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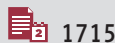
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Intracranial Parasitic Fetus in a Living Infant: A Case Study with Surgical Intervention and Prognosis Analysis

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Corresponding Author: Lan Yao, e-mail: yaolan@pkuh.edu.cn
Financial support: None declared
Conflict of interest: None declared**Patient:** Female, 1-year-old
Final Diagnosis: Parasitic fetuses
Symptoms: Large head circumference • delays in motor skills and speech development
Clinical Procedure: —
Specialty: Anesthesiology • Neurology • Neurosurgery • Pathology • Radiology**Objective:** Rare disease
Background: Fetus in fetu (FIF), or parasitic fetus, is a rare malformation that typically occurs in the retroperitoneum, but can be found in other unusual locations, such as the skull, sacrum, and mouth. The presence of a spine is necessary for diagnosis.
Case Report: Intracranial FIFs were retrospectively studied. Abnormalities were detected in the fetal head during a 33-week prenatal examination; however, MRI could not provide more information, due to space occupation. A baby girl was born via cesarean delivery at 37 weeks, with a large head circumference. She had delays in motor skills and speech development, only able to say “mom”. There was a large mass in the cerebral hemisphere, with a 13-cm maximum diameter, smooth boundary, and internal bone structure visible on head CT scan. Both ventricles and third ventricle had hydrops, with a fetal shape at a continuous level, along with apparent compression near the cerebral parenchyma. After performing preoperative examinations, laboratory tests, and surgical planning, craniotomy was performed on the FIF, under general anesthesia. Following complete mass resection, mouth, eye, arm, and hand shapes could be observed. The patient was unconscious after surgery and had seizures that were difficult to control. She died 12 days after surgery. Teratomas can be distinguished based on anatomy and imaging. Surgical resection is the only curative treatment and its prognosis is poor.
Conclusions: Intracranial FIF cases are rare and require early diagnosis and surgical treatment. Differentiating between FIF and teratoma is crucial, and monitoring alpha-fetoprotein levels after surgery can help detect recurrence.**Keywords:** Fetus-in-Fetu • Teratoma • Anesthesia • Magnetic Resonance Imaging (MRI) • Radiology • Pathology, FetusFull-text PDF: <https://www.amjcaserep.com/abstract/index/idArt/944371>

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Introduction

A fetus in fetu (FIF), also known as an inclusion fetus or parasitic fetus, refers to a fetus containing one or more additional fetuses within a complete fetus, [1-3]. Its unique diagnostic criteria involve the vertebral system and organoid formation [4], often accompanied by the formation of the ribs and quadrilimb bones [5]. Several hundred cases are reported every year [6,7], mostly in males (with an approximate male to female ratio of 2: 1) [8], and about two-thirds of cases begin before the age of 2 years [2]. Most FIFs live in the retroperitoneum (80%), but some can be found in the oral cavity, brain, mediastinum, thoracic cavity, pelvic cavity, sacrum, and scrotum [6-10]. Several manifestations, such as hydrocephalus in this case, can occur as a secondary effect of the mass on nearby organs. Early detection and surgical removal are the only effective treatments. The following is a case of humanoid differentiation in a FIF admitted to our Neurosurgery Department in 2021.

Case Report

A 1-year-old female patient, with a height of 70 cm and weight of 13.5 kg, was admitted to the hospital because of her inability to stand, along with an increasing head circumference.

At 33 weeks of gestation, a routine prenatal check-up revealed an enlarged fetal head circumference on abdominal ultrasonography. Owing to the inability to determine fetal characteristics through magnetic resonance imaging (MRI) in the intracranial space, a cesarean delivery was performed at 37 weeks because the fetus was in a breech position. Upon birth, the infant's head circumference was larger than that of a child of the same age. At presentation, she did not display any signs of nausea or vomiting. The child could only raise her head slightly, could not sit up or walk, and could only pronounce "mom". She exhibited poor fine motor movements in her hands and was incontinent. The patient's head circumference measured 56.6 cm. A head computed tomography (CT) scan revealed soft tissue, limb-like bone, and mixed bone tissue shadows in the intracranial area, as well as a mixed density mass of 15.0×13×12.5 mm in the intracranial area. Blood tumor markers show an alpha-fetoprotein level of 5.6 ng/mL and human chorionic gonadotropin level of less than 0.1 IU/L, leading to the diagnosis of an intracranial FIF, as shown in **Figures 1 and 2**. Craniotomy was performed under general anesthesia and tracheal intubation, revealing a white capsule within the brain tissue. Approximately 50 mL of the brown viscous capsule was extracted after opening the capsule, revealing a finger-like limb protruding from its opening (**Figure 3**). We removed an immature embryo, with visible vernix and organs, such as the mouth, eyes, fetal head, fetal hair, body, forearm, hands, and feet (**Figure 4**). Intraoperative head MRI confirmed the complete removal of the FIF (**Figure 5**). MRI of the FIF (**Figure 6**) allowed

