FETUS IN FETU: A CASE REPORT AND REVIEW OF LITERATURE

ATTA ULLAH JAN, QAMAR ZAMAN, UMAR ALI KHAN AND QAZI KHADIM MOHYUDDIN

Department of Surgery,
Postgraduate Medical Institute,
Lady Reading Hospital, and
Hayat Shaheed Teaching Hospital,
Peshawar.

A case of fetus in fetu in a boy, 50 days of age is presented. This baby had a mass abdomen noticed by his parents since birth. Surgical removal revealed more than 5 fetuses within a retroperitoneal sac.

Fetus in fetu as quoted by Lewis is a term first introduced by Meckels (Circa 1800) to describe a rare condition in which malformed parasitic twin was found inside the body of its partner, mostly the abdominal cavity. The pathogenesis, diagnosis and differentiation from retroperitoneal teratoma is still a disputed subject.¹

In most of the previously reported cases only one fetus was found but in this case report we are presenting more than five fetuses in the abdomen of a male child.

CASE REPORT

A 50 days old normally developed male baby weighing 14 lbs was the 6th born child of his parents. It was a normal full term home delivery. There was no contraceptive history of twinning or teratoma in the family. There was no history of pills or any other drug intake during pregnancy. The parents noticed abdominal swelling at time of birth, smaller to start with and gradually increased in size on till six days before presentation to surgical out patient department. It has rapidly increased in size and become painful. The baby was having yellowish vomiting which was not related to feeds. There was no history of diarrhoea, dysentery or constipation. No urinary symptoms were obvious.

Clinical examination under sedation revealed an intra-abdominal mass, mostly in the centre but extending towards the left side. The mass was firm to hard in consistancy, fixed, non-pulsatile and had no movements with respiration. The cardiopulmonary, genitourinary and central nervous systems were found normal. The results of Laboratory examination were as follows; Hb: 10.2 gm%, ESR- 90mm 1st hour, TLC-8500/cm (Poly:49%, Lymph :47%, Mono:2%, Eosin:2%) plain X-Ray abdomen showed areas of linear calcification in the area of abdominal mass. I.V.U normal and a Clinical diagnosis of Retroperitoneal teratoma was made.

The baby was operated on 23.1.1994. On operation a retro peritoneal mass in the left side of the abdomen extending towards midline was found. The mass

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Fig. 1. Largest developed Fetus-in-fetu showing fully developed limbs, stomodeum, umbilical cord, external genital tubercle and fine hair on the head which is incomplete.

Fig. 2. Radiographs of all Fetus-in-fetu. The fully developed one showing vertebral column, rib cage, base of skull and long bones.
was firm, nodular and partly cystic. On opening the membrane clear fluid drained. The sac contained multiple fetuses amongst which one was fully developed but anencephalic. (Fig. 3)

The others were in various stages of development. Four separate fetuses could be differentiated but one of them showed body parts of more than two or three conjoined fetuses. Fetuses were delivered and sac could easily be separated in most of its part. Post operatively the child had uneventful recovery and discharged home on 5.2.1984 i.e. 13th post operative day.

PATHOLOGICAL FINDINGS

The mass was found to have greyish-white fibrous sac which on opening revealed 4-fetal masses attached separately to the fibrous sac, the sac contained clear watery fluid.

No membranous separation was found between them. (Fig. 2) The large fetus measured 7cm X 6cm X 4.5cm and the conjoined triplets measured 7cm X 7cm X 6.5cm. The third one measured 5cm X 2.5cm X 1.5cm and fourth one measured 5cm X 0.75cm X 0.5cm. All the fetuses were anencephalic with fetal grey skin covering the body and fine black hair could be seen on top of stomodeum in large fetus. The larger one had all four limbs developed with stomodeum, external genital tubercle, umbilical cord, and digits could be differentiated but no nails. (Fig. 1)

The roentgenography of the fetuses (Fig. 2) revealed well formed long bones, spinal column, ribs and base of skull in single well formed fetus. Histology revealed keratinised stratified squamous epithelium and greater part of the fetus consisted of connective tissue.

The second large fetal mass consisted of multiple body parts fused together and the limbs could be clearly demarcated which belonged to more than two conjoined fetuses. At places intestine like structures could be clearly seen. (Fig. 3)

The third specimen had trunk and spad like hand but other parts could not be clearly seen. The fourth specimen was not fully developed.

