

CASE REPORTS

Lithopedion

LILLIAN A. CHASE, B.A., M.B., C.R.C.P.[C], *Toronto*

THE discovery of a lithopedion is as dramatic as it is rare. Although the condition was known and recognized in ancient times, only about 270 cases have been recorded in the literature. Most of these have been in this century; up until 1900 there had been only 38 recorded cases, and in the next 12 years only 12 more cases were added; the remainder have been reported since then.

Today the occurrence of a lithopedion is rarer than ever; it is unknown among women who receive adequate prenatal care or regular medical examinations, so it is usually found only where modern health services are unavailable or when a gross error has been made.

A 54-year-old Jamaican woman applying for a post on the domestic staff of Sunnybrook Hospital, Toronto, was found to have an abdominal mass during a pre-employment examination in the outpatient department on February 9, 1966. Although aware of the mass, she had no complaints. Her history included the following particulars: Her menarche occurred at the age of 15; since then her menses had been regular, occurring every 28 days and lasting for 5 days. The flow was scanty, but was never associated with pain. In 1933, at the age of 21, she had her first pregnancy and was delivered of a male child after a 14-hour labour. The second pregnancy occurred a year later and this delivery apparently was painless. During neither of these pregnancies nor at delivery was she attended by a physician. Her periods resumed their normal pattern after the second pregnancy. Although she never used contraceptives, she did not become pregnant again; she had not had any miscarriages.

In 1957, at the age of 45, she noticed that her abdomen was increasing in size and she was able to feel fetal movements. However, she did not think she was pregnant at this time, because she had had vaginal bleeding, which was occasionally regular but more often at irregular intervals. At times the bleeding lasted for as long as 15 to 21 days. It was accompanied by considerable lower abdominal pain and was frequently quite profuse, enough to soak the newspapers with which she attempted to control it. She said that she had not at any time passed

anything recognizable as tissue. She developed considerable dysuria at this time, and on two occasions, because of acute urinary retention, had to be catheterized at a hospital in Kingston, Jamaica. After some months she was no longer able to feel movements, but the bleeding and the pain continued. At no time was she told that she was or had been pregnant. After being on a waiting list for eight months she was finally admitted to hospital, where she underwent an abdominal operation. She did not know what type of procedure had been carried out and could only say that "a string was giving me trouble and they loosened it". Postoperatively she passed some clots, but the bleeding finally stopped and she no longer had any pain. She felt stronger than before and soon returned to work. Although her lower abdomen had decreased in size, she was still able to feel a mass there. She stated that her physician at that time had assured her that this was of no significance. Postoperatively she never again had a menstrual period, and although the mass appeared to get somewhat smaller, it was still present when she was seen at Sunnybrook Hospital.

In 1961 she emigrated to England from Jamaica and in 1965 she came to Canada; thus on at least two occasions she had been examined by immigration medical officers. Although these examinations included chest radiographs, at no time was she informed that anything was wrong.

She had no history of tuberculosis, pelvic inflammatory disease, venereal disease or urethral discharge, and no dysuria, nocturia or stress incontinence. She had had no recent vaginal bleeding and only a normal amount and type of vaginal discharge.

On physical examination this 135-pound woman, 5'5" in height, was found to be edentulous; her blood pressure was 140/80; her pulse was 65 and regular. A hard, stone-like, irregular mass was palpable, lying across the lower abdomen. A lower abdominal midline scar was present. The plain film of the abdomen showed a lithopedion, and it was therefore concluded that it was the head that was palpable in the left lower quadrant and the buttocks on the right side. The mass appeared to be quite superficial and was mobile; it measured 27 cm. in length and 10 cm. in width.

