

LITHOPEDION

(Review with a Case Report)

by

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History

The term lithopedion is applied to a foetus which has been retained within the maternal abdomen, and the tissues of which have become more or less completely infiltrated with calcium salts.

According to Dorland and Hubeney (1926), lithopedion formation had been recorded as early as the 16th century by Veneteiss (1595), Albo-sius (1597) and Densingus (1661). In the early days the discovery within a woman's abdomen of such a hard stony mass having the form of a child excited wonder and speculation, and was even viewed at times with considerable awe. Bainbridge (1912) quoted an interesting account of the first case (from Gould and Pyle, 1897). He said "Israel Spach in an extensive gynaecological work, published in 1557, figures a lithopedion drawn *in situ* in the case of a woman with her belly laid open. He decided this to be a calcified foetus, which he regarded as a reversion, following the

curious epigram alluding to the classical myth that after the flood the world was repopulated by the two survivors, Deucalion and Pyrrha, who walked over the earth and cast stones behind them which on striking ground became people". The epigram read as follows: "Deucalion cast stones behind him and thus fashioned our tender race from hard marble. How comes it that now-a-days by a reversal of things, the tender body of a little babe has limbs nearer akin to stone"?

Classification

In 1881 Kuchenmeister collected 45 cases of various forms of foetal calcification recorded during the 200 year period, antedating 1880. He pointed out that calcification in the products of conception may not be entirely limited to the foetus, but may also involve the membranes and the placenta or may be entirely limited to the latter structures. On the basis of these variations he proposed their division into three groups.

1. *Lithokelyphos* (Stone-sheath or egg shell), in which the membranes alone are calcified and form a hard shell surrounding the foetus. The foetus may undergo only slight

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change or it may completely be skeletonized, but it is not involved in the process of calcification. Kuchenmeister believed this to be a most common type and the result of the membranes remaining intact around the foetus at the termination of pregnancy.

2. *Lithokelphopaedion* (Stone-sheath child), in which both the membranes and the foetus are calcified. The amniotic fluid has escaped or has been absorbed.

3. *True lithopedion* (Stone child), in which the foetus is infiltrated with calcium salts and in which the calcification of the foetal membranes is negligible. This type, he believed, resulted when the foetus escaped unattached into the abdominal cavity, the membranes being either left behind or closely wrapped about the foetus.

While this classification has no clinical value, it provides a convenient method of grouping the specimens. The confusion resulting from the double use of the word lithopedion in referring to the general group as well as to the third group could be avoided by substituting the word lithotecnon when referring to the third group, the true lithopedion of Kuchenmeister, in which the foetus alone is calcified.

D'Aunov and King (1922) proposed another classification of retained abdominal pregnancies into four possible categories: (1) skeletonization or retention of a collection of foetal bones with absorption of foetal soft parts; (2) adipocere, or replacement of the soft parts by soap and fats; (3) suppuration, or abscess formation due to infection with destruction of the foetal tissues; and (4) true

lithopedion, or sterile calcification of the foetus. The above classification is based on modern pathological concept and is not limited to the retained calcified foetus in the abdominal cavity but indicates the various changes which can take place in the foetus in an intra-abdominal pregnancy.

Etiology

The factors leading to the calcification of the foetus are vague. Leopold (1881) did some experimental work to show the fate of rabbit foetuses, removed from the maternal uterus and left in the abdomen. He placed, embryos 2.5, 5 and 8 cms. in length in the abdominal cavities of adult rabbits. In some instances he used the foetus alone and in others the entire embryo with its membranes intact. There was no reaction on the part of the host except in those cases in which peritonitis developed by the second day; the 2.5 cms. embryos had almost completely disappeared and by the 9th or 10th week the 8 cms. foetus had been reduced to few remnants of the paws, the integument, and the skeletal system. In the presence of infection the process of destruction and absorption progressed much more rapidly. If the umbilical cord was ligated, absorption was slower. The miniature foetus meets the same fate as the one extruded into the abdomen of a woman on the termination of extra-uterine pregnancy.

