



A case report of complicated appendicular hydatid cyst mimicking an appendiceal mucocele

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Abstract

Retaining the etiology of a cystic lesion in the right iliac fossa can be difficult. Appendicular hydatid cyst is a very uncommon cause of a such lesion. In some cases, diagnosis is not obvious. It can radiologically mimic an appendix mucocele, a complicated ovarian cyst, an appendicular lymphangioma or an abscess. Our case highlights the difficulties encountered in this kind of situation and despite the contribution of imaging. We present a case of a 75 years-old woman presented with a right lower quadrant continuous pain. Abdominal CT-scan revealed a multilocular cystic and hydro-aeric mass. The diagnosis of an appendiceal mucocele complicated with gelatinous peritonitis was suspected. An open debulking surgery with right hemicolectomy was performed. The pathological exam has concluded to an infected appendicular hydatid cyst with thick calcified walls. The aim of this work is to report a case of an appendicular hydatid cyst that has imitated an appendicular mucocele to discuss the importance of differential diagnostic reflections and the appropriate treatment.

Keywords Hydatid cyst · Mucocele · Surgery · Appendix · Infection

Introduction

Lower right quadrant pain is a frequent symptom in surgery consultations. In some cases, diagnosis is not obvious. Many illnesses can imitate surgical conditions and may lead to unnecessary or inappropriate surgical treatment. Appendicular hydatid cyst is one of these rare conditions. The clinical signs are often insidious. It can radiologically mimic an appendiceal mucocele, a complicated ovarian cyst, an appendicular lymphatic malformation or an abscess. The aim of this work is to report a case of an appendicular hydatid cyst that has mimicked an appendicular mucocele to discuss the importance of differential diagnostic reflections and the appropriate treatment.

Case report

A 75-years-old woman presented in the Emergency Department with a right lower quadrant continuous pain and fever for 3 days. She had no associated signs, essentially, no metrorrhagia or gynecologic abnormalities. Abdominal examination revealed right lower quadrant tenderness with an abdominal mass. The liver and the spleen were not palpable. White blood cells count was 13,000 cells/ml, eosinophilia cells count was 160 cells/ml, hemoglobin level was 12.8 g/dl, and C-reactive protein was 264 mg/ml. The ELISA test of hydatid disease was not performed. Abdominal ultrasound revealed a cystic hypoechoic unilocular mass in the right iliac fossa. It was a heterogenous cyst measuring 110×45 mm (Fig. 1). Then an abdominal CT-scan was performed. Which revealed a cystic mass localized under the caecum and extending to the right vesical flank measuring 152×58 mm with no liver cystic lesions. This mass had a thin laminated membrane with calcifications and intraluminal gas along the margin, no filled by contrast medium and it was surrounded by lymph nodes (Fig. 2). Based on radiological features the diagnosis of an appendiceal mucocele complicated with gelatinous peritonitis was suggested. At laparotomy, there was a large infected right lower quadrant

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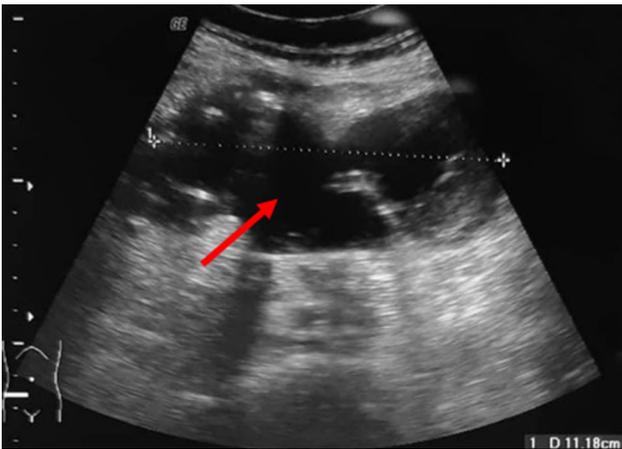


Fig. 1 Abdominal ultrasound revealed a cystic heterogenous hypo-echoic unilocular mass with intraluminal gas measuring 110×45 mm (red arrow)



Fig. 2 Abdominal CT-scan revealed a cystic mass (red star) measuring 152×58 mm with a thin laminated membrane with calcifications (red arrow) along the margin

mass fistulized to the caecum without purulent intra-peritoneal effusion. This mass had a calcified capsule and did not adhere to adjacent organs (Fig. 3), with no disseminated nodules, no peritoneal spread and no liver metastases. Open debulking surgery with right hemicolectomy was performed. The postoperative course was uneventful and the patient was discharged after 5 days. Pathological exam objectified a cyst containing folds of laminated chitinous layer, partly lining the wall, surrounding areas show chronic inflammation with sheets of foamy macrophages with Echinococcal scolex in the capsule and concluded to appendicular hydatid cyst with a thick calcified wall (Fig. 4). Patient was put on Albendazole regimen 10 mg/Kg/daily for 12 weeks after surgery.