The pelvic examination confirmed that the present mass appeared to be of uterine origin, filling the pelvis and extending into the abdomen. The cervix was healthy. Laboratory investigation revealed a hemoglobin of 12.8 g., a sedimentation rate of 34 mm. in one hour (Westergren), a white cell count of 6200 and a blood urea nitrogen of 12 mg. per

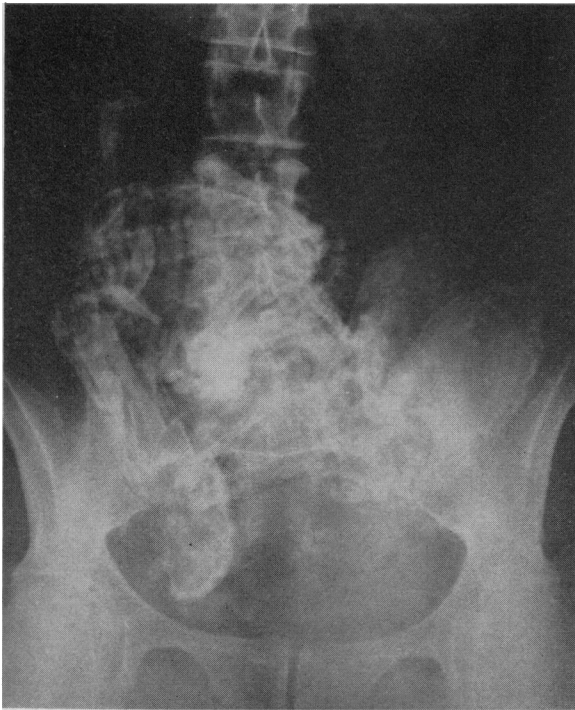


Fig. 1.—Anteroposterior radiograph of the abdomen.

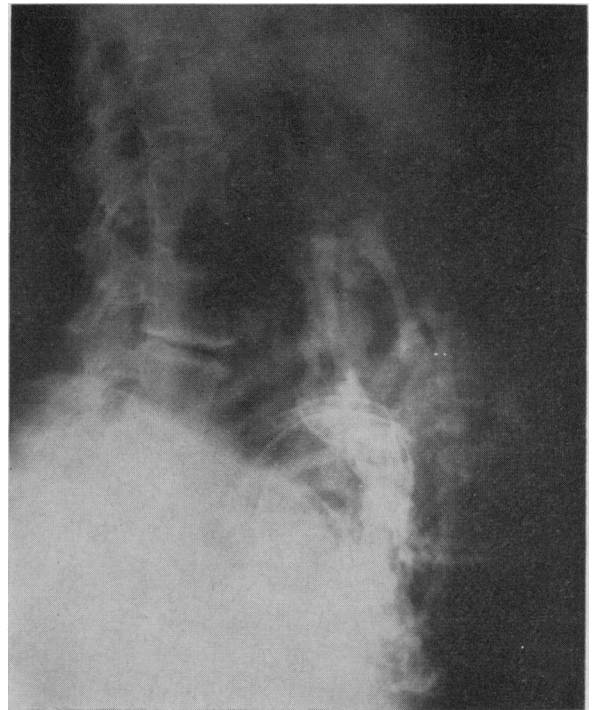


Fig. 2.—Lateral view of the abdomen seven days later.

100 ml. The prothrombin time and the fibrinogen level were both within normal limits. Urinalysis showed a specific gravity of 1.016, a pH of 5.5, a trace of albumen and one or two red blood cells per high-power field. The VDRL was negative. A smear and culture examination of her sputum revealed no abnormalities.

The plain x-ray film of the abdomen was reported as follows:

"A single A.P. view of the abdomen shows the presence of a fetus, the size of which would suggest that it has developed fairly close to term. There are many signs which indicate that the fetus is dead and that it has been dead for a considerable period of time. In addition there is evidence of calcification of the soft tissues adjacent to the fetus and probably of the fetus itself. The fetus occupies a comparatively high position in the pelvis, and this would be in keeping with an intra-abdominal pregnancy. The above findings are in agreement with the radiological diagnosis of a lithopedion" (Figs. 1 and 2).

