Fetus-in-fetu is a rare congenital malformation in which a vertebra fetus is enclosed within the abdomen of a normally developing fetus. Magnetic resonance imaging (MRI) appears to be an ideal tool for demonstrating the wide range of tissue within such lesions, but there are only a few reports of the use of MRI in the identification of this anomaly. We present a case of fetus-in-fetu in which the MRI findings were diagnostic clues of this unusual entity.

**Key words:** Fetus-in-fetu, Magnetic resonance imaging

Fetus-in-fetu was first coined by Meckel in the late 18th century and defined by Willis in 1935 as a fetiform mass in which organ-like tissues are arranged around a vertebral axis [1,2]. This pathology is rare with approximately 100 reports documented to date. Characteristic imaging features of this unusual entity on plain abdominal radiograph, ultrasound as well as computed tomography (CT) scan have all been well described [3-6]. The purpose of this case study is to describe the MRI appearances of this rare condition.

**CASE REPORT**

Obstetric sonography at 24 weeks of gestation revealed an abdominal mass in a male fetus. The mother was 36 years old, gesta 2, para 1, and no history of twin pregnancy. The mass lesion was located at the left abdomen, measured 30mm in diameter with heterogeneous internal structure and cystic component. The weekly follow-up sonography showed gradual growing of this cystic mass along with its internal echoic structures. At 39 weeks' gestation age, a male newborn weighing 3750 grams was delivered by elective caesarian section with Apgar scores of 9 and 9 at 1 and 5 minutes, respectively. The newborn did not present any evidence of abnormal clinical or dysmorphic features except for a distended left abdomen with a palpable mass.

Abdominal sonography at birth showed a well-circumscribed hypoechoic mass measuring 80mm in diameter, heterogeneous with some liquid areas and hypechoic foci resembling fetal vertebral bodies (Fig. 1). The plain abdominal film clearly demonstrated some coarse calcifications in the left abdomen, suggesting fetal skeletal structures (Fig. 2). The MRI examination was performed at a 1.5T unit (Magnetom Symphony; Siemens, Germany). The MRI protocol included an axial 2-D Fast Low Angle Shot (FLASH) plus fat suppression sequence, axial, sagittal and coronal true Fast
Imaging with Steady-state Precession (true FISP) sequence, axial and coronal T2-weighted Half-Fourier Acquisition Single-Shot Turbo SE (HASTE) sequence. MRI revealed a well-encapsulated mass with mixed high, intermediate and low signal intensities in the left abdomen. The mass contained areas of fluid that surrounded a central solid component with skeletal elements inside. The skeletal elements resembled a fetal vertebral axis and long bones, with intestine-like loop structures between them. A cord-like structure that suspended the solid component in the fluid was also clearly demonstrated (Fig. 3). Based on these findings, fetus-in-fetu was presumed to be the most likely diagnosis, and the patient was submitted to elective laparotomy at 3 days of age with excision of a well-encapsulated retroperitoneal mass. There was an anomalous blood supply from a branch of the inferior mesenteric artery to the mass.

Grossly, the mass was a sac covered by a thin fibrous capsule. It contained yellowish fluid and an anencephalic fetiform structure that attached to the sac by an umbilical cord of 1.5 × 0.2 cm (Fig. 4). The fetiform mass weighed 89 grams and measured 8 × 7 × 4 cm had skin covering its entire body and hair over the cephalic end. Rudimentary lower extremities were formed and the upper extremity buds could be identified. The gluteal cleft was present but no anal dimple was found. The mass also had clavicle and ribs formation. Vertebral bodies could be identified along the length of the fetiform mass. Histopathologically, the fetiform mass consisted of intestine, adrenal glands, primordial gonadal tissue and an umbilical cord containing 2 vessels. The postoperative course was uneventful and the patient was discharged on the tenth postoperative day.

**DISCUSSION**

Fetus-in-fetu is an extremely rare malformation. The exact incidence of this abnormality is unknown but an estimated frequency of 1:500,000 births is commonly reported in the literature [7]. According to Willis [8], fetus-in-fetu arises from an inclusion of a monozygotic diamniotic twin within the bearer. Presence of vertebral axis (which develops from the primitive streak) and appropriate arrangement of other organs or limbs with respect to the vertebral axis are the diagnostic features of fetus-in-fetu, which help in differentiating this entity from the highly differentiated teratoma [8]. Most fetus-in-fetu are detected before 18 months of age and present as an asymptomatic slow-growing abdominal mass [9].

**Figure 1.** Abdominal sonography shows hypoechoic mass with heterogeneous contents, and hyperechoic foci suggesting fetal vertebral bodies.

