

Case Report

First Case of Hepatic Polycystic Echinococcosis Involving the Gallbladder in Acre State, Brazil

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Abstract

Echinococcosis or hydatid disease is one of the most life-threatening zoonotic parasitic diseases worldwide. *Echinococcus vogeli*, the causative agent of human and animal polycystic echinococcosis, is distributed in Central and South American countries. Although hydatid cysts are mostly seen in the liver and the lung, they can be located in various other tissues. In the following report we present detailed clinical and pathological findings of the first case of hepatic polycystic echinococcosis involving the gallbladder.

ABBREVIATIONS

PE: Polycystic Echinococcosis; CE: Cystic Echinococcosis; US: Ultrasound; CT: Computed Tomography

INTRODUCTION

Echinococcosis or hydatid disease is a life-threatening disease caused by the larval stage (metacestode) of tapeworms, affecting livestock, wildlife and humans worldwide [1]. In South America, *Echinococcus granulosus* sensu lato (s.l.) and *E. vogeli* are causative agents of cystic echinococcosis (CE) and polycystic echinococcosis (PE), respectively [2-4]. In Brazil, *E. vogeli* is distributed in the northern region, where the life-cycle is based on a predator-prey relationship between bush dogs and lowland pacas (*Cuniculus paca*) [5].

It is likely that humans become infected with *E. vogeli* eggs released into the backyards by domestic dogs (*Canis familiaris*), after being fed with offal from infected pacas [5]. To date, more than 200 cases have been reported, evidencing that human PE is no longer a medical rarity [6]. Data from pathology and surgery departments of public hospitals show that metacestodes are most commonly found in liver and lungs [7]. Other sites such as pancreas, omentum, peritoneum, mesentery and abdominal wall have been reported [5-8].

PE is characterized by an initial asymptomatic incubation period and a chronic course, often with the onset of clinical symptoms [7]. Abdominal pain is the main clinical presentation

and physical examination may reveal a palpable abdominal mass and right upper quadrant pain [5]. The first case of hepatic and secondary gallbladder polycystic echinococcosis is reported here.

CASE PRESENTATION

Written informed consent was obtained from the patient. A 50-year-old woman was admitted to a tertiary teaching hospital complaining of a five-year history of right upper quadrant abdominal pain. She also complained of nausea, vomiting and fever. Her epidemiological history she had previously resided in Tarauacá municipality (state of Acre), where she engaged in hunting for game and owned a dog. Her clinical history revealed diffuse and intermittent abdominal pain with no radiation. The liver was palpable at 10 cm from the costal right edge with smooth and painless surface. Her past medical history was significant for HBV virus coinfection. The remaining medical history was unremarkable. By ultrasound (US), cystic lesions were seen and a computed tomography (CT) scan showed calcified cysts in segment V of the liver, also involving the gallbladder (Figure 1). A presumptive diagnosis of PE was made, and albendazole (10mg/Kg/day) was prescribed.

An extended cholecystectomy was performed, covering the edge of segment V, resecting the cysts completely (Figure 2). The right hepatic artery was enveloped by cysts in the retro hilar position. During dissection of the cysts attached to the hepatic artery, one of them was opened and its contents were emptied and treated with 10% hypertonic saline and then

