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Granulosa Tumor Simulating a Secondary Localization of a Hydatid Cyst: Case Report

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Abstract **Case Report**

The location of hydatid cysts in the pelvis as a secondary site is rare and misleading, with a disconcerting clinical symptomatology. Preoperative diagnosis of pelvic hydatidosis is sometimes difficult, and should be considered in the presence of any pelvic mass, especially in endemic countries. However, misleading cystic forms do exist, and differential diagnoses other than secondary localizations of hydatid cysts may be observed. We report a case of an adult granulosa tumor mimicking a secondary pelvic localization in a patient with a hydatid cyst of the liver.

Keywords: Liver, Cyst, Surgery, Cystic Fibrosis, Echinococcosis, Peritoneum, Adult-Juvenile-Ovarian Granulosa Tumor.

INTRODUCTION

Hydatidosis is a metacestodosis caused by the development in humans and many wild and domestic mammals of the larval form of a small dog taenia: Echinococcus granulosus. Hepatic localization is the most common (60%), followed by pulmonary localization, most frequently in the thoracic compartment. However, cysts can affect all organs [1]. Pelvic hydatid cysts in women remain rare and misleading, with a frequency varying from 0.3 to 4.27% of hydatid localizations [2].

The seriousness of hydatid cysts lies mainly in their complications, which are dominated by rupture in the bile ducts, infection and intraperitoneal rupture. Intraperitoneal rupture is a rare complication, but a real emergency, and represents a major turning point in the evolution of hydatidosis: immediately, through the risk of anaphylactic shock and hydatid peritonitis, and secondarily, through secondary peritoneal hydatidosis caused by seeding of this serosa by the echinococcus tapeworm. Les tumeurs de la granulosa sont des tumeurs rares appartenant à la famille des tumeurs du stroma et des cordons sexuels [3]. Ces tumeurs, difficiles à diagnostiquer en préopératoire, peuvent-elles être un diagnostic différentiel d'une localisation pelvienne d'un kyste hydatique ?

We report a case of a 43-year-old female patient who was admitted to the emergency department for abdominal pain associated with progressive enlargement of the abdomen and dyspnea, all evolving in a context of altered general condition.

Radiological examinations confirmed a hepatic cystic mass with a secondary pelvic localization on the right ovary. The diagnosis of pelvic hydatid cyst was questioned intraoperatively when a voluminous cystic mass was found depending on the right ovary. Treatment was based on surgical resection of the hepatic cyst and pelvic cystectomy involving the right ovary. The cystic mass measured 30cm long and weighed 8kg. Pathological examination and immunostaining showed an adult granulosa tumor. The evolution was favorable after surgery.

OBSERVATION

A 45-year-old female patient presented to the emergency department with abdominal pain of the heaviness type, progressive in onset over the past 10 months, followed by an abdominal mass, several episodes of vomiting and decubitus dyspnea, all evolving in a context of altered general condition.

Clinical examination revealed a hemodynamically stable, dyspneic patient with a highly distended, unscarred abdomen and, on palpation, a poorly defined, firm mass occupying the right hypochondrium and pelvic region.

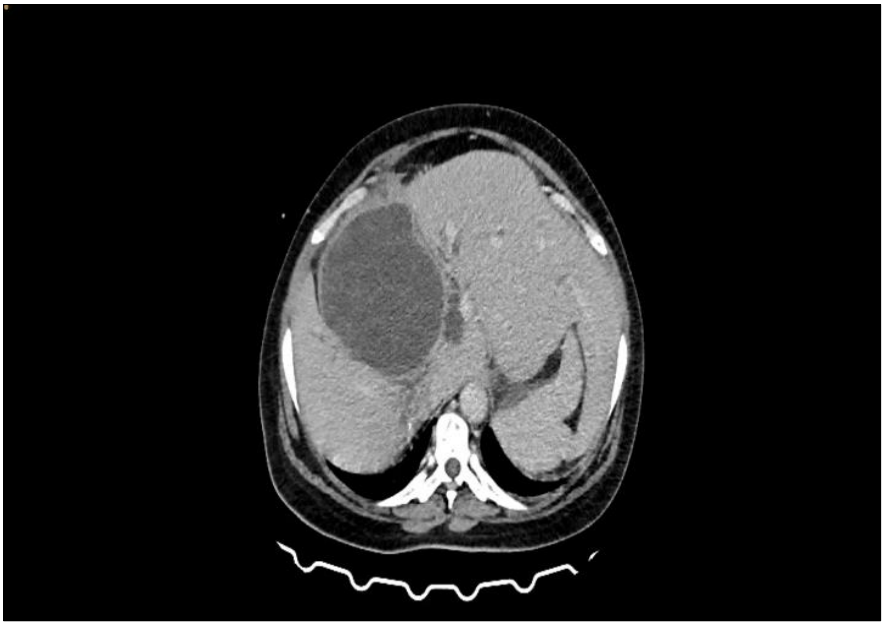
Biological workup showed predominantly neutrophilic hyperleukocytosis, WBC:16,000, anemia 6.2g/dl, CRP:135, liver function tests normal.

