FOETUS IN FOETU

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Foetus in foetu, a term coined by Meckel (quoted by Lewis, 1961), is applied to those cases where a parasitic twin is found included within its bearer. It is rightly regarded as a rarity. Since Young reported a detailed study of a case in 1809, only sporadic case reports appeared from time to time, only 13 having been traced since 1900. Lord (1956) reported 2 personally studied cases and reviewed 7 others from published reports. In most of these, only one foetus was found, and nearly all were intra-abdominal in situation (Lord, 1956; Lewis, 1961; Janovski, 1962). In this communication, an example of inclusion quadruplets is reported with unusual and perhaps unique features.

Case Report

A 2-month-old Chinese male infant was admitted to the Professorial Surgical Unit, Queen Mary Hospital, Hong Kong, on May 10, 1964. The complaint was that of gradual abdominal distension for 10 days with no symptoms of obstruction either in the gastro-intestinal tract or in the genito-urinary tract. He had been taking feeds well and weight gain had been normal. The child was the last sib in a family of 11. There was no history of twin pregnancy.

On examination the infant appeared well nourished and healthy. His weight was 12 lb. (5,443 g.). There was no sign of dehydration or anaemia. The abdomen was grossly distended across the midline with dilated superficial veins coursing vertically and upwards. An irregular but circumscribed firm mass was palpable in the abdomen extending into the pelvis. On rectal examination, the same mass was felt to the right of the rectum and was ballotable bimanually. It was slightly mobile but could not be pushed out of the pelvis. Emptying the bladder failed to alter the physical characteristics of the mass.

Investigations. Haemoglobin was 11.5 g./100 ml. WBC count 8,900/c.mm. Blood group A. Urinalysis was normal. A plain radiograph of the abdomen was taken; complete spinal columns and other skeletal parts were seen in a large soft tissue mass (Fig. 1).

Operation. Three foetuses were found bathing in clear fluid inside a single thin wall sac to which they were

attached by umbilical cords (Fig. 2). The sac itself was situated in the pelvis behind the bladder. The rectum was pushed to the right. On cutting the umbilical cords, arterial blood was found spurting from the distal ends and on tracing them further, they were found to lead to the mid-sacral artery, to which the sac was adherent. The three foetuses with the sac were easily removed. The abdomen was closed in layers without drainage. Post-operative course was uneventful and normal feeding was rapidly re-established. The infant was discharged on the 8th post-operative day. The patient has shown normal progress when seen in the follow-up clinic.

Pathology. A detailed study of the specimens was made by dissection and microscopy. The sac was found to be lined by one layer of endothelial cells. No placenta was found. The largest specimen, measuring 15 cm. $\times$ 6.2 cm. and weighing 1,262 g. was the most developed of the 3. It was covered by skin with hair-bearing areas in front and at the shoulders, 4 well-formed limbs were present, bearing 5 and 6 digits on the upper limbs and 7 and 6 on the lower limbs. There was no face or skull discernible, but a well-formed central nervous system with differentiation into cerebrum and cerebellum covered with meninges and in continuity with an exposed spinal cord was found. The skeletal system appeared to be of the highest development. The radiograph of the specimens (Fig. 3) and dissection showed a well-developed spine composed of a series of vertebrae with characteristic cranio-caudal differentiation, namely, well-formed thoracic, lumbar, and sacral parts. The thoracic part showed related ribs on either side and the sacrum was found attached to a well-formed pelvic girdle. Scapulae were well developed, so were the limb bones as distal as the digits. Thus, excepting the skull and facial bones, all other parts of the skeleton were as well developed as those of a full-term infant.

The nervous system also showed a high degree of differentiation (Fig. 4), two cerebral hemispheres with enclosed ventricles were found. The cerebellum was identified. The spinal cord, enclosed in a spinal canal lined by meninges, was exposed at its upper half. Spinal nerves with ganglia were found to be given off segmentally.

Loops of intestine were found suspended in mesentery within an endothelium-lined cavity (Fig. 5). The caecum and appendix were identified, and, therefore, small and large bowels were easily differentiated. No

FIG. 1.—Radiograph of abdomen showing well-formed spinal columns and other skeletal parts.