An analysis of the pregnancies responsible for the production of the foetus of lithopedion shows evidence that in 73 to 84% of cases there is clinical evidence of development to

or beyond the seventh month and in 60% to term. The average term of development was about 7 months, which shows that only the more mature fetuses undergo this transformation. It is impossible to postulate the types of extra-uterine pregnancy that are most likely to result in the production of lithopedion. In general it is the type in which the foetus is most likely to develop to near term, and while this may occur in practically every type, it is more common in some than in others.

The retention of the foetus in the abdomen, its development to the third month or more, are essential for the production of lithopedion. It is extremely doubtful if a foetus retained within a normal uterus ever undergoes this transformation because there the occurrence of infection leads rather to destruction of the tissues of the foetus and skeletonization. Oden and Lee (1940) listed the following conditions necessary for lithopedion formation:

1. There must be an extra-uterine pregnancy.
2. The foetus must survive in the abdomen over 3 months.
3. The pregnancy must escape medical notice.
4. The foetus must remain sterile.
5. Conditions necessary for the deposition of calcium must be present, namely sluggish circulation.

Mummification, adipocere and various types of fatty change have been described in these retained mature fetuses, but it is not known if these changes are entirely different processes or merely successive steps in the production of a lithopedion.

The exact chemical process by which calcium is deposited in the tissues or elsewhere in the body is not well understood.

Wells (1918) found that pieces of sterilized cartilage placed in the peritoneal cavity of a rabbit soon became calcified, having taken up lime from the fluid in which they bathed. He concluded that the process is a physical rather than a chemical one, the calcium and phosphorus being absorbed by the degenerated tissue. Mac Callum (1916) stated that there seems to be something peculiar about tissues, living or dead, which gives them power to catch up the calcium from the circulating fluid and hold it firmly in solid form. Later on, Mac Callum (1940) suggested that some local chemical process must be responsible for the precipitation of calcium in the dead materials exposed to the circulating fluid, as well as in bone. He stated that iron is practically always demonstrable in areas of calcification, but it is possible that it is merely absorbed by the calcium salt or precipitated by phosphoric acid liberated in that position. Nesbitt (1955) suggests that calcium is deposited in dying or dead tissue without reference to the blood calcium. Fatty degeneration occurs initially, then hydrolisation takes place and a fatty acid is liberated. The fatty acid is replaced by carbonic acid and phosphoric acid in the blood to form calcium carbonate and calcium phosphate. Schumann (1921) categorized the final results of ectopic gestation as follows:

Resorption, mole formation, haematocele, suppuration, skeleto-

nization, adipocere and lithopedion formation.

Owens (1962) stated that the foetal tissue undergoes aseptic autolysis and circulating calcium from maternal blood is precipitated locally in the acidic necrotic tissue, as a passive physico-chemical process, to produce a lithopedion.

Diagnosis

The diagnosis of lithopedion can be made from the history but its presence is frequently not recognised because the condition is not borne in mind and the various events in the history are not correlated until the diagnosis has been made at operation. The symptoms may be divided chronologically into four groups corresponding to the successive stages in the development (Masson and Simon, 1928).

1. The onset of pregnancy. The patient gives a history of a pregnancy which had been atypical in some respects. The symptoms may be those of ruptured tubal gestation which continues as a secondary abdominal pregnancy. In such cases the symptoms of rupture subside but the abdomen continues to enlarge and the other evidences of pregnancy persist. In other cases the symptoms may be mistaken for those of miscarriage but the inquiry discloses the fact that a foetus was not passed and that instead of usual sequence of events following a miscarriage, the abdomen continued to enlarge and the other evidences of pregnancy persisted.

In the event of primary abdominal or ovarian gestation, the patient considers herself pregnant and may notice nothing abnormal during the

early months except possibly pain or unusually active foetal movements.

2. The termination of pregnancy. The later months of the pregnancy are usually uneventful and at or near term, labour is initiated in an apparently normal manner. The pains continue to be mild; however, after 24 to 48 hours, they cease entirely without the delivery of the child. This is commonly referred to as "false" or "missed" labour. The patient is no longer aware of foetal movements. She gradually recovers, menstruation returns and the abdomen decreases in size.