The patient was admitted to Sunnybrook Hospital, and on February 15, 1966, under general anesthesia, a laparotomy was performed using a sub-umbilical right paramedian incision. A partially calcified fetus was found lying obliquely in the abdomen with the head in the left lower quadrant and the buttocks on the right at a slightly higher level (Fig. 3). The omentum surrounded and was adherent to the superior and anterior aspects of the mass. After division and separation of the adhesions the fetal mass was removed. The uterus, enlarged to about the size of a 12-week pregnancy, contained multiple fibroids, some of which were

calcified. Attached to the right cornu in front of the right fallopian tube was a smaller calcified mass, firmly adherent to the adjacent structures. This mass was also removed after separation and division of the many adhesions. Except for a dilated esophageal hiatus, no other abnormalities were found in the abdomen.

Regrettably, no pathological report regarding the fetus or its membranes was available, and the weight was not recorded. However, Figs. 4 and 5 display the lithopedion immediately after it was removed from the abdomen. The pathological report of the smaller mass was recorded as follows:

"A calcified mass measuring 8 x 4 x 3 cm. with attached fat measuring 9 x 5 x 1 cm. was received. The calcified mass is oval in shape. One surface is hard and contains calcific material; the other surface is much softer and is composed of a capsule which is yellowish in colour and is nodular. It contains yellow debris which is quite soft. Microscopic examination shows stretches of dense cellular collagen in which there are irregular zones of granular and partially calcified debris. At the margins there appears to be a capsule containing some lymphocytes and near the margins in a loose stroma there are some collections of vascular channels. There is no evidence of malignancy. The pathological diagnosis is calcified omentum and degenerate placenta."

DISCUSSION

A lithopedion usually results from an abdominal pregnancy, either primary or, more usually, secondary, i.e., where the gestation

Fig. 3

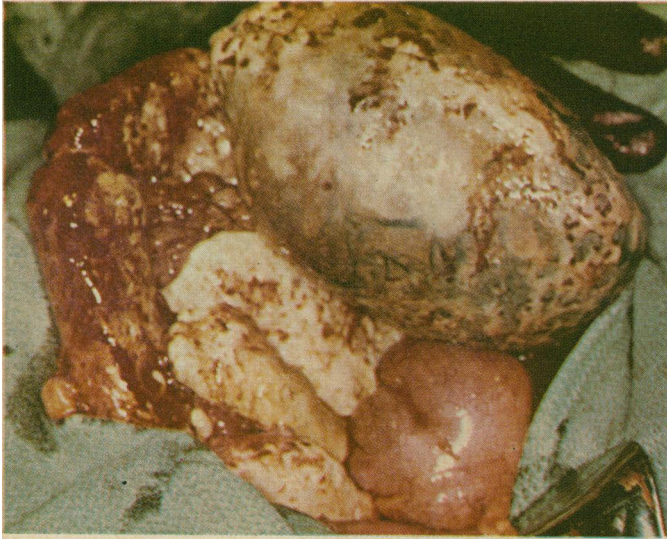


Fig. 5



Fig. 4

Fig. 3.—Lithopedion as it lies in the wound. The surgeon's gloved hand appears on the patient's left side.

Fig. 4.—Lithopedion lying in the surgeon's hand. The head of the calcified fetus can be seen on the right side.

Fig. 5.—Lithopedion after operative removal.

actually occurs free in the peritoneal cavity, even though it may have originated in a fallopian tube or even in the uterus.¹⁶ The circumstances under which lithopedions are usually found are listed by Cave¹¹: the pregnancy is extrauterine; the fetus has developed for more than three months; the conditions for the deposition of calcium must be present, i.e., the local circulation must be sluggish and the fetus must remain sterile. Only in a few patients can a placenta be identified.