Abdominal ultrasonography showed an uncomplicated hepatic hydatid cyst of segments IV and VIII, classified as Gharbi III, and a pelvic mass probably dependent on the ovary, suggesting a secondary location of the hydatid cyst.

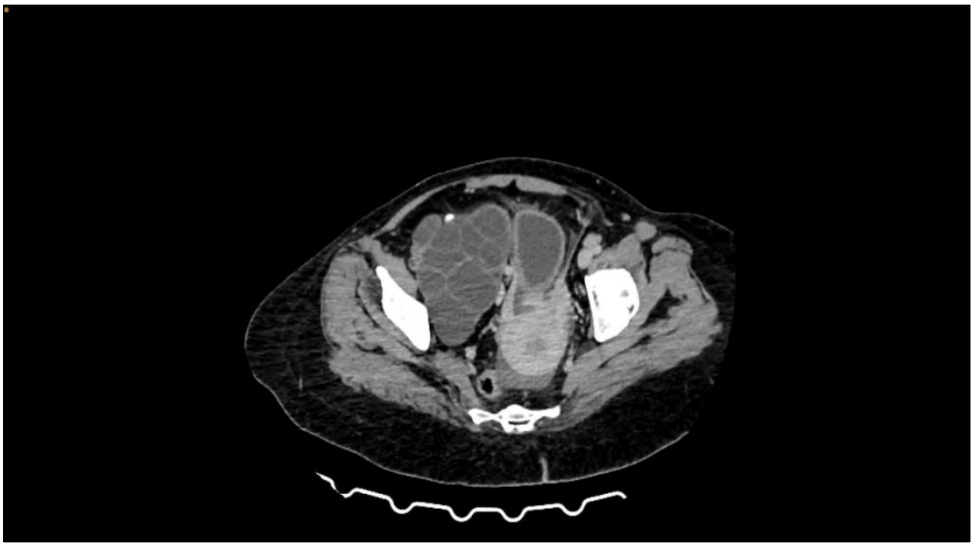
An abdominal CT scan showed a CE2b cystic mass at the junction of segments IV and VIII, and multiple confluent, multipartitioned intraperitoneal cystic masses creating a voluminous peritoneal mass measuring 24x19cm, indicative of peritoneal hydatidosis.

Based on the results of the patient's clinical examination and CT scan, the patient was prepped for surgery.

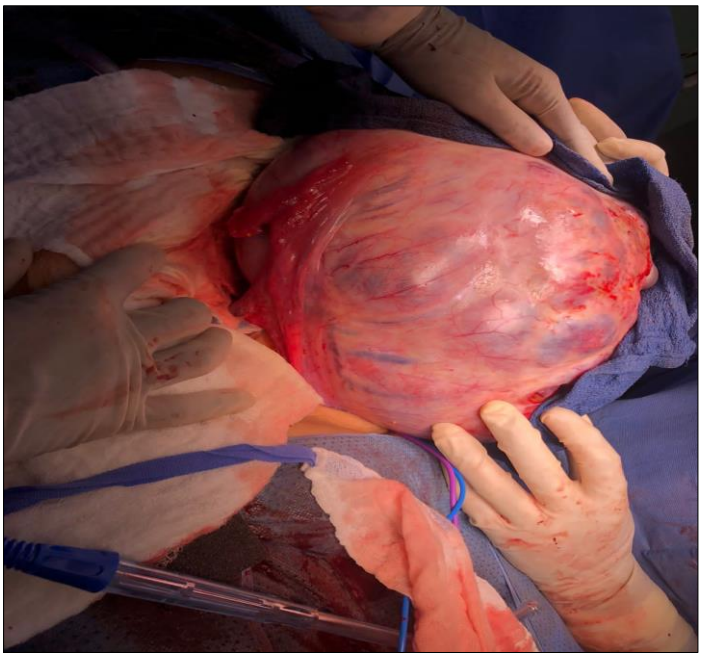
Exploration revealed a hydatid cyst, which was evacuated, and a large cystic mass dependent on the right ovary, which was resected in monobloc with adnexectomy.



