



Fetus-in-Fetus: Our Case Series and Review of the Literature

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Abstract

Fetus in fetu is a rare condition commonly presenting as an epitomized pedunculated vertebrated fetiform mass in a newborn infant. Different kinds of presentation and locations have been reported in the hosts. Although numbers of instances have been reported the fascination and curiosity about other presentations and genesis of fetus in fetu remains confirmed. In Tabriz pediatric surgery ward we have had five different presentations of fetus in fetu. The aim of this case series and a subtle literature review is gathering these interesting presentations.

Keywords

- Fetus in fetu
- Adominal mass
- New born

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Introduction

Fetus in fetu is a rare condition commonly presenting as an epitomized pedunculated vertebrated fetiform mass in a newborn infant. Few cases of fetus in fetu have been presented and the great majority of them have been revealed after a surgical procedure. With advanced imaging techniques the extraordinary features of calcified vertebral segments and bones of the fetus in fetu can nowadays be diagnosed

preoperatively by pediatricians and pediatric surgeons. Some reviews of fetus in fetu have mentioned recognizing it prenatally. (Sada et al, 1986; Chitrit et al. 1990)^{1,2} and the identification thus far has been limited only to an appropriately characterized epitomized mass with a solid calcified segment encompassed by fluid within the fetal abdomen. The revelation of a fetus inside a toddler or a male young adult is a particularly interesting

Erratum:

The name "Mohammad A Baky Fahmy" and "Sina Zarrintan" were mistakenly omitted in the publication of this article and hereby are added.

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finding that records of these occasions has existed for quite a long time.¹⁻⁴ Inside the more seasoned literature, disarray existed between fetus in fetus and more common diagnoses such as teratoma.

Prevalence

The prevalence is unknown. Less than 200 cases have been stated;⁵ however this varies based on how strictly identification standards are used. Numerous cases are officially identified as fetus in fetus, even though completely dismissed by others.⁶ Some cases have been diagnosed in uterus⁷ An expected frequency of 0.02:10000 is normally stated within the literature; however this number is primarily based on the unsubstantiated assumption that fetus in fetu represents 5% of conjoined twins. The male-to-female ratio within the 39 reports that we reviewed was M:F=1.3: 1, that is in contrast with conjoined twins, which happens predominantly in girls.

Localization

Most cases of fetus in fetus are retroperitoneal,⁴ however some have been located within the mesentery,^{8,9} adrenal,¹⁰ cranial cavity,¹¹ lateral ventricles, pelvis,⁸ inguinal area,¹² retroperitoneum,¹³ and oropharynx,¹⁴ scrotum.¹⁵

This shows that many of the resting locations are retroperitoneal or on the route that the germ cells follow on their way back from the yolk sac to the retroperitoneum and to the gonads. When placed on ectopic testes, they may be intraperitoneal.¹⁶ The affected testicle is usually ectopic or undescended, possibly because the introduced bulk hinders the migration of the cells.

Age and weight and Number at detection

Only few cases have been detected parentally.⁷ Many cases are now detected in neonates or toddlers. The weight of cases range from as low as 2000g⁸ to 4000g.¹⁷ There's some estimation regarding the weight of the cases since a few reviews consider the weight of the entire tumor, at the same time others report the weight of the fetus in fetu alone.

Commonly one fetus is discovered, however numerous cases of two,¹⁸⁻²⁰ three,⁸ five⁶ or even more were identified. At the point when several fetuses are available, they typically share an identical sac.⁸

Microscopic representation

At surgery, the embryo in hatchling appears as an appropriately encircled mass limited by a fibrous membrane.