Küchenmeister,¹ to whom we owe so much for his investigation of ancient records of this condition, classified three types: *lithokelyphos*, in which only the fetal membranes are calcified and the fetus degenerates within them; *lithokelyphopedion*, when both the fetus and the surrounding membranes show calcium deposits, and *true lithopedion*, where only the fetus is

calcified (without calcification of the membranes).²

D'Aunoy and King³ have listed four changes which an abdominal fetus may undergo if it is not removed: (a) *skeletonization*, where only the bones of the fetus remain following the disintegration and absorption of the soft parts; (b) *adipocere*, where the soft parts are replaced by fatty acids, soaps and salts of palmitic and stearic acids; (c) *suppuration*, where the fetus is destroyed after an abscess has formed, usually due to *E. coli* infection. Under these circumstances the abscess may rupture into the vagina, the rectum or other parts of the bowel, and fetal bones may be discharged through any of the body's orifices, including the mouth. (d) *True lithopedion formation* occurs if the fetus remains sterile and to varying degrees becomes infiltrated with calcium salts.

The case reported here has the classical symptoms and signs of an abdominal pregnancy—suspension of regular menses, development of irregular uterine bleeding, lower abdominal pain, dysuria and the appearance of the abdominal mass; even fetal movements were noticed. Occasionally the condition can cause remarkably few symptoms and these are due mainly to pressure, such as dyspareunia and bladder symptoms. Once the lithopedion forms the only evidence may be an abdominal mass, as in this case. In this era the diagnosis is usually made by a radiologist, but if the clinician fails to order an x-ray examination of the abdomen, the diagnosis will then be made only at laparotomy. It seems likely that this patient had her abdominal pregnancy in 1957, but it is difficult to explain why the laparotomy at that time failed to reveal it.

Cases have been recorded where the normal development of an intrauterine pregnancy has occurred simultaneously with the formation of a lithopedion from a concomitant extrauterine pregnancy.² Normal deliveries have been observed in women who at the same time have carried a lithopedion.

Although at one time the maternal mortality from abdominal pregnancy was recorded as 35% (1919), it is now so rare that it is not recorded separately.

One of the earliest recorded cases of lithopedion occurred 1000 years ago. It was reported by Albucasis, the greatest surgeon of the Arabic era, who flourished in Cordova, Spain (A.D. 936-1013).¹ This article, one of the earliest medical writings, was first published in Venice shortly after the advent of printing in 1497; it was republished later in Oxford in 1779. The record says that the mother passed fetal bones through the abdominal wall but recovered.

In 1557 Israel Spach, quoted by Bainbridge⁴ and others, showed the figure of a lithopedion drawn *in situ* with the mother's belly laid open. Spach related this to the myth that after the flood the world was repopulated by the two survivors, Deucalion and Pyrrha, who walked over the earth, casting stones behind them which, on striking the ground, became people. The epigram reads: "Deucalion cast stones behind him and thus fashioned our tender race from hard marble. How comes it, that nowadays, by a reversal of things the tender body of a little babe has limbs nearer akin to stone?"

Other early cases are those reported by Venetiss in 1595 and by Denesingus in 1661. A famous lithopedion occurring in the city of Sens was reported in Gynarcorium by Cordaeus in the early part of the sixteenth century; when

the patient died the fetus was found. In 1540, Christopher Bain, the "travelling surgeon", operated on an Italian woman and extracted the skeleton of a male child; the patient survived to have four more children. In 1721 Anna Mullern, 46 years before her death, declared herself pregnant; the swelling remained until she died at the age of 94. She desired that her body be opened after death; the surgeon broke open the mass with a hatchet—a lithopedion.

The first operation in North America for removal of a fetus from an abdominal pregnancy was done by John Baird of New York in 1759. Details of this pregnancy were recorded by Fothergill.⁵

"Memoir on Extra-uterine Gestation", published in Edinburgh in 1840,⁶ is a mine of information. Up to that time the condition was regarded more as a medical curiosity than as "one of the most dreadful calamities to which a woman can be subjected".⁵

In 1960, Woodbury and Jarrett² reported a patient in whom an intra-abdominal lithopedion was retained for 13 years. They also refer to a report of an intrauterine lithopedion delivered while "wrapped around the neck of a living twin".⁵

Masson and Simon⁷ in a comprehensive article reported 9 lithopedions in 44 cases of extrauterine pregnancy at the Mayo Clinic from 1903 to 1926, and added 9 more cases that were seen there.

