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HYDATIDOSIS FISTULIZED TO THE ABDOMINAL WALL: A RARE MODE OF PRESENTATION

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Abstract

In countries of the South, hydatid cyst disease is an endemic parasitic condition that can sometimes lead to serious complications. Spontaneous rupture of the cyst through the abdominal wall is extremely rare, and very few cases have been reported in the medical literature. We report the case of a 47-year-old man with multiple, previously unrecognized hydatid cysts, one of which spontaneously fistulized to the abdominal wall, revealing the disease. The diagnosis was established based on clinical examination, radiographs, and serological tests. The patient underwent surgical treatment and received perioperative albendazole chemotherapy, with uncomplicated postoperative recovery.

Keywords: Echinococcosis, Fistula, Complication of the Hydatid Cyst, Abdominal Wall.

INTRODUCTION

Hydatid disease, or cystic echinococcosis, is a zoonotic infection caused by the larva of Echinococcus granulosus, which accidentally infects humans through the oro-fecal route. The liver is the preferred site for the development of hydatid cysts due to the intestinal absorption of the parasite. Often, the disease is asymptomatic, and the typical cystic lesion may be described as an incidental finding, sometimes revealed by a complication such as cyst rupture (spontaneous, traumatic, or iatrogenic) or occasionally by secondary superinfection. The most frequent complication is opening into the bile ducts; spontaneous cutaneous fistulization of a hepatic hydatid cyst (cyst-cutaneous fistula) is the least encountered presentation. The patient's medical history, specific serological tests, and imaging play a significant role in describing a comprehensive clinical picture.

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A 47-year-old patient, with no notable medical history, presented over the past year with a progressively enlarging and painful mass in the right hypochondrium, which subsequently fistulized to the skin. Clinical examination revealed an afebrile patient with a mass in the right hypochondrium extending to the flank, painful, with a lateral fistulous orifice measuring 20mm, draining a few cc of clear fluid with no evident hydatid material.

Ultrasound, complemented by computed tomography (Figure 1 and 2), revealed the presence of a hypodense formation in segment VII of the liver measuring 50mm in length and 20mm in thickness, with collapse of the hepatic contours in the corresponding area, and a perihepatic collection in segment VIII adjacent to the collapsed liver contour measuring (105x95x31) mm, with discretely enhanced thickened wall after contrast injection. This collection extended into an intramuscular collection in the right hypochondrium subcostally measuring (102x100x50) mm. Additionally, two formations suggestive of hepatic hydatid cysts were noted in segments IV (55x35) mm and V (45x35) mm, classified as Gharbi stage III, along with a unilocular cystic formation subphrenic and left lateral to the interhepatogastric area measuring (90x63x42) mm, with a thin wall and liquid density, not calcified and showing no enhancement after contrast injection. Analysis of the cystic fluid and fistulography were not performed. Serology for hydatidosis was strongly positive. The diagnosis of multiple hydatidosis with a cyst fistulized to the skin was established.