**Figure 2.** Plain abdominal film shows a mass lesion with local mass effect in the left hemiabdomen. It contains some coarse calcifications inside which resembling fetal bones.
Fetus-in-fetu: MR appearance

Delayed presentation has been documented and on occasion fetus-in-fetu has been found at autopsy [6]. There have been a few previous occasions recorded in which this entity has been detected prenatally [6]. However, the increasing use of obstetric ultrasound has made prenatal diagnosis of this condition become more possible [10]. In the antenatal period, the diagnosis of fetus-in-fetu can be suggested when a well-defined encapsulated cystic mass containing solid and calcified components is found. The mass often increases in volume throughout the gestation period and the calcified components take on the appearance of fetal skeletal bones [11].

Most fetus-in-fetu are cases of pedunculated vertebrate masses within a capsule containing fluid and with an umbilical cord composed of only 2 vessels [9]. They are often presented in the retroperitoneal space [9]. Rare locations like the skull, liver, pelvis, scrotum, sacro-coccygeal region, mouth, adrenal gland, mesentery and right iliac fossa are also reported [6, 9, 12]. Usually a fetus-in-fetu is one in number, but a few cases of more than one have been reported [12]. The recommended treatment for fetus-in-fetu is complete surgical resection.

The diagnosis of this rare condition is based on confirmation of a spinal column along with the complex and well-differentiated tissues [10]. Occasionally, an underdeveloped and markedly dysplastic spinal column prevents the identification of the vertebral bodies at imaging [3]. A recent review on the subject reported that a preoperative diagnosis of fetus-in-fetu was made in only 17.67% of all the cases till 1980 and the vertebral column was not

Figure 3. a. The sagittal true FISP 6ms/3ms/70 (TR/TE/flip angle) image of the newborn reveals an encapsulated huge intraabdominal mass with heterogeneous components. The predominant component of the mass is liquid and the solid component is suspended in the fluid by a cord-like structure (black arrow). b. The coronal true FISP 6ms/3ms/70 (TR/TE/flip angle) image shows the solid component that contains areas of skeletal elements resembling a vertebral axis and long bones with intestine-like loop structures (black arrow) in between them.

Figure 4. The photograph of the surgical specimen.
identified in about 9% of cases even after the pathological examination [9]. Thus, nonvisualization of the vertebral axis on radiography or on computed tomography scan does not exclude the diagnosis of fetus-in-fetu [9]. Though MRI has developed into a powerful diagnostic imaging tool, there are only a few reports of the use of MRI in the identification of fetus-in-fetu [11, 13, 14, 15]. MRI is characterized by great inherent contrast, excellent spatial resolution, and exquisite anatomic display. MRI has some obvious advantages compared to the other scanning methods like CT scan in evaluating this rare condition: (1) It does not rely on calcification for demonstrating tissues and this helps in identifying insufficiently calcified vertebrae and vertebral axis [13]. (2) No ionizing radiation is involved. (3) Bone distortion is non-existent. (4) The iodine-based intravenous contrast agents used in contrast-enhanced CT have considerable risks of kidney damage and allergic reactions. In addition, the lack of ionizing radiation in both sonography and MRI make them ideal prenatal imaging modalities. Given its low cost, abundant availability and real-time imaging, sonography has been the method of choice for in utero imaging and prenatal detection of fetal structural and growth anomalies. However, because of the intrinsic limitation of sonography with regard to tissue contrast, especially in the presence of maternal obesity or oligohydramnios, MRI has been explored as an excellent and safe alternative technique. It offers a large field-of-view for better evaluation of the spatial relationships of anatomic anomalies and its visualization is not uniquely operator- and interpreter-dependent.

The recent popularity of fetal MRI has been attributed to the development of ultrafast MRI techniques such as the single-shot fast spin-echo sequence. This ultrafast MRI method allows minimal image degradation by fetal motion and high-quality visualization of fetal organs without the need for fetal or maternal sedation. There are no known or reported adverse biologic effects caused by MRI during pregnancy; therefore, it is considered a safe imaging modality even in pregnancy.

In conclusion, we have reported a case of fetus-in-fetu in which the diagnosis was made preoperatively and confirmed by pathology. In our patient the MRI findings were diagnostic for this entity. MRI provides high-quality images of human anatomy, displaying high tissue contrast and spatial resolution with no ionizing radiation. Therefore, MRI is helpful to corroborate and refine ultrasound diagnoses and facilitates perinatal management and family counseling for such anomalies. We propose that MRI is a valuable adjunct to ultrasound for the diagnosis and surgical planning for fetus-in-fetu.

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『胎中胎』之磁振造影影像：病例報告

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“胎中胎”或稱“胎兒中胎兒”是一種少見的先天性異常，為一脊椎胎兒被另一正常發展的胎兒包裹於腹中。此腫塊內可能含有骨頭、軟骨、牙齒或中樞神經系統的組織成分。與畸胎瘤不同之處在於它有可辨識的軀幹或四肢。磁振造影因可呈現出在此病灶內不同的組織變化，所以為一理想的診斷工具。但是只有少數的文獻記載使用磁振造影來診斷此病灶。在此呈獻此少見疾病的磁振造影特徵。

關鍵詞：胎中胎，磁振造影