Case Report

Primary peritoneal hydatid cyst with gastric fistula complicated by spontaneous pleural and bronchial fistula: report of a rare observation✩✩

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A B S T R A C T

Hydatid cyst is a parasitic infection widespread in Morocco. Hydatid disease mainly affects the lungs and liver. Primary peritoneal hydatidosis has a stealth clinical evolution until it reaches complicated stages. Complications may include mass effect, rupture, allergic reactions and secondary infection. We report a very rare case of an isolated primary peritoneal hydatid cyst, first complicated by a gastric fistula and secondarily by a spontaneous pleural and bronchial fistula in a patient, who presented with isolated upper abdominal pain.

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Introduction

Hydatid disease is a zoonotic disease caused by the echinococcus parasite, which belongs to the family of Taeniidae. Although the liver and lungs are the most commonly affected organs, peritoneal echinococcosis, whether primary or secondary, is an unlikely but potential manifestation of the disease. Hydatid cysts can cause a variety of symptoms may that be by direct mass effect, or complications due to cyst rupture or secondary infection [1]. We report a very unusual case of primary peritoneal hydatid cyst without the involvement of other organs, complicated with gastric fistula and peribronchial fistula in a patient presenting an acute upper abdominal pain.

Case report

We report the case of a 71-year-old male patient, without significant history, who presented with an upper abdominal pain, fever and vomit. The initial clinical examination showed a soft epigastric mass with moderate tenderness.

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A laboratory analysis was performed showing marked anemia with a hemoglobin: 6.2 g/dl (normal value: 14-16 g/dl); hyperleukocytosis at 13000μl (normal value: 4500-11000 μl) and elevated CRP levels at 42 mg/l (normal value inferior at 5 mg/l).

The thoracic and abdominal computed tomography (Fig. 1) showed a cystic lesion of the left hypochondrium with thickened wall and focal peripheral calcification as well as air bubbles. A fistulous tract was also revealed connecting the cyst to the stomach, without any other organ involvement. Conclusive diagnosis was peritoneal hydatid cyst complicated with gastric fistula.

Serologic tests for hydatidosis were positive. Upper gastrointestinal endoscopy showed an extrinsic compression of the major gastric curvature with individualization of a millimetric fistulous path with pus outflow.

Latter on the patient developed dyspnea. The control CT scan revealed a pleural and bronchial fistula of the peritoneal hydatid cyst (Fig. 2).

The final diagnosis was primary peritoneal hydatid cyst with gastric fistula secondarily complicated by pleural and bronchial fistula.

The patient was preoperatively prescribed albendazole 400 mg twice a day for 3 weeks, followed by surgery, with simple postoperative course.

Discussion

Hydatid disease is a zoonotic disease caused by the parasite echinococcus, which belongs to the family Taeniidae. The most common species of echinococcus that cause hydatid disease in humans are Echinococcus granulosus (cystic echinococcosis) and Echinococcus multilocularis (alveolar echinococcosis). Human transmission occurs through ingestion of infected food or water, or through direct contact with animal hosts. Dogs and sheep are the most common primary and intermediate hosts respectively. 95% of all hydatid disorders are caused by cystic echinococcosis, commonly known as hydatid cysts [1]. Hydatidiasis can affect any organ system or anatomical part of the human body, the liver (50%-77%) and lungs (15%-47%) are the most frequently affected organs [2]. Peritoneal hydatidosis is often secondary to liver or spleen involvement of the disease [3-4]. Primary peritoneal hydatid cyst accounts for approximately 2% of reported cases [3-5].

The natural physical barriers to hematogenous diffusion of cysts formed by the liver and lungs may account for the low incidence of primary peritoneal infection [6]. Various pathways have been suggested in the pathogenesis of peritoneal localization of the cysts. Up to 15% of parasites escape from being
filtered in the liver and lungs and enter the systemic circulation to implant in various sites [6,7].

The clinical course of peritoneal hydatid cyst is frequently non-specific, and depends on the location, cyst size, and the effect of larger cysts on adjacent structures [1,2]. It is, therefore, not uncommon to encounter the presence of an inert mass without any symptoms or deterioration in the patient's condition [5,8]. In a similar case, a painless multicystic mass was discovered in our patient, who had no major complaints other than abdominal pain.

Spontaneous rupture of an abdominal hydatid cyst is a rare complication that typically affects younger patients unlike our case. The rupture usually occurs into the biliary ducts, pancreatic duct, or the pleural and peritoneal cavities. Rupture into the hollow viscera is a very uncommon clinical entity. There are only 3 abdominal hydatid cases complicated with gastric fistula [4,6,9]; our patient seems to be the forth. This uncommon complication is due to the mass effect and high cyst pressure that results in the ulceration and erosion of the gastric wall leading to the fistula.