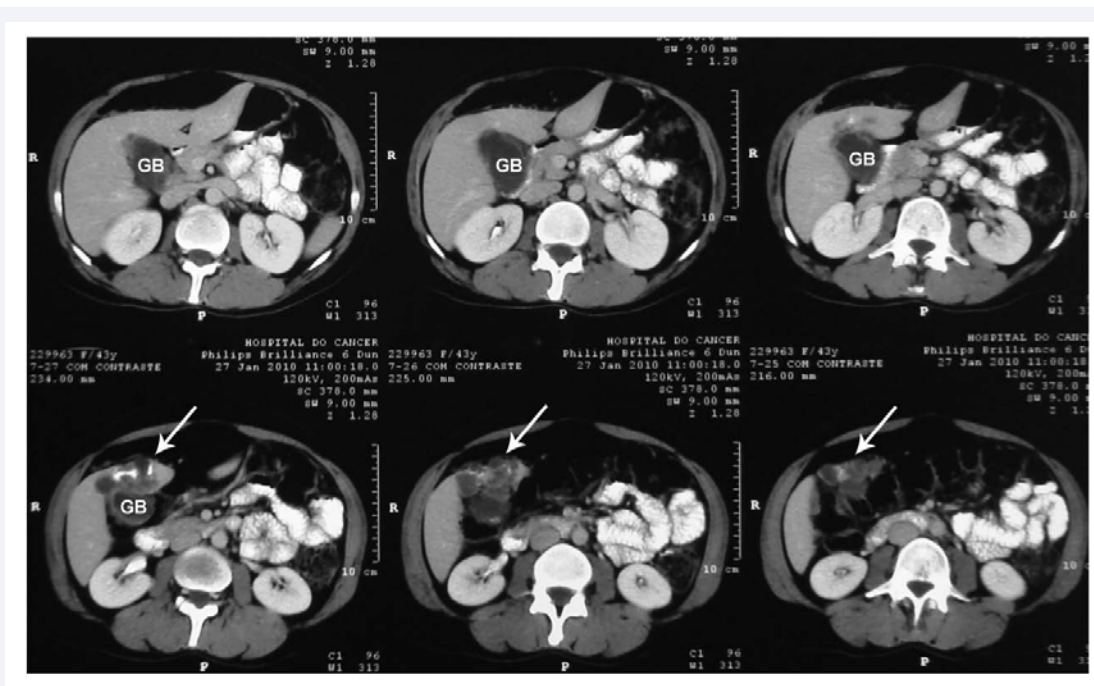


Figure 1 Computed tomography images of the liver showing the gallbladder (GB) with calcified cystic lesions (arrows).

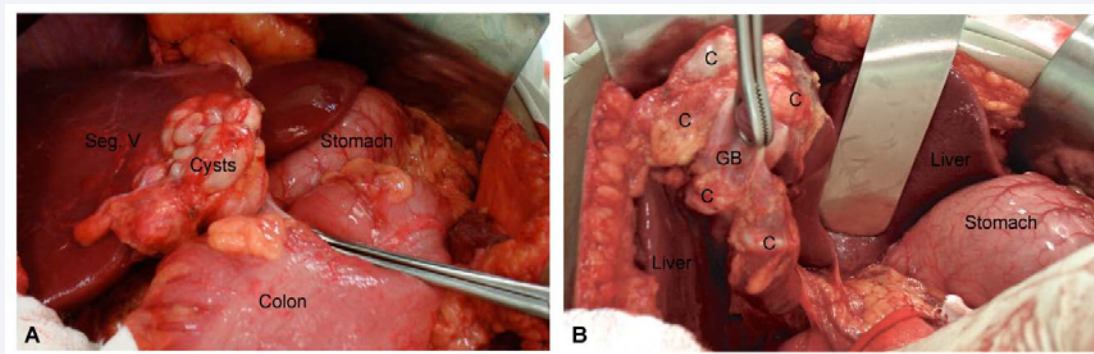


Figure 2 Intraoperative findings showing liver and gallbladder cystic lesions. (A) Cysts on the edge of the V segment of the liver, involving the gallbladder; (B) Gallbladder (GB) pulled anteriorly subsumed by various cysts (C).

70% ethanol. The cavity was protected with compress soaked in 10% hypertonic saline. The patient's postoperative course was uneventful. No recurrence was observed in CT scan at the thirteen-month follow-up.

Cyst histopathology was examined by light microscopy (Figure 3). Stained sections revealed parasitic structures composed of a host-origin layer (adventitia), an outer laminated layer, an inner germinative layer (Figure 3A), invaginated protoscolexes with different densities of calcareous corpuscles (Figure 3B) and free hooklets [$41.2 \pm 2.3 \times 13.0 \pm 0.9 \mu\text{m}$ (large hooks) and $32.4 \pm 3.1 \times 10.6 \pm 1.3 \mu\text{m}$ (small hooks)], consistent with PE (Figure 3C). There were many fibrous collagens in the laminated layer and a small amount of leucocyte infiltrate, including eosinophils and neutrophils. The presence of bile inside the cyst was also noted (Figure 3D).

DISCUSSION

It is a consensus that the clinical history and epidemiological information (geographical origin of the patient, paca hunting, dog ownership, feeding raw paca viscera to dog and close contact with these dogs) are important to achieve correct diagnosis of PE [5,7]. It is well known that *Echinococcus* sp. metacestodes determine significant morbidity [5,7,9]. In the present case, the clinical history and physical examination revealed diffuse abdominal pain and hepatomegaly, respectively, which are the mostly frequent clinical symptoms [7].

The involvement of liver and mesentery [5] and exclusively the mesentery [10] are the most reported PE clinical presentation types. Our patient had liver and gallbladder involvement, as previously reported in cystic echinococcosis [11-15].

