

Review

An unusual cause of intra-abdominal calcification: A lithopedion

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Received 17 September 2014; accepted 30 September 2014

Available online 16 October 2014

Abstract

We report a case of a 77-year-old female who was admitted to the emergency department complaining of diffuse abdominal pain for five days, associated with nausea, vomiting and constipation.

Physical examination disclosed a large incarcerated umbilical hernia, which was readily apparent on supine abdominal plain films. These also showed a calcified heterogeneous mass in the mid-abdominal region, which was further characterized by CT as a lithopedion (calcified ectopic pregnancy). This is one of the few cases studied on a MDCT equipment, and it clearly enhances the post-processing abilities of this imaging method which allows diagnostic high-quality MIP images.

Lithopedion is a rare entity, with less than 300 cases previously described in the medical literature. However, many reported cases corresponded to cases of skeletonization or collections of fetal bone fragments discovered encysted in the pelvic region at surgery or autopsy. It is thus estimated that true lithopedion is a much rarer entity.

The diagnosis may be reached by a suggestive clinical history and a palpable mass on physical examination, while the value of modern cross-sectional techniques is still virtually unknown. Ultrasonography may depict an empty uterine cavity and a calcified abdominal mass of non-specific characteristics, and computed tomography or magnetic resonance imaging are able to reach a conclusive diagnosis and may additionally define the involvement of adjacent structures.

The differential diagnosis includes other calcified pathologic situations, including ovarian tumors, uterine fibroids, urinary tract neoplasms, inflammatory masses or epiploic calcifications.

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Keywords: Lithopedion; Abdominal plain film; Computed tomography; Calcification; Abdomen

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1. Introduction

Lithopedion is a term designating an ectopic pregnancy that evolves to fetal death and calcification [1]. It is a rare occurrence, its incidence being reported as 1.5–2.0% of all ectopic pregnancies, and the incidence of ectopic pregnancy is 0.3–1.0% of the totality of gestations. As a consequence, less than 300 cases

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have been described in the medical literature [2–5]. However, many reported cases of lithopedion corresponded to cases of skeletonization or collections of fetal bone fragments discovered encysted in the pelvic region at surgery or autopsy [6]. It is thus estimated that true lithopedion is a much rarer entity.

If on one side the incidence of ectopic pregnancy is raising due to an increase in pelvic inflammatory disease, tubal surgery and intra-uterine devices, on the other lithopedion formation should become rarer since there is nowadays an easier access to improved pre-natal care with a consequent possibility of an early diagnosis and treatment of patients [1,2,4].

2. Case report

A 77-year-old female of poor socio-economical status was admitted to the emergency department of our hospital with a history of diffuse abdominal pain with an evolution of five days, associated with nausea, vomiting and constipation in the last two.

Her personal and family history was unremarkable. She was nulliparous and did not recall ever being pregnant.

Physical examination disclosed a huge incarcerated umbilical hernia. Bowel sounds were maintained outside the herniary formation.

Laboratory findings (blood counts, biochemical parameters, blood gases) were within normal limits.

An abdominal plain film was obtained in the supine position, both with vertical and tangential X-rays. It clearly showed the hernia with some air-containing bowel loops, and also a calcified heterogeneous mass in the mid-abdominal region (Fig. 1).

The hernia required urgent surgical correction (herniorrhaphy). It contained right and transverse colon, terminal ileum, epiploic fat and also the cecal appendix.

In an attempt to further characterize the calcified lesion, which was mistaken for a retroperitoneal mass during surgery, an abdominal and pelvic computed tomography (CT) examination was requested and performed two days after surgery, in a 4-row multidetector CT equipment (BrightSpeed, GE Healthcare, US) using a non-enhanced acquisition protocol (slice thickness: 2.5 mm, pitch: 1.5, reconstruction intervals: 1.25 mm). It disclosed a lithopedion (calcified ectopic pregnancy), depicting in great detail the fetal anatomy, especially on tridimensional MIP reconstructions (Fig. 2). The measurement of the femur length allowed determining that the gestation proceeded until the 30th week (Fig. 2f).

Due to the advanced age of the patient and because she was asymptomatic, it was decided that the calcified fetus should be left in place and no further surgery would be recommended.

The patient died few weeks after surgery due to a post-operative complication (a nosocomial pulmonary infection that was acquired shortly after surgery).

