its long axis, extending from the costal margin and right hypochondrium to the right iliac fossa, and 16 cm in its transverse axis, from the paravertebral line in the lumbar region to the midline above the umbilicus.

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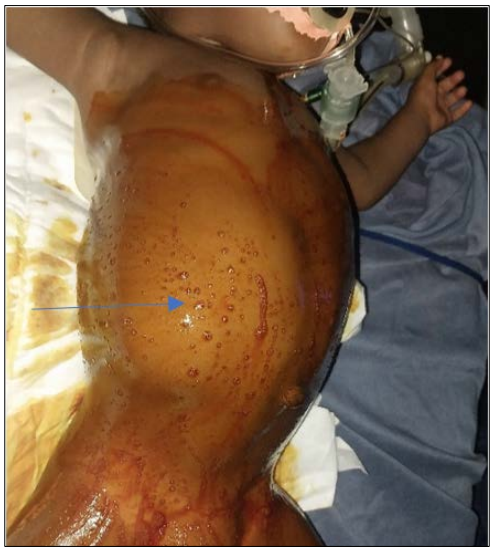

Fig 1: Large right abdominal mass with a clearly visible bulge in the supine position, tilted to the left, showing the various boundaries.

A second ultrasound performed at the Donka National Hospital revealed a large abdominal tumor, prompting a CT scan that identified a massive mass with osseous structures, including the pelvis, limbs, and teeth. This mass compressed and displaced the liver, right kidney, spleen, lungs, mesentery, and digestive loops. The mass measured 107x105x103 mm, indicative of a teratoma, and the alpha-fetoprotein level was 27,122 UI/ml, which reinforced the teratoma diagnosis.

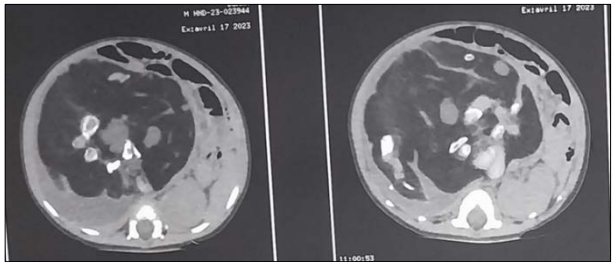

Fig 2: Abdominal CT scan showing a large compressive mass with osseous structures suggestive of a teratoma.

After performing a complete blood count, a normal hemostasis assessment, and a validated pre-anesthetic consultation, we decided to operate on the infant. Under general anesthesia, a right transverse incision revealed a large retroperitoneal mass displacing all the viscera to the left. After accessing the retroperitoneal space, we performed a total excision of a fetal mass covered by an intact membrane, attached by a nourishing vessel resembling a placenta, without any complications. Opening this membrane postoperatively revealed a parasitic fetus weighing 950g, with a brain exhibiting underdeveloped convolutions, a curved dorsal region with a cartilaginous vertebral column, rudimentary upper limbs, a pelvis with two cartilaginous iliac wings, lower limbs (thigh, leg, and feet), a rudimentary anus, and a primitive intestine on the ventral surface, which contained meconium. This immature parasitic fetus appeared well-vascularized and was covered with vernix caseosa.

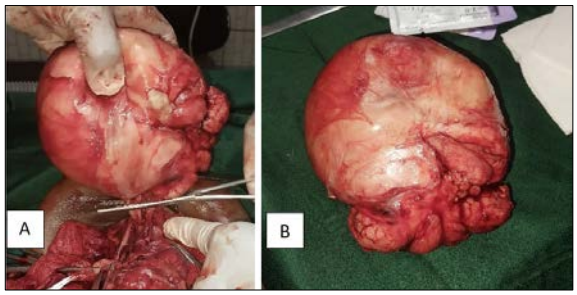

Fig 3: A: Complete dissection without breach, followed by clamping of the nourishing vessel of the fetal mass covered by an amniotic membrane (B).