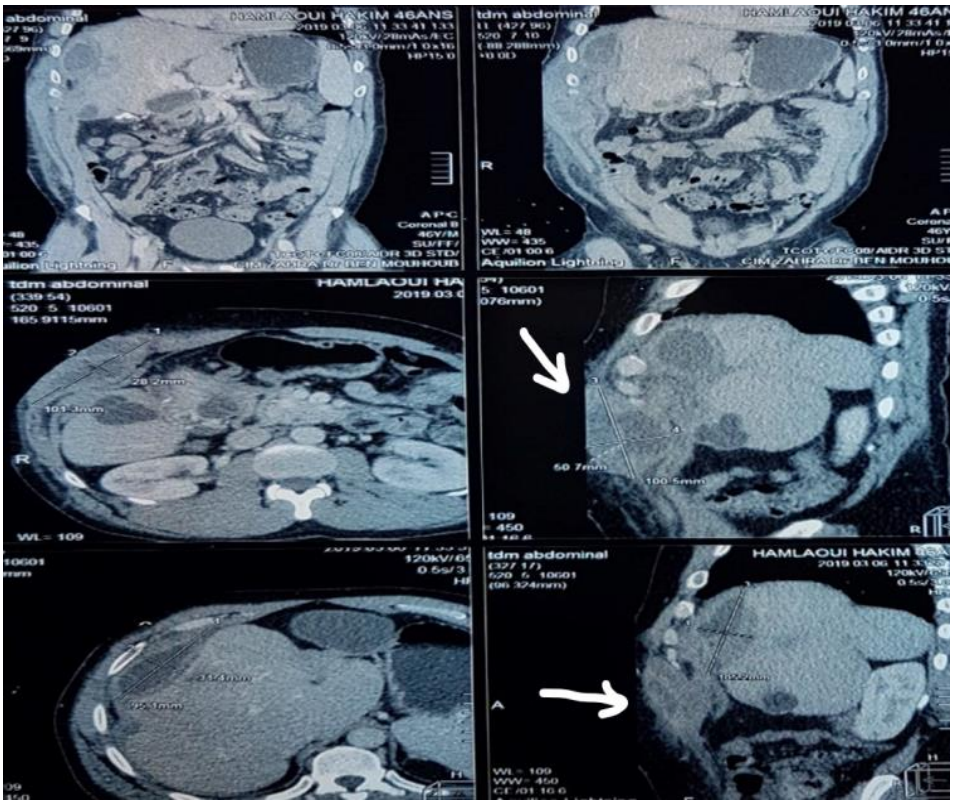

Figure 1: Multiple hydatid cysts of the liver

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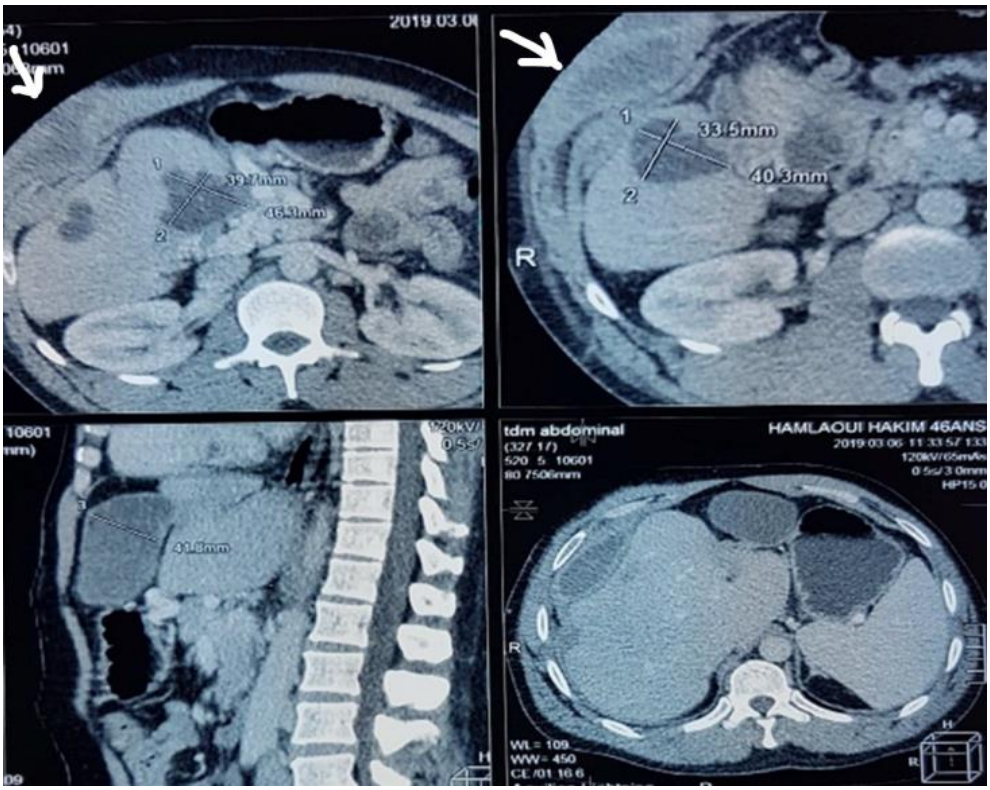

Figure 2: Hydatid cyst cutaneous fistula

The patient was treated with Albendazole at a dose of 12 mg/kg/day for 15 days, followed by surgery via an extended right subcostal approach to the left (Figure 3). Surgical exploration revealed a well-defined cystic formation of approximately 80 mm located inter-hepatogastric, resting on the pars flaccida, two hepatic cystic formations in segments IV and V measuring approximately 60 mm and 40 mm, respectively, and another cystic formation in segment VII adherent to the abdominal wall and fistulized to the skin. Initially, the operative field was protected with compresses soaked in hydrogen peroxide. After complete mobilization, cysto-parietal disconnection with aspiration of cystic fluid and daughter vesicles was performed, followed by sterilization of the cyst, resection of the protruding dome, and complete resection of the parietal fistulous tract. Interposition of the omentum between the cyst and the abdominal wall was then performed, followed by drainage of the residual cavity and the cutaneous cavity. The inter-hepatogastric cyst was treated by total closed cyst pericystectomy (Figure 4). The cysts in segments V and IV were treated by puncture, aspiration, and sterilization with hydrogen peroxide, followed by resection of the pericysts and drainage of the residual cavities. No communication with the bile ducts was observed. The postoperative course was uneventful, with drain removal erformed after eight days. The patient was discharged on postoperative day 9 with a prescription for a 3-month course of Albendazole chemotherapy.

