

Twin lithopaedions: a rare entity

Mishra J M, Behera T K, Panda B K, Sarangi K

ABSTRACT

Lithopaedion (stone baby) is the name given to an extrauterine pregnancy that evolves to foetal death and calcification. There are around 300 cases reported in the world medical literature to date. We report the case of a 40-year-old woman who presented with features of acute intestinal obstruction (abdominal distention, vomiting and absolute constipation) for a week. She had a past history of a missed abortion in the fifth month of gestation, eight years prior to this presentation, one which we thought to be irrelevant to the present illness. However, complementary investigations, including scout abdominal radiographs and ultrasonography of the abdomen and pelvis, were done before the operation. The abdominal radiograph showed two opaque globular masses on either side of the lower abdomen with distended small intestinal loops. Exploratory laparotomy was performed and a portion of strangulated small bowel attached to a solid globular mass behind the left ovary was removed, with a subsequent resection of the gut and end-to-end anastomosis. Upon removal of a thick membrane from the globular mass, a dead five-month-old calcified foetus was recovered. In the right iliac fossa, a similar mass was removed and another dead calcified foetus of similar age was recovered.

Keywords: abdominal pregnancy, foetal death, intestinal obstruction, lithopaedion

Singapore Med J 2007; 48(9):866–868

INTRODUCTION

Lithopaedion (litho: stone, paedion: child) is the name given to an extrauterine pregnancy that evolves to foetal death and calcification. It is a rarely encountered consequence of an undiagnosed and untreated advanced abdominal pregnancy, where the dead foetus is retained in the maternal abdominal cavity and calcification ensues. The incidence of abdominal pregnancy is around one in 11,000 pregnancies and lithopaedions occur in 1.5%–1.8% of these cases.⁽¹⁻³⁾ So far, approximately 300 cases of lithopaedions have been



Fig. 1 Operative photograph shows adherence of the greater omentum to the right globular mass.

reported in 400 years of world medical literature.^(2,4,5) Due to an increase in the incidence of pelvic inflammatory diseases and uterine tubal surgeries, there has been a spurt in the incidence of ectopic pregnancies. Incidence of intestinal obstruction due to adherence of gut to the inflamed abdominal pregnancy is a rather rare finding. Further, twin lithopaedions, a result of the evolution of a twin abdominal pregnancy, has yet to be reported.

CASE REPORT

A 40-year-old woman was admitted to our hospital with features of intestinal obstruction. She had abdominal distention, vomiting and absolute constipation. Her past history revealed mild to moderate pain in the lower abdomen on both sides for the last eight years. This pain subsided after taking analgesics from a local doctor. A gynaecological examination did not reveal anything except a history of missed abortion in the fifth month of gestation, eight years prior to this presentation. Investigations included blood parameters, radiographs of the abdomen and pelvis in erect posture, and ultrasonography (US) of abdomen and pelvis. Two radiopaque, calcified, globular shadows were detected on both sides of the lower abdomen, with features of small bowel obstruction. The US showed two oval calcified areas on both sides of the lower abdomen.

Keeping these unusual radiographical and US findings in mind, an emergency laparotomy was conducted under general anaesthesia. A devitalised portion of ileum was found to be morbidly adhered

Post Graduate
Department of
General Surgery,
VSS Medical College
and Hospital,
Burla,
Orissa 768017,
India

Mishra JM, MBBS,
MS
Professor

Behera TK, MBBS,
MS
Lecturer

Panda BK, MBBS,
MD
Lecturer in
Anaesthesiology

Sarangi K, MBBS
Postgraduate

Correspondence to:
Prof JM Mishra
QR / No- 3R / 20,
Doctors' Colony,
Burla,
Orissa 768017,
India
Tel: (91) 943 733 9326
Fax: (91) 663 243 0151
Email: kiranmoy123
@yahoo.co.in;
jmmishra123@
yahoo.com

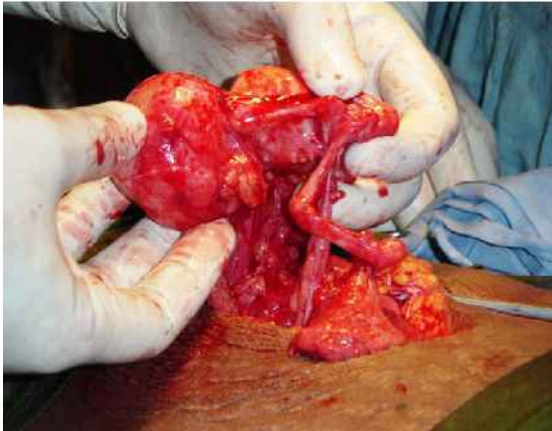


Fig. 2 Operative photograph shows a lithopaedion that was evident after dissection of its overlying membrane.



Fig. 3 Postoperative photograph of the twin lithopaedions.

to a globular mass in the left flank, while the greater omentum was attached to another globular mass on the right side (Fig. 1). Subsequently, a small gut resection and anastomosis were performed, and both the globular masses were carefully removed. Both the ovaries and the uterus were found to be normal and healthy. On dissecting the thin but tough tissues covering these globular masses, two mummified and calcified foetal skeletons were recovered (Fig. 2). Both skeletonised foetuses were of the same age (around five months old) (Fig. 3). After the surgery, a detailed gynaecological history revealed a missed abortion at the fifth month of gestation in 1997, as diagnosed by a local doctor, but without any resultant expulsion of the products of conception. Thereafter, she had intermittent pain in both sides of the lower abdomen, which subsided after taking analgesics. Due to the scarcity of radiographical and US facilities in the local area, the cause of her abdominal ailment could not be ascertained and the abnormalities remained undetected till she developed an intestinal obstruction and presented to us.