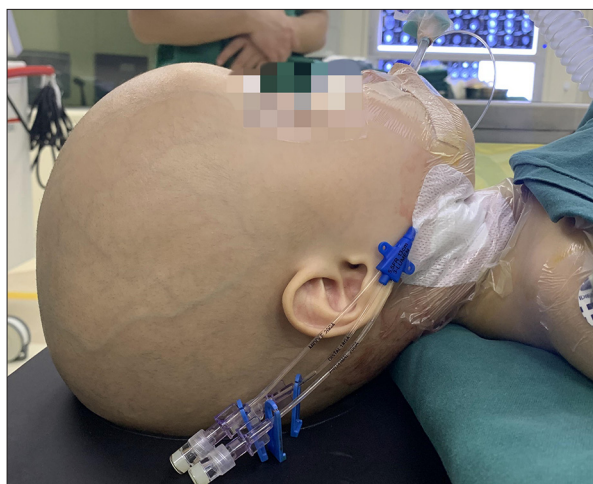


Figure 1. Head of the child after anesthesia.

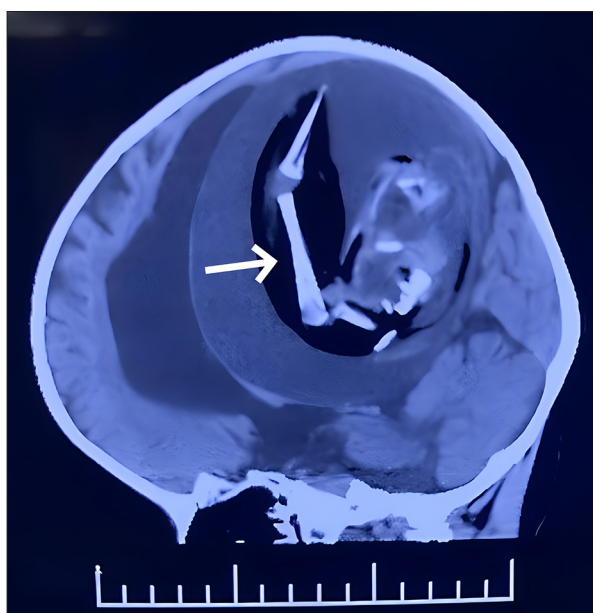


Figure 2. Preoperative computed tomography (CT) image of the head. CT reconstruction in the sagittal position showed a large mass in the cerebral hemisphere with a maximum diameter of 13 cm, clear and smooth boundary, and internal bone structure. Fetal shape was observed in multi slice, with obvious compression near the cerebral parenchyma, and hydrosis of both ventricles and the third ventricle. The arrow points to the long bone.

visualization of the structures of the spine and long bones. The pathology report detailed 1 embryo, 18 cm in length, exhibiting malformed features and limbs (**Figure 4**). The fibrous capsular skin was examined, revealing the interior covered with squamous epithelium with hyperkeratosis (**Figure 7**), consistent with a FIF. Three days after the surgery, the cystic fluid was found to contain amniotic fluid (**Figure 8**). A-fetoprotein was measured at 1.6 ng/mL and human chorionic gonadotropin at 0.1 IU/L.

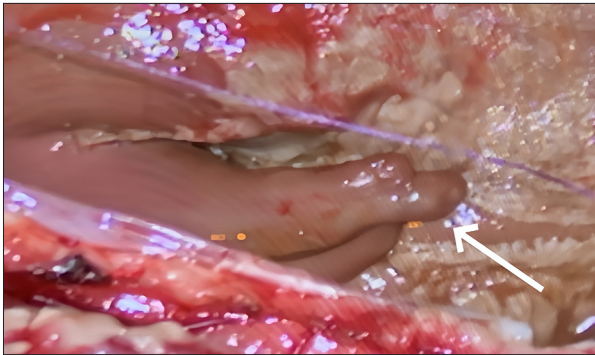


Figure 3. A finger-like limb (the white arrow) can be seen when the intracapsular membrane of the brain tissue was opened.