3. Discussion

Lithopedion is a term derived from the Greek words *lithos* (meaning stone) and *paedion* (meaning child) and describes an extra-uterine dead fetus that has become calcified [5,6]. This rare

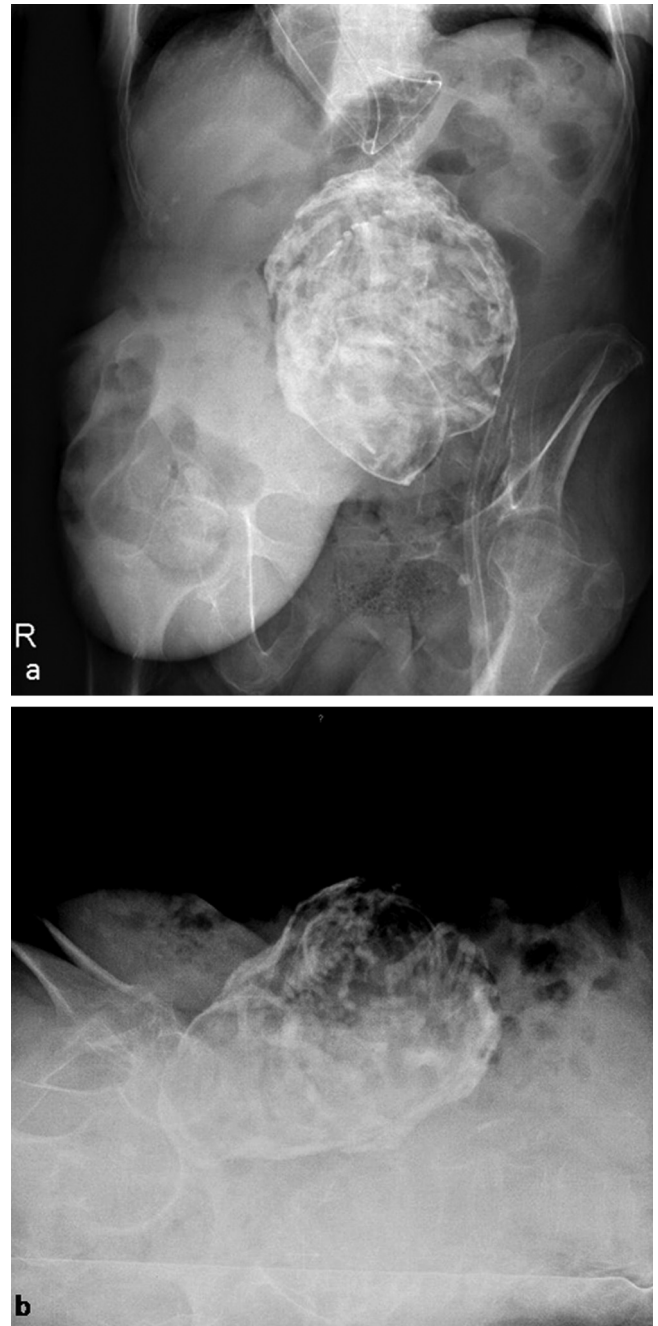


Fig. 1. Plain abdominal film obtained in the supine position, both with vertical (a) and tangential (b) X-rays. It clearly depicts a large abdominal wall hernia with some air-containing bowel loops, and also a calcified heterogeneous mass in the mid-abdominal region corresponding to a lithopedion. Some details of the fetal anatomy may be recognized, such as the head and the rib cage.

condition was first described in the 10th century by Albucasis, a surgeon of the Arabic era of medicine [6].

Abdominal pregnancies result from rupture of a tubal or ovarian pregnancy with implantation in the abdominal cavity [2,3]. They can have a complex course, and sometimes undergo calcification instead of being absorbed [7]. Requisites for the development of a lithopedion include an extra-uterine pregnancy that has escaped medical detection, fetal death after 3 months of pregnancy, a fetus that has remained sterile, and local