Inside the mass the baby in hatchling is suspended in straw-colored fluid with the guide of a pedicle. Two vessels (one artery and a vein) travel along the pedicle. The liquid is normally no longer in great supply and had been characterized as containing sebaceous material. The wellspring of this liquid is uncertain. Various authors have called attention to that the layers of an ordinary undeveloped embryo are not responsible for the creation of amniotic fluid. They could be considered as a semi-permeable membrane. In fetuses past 12 weeks, the urinary tract creates the liquid and the gastrointestinal tract reabsorbs it. taking into account that no fetus in hatchling has ever been characterized to incorporate a urinary tract, and

the segments of gastrointestinal tract which can be noticed, are too deficient to have any reabsorptive abilities, the liquid is most likely kept up inside the amniotic hollow space totally by means of osmotic and oncotic pressure. The presence of chorionic villi has solely been reported in a single case²¹ aside from one statement of a 15 years old pronouncing ‘Mother, do come to me, i have something alive in my body’ and the mom being cited at declaring that she felt something comparable to “the motion of a child in the course of gestation”, not any fetal motions have ever been reported in a fetus in fetu. The above report of motion is relatively dubious on account that striated muscle mass have not often been located around joints. However this host is perhaps the most seasoned host reported.

What is included?

Fetus in fetu resembles poorly shaped acardiac twins. Most of the organs found had been in numerous levels of development. The remarkable exception is the urinary system, which does not seem to be identified in any of the instances that we reviewed.

A few structures such as ribs,²² intrathoracic organs (lungs, heart, thymus)⁹ and retroperitoneal organs liver, spleen, kidneys, adrenal glands, pancreas and gonads^{9, 1, 23} are hardly ever described. An incomplete heart has been discovered in a single case^{6, 9, 23-26} a primary two chambers heart was observed with the atrium in the caudal position and the ventricle in the cranial position, that is the stage typically reached in a 22 days embryo.¹⁸ The cord that connects the hatchling to the layer has exceptional features compared with a regular cord:

it Carries vasavasorum and nerve fibers.

The development of the baby in hatchling is stopped at the first trimester, and further development is through mass growth more than by means of improvement. Normal frameworks got from the ectoderm are better developed than frameworks got from the other two layers. The mesoderm collaborates in making of the musculoskeletal framework which is often appropriately visible; anyway the various subordinates (the vascular and urogenital framework, the spleen and adrenal organs) are only noticed as a rare exception. The most normally developed structure from the endodermal layer is the gastrointestinal tract and the liver and pancreas are always noticeable.

Our cases

A 20 weeks pregnant 23 years old lady came for assessment of fetal growth and development by sonography. Two fetuses were detected: one male and one female. The amniotic fluid volume, fetal limb and cardiac activities were reported to be normal **Figure 1, Figure 2**.

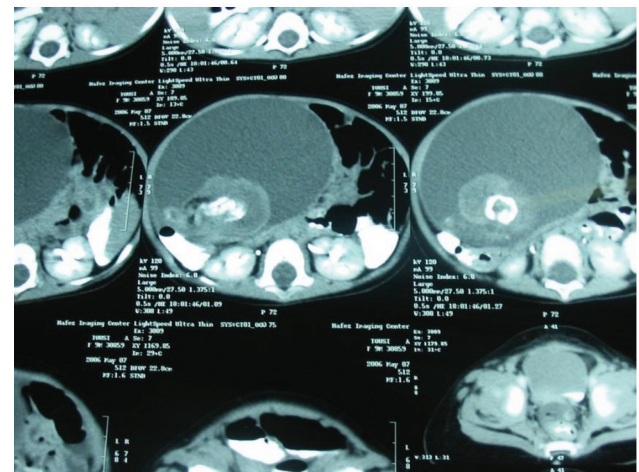


Figure 1: Vertebrated bony mass within the abdominal cavity of newborn



Figure 2 : Fetus in fetus with lower limbs and blood supply originated from host's omentum

Both fetuses had normal anatomy, the only exception being an abdominal mass. Both babies were born at 39th week of gestation, (male: 2600gr, female: 3000gr). Forty three days after delivery one of the twins was brought with a complaint of a large mobile abdominal mass. The radiological reports mentioned teratoma or neuroblastoma as possible differential diagnosis. Yet, since we had found a bone like structure in the mass we considered the potential diagnosis of fetus in fetu. At laparotomy, there was a fetus with lower limbs, an almost normal abdomen and external genitalia, small and large bowel and without any trunk and head which were attached to the host infant's omentum by small vessels. The pathologic report confirmed it to be Fetus in fetu. Nine months after the operation there were no complications.