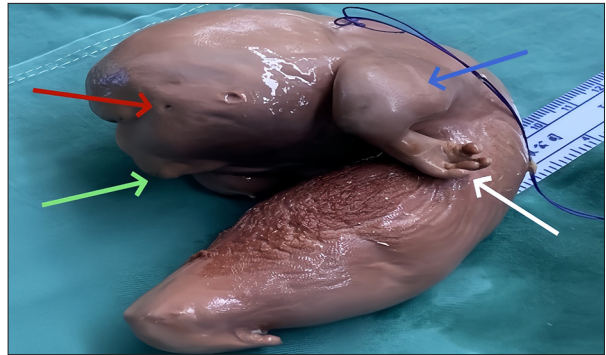


Figure 4. The fetal body can be seen, organs such as the mouth (green arrow), eyes (red arrow), trunk, forearm (blue arrow), and hand (white arrow) are visible.

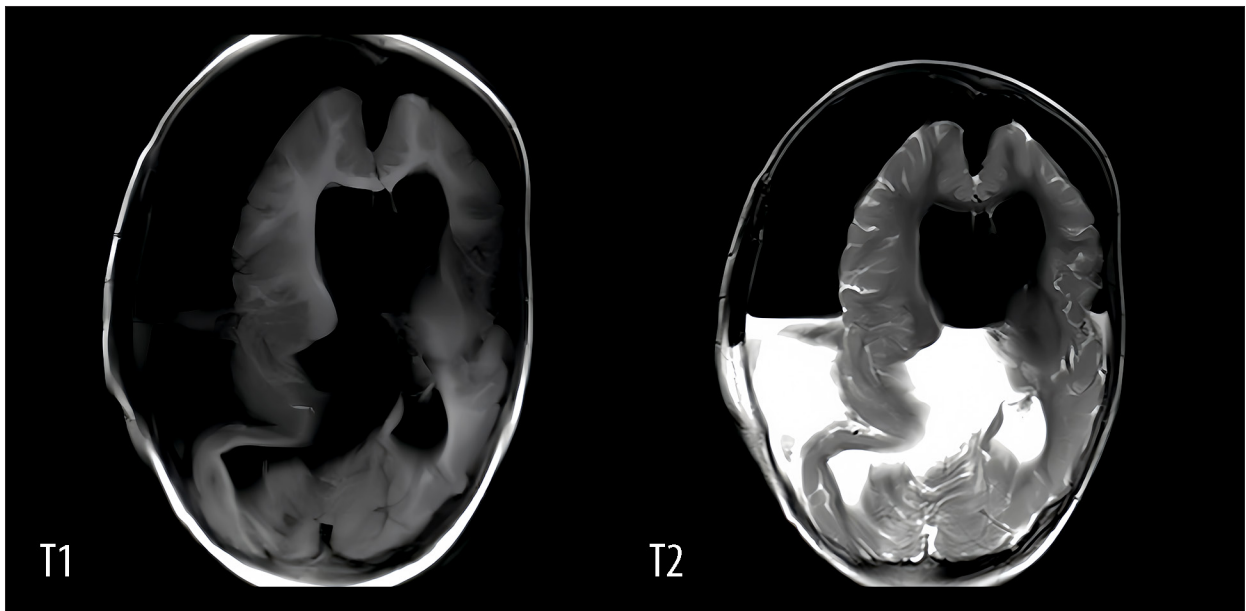


Figure 5. Following brain tumor surgery, the intracranial and lateral ventricles in axial T1 and T2 images displayed flattened air and liquid, increased ventricle size, reduced brain parenchyma volume, and absence of brain tumors.

Discussion

Intracranial FIFs exhibiting human-caused variations in infants are uncommon. Willis described them in 1935 as masses including the spinal column and other visible organs or limbs [4]. A FIF consists of well-developed fetal organs, spine, and limbs. A spine indicates that the fetus has passed the primary stages of gastrulation, including neural tube formation, polymerization, and symmetrical growth around the vertebral axis [11]. In the present case, a head CT scan of the child revealed a clearly defined spinal structure and long bone structure, further confirming the diagnosis of parasitic pregnancy. It has been reported that the presence of fingers in FIFs facilitates the diagnosis [12]. FIFs remain a mystery, and their causes and mechanisms may be related to environmental pollution, genetics, low temperatures, oocyte senescence, pesticide

exposure during early pregnancy, and other factors [13,14]. FIF is often detected in the late stages of pregnancy or after birth, making early diagnosis challenging [10]. In this study, the mother received a diagnosis during a prenatal examination at 33 weeks of gestation. It has been reported that 89% of cases are detected before 18 months of age [15], possibly due to the disease's tendency to develop in late gestation, making it rarely detectable during routine screening in the second trimester. Prenatal ultrasound examination is the best method for diagnosing a FIF during pregnancy [16,17], highlighting the importance of regular prenatal check-ups.