On imaging, they appear similar to hydatid cyst at any other location [10,11]. Preoperative diagnosis of gastric fistula in hydatid disease is usually revealed by dramatic clinical signs such as hydatiemesis and hydatidorrea. Gastrointestinal Gastrografin studies, ultrasonography, and computed tomography are helpful tools that may show the site of communication with air fluid levels containing floating hydatid membranes [4,6,12]. Sequential computed tomographic scans can reveal partial cyst drainage of hollow viscous fistulas. However the narrow diameter of the fistula or the obstruction with hydatid material may obscure the diagnosis [4].

According to some reports, serology may not always be beneficial in diagnosing primary and unconventional hydatidosis (other than liver and lung) [13,14], nevertheless, this was not the case in our report. As in other reported cases, imaging evaluation including US and CT were very useful in diagnosis and treatment follow-up.

All of these investigations are useful resources that allow us to navigate through the other differential diagnoses such as gastrointestinal duplication cyst, abdominal abscess, or a mesenteric cyst from echinococcosis [4].

According to the literature, medical treatment for certain types of hydatid cysts spans from aspiration, injection, and percutaneous re-aspiration to surgery, which is still the treatment of choice [2].

**Conclusion**

There are currently few reports of primary peritoneal hydatid cyst disease with no other organ involvement. It is an asymptomatic disease until serious complications occur. The diagnosis of gastric, pleural and bronchial fistula requires a precise assessment not only by imaging but also the fibroscopy allowing the accurate localization of the lesion before any further surgical management.

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**Fig. 2** – Contrast-enhanced computed tomographic scan of abdomen and chest in the mediastinal parenchyma window, in axial section (A, C), and coronal reformatted scan (B), showing the hydatid cystic peritoneal of the left hypochondrium (white arrow) with thickened wall and focal peripheral calcification (blue arrow) as well as air bubbles (red arrow), with individualization of trans diaphragmatic fistulous communicating with a pleural lesion (white star) containing a liquid level, that drains into a bronchus (yellow arrow), associated with pleural effusion (black arrow).
Author's contributions

All authors contributed to this work. All authors have read and approved the final version of the manuscript.

Patient consent

Written informed consent for publication was obtained from patient.

REFERENCES

[1] Eckert J, Deplazes P. Biological, epidemiological, and clinical aspects of echinococcosis, a zoonosis of increasing concern. Clin Microbiol Rev 2004;17(1):107–35.
[2] Yuksel M, Demirpolat G, Sever A, et al. Hydatid disease involving some rare locations in the body: a pictorial essay. Korean J Radiol 2007;8:531–40.
[3] Tsaroucha AK, Polychronidis AC, Lyrantzopoulos N, et al. Hydatid disease of the abdomen and other locations. World J Surg 2005;29:1161–5.
[4] Selim YY, Huseyin B, Suleyman H. A rare complication of intraabdominal hydatid disease: gastric fistula and recurrent gastric bleeding. Am J of Surg 2010;200:e59–60.
[5] Pandya JS, Bhambare MR, Waghmare SB, Patel AR. Primary hydatid cyst of peritoneum presented as abdominal lump: a rare presentation. Clin Case Rep 2015;3(5):331–2.
[6] Jain R, Sawhney S, Berry M. Hydatid disease: CT demonstration and follow-up of a cystogastric fistula. AJR Am J Roentgenol 1992;158:212.
[7] Hegde N, Hiremath B. Primary peritoneal hydatidosis. BMJ Case Rep 2013;2013 bcr2013200435. doi: 10.1136/bcr-2013-200435.
[8] ALmasri LA. Rare isolated primary peritoneal hydatid cysts: a case report from Syria. Qatar Med J 2016:13. doi: 10.5339/qmj.2016.13.
[9] Placer C, Martin R, Sanchez E, Soleto E. Rupture of the abdominal hydatid cyst. Br J Surg 1988;75:157.
[10] Mehta P, Prakash M, Khandelwal N. Radiological manifestations of hydatid disease and its complications. Trop Parasitol 2016;6(2):103–112. doi: 10.4103/2229-5070.190812.
[11] Pedrosa I, Saiz A, Arrazola J, Ferreiros J, Pedrosa CS. Hydatid disease: radiologic and pathologic features and complications. Radiographics 2000;20;3:795–817.
[12] Turgut AT, Altun L, Topçu S, Küçükoğlu B, Altnok T, Kaptanoğlu E, et al. Unusual imaging characteristics of complicated hydatid disease. Euro J Radiol 2007;63(1):84–93.
[13] Erikci V, Högör M, Aksoy N. Primary abdominal wall hydatid cyst: a case report. Turk Pediatr 2014;56(2):183–5.
[14] Sadjjadi SM, Abidi H, Sarkari B, et al. Evaluation of enzyme-linked immunosorbent assay, utilizing native antigen B for serodiagnosis of human hydatidosis. Iran J Immunol 2007;4:167–72.