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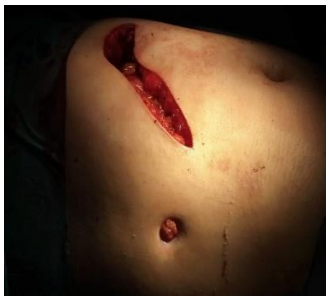

Figure 3: Cystocutaneous fistula

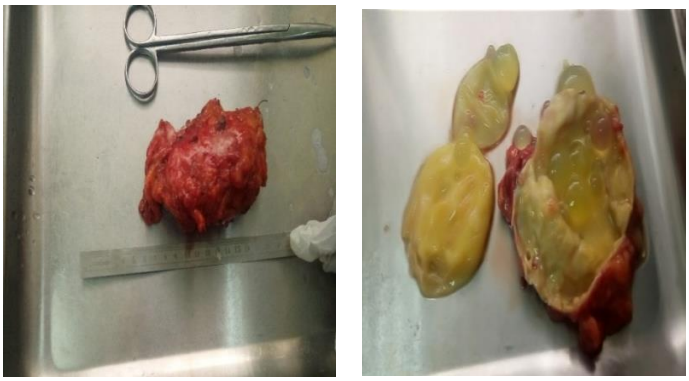

Figure 4: Total closed cyst pericystectomy

DISCUSSION

Cutaneous fistulization of a liver hydatid cyst is a rare complication, and our observation represents one of the few cases reported in the literature [1-4]. Hydatid disease, which is endemic in certain regions, is caused by the tapeworm Echinococcus granulosus and primarily affects the liver (50%-77%) [5, 6]. The progressive course of the disease can result in asymptomatic manifestations over several years or may be revealed by complications (in 20% of cases). Liver hydatid cysts typically rupture into the bile ducts, gastrointestinal tract, bronchi, peritoneal cavity, or pleura. Rupture into the bile ducts is the most frequent complication, occurring in 10 to 15% of cases [7, 8].

This complication mainly affects peripheral, thick, and calcified cysts in the right lobe of the liver. Two mechanisms, inflammatory and mechanical, appear to be involved in the development of fistulas [9]. In our case, the fistulized cyst was posterior, exophytic, with a thick pericyst suggestive of inflammation. Additionally, the mechanical effect of cyclic respiratory movements likely contributed to the progressive erosion of the cyst and fibrosis of adjacent tissues. Three stages have been identified in the genesis of these fistulas [10]: Stage I: Involvement of the innermost layer of the abdominal or thoracic wall muscle; Stage II: Extension beyond the muscle layer, protruding into the subcutaneous tissue; Stage III: Passage of lesions into the subcutaneous tissue and fistulization to the skin.

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These lesions are often asymptomatic; however, clinical symptoms may vary depending on the cyst location, size, stage of development, and the presence or absence of complications. Abdominal pain is the most frequent clinical symptom [11]. The difficulty of diagnosis and the severe progression of hydatid disease make cystic hydatid disease with cutaneous fistulization of particular clinical interest. The rarity of hydatid cyst fistulization may lead to misdiagnosis, emphasizing the importance of considering this possibility, especially in endemic areas. The combination of clinical, radiological, and serological data facilitates diagnosis. CT scans provide a comprehensive description of the cyst and its fistulous tract [12]. Ultrasonography, thoracic CT, MRI, and fistulography [13-18] are commonly used radiological modalities for diagnosis. ERCP, PTC, and endosonography are invasive radiological methods that may be used for both diagnostic and therapeutic purposes. Enhanced fistulography appears to be the most useful modality for cutaneous complications of hydatid disease. It specifies the extent of the fistula, its location, the size of the fistulized lesion, and its relationship with the biliary tract.

Histopathological examination of the fluid drained from the external fistulous orifice may, in some cases, reveal the parasite and thus confirm the diagnosis. Positive hydatid serology provides additional support in cases with uncertain imaging findings. However, some liver abscesses can also fistulize to the skin, although this remains an exceptional phenomenon. In our patient, the diagnosis was primarily based on radiographic findings.

Surgical treatment is clearly indicated for complicated cases of liver hydatid cysts. It involves elective surgery for parietal complications, preceded by neoadjuvant benzimidazole treatment for 2 to 4 weeks. In stage I and II parietal complications, surgical treatment consists of total resection of the protruding hydatid cysts. In stage III parietal complications, cystic resection should be supplemented with excision of the fistulous tract to the adjacent skin [19, 20]. In our case, the patient had a cystocutaneous fistula (Stage 3), and he received neoadjuvant Albendazole followed by conventional conservative surgery for the liver hydatid cyst, which included total resection of the fistulous tract and postoperative medical treatment. Major radical surgeries such as segmentectomy and hepatic lobectomy are less prone to recurrence but are associated with increased morbidity compared to conservative surgical approaches. Adjuvant treatment is indicated for complicated cases treated with conservative surgery, such as partial cystectomy. The cystic cavity should be drained and reduced in size, and benzimidazole treatment should be administered for 4 to 12 weeks.

CONCLUSION

Spontaneous cystocutaneous fistulization of a hydatid cyst is an exceptional complication of hydatid disease. Diagnosis can be challenging, especially in the absence of hydatid material discharge from the external fistulous orifice. However, it should be considered in the differential diagnosis of patients presenting with a cutaneous fistula, particularly in endemic areas of hydatid disease. Imaging plays a crucial role in resolving these diagnostic challenges. Surgical treatment aims not only to treat the liver hydatid cyst but

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also to disconnect the cyst from the abdominal wall. Chemotherapy with anthelmintic agents remains a valuable alternative to reduce hydatid recurrence.

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