Fig. 3 Intra-operative view of the right lower quadrant intra-peritoneal mass with calcified capsule

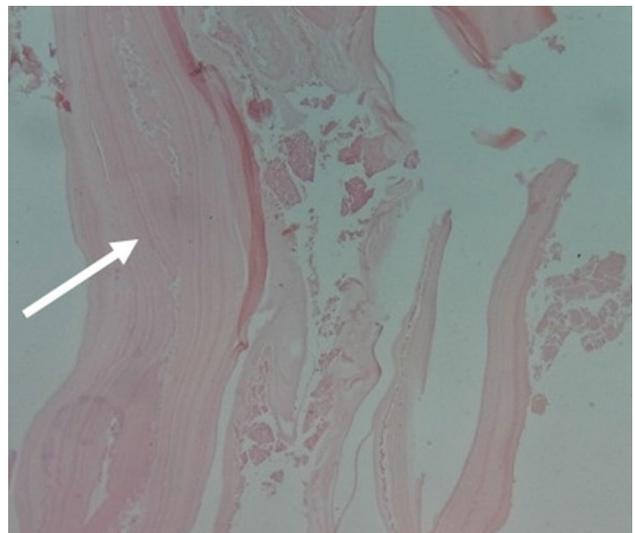


Fig. 4 Acidophil laminated hydatid membrane (white arrow) (HEX40)

The patient was followed for 3 years, no recurrence was observed.

Discussion

Many surgical affections can be discovered by a cystic lesion in the right iliac fossa. In some cases, diagnose is difficult. Our case highlights the difficulties encountered and the radiological contribution. It includes ileal, caecal, ovarian, peritoneal and parietal disease. In our case, the dilemma was between an appendicular hydatid cyst and appendicular mucocele, regarding the uncommon bowel localization, especially appendicular, of hydatid cyst and insidious

clinical signs. The incidence of mucocele of the appendix is reported to be 0.2%–0.3% [1]. Primary hydatid cyst of appendix is extremely rare [2], in a review of the literature, only nine cases have been reported [2]. In 25%–50% of cases, mucoceles are asymptomatic and found incidentally at physical examination or during abdominal imaging or surgery [1]. Generally, appendiceal mucocele and appendix hydatid cysts are more symptomatic when they are complicated by infection or rupture in the gastrointestinal tract [3]. In many cases, it evokes clinical and radiological issues. Despite the increasing sophistication of cross-sectional imaging with very rapid development and integration of interventional radiology, the diagnosis of some conditions remains difficult. In the case of an appendicular hydatid cyst, ultrasound, CT-scan and/or MRI could demonstrate a characteristic pattern of hydatid disease by showing these signs:

- Daughter cysts sign [4].
- Rim sign [5]: the presence of a low-signal-intensity rim separating the parasitic cyst from the patient's tissue.
- Serpent sign [6]: the collapse of parasitic membranes occurs secondary to damage or degeneration of hydatid cysts with rupture of the endocyst.
- Air bubble sign [7]: most ruptured cysts become infected. Air may enter the cyst by dissection of pericyst and parasitic membranes.
- Cyst wall sign [4]: the wall of a hydatid cyst can be visualized if several embryos are embolized close together, they may first develop as individual cysts.
- Ring enhancement sign [4]: due to vascularization of the pericyst and there may be ring enhancement secondary to contrast enhancement.
- Sunburst sign [8]: Old, damaged, or ruptured cysts may display a 'sunburst appearance'.

None of these radiological signs were found in our patient. In addition, these radiological exams can clearly identify the cyst wall calcification, infection and peritoneal seedling [9]. In our case, the diagnosis of appendicular mucocele was suspected. Because a cystic mass in the expected region of the appendix is the typical radiological finding of an appendiceal mucocele. Ultrasound had shown a unilocular cystic structure with no distinct cyst wall suggesting an appendiceal mucocele. Typically, abdominal ultrasound shows an oblong, heterogeneous well-encapsulated cystic mass containing mucin [10]. Excellent through-transmission and posterior enhancement are usually present [11]. Abdominal CT-scan objectified a low-attenuated and well-encapsulated mass with smooth regular walls. Furthermore, the presence of wall calcification in right iliac fossa mass, but it is detectable in less than 50% of cases [1], with intraluminal gas allows one to differentiate an infected mucocele from an abdominal abscess cavity [11].

Diagnosis of primary abdominal hydatid cyst presenting in emergency as appendicular mass is difficult due to non-specific imaging findings, no past medical history of hydatid cyst disease, absence of specific clinical symptoms, absence of serological test like ELISA feasible in emergent condition and contraindication of radiological guided biopsy. This diagnosis is easier when appendicular cyst is associated with concomitant liver hydatid cyst or previous hydatid disease [2, 12]. In addition, even in high index of clinical suspicion the confirmation of the diagnose remain by pathological examination. With respect to treatment of this hydatid cyst localization, radical surgery is the best alternative. Surgical treatment, using open or laparoscopic approach, is feasible and safe in case of appendiceal hydatid cyst and appendiceal mucocele [13]. But it is commonly admitted now that the laparoscopic approach is associated with a higher rate of complications and recurrence in case of hydatid cyst [14]. The post-operative delay is generally uneventful, like in our case. But, it is recommended to associate medical treatment, Albendazole, after surgery [15]. Since recurrence rate remains very variable from 0 to 25% [16].

Conclusion

Cystic lesion in right iliac fossa imposes a systematic and meticulous confrontation between clinical and radiological data to approach diagnosis and to avoid an inappropriate treatment.

Compliance with ethical standards

Conflict of interest Mohamed Ali Chaouch, Mohamed Wejih Dougaz, Mehdi Khalfallah, Hichem Jerraya, Ramzi Nouira, Ibtissem Bouasker, Chadli Dziri had no conflicts of interest to disclose.

Human/animal rights All procedures followed were in accordance with the ethical standards of the responsible committee on human experimentation (institutional and national) and with the Helsinki Declaration of 1964 and its later amendments.

Informed consent Informed consent was obtained from all patients for being included in the study.

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