Thakral et al [18] reported that the incidence of FIF was equal in males and females when reviewing the literature, yet Patankar et al [19] reported a male to female advantage of 2: 1, compared with teratomas. Most FIFs were found to be

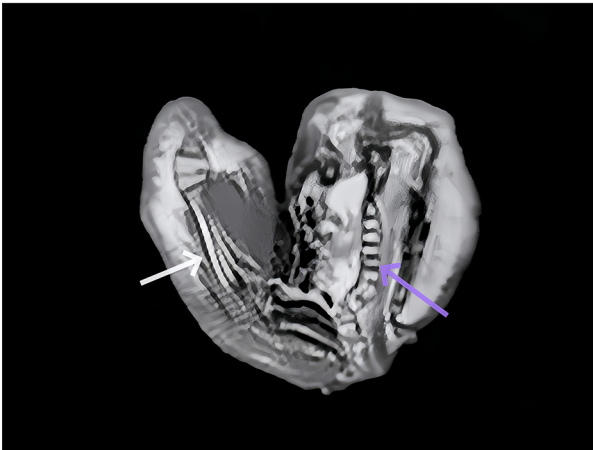


Figure 6. The spinal column (purple arrow) and long bone (white arrow) morphology were shown on magnetic resonance imaging of the fetus in fetu.

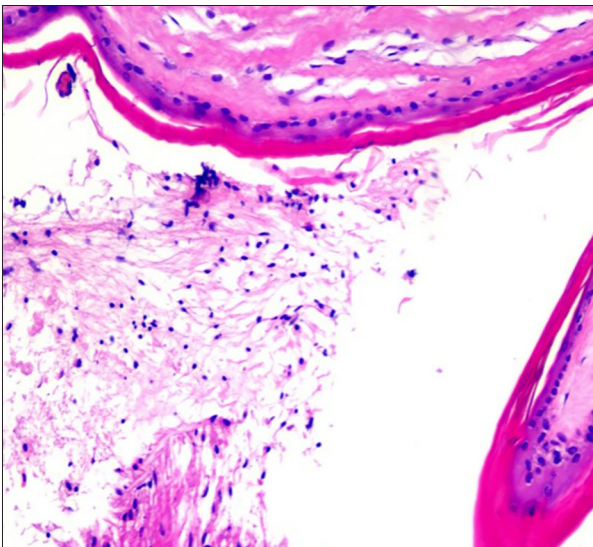


Figure 7. Squamous epithelium with hyperkeratosis.

male (47: 35) [20]. FIFs are primarily located in the retroperitoneum and abdominal cavity and can grow alongside with the fetus [21]. Many patients can experience mass effects, including abdominal distension, dysphagia, vomiting, difficulty eating, and jaundice [22]. These symptoms depend on the location, growth rate, and size of the FIF. The FIF consists of a fetus suspended from the umbilical cord and fluid containing thin fibrous membranes (the chorio-amniotic complex and amniotic fluid). In the present case, an intracranial parasitic fetal membrane and intracellular fluid were detected pathologically. Squamous epithelium with hyperkeratosis was observed, and amniotic fluid was found in the intracellular fluid. These findings confirmed the diagnosis of a FIF. Most FIFs are treated surgically, and their prognosis depends on the parasitic site [16]. Abdominal FIFs generally have a good prognosis; however, intracranial FIFs carry a nearly fatal prognosis [10]. Since

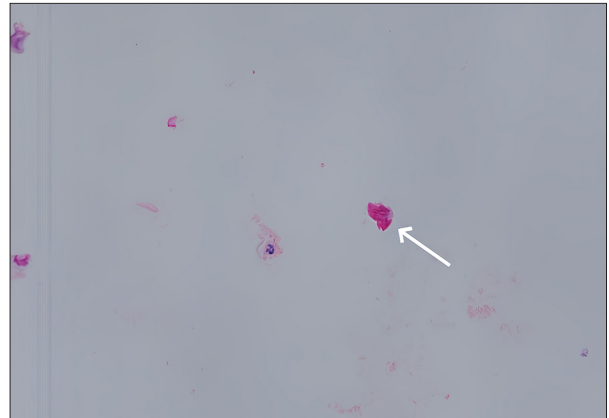


Figure 8. Brown capsule fluid containing amniotic fluid. The white arrow shows amniotic epithelial cells.