3. Latent period. The foetus now ceases to exist as such. It becomes a parasite in the maternal abdomen and while it may continue to derive some blood supply from the mother, it leads only a passive existence. Its bulk decreases with the absorption of the amniotic fluid. Dehydration occurs and tissues begin to be infiltrated by calcium salts. During this period the patient is free from symptoms except that she may still be aware of the presence of a mass in the lower abdomen. From two to many more years may lapse in this manner while the foetus is being gradually transformed into a lithopedion.

4. Late symptoms. In about 65% of cases, if the patient lives, late symptoms eventually develop. They consist of mild abdominal pains or a consciousness of fullness in the lower abdomen frequently associated with a persistent foul vaginal discharge. Symptoms of bladder irritation may result from pressure of the mass on the bladder or from secondary cystitis. If the patient is untreated the lithopedion may rupture into the

adjacent viscera or through the abdominal wall.

The general health of the patient may be unimpaired or there may be loss of weight. A hard mass usually associated with slight tenderness is present in the lower part of the abdomen. On vaginal examination this mass may be palpated on one or the other side of the pelvis and is frequently attached to the uterus.

X-ray evidence is characteristic. Kirklin and Simon (1928) have pointed out that the foetus appears much more distinctly than in a normal pregnancy because of its increased density. The foetus may also occupy an abnormal position. Other tissues besides the skeleton, which have become infiltrated with calcium salts, will produce an extra-skeletal shadow of varying density and outline, depending on the extent of calcification of the products of conception.

The conditions with which lithopedion are most frequently confused are calcified fibromyomata and ovarian dermoids. The fibromyomata, when sufficiently calcified, rarely produce symptoms. Ovarian dermoids may be a source of confusion except when they are bilateral. Previous miscarriage or atypical pregnancy is not an essential part of the history of ovarian dermoid.

Incidence

The true incidence of abdominal pregnancy is difficult and varies widely in different series. The occurrence of lithopedion formation has been reported as from one to two per cent of all extra-uterine pregnancies. Masson and Simon (1928) reported an in-

cidence of 2.0 per cent, Schumann (1921) 1.5 to 1.8 per cent and Anderson *et al.*, (1951) 0.81 per cent in 370 cases of extra-uterine pregnancy. Clark and Ellison (1959) reported an incidence of 0.29 per cent based on 351 ectopic pregnancies operated during the period 1947-1957. Bland *et al.*, (1933) brought the total aggregate of reported cases to 197. Later Anderson *et al.*, (1951) reported a total of 252 cases of authentic lithopedion formation in the world's literature. Subsequently, various authors have reported cases: McClure and Epperson (1952); Schwarz (1952); Roberts (1952); Sordo Noreiga (1952); Herrera and Casimiro (1954); Menon (1954); Nesbitt (1955); Gupta (1955); Steinberger and Pogue (1956); Parikh (1956); Clark and Ellison (1959); Temple and Hester (1959); Woodbury and Jarrett (1960); Benerjee (1961); Owens (1962) and O'Leary and Bepko (1963): These cases with the one herein described make an aggregate of 269.

There are innumerable instances where women carrying lithopedion or retained foetal parts have had concomitant normal pregnancies and deliveries. Williams (1946) reported a case of an intra-uterine lithopedion being delivered wrapped around the neck of a living twin.

The discovery of a lithopedion in the second half of the twentieth century implies that either the patient involved has had no medical attention whatsoever, or rather some serious mistakes in medical judgement have been made. Obviously, a retained abdominal pregnancy should be re-

cognised eventually in any modern obstetrical practice. However, with close medical supervision and improved diagnostic methods ectopic gestation escapes surgical treatment less frequently, so that lithopedion development is now a truly rare occurrence. This case is therefore reported because of the rarity of its type.

Case Report

Mrs. L., aged 30 years, married for 12 years, came to the hospital on 10th April, 1963, for sterility, scanty menstruation and a lump in the abdomen of 5 years' duration.