FIG. 2.—The three foetuses with their ruptured sac. Arrows show the umbilical cords attached to the foetuses.

FIG. 3.—Radiograph of the specimens showing spinal columns and other skeletal parts, including thoracic cages.

derivatives of the genito-urinary tract were discovered, though what appeared to be a genital tubercle was found in the perineum. The organs described were all well supplied with blood vessels, but no heart or aorta was found, though a particularly large artery was found in the umbilical cord.

The other two foetuses were much less developed,

weighing 52 g. and 13 g., respectively; nevertheless, they showed well-formed skeletal parts, coelomic cavities with intestinal loops, umbilical cords, and nervous tissues. There was little doubt that they were foetuses of a more primitive development than their companion. All three were included in a single sac and all three had the same blood group as their bearer.

FIG. 4.—Dissected specimen showing well-formed central nervous system, including cerebrum, cerebellum, spinal cord, and their meninges. Arrow points to a part of the cerebellum.

FIG. 5.—Dissected specimen showing loops of intestine in an abdominal cavity as shown by the arrow. The umbilical cord shows two large vessels and several intestinal loops. A scrotum-like nodule seen at the perineum.

Discussion

Willis (1935) adopted the criterion of diagnosing foetus in foetu, in distinguishing it from teratoma by the presence of a vertebral axis and by an appropriate arrangement of other organs or limbs with respect to the axis. When this criterion was adopted, probably many of the recorded cases were not true examples of foetus in foetu. Lord (1956) found 31 reports of alleged cases before 1900 and 11 reports since that year. Of the 11 more recent cases, only 4 had unequivocal evidence of the condition. Most of these were intra-abdominal foetus in foetu. Examples of foetus in foetu at other sites were less common. Kimmel, Moyer, Peale, Winborne, and Gotwals (1950) described a cerebral tumour in which 5 foetuses were found. Older writers customarily subdivided cases of abdominal foetus in foetu into two groups, extra- and intra-abdominal. But those reports of allegedly extra-abdominal origin (Taruffi, 1885; Curtis, 1826) are far from clear in their descriptions.

In all cases of intra-abdominal foetus in foetu in which the sites of origin were specified, the retroperitoneal tissues in the upper abdomen were found to be the sites. In many of the surgically treated cases, the site was not ascertained. In the present

case the sac was found to be attached to the mid-sacral artery by a pedicle in the retroperitoneal space in front of the sacrum. It appears to be the only case of pelvic origin.

Another unique feature in this case was the number of foetuses found within a sac. Gross and Clatworthy (1951) reported a case of twin foetuses. All other examples of intra-abdominal foetus in foetu consisted of only one foetus. All three foetuses in this case showed separate umbilical cords, all showed well-formed spinal columns and skeletal parts, and all had well-developed intestinal elements.

Pathogenesis. Many theories have been advanced to elucidate the pathogenesis of foetus in foetu. It has been repeatedly emphasized not to confuse them with highly differentiated teratomata. Teratoma, though it may contain highly organized structures, is a true neoplasm (Willis, 1953) which arises from embryonic pluripotential cells with benign or malignant propensities. Whereas a foetus in foetu, as its name implies, is a parasitic twin within its fellow. The most plausible explanation is that it is a monozygotic twin of its bearer. White (1956), as quoted by Lord (1956), suggests that it is included as a consequence of the anastomosis of the vitelline

circulations. In the present case, though the sex of the malformed fetuses is not externally discernible, they are all confined in one sac and have the same blood group. The two smaller masses are examples of amorphous twins which had been overshadowed in development by their larger co-twins.

Summary

A unique example of foetus in foetu is described. Three fetuses of various stages of development in a single sac were found at operation in the pelvis of a 2-month-old Chinese male infant. The child survived.

Careful dissection of the fetuses revealed well-formed skeletal, intestinal, and nervous parts in all three members, all of which had umbilical cords attached to the host.

The published material is briefly reviewed and the diagnosis and pathogenesis of the condition are discussed.

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