

## Cystocutaneous fistula, an exceptional complication of the hydatidcyst of the liver: About a case

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### Abstract:

Hydatid disease is a parasitic disease that rages in an endemic state in Algeria, spontaneous cutaneous fistulization of a hydatid cyst of the liver remains an exceptional complication, to date very few cases have been reported in the literature. We report the observation of a 47 year old patient with multiple hydatidosis revealed by a spontaneous cystocutaneous fistula from segment VII of the liver. The diagnosis was established on clinical, radiological and serological data. The patient received surgical treatment and antiparasitic chemotherapy in the perioperative period, based on Albendazole with simple postoperative consequences.

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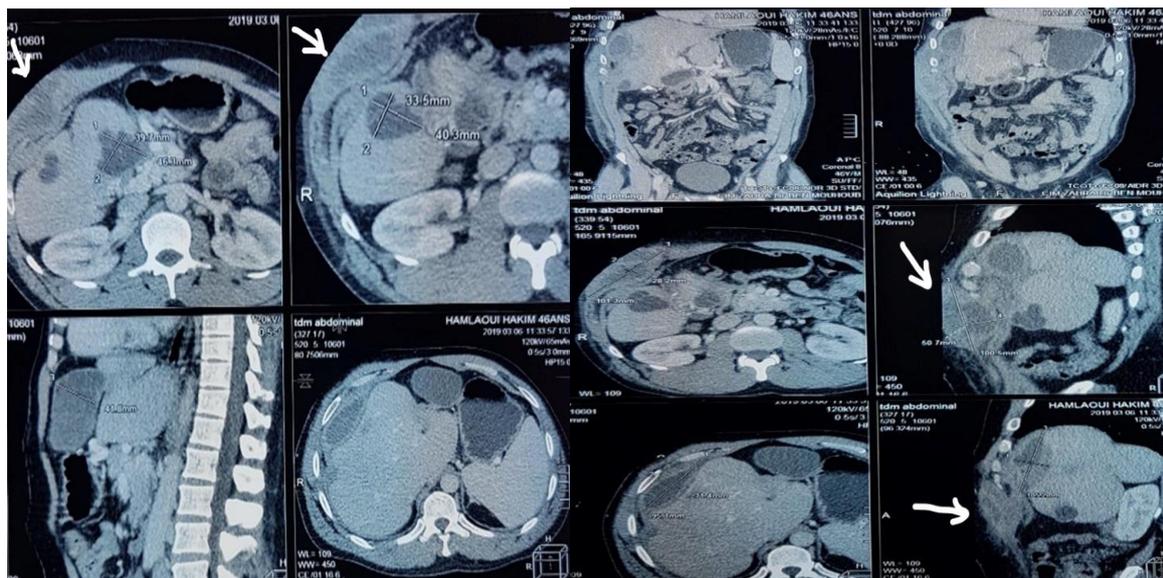
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### I. Introduction

Hydatid disease, or cystic echinococcosis, is a zoonotic infection caused by the larva of echinococcusgranulosus, which accidentally infects humans via the oral-fecal route. The liver is the preferred site for the development of the hydatid cyst, due to the intestinal absorption of the parasite. Most often, the disease is asymptomatic and the typical cystic lesion can be described as an incidentaloma, sometimes revealed by a complication such as rupture of the cysts (spontaneous, traumatic or iatrogenic) or sometimes by secondary secondary infection. The most common complication is the opening in the bile ducts, the spontaneous cutaneous fistulization of a hydatid cyst of the liver (cystocutaneous fistula) is the least encountered presentation. The patient's clinical history, specific serological tests and imaging are of interest in describing a complete clinical picture.

### II. Clinical case:

A 47-year-old patient, with no notable pathological history, who has had a progressively increasing and painful mass of the right hypochondrium for a year, which has subsequently fistulated on the skin. The clinical examination found an afebrile patient who had a mass of the right hypochondrium extended to the side, painful with the presence of a 20mm lateral fistulous orifice, draining a few cc of clear liquid with the absence of obvious hydatid material. Ultrasound supplemented by computed tomography (Fig 1,2), revealed the presence of a hypodense formation of segment VII of the liver 50mm long axis and 20mm thick with sagging of the hepatic contours facing, perihepatic collection of segment VIII opposite the sagging of the liver contour of (105x95x31) mm, with a thickened wall discretely enhanced after injection of contrast product. This collection continues with an intramuscular collection of the right hypochondrium by extension under parietal under right rib which measures (102x100x50) mm. We also note the presence of two formations in favor of hydatid cysts of the liver at the level of segments IV (55x35) mm, V (45x35) mm, classified stage III of Gharbi with also the presence of a cystic uniloculate formation under lateral phrenic to left inter hepato gastric (90x63x42) mm, with thin wall and liquid density, not calcified and not enhanced after injection of contrast product. The examination of the cystic fluid as well as the fistulography was not carried out. The hydatidosis serology was strongly positive. The diagnosis of multiple hydatidosis with fistulized skin cyst was accepted.