Previous medical and surgical histories were not significant. History of present illness — 5 years ago she had a period of amenorrhoea for 7 months and she also felt foetal movements followed by a bout of vaginal bleeding along with acute abdominal pain, after which the foetal movements stopped. She had no complaint and the size of abdomen also reduced, her periods had been regular but scanty. She had her last period 15 days ago. One F.T.N.D. — 7 years ago but child died at the age of 5 months. General examination revealed nothing abnormal in her heart and lungs. Her blood pressure was 110/70 mm. Hg., her haemoglobin 13 gm. and urine normal.

Clinical Examination

Abdominal examination revealed a hard lump arising from the pelvis and filling up the lower abdomen almost up to the umbilicus. The margins were well defined all round except where the mass dipped deep into pelvis; the surface was smooth. There was no tenderness. Liver and spleen were not palpable.

Per vaginam, body of uterus was anteverted and anteflexed, and was of normal size, but pushed to the left and upwards by an irregular and hard mass filling the pouch of Douglas and extending up into the lower abdomen to about the level of the umbilicus, the mass was not tender. A provisional diagnosis of secondary abdominal preg-

nancy or calcified fibroid in the broad ligament was made.

Skiagram showed a small foetus with over-riding of bones of skull. Spalding's sign, was present. The spine and extremities were crowded in a small mass—dead foetus (Fig. 1). The diagnosis of a lithopedion

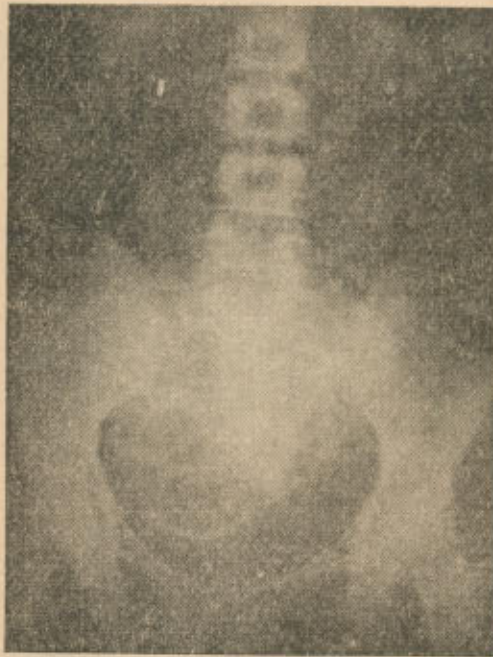


Fig. 1

Skiagram showing the shadow of lithopedion.

was made before the operation as skiagram was absolutely conclusive.

Operation: Under spinal anaesthesia the abdomen was opened by a median subumbilical incision. A hard irregular encapsulated mass was seen. It was adherent anteriorly to the omentum, to the parietal peritoneum on the right side, and posteriorly to the mesentery and small bowel. All adhesions were separated by blunt and sharp dissection, the upper pole of the sac got opened up and foetal limbs could be visualised. There was no liquor amnii in the sac. The lower pole of the sac was deep between the layers of the broad ligament on the right side and low down in the pouch of Douglas. The sac along with the

foetus was excised after applying a clamp at the uterine end. Raw area could not be peritonised as healthy peritoneum was wide apart. Placenta could not be visualised separately. The abdomen was closed in layers. Post-operative period was uneventful.

Specimen: Showed a foetus about the size of 7 months. Evidences of skeletal portions of the foetus were found. Details of lithopedion are shown in Fig 2. Sac was



Fig. 2
Showing details of lithopedion.

adherent to the body of the foetus; features of foetus were well marked and defined. All body parts were well developed.

Summary

1. The present case of lithopedion formation brings the total of reported cases in the world's literature to 269, including the cases reported in this country.

2. The lithopedion developed over a period of 5 years. The initial attack of acute abdominal pain was undoubtedly due to rupture of an extra-uterine pregnancy with extrusion of the unattached foetus into the broad ligament where it subsequently became calcified. The patient came to the hospital for treatment of sterility.

3. A brief review of the literature of lithopedion is given.

4. The condition has become rare over the decade due to close medical supervision and improved diagnostic methods in the very early stages of the condition.

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