DISCUSSION

First case of fetus in fetu was reported by Young in 1809\(^{(1)}\). Lord in 1956 after an extensive review noted 30 alleged cases prior to 1900 and 11 cases including her own from 1900-1956. Out of these 11 cases only 4 cases had unequivocal evidence of the condition. Subsequently till March 1989, 19 more cases were reported in the literature. Alpers and Harrison\(^{(17)}\) found less than 30 generally accepted cases recorded in literature till 1985. While according to Montupet P et al\(^{(26)}\) only 22 cases were quoted in the literature including two cases of his own till Jan 1985. According to Hing A et al over 50 cases of Fetus-in-fetu have been recorded since 1800 to Sep 1993.\(^{(22)}\) The incidence of fetus in fetu is 1:500,000 deliveries.\(^{(22)}\)

Fetus in fetu is usually discovered in early infancy as retroperitoneal mass but there are cases on record where it was seen at age of 17,15.9 and even 47 years old man, by Highmore Know and Webb as quoted by Hock Liew Eng\(^{1}\) and Degradi AD et al\(^{(33)}\) respectively. An interesting case was reported by Grosfeld et al in 1974, on an autopsy of an old man killed in road traffic accident. \(^{(4)}\) In our case the tumor was first noted at birth which gradually increased in size suggesting post natal growth of fetus in fetu, a phenomenon also studied by Kim OH and Shin KS, detected by series of radiographs.\(^{21}\)
Fig. 3. (a) Showing Fetus-in-fetu as seen in operating field. Digits of the feet can be clearly counted. (Right)

Fig. 3. (b) All four specimen. From L to R, conjoined fetus-in-fetu showing body parts of more than two fetuses. Intestine like structures can be seen in the right part of the specimen. The single fully developed fetus-in-fetu showing parts as seen in Fig. 1. The third one shows all four limbs & trunk with umbilical connection protruding from the trunk toward right. Fourth one incompletely developed fetus in fetu. (Below)
In mostly previously reported cases and in our case fetus in fetu was present intra-abdominally and in upper part of retroperitoneal space, but less frequently it can occur in the pelvis and ileal mesentery. The unlikely sites of occurrence reported in literature include cranial cavity, scrotal sac, and testis sacrococcygeal, inguinal region, liver, mouth and adrenal.

The number of fetuses in fetu is usually one but two to five have been reported. In our case more than five were found as four separate masses but one conjoined fetus had body parts of more than two fetuses. Thus it is the second case report with regard to number of fetuses in one case. The highest number reported is twenty one by Povysilova.

Only one fully developed fetus in fetu had umbilical cord attached to the mesentery of transverse mesocolon but no definite placental tissue was found. The other fetuses had fibrous connection to the root of mesentery of small intestine. Only five other cases were reported to have definite vascular connections, otherwise intra-abdominal fetus in fetu is usually suspended by a pedicle within a complete sac containing little fluid. Hock Liew Eng is of the opinion that the lack of independent vascular supply means subsequent fetal growth retardation. Only one case has been reported where chorionic villi were demonstrated at the point of attachment to abdominal wall.

Many theories have been presented by various research workers at different times regarding the pathogenesis of fetus-in-fetu. Some workers have put forward the hypothesis of modified process of twinning and have traced natural progression from normal twins to conjoined asymmetrical twins, through parasitic fetuses and fetal inclusions and finally to teratoma. (Gross RE and Grant P quoted by Hock Liew Eng).

On the other hand Willis in 1935 in his article "The Structure of Teratoma" discarded the above theory. His view is stated here as such as quoted by Hock-Liew-Eng et al. "The fetus in fetu most likely arise from inclusion of a monozygotic, diamniotic twins within the bearer." He adopted the presence of vertebral axis (indicating development through the stage of primitive streak) and an appropriate arrangement of other organs or limbs with respect to vertebral axis as the criterion to distinguish fetus in fetu from highly differentiated teratoma. He regarded teratoma as a discordant congregation of pluripotential cell that demonstrate no vertebral segmentation or systemic organogenesis. Lacking a primary organiser, embryonic tissue composed of teratoma can not pass through an organised primitive streak stage, which endows the developing organism with its fundamental vertebral pattern. In addition, teratoma occurs in many extraperitoneal locations and has definite malignant potential except a very highly differentiated teratoma. In contrast the fetus in fetu is almost always benign in nature and retroperitoneal in location. Though Willis theory is now generally accepted and has been supported by genetic, cytogenetic and serological assessment, yet the pathogenesis of fetus in fetu in other locations is controversial.

Because of the presence of definite vertebral column, ribs, long bones and base of skull (Fig. 2) we consider our case as true fetus in fetu as it fulfils the requirements of Willis theory and so is not a teratoma. We based our diagnosis on operative findings and radiology,
though antenatal diagnosis have been reported in certain cases.\textsuperscript{6,7,8,9}

REFERENCES


3. Lewis PH: Foetus in Foetu and the retroperitoneal teratoma. Arch Dis Child 1961; 36;