**Fig1, 2:** Multiple hydatid cysts of the liver including one fistulated on the skin (segment VII) ;Interhepatogastrichydatid cyst

The patient was treated with Albendazole, at a dose of 12 mg / kg / day for 15 days, then operated on by a right costal approach enlarged on the left (Fig 3). Surgical exploration finds a well limited cystic formation of 80mm inter hepatogastric resting on the pars flacida, two hepatic cystic formations at the expense of segments IV and V measuring 60mm and 40mm approximately, another cystic formation of segment VII, adherent to the abdominal wall and fistulated on the skin. Protection of the operating field was first performed with compresses soaked in oxygenated water. After complete mobilization, a cysto-parietal disconnection with aspiration of the cystic fluid and the daughter vesicles was performed, then sterilization of the cyst was carried out, followed by resection of the protruding dome and complete resection of the parietal fistulous path. an interposition of the omentum between the cyst and the abdominal wall was performed, then we ended with drainage of the residual cavity and the skin cavity. The inter hepatogastric cyst was treated with a total pericystectomy with a closed cyst (Fig 4). The cysts of segments V and IV were treated by puncture, aspiration and sterilization with hydrogen peroxide then resection of the perikysts and drainage of the residual cavities. No communication with the bile ducts was found. The consequences were simple, the drains were removed after eight days and the patient was discharged on D9 postoperatively with antiparasitic chemotherapy (Albendazole) prescribed for 3 months.



**Fig 3:** Cystocutaneous fistula



**Fig 4:** Total peri-cystectomy with closed cyst

Cutaneous fistulization of a hydatid cyst of the liver is a rare complication, our observation represents one of the rare cases reported in the literature [1, 2, 3, 4]. Hydatidparasitosis, which rages in an endemic state, is due to the tæniaEchinococcusGranulosus, localized mainly in the liver (50% -77%) [5,6]. The progressive evolution of the disease means that it can remain asymptomatic for years or sometimes be revealed by a complication (20% of cases). The hepatic hydatid cysts rupture mainly in the bile ducts, the gastrointestinal

tract, the bronchi, the peritoneal cavity and the pleura. Rupture in the bile ducts is the most frequent complication, it is encountered in 10 to 15% of cases [7,8]. Above all, it complicates the thick, calcified peripheral cysts of the right liver. Two mechanisms (inflammatory and mechanical) seem to be involved in the genesis of fistulas [9]. In our observation the fistulized cyst was posterior exohepatic with a thick pericyst and inflammatory in appearance, to which is added the mechanical effect of the cyclic respiratory movements, causing in the long term the progressive erosion of the cyst as well as the fibrosis of the neighboring tissues. Three stages have been identified for their genesis [10]: Stage I: Involvement of the innermost muscular layer of the abdominal or thoracic wall; Stage II: Damage beyond the muscle layer, protruding into the subcutaneous soft tissue; Stage III: Passage of lesions in the subcutaneous tissue and their fistulization in the skin

The lesions are most often asymptomatic, however clinical symptoms may be observed. Often variable, it depends on the location of the cyst, its size, its stage of development and the presence or absence of complications. Abdominal pain remains the most frequent clinical symptom [11]. The difficulty of diagnosis and the severe course of hydatid disease make the hydatid liver cyst complicated by cystocutaneous fistula of particular clinical interest. Indeed the rarity of fistulization of KHF can lead to a wrong diagnosis, hence the interest to think about it especially in endemic areas. The combination of clinical and radiological data and serological tests facilitate diagnosis. CT allows a complete description of the cyst and its fistulous course [12]. Ultrasound, chest scanner, MRI and fistulography [13,14,15,16,17,18] are the most commonly used radiological means for diagnosis. Endoscopic retrograde cholangiopancreatography (ERCP), percutaneous transhepatic cholangiography (PTC) and endosonography are invasive radiological means that can be used for diagnostic and therapeutic purposes. Enhanced contrast fistulography appears to be the most useful means for skin complications of hydatid disease. It specifies the extension of the fistula, the location, the size of the fistulized lesion as well as its relationship with the bile ducts.

Histopathological examination of the liquid drained by the external fistulous orifice, in certain cases makes it possible to highlight the parasite and thus make the diagnosis. Positive hydatid serology brings an additional argument in doubtful cases to imaging. Nevertheless certain abscesses of the liver can also fistulise on the skin even if this remains an exceptional phenomenon. In our patient, the diagnosis was mainly made on the radiographic results.

Surgical treatment remains clearly indicated in complicated forms of KHF. It consists of elective surgery for parietal complications, preceded by neo-adjuvant treatment with benzimidazole for 2 to 4 weeks. In stage I and II wall complications, surgical treatment consists of the total resection of the protruding hydatid cysts. In stage III wall complications, cystic block resection must be supplemented by excision of the fistulous tract to the surrounding skin. [19,20]. In our observation, the patient presented with a cystocutaneous fistula (Stage 3), he benefited from neo-adjuvant treatment based on Albendazole followed by conventional conservative surgery of KHF, supplemented by total resection of the fistulous path associated with treatment postoperative medical. Major radical surgery, such as segmentectomy and hepatic lobectomy, are less recurrent treatment options, although they are associated with increased morbidity compared to approaches conservative surgery. Adjuvant therapy is indicated for complicated cases treated with conservative surgery such as partial cystectomy. The cystic cavity must be drained and reduced in size. Benzimidazole treatment should be administered for 4 to 12 weeks

### III. Conclusion

Spontaneous cystocutaneous fistulization of the hydatid cyst is an exceptional complication of hydatid disease. Sometimes difficult to diagnose in the absence of drainage of hydatid material from the external fistulous orifice, it must be taken into account in the differential diagnoses of patients with cutaneous fistula, in particular in areas endemic for hydatid disease. Imagery is of great help in solving these diagnostic problems. Surgical treatment essentially aims to treat not only the hydatid cyst of the liver but also it aims at the cystoparietal disconnection. Anthelmintic-based chemotherapy remains a good alternative for reducing water recurrences

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