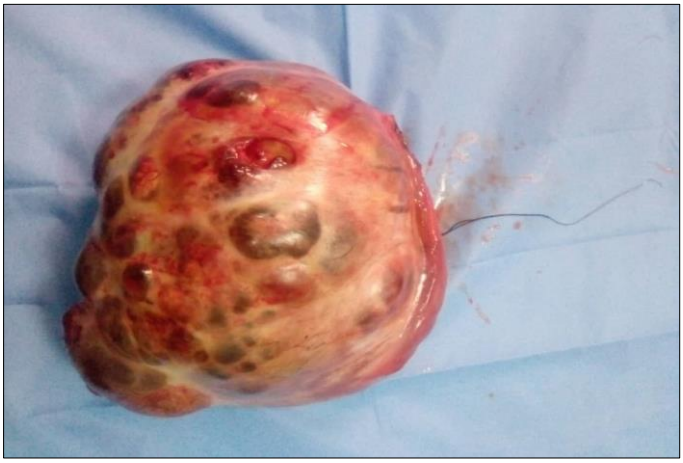
a. Hydatid cystic mass type CE2b of liver segments II, III et IV



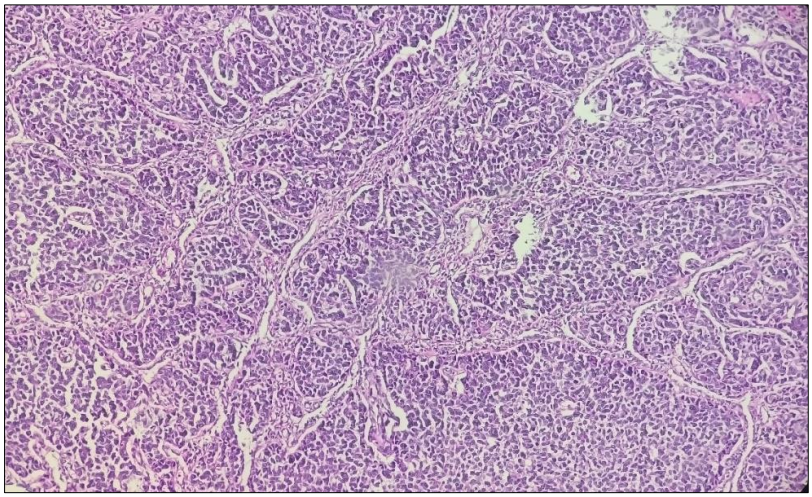
b. Intraperitoneal confluent cystic mass with multiple septations, creating a honeycomb appearance measuring 24x19cm, indicative of peritoneal hydatidosis.