Imaging diagnostic tools such as ultrasound and computed

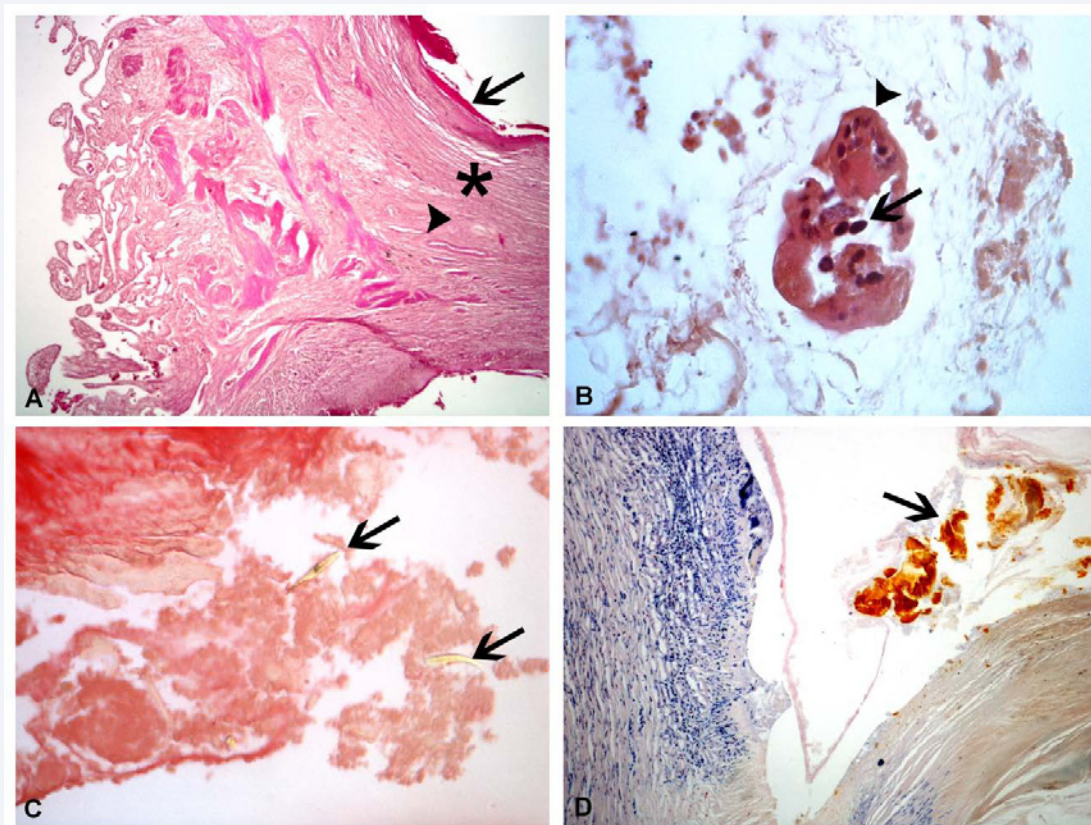


Figure 3 Representative images of polycystic echinococcosis in gallbladder tissue. (A) Germinal layer (arrow), a laminated layer (♣) and an adventitial layer (arrowhead), H&E stain (40x); (B) Invaginated protoscoleces (arrowhead) with different densities of calcareous corpuscles (arrow), H&E stain (400x); (C) Free hooklets in hydatid sand (arrows), H&E stain (400x); (D) - Bile inside the cyst (arrow), Lennert's Giemsa stain (100x).

tomography are very helpful in the diagnosis of hydatid cysts [14,16-18]. However for unusual localizations, ultrasound may be useful but with a very lower sensibility rate. In such cases computed tomography scan is often required [19].

PE is a life-threatening disease with high morbidity but low lethality [7]. Therefore, surgery combined with chemotherapy can improve the quality of life of patients with polycystic echinococcosis [8]. In this study, the exploratory laparotomy not only revealed the sites of the cysts in hepatic segment V, but also showed they were also attached to the right hepatic artery. The histopathological examination of the surgical material revealed cysts composed of a host-origin layer, an outer laminated layer, an inner germinative layer, invaginated protoscoleces with different densities of calcareous corpuscles, hooks and fibrous collagens in the laminated layer, a small amount of leucocyte infiltrate, including eosinophils and neutrophils. These data are in agreement with our previous study, where the infection had a common location [20], and other reported cases of gallbladder involvement in cystic echinococcosis [14,16-18]. The presence of bile inside the cyst was found in this study and in cases of cystic echinococcosis in Greece [21].

Although Brazil is an endemic area for cystic and polycystic echinococcosis, they do not overlap. *E. granulosus* (s.l.), the causative agent of cystic echinococcosis, is distributed in areas of

extensive cattle and sheep farming in southern states bordering Argentina and Uruguay [22]. In turn, PE cases have been reported in several municipalities (Sena Madureira, Assis Brasil, Feijó, Plácido de Castro, Brasiléia and Tarauacá) in Acre state, northern Brazil [7,10,22].

Although hydatid cysts are mostly seen in the liver and lung, they can be located in various other tissues [23]. In conclusion, we present the first case of a rare location of polycystic echinococcosis in Brazil. Even though considered a rarity, hydatid cysts should be included as diagnostic differential in cases with liver and gallbladder involvement, especially in patients who have spent time in endemic areas.

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