King,⁸ in a very interesting, well-illustrated article, wrote of his experiences with 12 cases of advanced extrauterine pregnancy during his 25 years in China. One patient passed what she thought was a chicken bone by rectum; it was a fetal femur. His illustration shows the fetal bones neatly arranged, apparently all of them having been passed by rectum.

Auvray⁹ has reported the only case of a lithopedion in which a carcinoma developed.

This condition has also been a theme for a novel. In 1944 Samuel Hopkins Adams¹² published "Canal Town", which tells of a young doctor who diagnosed pregnancy in the unmarried daughter of the town's leading citizen; no baby was born, but after her death from cholera her body was exhumed, and the doctor, previously castigated by the father, showed the lithopedion to him. This story is based on a specimen which is now in the museum of the Albany Medical School of New York. The factual account of this case, based on the report of Dr. Davis Little (1768-1852) to the Albany County Medical Society, appeared in *The New Yorker* of December 10, 1938.

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Licorice and Hypertension

ROBERT E. LEFEBVRE, M.D.* and JULIEN MARC-AURELE, M.D.,†
Montreal

THIS case report is intended to illustrate that a complete history is still invaluable in providing clues as to the nature of disease and remains the corner stone for subsequent logical and warranted investigation.

Mrs. P.F., a 34-year-old gravida 3, para 2, aborta 1, white woman, presented with a history of known hypertension of about two years' duration.

The patient's symptoms first began about December 1965, when she complained of intermittent headaches, mostly frontal in location, of variable duration and described as a sensation of heaviness. They occurred most often in the morning on rising and were not relieved by acetylsalicylic acid. No medical attention was sought at this time.

In April 1966, the patient was hospitalized for a dilatation and curettage following a spontaneous abortion. On admission, her blood pressure was noted to be 170/100. Because the blood pressure remained elevated, a full renal and hypertensive investigation was undertaken. All investigated parameters were found to be normal. Approximately one week after her admission the blood pressure spontaneously reverted to normal. She was asymptomatic and was discharged with a diagnosis of essential hypertension of neurogenic origin.

Approximately two weeks after discharge, the headaches recurred and were accompanied by pal-

pitations, anxiety and episodes of dyspnea. After consultation with her doctor, who noted her blood pressure to be 180/120, she was treated with phenobarbital.

Despite this therapy her symptoms did not subside and in June 1966 she underwent a complete thyroid investigation the results of which were within normal limits. The patient was subsequently given other types of sedative without relief of her complaints.

Convinced that her symptoms and hypertension were emotional in origin, the patient decided to return to her occupation of nursing. While working in a postoperative recovery room, she would have her blood pressure taken regularly and noted it to be consistently elevated between 200/100 and 240/140. Disturbed by this persistent high blood pressure, she consulted us.

The family history disclosed that the father had died at 54 years of age from a cerebrovascular accident, and that the mother had been slightly hypertensive since her menopause.

The patient's own history was non-contributory; there was only one hospitalization at 22 years of age for investigation of fever of unknown origin.

During questioning, the patient volunteered the information that she had developed a peculiar habit—that it was not unusual for her to eat at least two pounds of fresh licorice a week.

Physical examination at this time revealed only an elevated blood pressure of 220/115, and there were no signs of neuromuscular disturbance. Preliminary investigation, including a rogitine test, was negative.

In the ensuing days, the patient on her own accord decided to stop eating licorice. She continued to have her blood pressure taken regularly at work,

From the Department of Medicine, Section of Nephrology, Hôtel-Dieu de Montréal, Montreal 18, Quebec.

*Assistant Resident II, Department of Medicine, Hôtel-Dieu de Montréal.

†Assistant Professor of Medicine, Department of Medicine, University of Montreal, and Nephrologist, Hôtel-Dieu de Montréal.

Reprint requests to: Dr. J. Marc-Aurèle, Department of Medicine, Hôtel-Dieu de Montréal, 3840 St-Urbain Street, Montreal 18, Quebec.