A RARE CAUSE OF OBSTRUCTED LABOUR

Diffuse Lymphangiectasis with multiple lymphangiomas

(A Case Report)

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Diffuse lymphangiectasis is a very rare condition and it is still rarer to find it to be a cause for obstructed labour. Potter (1962) had described this condi ion among abortuses and in neonates. Review of the literature give enough information about this condition, but no case has been reported resulting in obstructed labour. Hence this case has been reported due to rarity of the condition.

CASE REPORT

Mrs. K. aged 35 years, was admitted on 11.8.1972 at 7.30 a.m. as a case of obstructed labour. She was a fifth gravida with previous four full term normal vaginal deliveries. She was full term pregnant and was in labour since previous night. She was being attended by 'dai' at home. Membranes had ruptured at 3.00 a.m. Head and shoulder had delivered at 5.00 a.m. (2½ hours before admission), and further delivery of the foetus was arrested.

On examination she was exhausted, anaemic B.P 130/90 mm. Hg., pulse 104/mt. regular, good volume; temperature was 37°C.

Respiratory and cardiovascular systems were normal.

On abdominal palpation uterus was upto xyphisterum, foetal parts were felt but foetal heart sounds could not be localised. There was marked distension on the right side. She was having strong bearing down pains. Vaginal examination revealed that head and shoulders were lying outside the introitus and further examination was not possible since foetal mass was impacted in the vagina. She was taken to the operation theatre for examination and extraction of the foetus under general anaesthesia.

Operation: Under general anaesthesia vaginal examination revealed oedematous thick abdominal wall of the foetus giving an impression of foetal ascites. An attempt was made to tap the foetal ascites but no fluid was aspirated. Evisceration was done to help the delivery of the foetus but it did not reduce its size. Finally, it was decided to decapitate the head and deliver the body by the abdominal route. The abdomen cavity was opened by subumblical midline incision. The lower uterine segment was found to be overstreched. The foetus was delivered by lower segment caesarean section. The uterus was stiched in layers and both the tubes were ligated by modified Pomerov's technique, as she was a multiparous patient. Female baby weighing 14 lbs was extracted. On gross examination, generalized solid oedema of the body was noted with localized swellings in the iliac and groin regions. X-ray of the foetus was taken and the body was sent for post-mortem examination. During postoperative period mother had high intermittent pyrexia which responded to chloromycetin. In view

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prolonged obstructed labour self retaining catheter was left in for 7 days.

Investigations: X-ray report of dead foetus showed excessive soft tissue density involving the thorax, the abdomen and the extremities. Multiple fractures of the bones were visulized. (Traumatic). (Fig. 1).

Post-Mortem Examination: The foetus was swollen. The swelling was more marked in the left groin and thigh where it was like a bag of fluid. On cutting open the body, the subcutaneous tissue showed honeycombed appearance made up of dilated channels (Fig. 2) which were filled with brown coloured fluid. Besides the swelling in the left groin region, similar swelling was noted on the right side occupying the whole of iliac fossa. The cut surface of these swellings showed cystic spaces of varying sizes filled with brownish fluid. The thoracic and abdominal cavities, each contained 100 ml. of brown coloured fluid. All the organs were situated in normal position, except the liver, which was pushed in the thoracic cavity as a result of dificient dome of right diaphragm The lungs were heavy with patchy consolidation. No congenital gross abnormality of any other viscera was noted.

Microscopic Examination from the swollen skin revealed dilated channels (lymphatics) deep to the subcutaneous fat. These were lined by single layer of endothelial cells (Fig. 3). Section from the swelling in the groin and iliac region show cavernous spaces separated by thin connective tissue septae containing collections of lymphocytes (Fig. 4). Both the lungs showed patchy pneumonia. Sections from the other organs revealed variable amounts of congestion The diagnosis given was diffuse lymphagiectasis with multiple lymphangioma.

Discussion

Allen (1934) studied 300 cases of lymphoedema and classified them as non-inflammatory and inflammatory. Non-inflammatory group includes congenital type and lymphoedema praecox. The congenital variety is probably due to abnormal development of lymphatic vessels causing defective lymph drainage. Goetsch (1938) concluded that cavernous type of lymphangiomas were due to sequestration of lymphatic tissue in early embryonic life. The congenital type may be simple, affecting only one member of the family or it may be hereditary type called "Milroy's disease." This was described in 1892, affecting number of blood relatives indicating that fault may be due to disturbance is genes. In simple congenital form swelling is visible at birth or may appear within 24 hours. It is not progressive. It is very rare to find diffuse cystic dilation of lymphatic channels involving widespread portions of the body. Potter (1962) observed one infant who appeared normal on external examination, except for mild generalized oedema. This was caused by diffuse dilatation of lymphatic channels in the subcutaneous tissue and between the muscle bundles. He also noted many aborted foeti with diffuse lymphagiectasis and bilateral cystic hygroma of neck. The case described herein presented with diffuse lymphangiectasis and multiple lymphangiomas. Lymphangiomas may produce localized or generalised swelling. When diffuse enlargement affect region like the tongue macroglossia, in lips macrochelia, and in neck cystic hygroma. The cut surface reveals spongy translucent tissue of considerable width lying between the skin and deep fascia. The foetus studied by us had a diffuse swelling with localized multiple lymphangiomas in the iliac and groin regions. As the trunk was very broad due to marked subcutaneous oedema, it resulted in obstructed labour. Usually the foetal with diffuse lymphangiectasis are aborted at an early stage and in them no problem of obstructed labour arises. If it goes upto term the big size of the foetus is likely to lead to obstructed labour, as in the present case.

Summary

A rare case of diffuse lymphangiectasis leading to obstructed labour which was dealt by caesarean section is presented 2. X-ray photograph and post-mortem findings are discussed.

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See Figs. on Art Paper XII