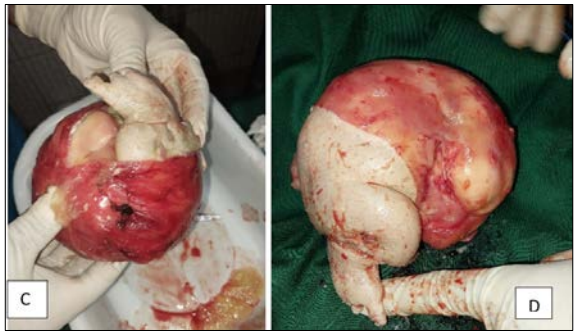

Fig 4: (C) Amniotic fluid leakage upon opening, revealing the fetus's body covered with vernix caseosa and a complete right lower limb (D).

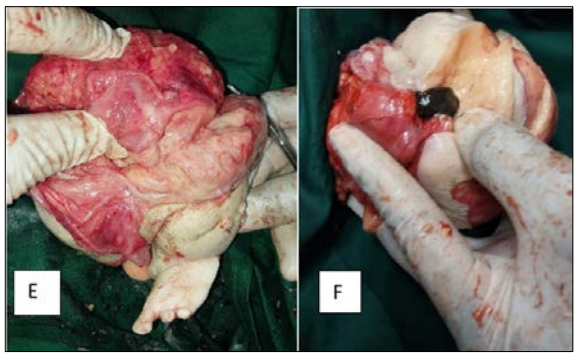

Fig 5: (E) Coronal section showing the left lateral face; cerebral convolutions between the surgeon's thumb and index finger extend to a vertebral column (arrow); left pelvic limb at the bottom. Primitive intestine with meconium output (F).

Postoperative Course

The postoperative course was uncomplicated. We monitored the biological markers, and the alpha-fetoprotein level at one month postoperative had significantly decreased to 96 UI/ml. At the last follow-up at 6 months, the patient had normal growth and development, and the alpha-fetoprotein level had normalized.

Discussion

Fetus in fetu (FIF) is a fetal malformation characterized by the presence of a parasitic monozygotic fetus within the body of its twin. Willis and Lord identified the presence of a vertebral column and extremities with surrounding organs as the basic diagnostic criteria for FIF [1]. In addition to these features cited by these authors, our specimen also included a brain and a primitive intestine containing meconium. Many authors believe that FIF represents a well-differentiated and highly organized teratoma [1]. Various theories with a common point have been proposed to explain teratomas. These tumors may contain tissues of varying differentiation levels, derived from endoderm, mesoderm, and ectoderm. Teratomas are thought to originate from multipotent cells [7]. The twinning theory would explain the occurrence of FIF due to the development of a bifid primitive streak. One branch evolves normally, while the other, due to mechanical or circulatory reasons, undergoes monstrous development leading to a teratoma, a true twin of its host: a twin in short or fetus in fetu [7, 8]. The location is 80% retroperitoneal. However, few cases have been reported involving locations in the head, sacrum, scrotum, and mouth. Despite the requirement for the presence of a vertebral column for diagnosis, there are reports of cases without a vertebral column [9]. Symptoms of this pathology arise due to its mass effect, such as the displacement of all viscera to the left in our case, related to the volume and weight of the fetal mass, which weighs over one kilogram. Sometimes, the anomaly may be asymptomatic [2].

The diagnosis of FIF relies on radiological results and biological markers as in our case. Sometimes, a simple abdominal X-ray can be useful for diagnosis in nearly half of cases, showing the presence of a vertebral column and axial skeleton. 3D ultrasound and CT scans have further improved preoperative diagnostic accuracy. Recently, magnetic resonance imaging (MRI) has also been used for diagnosis [10].

The size and weight of the fetus are likely related to its blood supply. Fetuses with distinct vascular connections to the host are larger and have better-developed characteristics [1]. Our specimen also exhibits these features.

During FIF excision, it is necessary to remove the entire mass, including its capsule. This complete excision, which requires meticulous dissection, is curative and allows for diagnostic confirmation [2]. While FIF is considered a benign condition, some researchers have suggested that it is possible to leave some remnants of the capsule in place, and in one case, the mass recurred as a yolk sac tumor after 4 months. This was attributed to the presence of immature tissues in small areas and remnants of the mass capsule [1].

Conclusion

FIF is an extremely rare entity. Although there is a consensus on its benign nature, and given the potentially malignant characteristics of teratomas, it is necessary to completely remove the mass, including its capsule. Additionally, evaluating postoperative tumor markers and conducting periodic ultrasound examinations are appropriate approaches.

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