the FIF grows within the host's brain tissue, it leads to severe brain tissue compression and accumulation of cerebrospinal fluid, resulting in cranial hypertension. Our patient, who was placed in the Postoperative Intensive Care Unit for treatment after resection of an intracranial FIF and continued to have large epileptic seizures, presenting a state of unconsciousness. The family discontinued treatment 12 days after the operation.

Clinicians have difficulty differentiating mature teratomas from FIFs. It is debated whether a FIF is a distinct entity or a well-organized teratoma [23]. The 7 main distinguishing points between the two are as follows. (1) Because the FIF has axial bone, it indicates that it has entered the primitive notochord stage, whereas the teratoma has a small amount of bone. (2) A FIF is anatomically more distinct, has different degrees of development, and presents benign lesions. There are normally 3 germ layers in the teratoma, and the teratoma cannot form an entire organ system [24]. Teratoma is potentially malignant as well. (3) The FIF usually appears like a typical fetus, covered with skin, in an amniotic sac lined with epithelium or squamous epithelium [25]. The teratoma is characterized by multiple cystic cavities, poor tissue differentiation, and little to no forming tissue, such as hair, skin, or teeth. (4) Most FIFs were found in the abdominal cavity or retroperitoneum, followed by the sacrococcygeal tail or the thoracoabdominal wall, and fewer in the head and neck. Generally, teratomas occur in the lower abdomen, pelvic cavity, and sacrococcygeal tail. (5) Ultrasound images of FIFs show cystic solid mixed echo masses with clear boundaries and fine echoes in the cystic region. Ultrasonic differential diagnosis relies mainly on strong echoes detected from the mass, such as the long bone and skull, or visible cord-shaped blood flow signals [26]. Generally, teratomas present a mixed echo with a distinct boundary and envelope, a strong echo or no echo signal, dense spot hyperecho, and lipid stratification. Strong echo is mostly observed in the mass, nodule, or short line shape, with poorly differentiated organ and limb soft tissue. The envelope and inside of the teratoma

cannot display blood flow signals, due to the lack of blood flow signals in color Doppler flow imaging. (6) During a CT examination, teratomas and endoparasites present mixed densities, including bone, soft tissue, fat, and fluid. However, the endoparasite exhibits long bones, vertebrae, skulls, and ribs. Using 3-dimensional recombination, a full picture of the bones and spine of the endoparasite was more clearly shown [27]. More than half of the masses are blood vessel-based. Finally, (7) the MRI revealed a large mass containing bone, highly differentiated tissues and organs, and a fat signal shadow. An under-calcified intervertebral disc enhanced the accuracy of the FIF diagnostic test [28].

Ultrasound during pregnancy is the preferred method for detecting a FIF, offering benefits such as being noninvasive, radiation-free, and easy to perform; however, the rate of successful prenatal diagnosis is below 20% [29]. MRI can provide more information than ultrasound, such as the anatomical location and tissue characteristics of the mass [30]. FIFs and teratomas can be differentiated with the help of CT examination, which is the main method for diagnosing FIFs. However, despite its radiation risk, CT is suitable for pre-surgery diagnosis [31] and it is not permitted for use during pregnancy.

The residue after removing the FIF has been reported to contain malignant changes [32]; in the present case, blood alpha-fetoprotein levels decreased significantly. Monitoring of tumor markers, such as alpha-fetoprotein and human chorionic gonadotropin, can be used for diagnosis, follow-up observation, and determination of a FIF cure [29,30], and long-term follow-up after surgery is recommended.

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Conclusions

The occurrence of intracranial FIFs is exceedingly rare and requires prompt differential diagnosis and aggressive surgical treatment. According to previous literature, we identified the key differences between FIF and teratoma from anatomical and imaging perspectives. A measurement of alpha-feto-protein was performed after surgery to diagnose recurrences.

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Declaration of Figures' Authenticity

All figures submitted have been created by the authors who confirm that the images are original with no duplication and have not been previously published in whole or in part.

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