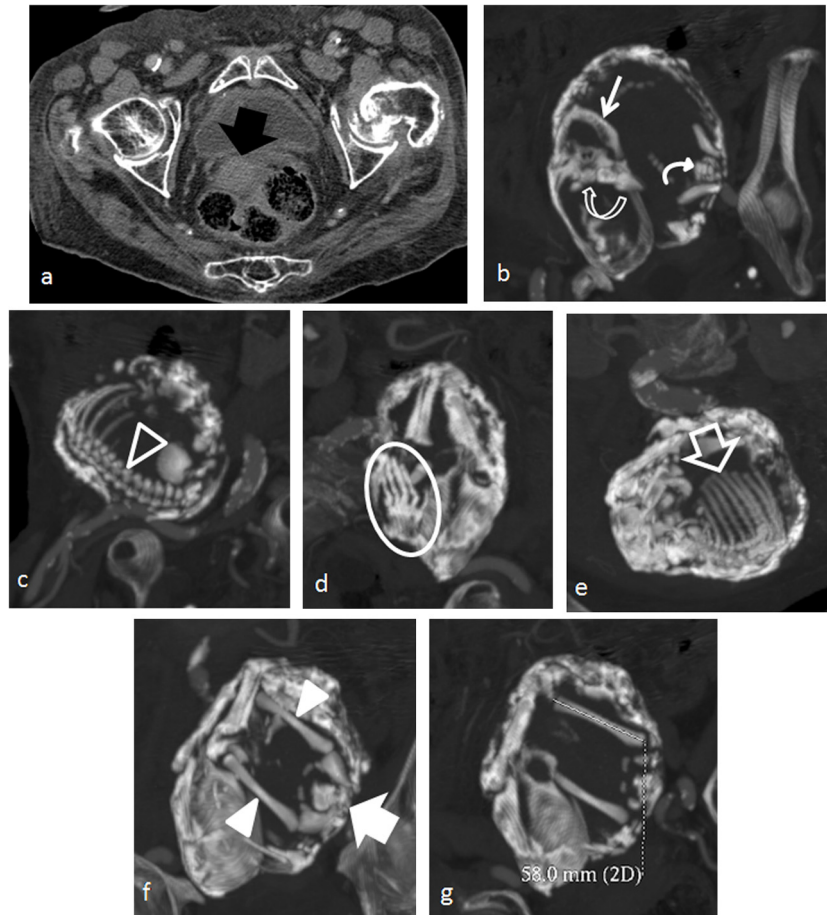


Fig. 2. CT: unenhanced study demonstrates an atrophic uterus with an empty cavity (black arrow in a); MIP reconstructions depict in great detail the fetal anatomy, such as the jaw bone (thin arrow in b), the sphenoid bone (open curved arrow in b) and the sacral bone (curved arrow in b), the dorsolumbar spine (open arrowhead in c), a hand (circle in d), the rib cage (open arrow in e), the femurs (arrowheads in f) and the bony pelvis (arrow in f); the measurement of the femur length allowed to determine that the gestation proceeded until the 30th week (g).

conditions that favor calcium deposition [2,5,8]. The pregnancy continues to develop in its intra-abdominal environment until fetal death, that occurred between 3 and 6 months of pregnancy in 20% of the cases, between 7 and 8 months in 27% and at full term in 43% of the cases previously reported in the literature [2,4]. In this particular patient, the measurement of femur length allowed to determine the fetal death at the 30th week of gestation.

Even if generically an abdominal calcified pregnancy is called a lithopedion, three different forms can be found, according to whether there is calcification of the fetal structures, of the placenta and membranes or both: true lithopedion (43%) in which the fetus is calcified but not the ovular membranes; lithokeliphos (26%) if the membranes are infiltrated and envelop the fetus whose calcification is negligible; and lithokeliphopedion (31%) when both the fetus and the membranes are involved in the process of calcification – the present case report is an example of this particular form [9].

The age of patients at the moment of diagnosis ranged from 23 to 100 years old in the related cases, with two-thirds of them being over 40 years old. The period of fetal retention varied from 4 to 60 years [2,4]. In this case, the exact retention period

is unknown, but it is reasonable to presume that it would be 30 years at least.

Although symptoms such as pelvic pain, abdominal tenderness and compressive symptoms to the urinary bladder and rectum may occur, most cases remain asymptomatic during large periods of time and represent incidental findings on imaging studies, surgery or necropsy [6].

The diagnosis may be reached by a suggestive clinical history and a palpable mass on physical examination. The rare previous imaging descriptions concern essentially the abdominal plain film and emphasize the value of this imaging method in the context of a lithopedion. Although it is usually enough to confirm the diagnosis, it cannot reliably differentiate from the different forms of lithopedion [10].

Due to the rareness of this situation, the value of modern cross-sectional techniques is still practically unknown. Ultrasonography may depict an empty uterine cavity and a calcified abdominal mass of non-specific characteristics [2]. Computed tomography or magnetic resonance imaging clearly are able to reach a conclusive diagnosis and may additionally define the involvement of adjacent structures such as rectum or urinary bladder [2–4]. Some authors also suggest that this last purpose

may be achieved by performing a barium enema or an excretory urography [1].

In this specific clinical case, since the diagnosis was not reached upon the abdominal plain film (although it could be achieved retrospectively), computed tomography was extremely important since it yielded the final diagnosis, by providing a clear-cut definition of the fetal anatomy with great accuracy; MIP images proved to be especially valuable in this regard. To the best of our knowledge this is one of the few cases studied on a MDCT equipment, and it clearly enhances the post-processing abilities of this imaging method which allows the obtention of diagnostic high-quality MIP images.

The differential diagnosis includes other calcified pathologic situations, including ovarian tumors, uterine fibroids, urinary tract neoplasms, inflammatory masses or epiploic calcifications [4].

The management of these situations is difficult, since complications such as bladder or rectal perforation, cecal volvulus, intestinal or urinary obstruction and abscess formation may arise [2,3,7]. As a result, some authors advocate surgical extirpation of a lithopedion, even if asymptomatic [7]. Others, on the contrary, adopt a more expectant attitude because some reports have demonstrated the stability of a lithopedion [2]. It is also known that there is an increase in morbidity when surgery is performed in elderly patients [7], rendering it necessary to evaluate the risk/benefit relationship of an operative approach in these cases. In this particular case, it was considered that the risk of excision overcame its benefits, so the lithopedion was left in place.

Retained non-viable abdominal pregnancies found at the 21st century imply either an absence of adequate medical attention or a serious mistake in medical judgment. Thus the present case

reflects not only a medical problem, but also a consideration about socio-economical and cultural attitudes.

Conflict of interest

The authors wish to confirm that there are no known conflicts of interest associated with this publication and there has been no financial support for this work that could have influenced its outcome.

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