The second case, we had a similar patient who was referred to us at her 21st day of birth with a large abdominal mass which was discovered by her mother during bathing. The baby was the result of an NVD and was the third baby of

the family. We had a US which showed a mixed cystic and solid mass in the left upper part of the abdomen just in front of her left kidney. During the laparotomy a mass containing an incomplete fetus was discovered which consisted of intestinal tract and kidneys and urinary system and almost normal lower limbs. We simply removed the mass by ligation of small vessels.

The third case was a 3 month-old girl with a solid encapsulated round mass in her abdominal cavity and preoperative diagnosis of teratoma (according to X-ray and ultrasonography and tumor markers). During the operation we observed a round capsulated mass which was attached to the omentum through a vascular band. After opening the capsule we found a fetus-like mass inside it that was completely removed. Further the pathologic exam affirmed the finding of fetus in fetu.

The fourth case was a male newborn with a fetus like mass in his oral cavity which was attached to his hard palate accompanied by a hard & soft cleft palate which had not been diagnosed prenatally. Surprisingly the newborn didn't have any signs of respiratory distress. The patient was intubated immediately and went under operation. During the operation we simply removed the mass without any technical problems. After 9 months the cleft palate was repaired and now he is well with no feeding difficulties.

The fifth case was a 28-day neonate with poor feeding and an abdominal mass in ultrasonography which was referred to our center. The patient also had monorchism and the right testis was not present in the scrotum. Computed tomography (CT) scan

was suggestive of a solid intraperitoneal mass which contained calcified regions presenting as bony structures. Surgical resection of the mass was planned and during operation the mass was totally resected. The resected mass contained two fetuses and the macroscopic and pathologic study of the mass were suggestive of an abdominal fetus-in-fetus.

Discussion

Meckel was the first to describe the term Fetus in fetu; in order to explain the existence of a foetus inside a fetus.²¹ It describes a vertebrate fetiform mass with an axial skeleton and a vertebral column.^{9,21,24} The popular theory is that fetus in fetu is a kind of twin, resulting from early stage duplication (a blanketed twin).^{21,24} A main difference between fetus in fetu and teratoma is the presence of an axial skeleton^{21,23,25, 26} though the embryological basis for this difference has not been proven. Teratomas with normal developed extrimities, digits or different properly-prepared organs without an axial skeleton were mentioned in numerous reports,⁷ showing the similarity of a progress and development among teratoma and fetus in fetu. Because of the presence of both teratoma and fetus in fetu in the same patient and the extra findings showing impeccable developments and growth of the specimen,¹ it is hypothesized that many cases of fetus in fetu are astoundingly perplexing, appropriately separated, fairly organized teratomas. Most of mentioned cases of fetus in fetu are placed within the retroperitoneal region. Sometimes the fetiform mass may be observed in the skull, sacrococcyx, pelvis, proper iliac fossa, mesentery, adrenal gland

, liver, undescended testes, and scrotum.^{2,6,8,12,15,27-29} Fetus in fetu frequently occurs singly, besides for a few stated instances with more than one fetuses which include 21 minute rudimentary fetuses, five fetuses, three, and two.^{6,7,8,17} Most cases of fetus in fetu are identified in infancy; only four instances up to now were stated in adolescence or in adulthood.^{21,26,30,31} The fetiform mass tends to develop slowly and asymptotically within the postnatal duration; even though vomiting, external compression at the digestive and urinary structures, and intense complications such as intraperitoneal hemorrhage and duodenal occlusion have been reported.²¹

Experiencing the above mentioned cases, we recommend that when observing any infant/newborn with an abdominal mass especially with calcified elements, bear in mind that it may be a fetus in fetu.

Ethical Consideration

This study was approved by Tabriz University of Medical Sciences with code number IR.TBZMED.REC.1399.433.

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Conflict of interests

There is no conflict of interest

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