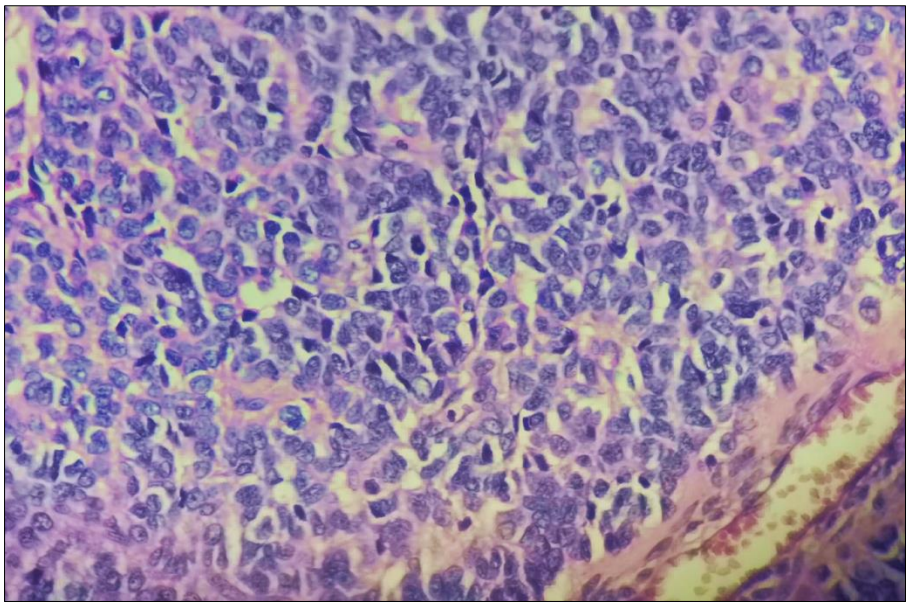
c. Pelvic cystic mass with appendix



d. Large cystic mass with associated adnexectomye



e. Histological section of an adult Granulosa cell tumor showing microfollicular architecture with "Call-Exner" bodies (HES x 40), [Pathological Anatomy Laboratory, CHU Hassan]



f. Histological section of an adult Granulosa cell tumour showing tumour cells with a kidney-like appearance (HES x 400), [Pathological Anatomy Laboratory, Hassan II University Hospital, Fez]

DISCUSSION

Peritoneal hydatidosis is one of the most serious complications of hydatid disease. Its frequency among operations varies between 4.5% and 6.9% depending on the series [4-6]. Rupture of the primary cyst is spontaneous in 78% of cases [7].

It is favoured by the superficial location of the cyst, its large size, thin wall and high intracystic pressure. Traumatic rupture is most often caused by iatrogenic surgery (14% of our patients underwent surgery for KHF) or diagnostic procedures (PBF, trans-hepatic cholangiography, etc.). It may also be secondary to abdominal contusion or a road accident. The stages following rupture reflect the reaction of the peritoneal sereosa to hydatid aggression. They take the form of simple hydatid ascites when the cyst is monolocular, free hydatids in the large peritoneal cavity when the cyst is muulti-vesicular, or hydatid peritonitis if the cystic contents are infected. In the absence of treatment, these early forms can evolve in two ways:

- Subserosal grafting with development of peritoneal cysts is by far the most frequent. It can be either a multiple generalized form, sometimes with more than 100 cysts, or a form localized to a region of the abdomen
- Encysted collections are characterized by the existence of a neofo rmed encystment membrane isolating the ruptured cystic contents from the rest of the peritoneal cavity.

The diagnosis of pelvic hydatidosis is rare, and is made in the presence of any pelvic cystic formation in a patient known to have a hydatid cyst, especially in an endemic area [8].

Granulosa tumours are rare malignant tumours of the ovary (2-3% of ovarian cancers) [9, 10]. Their main diagnostic features, although not very specific (abdominal pain, mass, abdominal distension, and possible digestive or urinary signs if the mass is large), may raise suspicion of a secondary localization of hydatid cysts in carrier patients, especially in endemic areas.

Histologically, two main forms can be distinguished: the adult form, which is the most frequent (95% of granulosa tumors), and the juvenile form, which is more aggressive. Adult granulosa tumors generally occur after the age of 30, often after the age of 50, whereas juvenile tumors are common before the age of 30.

Diagnosis of granulosa tumors is based on a combination of imaging, biological and pathological examinations, which remain the only definitive test.

Treatment options depend on the degree of extension of the tumour, e.g. into neighbouring organs. Surgery remains the cornerstone of curative treatment of granulosa tumours, and is frequently performed laparoscopically, thus limiting scarring. In the early stages (limited to the ovary, as in our patient's case), surgical treatment is based on adnexectomy (removal of the ovary and tube) with complete peritoneal staging (exploration of the entire abdomino-pelvic cavity with multiple biopsies) and uterine curettage [3-10].

Total hysterectomy is generally performed in women who have reached menopause or no longer wish to retain their fertility. There is no need for lymph node dissection. In advanced stages, surgery is based on complete excision of all tumour sites, such as epithelial ovarian cancer.

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Declaration of Competing Interests: The authors declare that they have no conflicts of interest.

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