Case report

Hydatid disease of the liver with portal vein invasion and cavernous transformation: A case report and literature review

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\begin{abstract}
Introduction: Hydatid cyst disease is a zoonosis caused by the parasite Echinococcus Granulosus. It may infest any organ of the body, but it most frequently involves the liver and lungs. Portal vein involvement by hydatid cyst disease is extremely rare with only six cases published to our knowledge.

Case presentation: We present a 47-year-old male with abdominal pain. His laboratory tests were in normal ranges. Upper gastrointestinal endoscopy demonstrated esophageal and gastric fundal varices. The ultrasonography (US) and computed tomography (CT) findings hydatid disease of the liver with portal venous thrombosis and cavernous transformation. It had been treated with albendazole.

Discussion: Hydatid cyst disease is a zoonosis caused by Echinococcus granulosus. It can infest various organs of the body with a particular predilection of the liver and lungs. It still continues to be a health problem in a significant part of the world. The symptoms are frequently non-specific and patients may even be asymptomatic. The portal vein thrombosis is usually accompanied with portal cavernous transformation. Patients with hydatid liver cysts and portal cavernous transformation require treatments for both hydatid cysts and portal hypertension.

Conclusion: Invasion of the portal vein and cavernous thrombosis by the echinococcus cysts is a very rare complication. Treatment should consist of both hydatid liver cyst and portal hypertension.

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\end{abstract}

Introduction

Hydatid cyst disease, a zoonosis caused by Echinococcus granulosus, still continues to be a health problem in a significant part of the world. It may infest any organ of the body, but it most frequently involves the liver (60–70\%) and lungs [2]. The symptoms are frequently non-specific and patients may even be asymptomatic. The most frequent complication is cysto-biliary communication.

Here, we aimed to present a very rare complication of a hydatid liver cyst Portal vein involvement by hydatid cyst disease and cavernous transformation.

Case report

A 47-year-old man was admitted to the hospital with abdominal pain. Patient had a hemoglobin of 13 mg/dl, CRP of 5 mg/L. Otherwise biochemical tests including renal and liver functions were normal. Abdominal ultrasonography (US) was ordered and it demonstrated multiple hydatid cysts of the right and left hydatid thrombosis of the portal vein.

Computed tomography (CT) clearly demonstrated a multiple hydatid cysts of the right and left liver of varying size and stages with communication between the multiloculated lesion and the portal vein and the multiple daughter vesicles obstructing the portal vein, portal cavernoma with signs of portal hypertension (Figs. 1 and 2, 3).

MRI has shown multiple cystic masses of variable size involving both lobes and of varying stages with hydatid thrombosis of the portal vein and its dividing branches portal cavernoma with signs of portal hypertension (Fig. 4).

Upper gastrointestinal endoscopy demonstrated the esophageal and gastric fundal varices.

He was discharged with anticoagulant therapy and albendazole therapy.

The patient is in follow-up.

Discussion

Hydatid cyst disease is a zoonosis caused by Echinococcus granulosus. It can infest various organs of the body with a particular predilection of the liver and lungs [1,4]. Parasite

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infestation starts through the intestines; initially duodenum, subsequently the portal venous system or lymphatic system, and ultimately the liver. Most larvae become trapped and encysted. Even though all echinococcus parasites pass through the portal vein, the portal vein invasion and obstruction is rarely encountered, likely due to the constant high pressure in the portal vein. The portal vein thrombosis is usually accompanied with portal cavernous transformation, which can be described as the neo-formation of venous collaterals around the occluded portal vein [1,2]. Cavernous transformation occurs in the process of time as a secondary portal venous formation to convey the mesenteric blood to the liver. These compensatory venous collaterals usually cannot completely resolve the portal hypertension symptoms and its complications [2,5]. Hydatid cysts may lead to potentially serious and life-threatening complications such as cyst rupture, wide spread infections, and perforations to the biliary tree [3,7].

Portal vein thrombosis and the cavernous transformation is a rare complication of hydatid liver cysts. There have been total six cases reported yet and all these hydatid cysts were located close to the hilum of the liver (Table 1) [5,8]. Previously reported cases were all from endemic areas of echinococcosis, such as Spain, Turkey, Greece and Chile. Four of all the cases were males (66%).

Patients with hydatid liver cysts and portal cavernous transformation require treatments for both hydatid cysts and portal hypertension. In the previously reported six cases, treatment of hydatid cysts was done with surgery only in two case. Another case was treated with endoscopic retrograde cholangiography. Three patients were treated with albendazole [2,4,13]. No treatment for portal hypertension was done in four cases and only one case was treated with propranolol and one case with surgery. Drugs decreasing the portal venous pressure, endoscopic band ligation, sclerotherapy for varices or percutaneous Trans jugular intrahepatic Porto systemic shunt (TIPS) procedures are efficient to treat the portal hypertension. However, surgical shunts are still one of the most effective ways to avoid the bleeding [1,2,12].

In conclusion, although rarely reported in the literature, vascular complications of hydatid cyst of the liver should not be overlooked. It requires multiple exams looking for predictive signs of vascular communication. The decision must be discussed in multidisciplinary staff to avoid intraoperative accident. Treatment
should consist of both hydatid liver cyst and portal hypertension [8,12,15].

**Consent for publication**

The patient gave written consent for their personal or clinical details along with any identifying images to be published. All participants gave written consent for their personal or clinical details along with any identifying images to be published in this study.

**Ethical approval**

The study was approved by Ethics Committee of Hospital Fattouma Bourguiba Monastir.

**CRediT authorship contribution statement**

Hiba Ben Hassine: Data curation, Writing - review & editing.

**Declaration of Competing Interest**

The authors declare that there is no conflict of interests regarding the publication of this paper.

**Table 1**

| Author                  | Year of the report | Country of the patient | Age of the patient | Gender of the patient | Treatment of hydatid cyst | Treatment of portal hypertension | Author                  | Year of the report | Country of the patient | Age of the patient | Gender of the patient | Treatment of hydatid cyst | Treatment of portal hypertension |
|-------------------------|--------------------|------------------------|--------------------|-----------------------|---------------------------|-------------------------------|-------------------------|--------------------|------------------------|---------------------|-----------------------|---------------------------|-------------------------------|
| Gil Egea et al.         | 1998               | Spain                  | 84                | F                    | Albenzazole               | None                          | Kayacetin et al.        | 2004               | Turkey                 | 63                  | M                    | Surgery                  | None                         |
| Kayacetin et al.        | 2006               | Turkey                 | 74                | F                    | Surgery                  | None                          | Spanou et al.           | 2006               | Greece                 | 62                  | F                    | Albenzazole               | None                         |
| Spanou et al.           | 2006               | Greece                 | 74                | M                    | ERCP                      | None                          | Moisan et al.           | 2012               | Chile                  | 62                  | M                    | Surgery                  | Propranolol                  |
| Moisan et al.           | 2012               | Chile                  | 74                | M                    | ERCP                      | None                          | Kirmizi et al.          | 2016               | Turkey                 | 33                  | M                    | Surgery                  | Albenzazole                  |
| Kirmizi et al.          | 2016               | Turkey                 | 33                | M                    | ERCP                      | None                          | G. Ertan et al.          | 2019               | Turkey                 | 77                  | M                    | Surgery                  | None                         |

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