DISCUSSION

In this case, a calcified twin abdominal pregnancy was detected after eight years of evolution because it was causing acute intestinal obstruction. According to a literature search in this field, the age of patients on the date of diagnosis varied from 23 to 100 years,⁽⁶⁾ two-thirds of them being older than 40 years of age. The foetal retention period varied from four to 60 years; foetal death occurred between the third and sixth month of pregnancy in 20% of cases, between the seventh and eighth month in 27%, and at full term in 43% of cases.^(4,5) Abdominal pregnancy results from the rupture of either ovarian or tubal pregnancy, with

subsequent implantation in the abdominal cavity.^(1,4) Certain conditions prevail for the development of a lithopaedion, such as: (1) extrauterine pregnancy; (2) foetal death after three months of pregnancy; (3) the egg must be sterile; (4) there cannot be any early diagnosis; (5) local conditions must exist for calcium precipitation and deposit.^(1,4,5) The development of this pregnancy is the same as for an intrauterine pregnancy until foetal death, after which dehydration of tissues and calcium infiltration occurs.^(1-3,5)

An abdominal pregnancy that calcifies can be classified into three subtypes according to the involvement of calcification to the membranes and the foetus: (1) Lithokelyphos (litho: stone, kelyphos: shell) – in which membranes alone are calcified; (2) Lithokelyphopaedion – in which both membranes and the foetus are calcified; (3) True Lithopaedion or Lithotecnon – in which the foetus is calcified and the membranes are negligible.^(5,7) Most cases of lithopaedions are detected incidentally while taking abdominal radiographs for various reasons, or when a palpable abdominal mass is felt during pelvic examination. Those patients with asymptomatic cases which remain undetected for years may have weight sensation in the abdomen, pelvic pain, compressive urinary bladder and rectal symptoms.^(2,4) Even dreaded complications, such as urinary bladder and rectal perforations;⁽⁸⁾ extrusion of foetal parts through abdominal wall, rectum or vagina; intestinal obstruction (due to collision of foetal parts with the intestine or adherence); volvulus; fistula formation; pelvic abscess⁽⁹⁾ and cephalopelvic disproportion of a concomitant pregnancy, have been reported from different parts of the world.^(2,3,5,7)

The diagnosis is revealed by a suggestive clinical history, a solid pelvic mass on local examination and

often, radiographs of the abdomen is adequate to confirm it.^(2,3,5,7) However, the US shows an empty uterine cavity with a non-specific appearance of the abdominal mass, confusing the diagnosis. Computed tomography and magnetic resonance imaging clearly define the pathology and help to diagnose adherence and other organs affected, although this is not a constant finding.⁽⁷⁾ In the world medical literature, most of the reported cases of lithopaedions or their variants were diagnosed only intraoperatively or postoperatively. The differential diagnosis may include calcified masses such as ovarian tumours,⁽¹⁰⁾ myomas, inflammatory masses, epiploon calcifications, and urinary tract or bladder tumours.⁽⁵⁾ Due to the possibility of future complications, even after years of evolution, the proper procedure is surgical removal.⁽³⁾

The surgery for this condition is usually simple with mild bleeding. No intraoperative deaths have been reported, even in elderly patients.^(2,5) As most cases are diagnosed perioperatively, the clinician must be aware of this rare condition. From this case, we learn that it is wrong to assume that a pregnancy has ended in spontaneous miscarriage unless definite products of conception can be seen. The lithopaedion could also have been avoided if there was access to US during this patient's pregnancy, as it would have revealed

an abdominal pregnancy. On the other hand, the occurrence of abdominal pregnancy and lithopaedion formation have become even rarer due to medical and prenatal care becoming more accessible to the population, providing the possibility of early diagnosis and treatment of the entity.^(1,5)

REFERENCES

1. Costa SD, Presley J, Bastert G. Advanced abdominal pregnancy. *Obstet Gynecol Surv* 1991; 46:515-25.
2. Irick MB, Kitsos CN, O'Leary JA. Therapeutic aspects in the management of a lithopedion. *Am Surg* 1970; 36:232-4.
3. Passini R, Knobel R, Parpinelli MA, et al. Calcified abdominal pregnancy with eighteen years of evolution: case report. *Sao Paulo Med J* 2000; 118:192-4.
4. Frayer CA, Hibbert ML. Abdominal pregnancy in a 67-year-old woman undetected for 37 years. A case report. *J Reprod Med* 1999; 44:633-5.
5. Spirtos NM, Eisenkop SM, Mishell DR. Lithokelyphos: A case report and literature review. *J Reprod Med* 1987; 32:43-6.
6. Lachman N, Satyapal KS, Kalideen JM, Moodley TR. Lithopedion: A case report. *Clin Anat* 2001; 14:52-4.
7. Alymlahi EK, Chat L, Chellaoui M, Dafiri R. Lithopaedion: stone baby. {online} EURORAD (Case 2439) 2003. Available at: www.eurorad.org/case.php?id=2439. DOI: 10.1594 / EURORAD / CASE 2439. Accessed July 2006.
8. El Hag IA, Ramesh K, Kollur SM, Salem M. Extrauterine placental site trophoblastic tumour in association with a lithopedion. *Histopathology* 2002; 41:446-9.
9. Jain T, Eckert LO. Abdominal pregnancy with lithopedion formation presenting as a pelvic abscess. *Obstet Gynecol* 2000; 96:808-10.
10. Kim MS, Park S, Lee TS. Old abdominal pregnancy presenting as an ovarian neoplasm. *J Korean Med Sci* 2002; 17:274-5.