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Intracystic bleeding of a solitary hydatid cyst: A rare complication of a rare disease in central Africa. A case report

Ankouane Andoulo Firmin[1*], Ngo Nonga Bernadette[2], Monabang Catherine[3], Djapa Roger[1], Tchoumi Eric[1], Njoya Oudou[1], Ndjitoyap Ndam Elie Claude[1], D. P. McManus[4], Malonga Eimo[2]

[1]Department of Internal Medicine, Faculty of Medicine and Biomdical Sciences, University of Yaounde I, Yaounde, Cameroon;
[*]Corresponding Author: ankouaneandoulo@yahoo.com
[2]Department of Surgery, Faculty of Medicine and Biomdical Sciences, University of Yaounde I, Yaounde, Cameroon
[3]Department of Radiology, Faculty of Medicine and Biomdical Sciences, University of Yaounde I, Yaounde, Cameroon
[4]Donald Peter McManus Infectious Disease Division, Queensland Institute of Medical Research, Brisbane, Australia

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ABSTRACT

Splenic cysts are rare lesions and most of them are hydatid in origin. Hydatid disease is very rare in central Africa although it is cosmopolitan in North Africa. We are presenting a case of intracystic bleeding complicated with shock in a rural based Cameroonian and owner of hunting dogs treated in our university teaching hospital.

Keywords: Hydatid Disease; Cyst; Bleeding; Splenectomy

1. INTRODUCTION

Splenic cysts are rare lesions. Though most often asymptomatic, they can, however, present with pain in the left hypochodrium, which generally herald a complication. In 60% - 70% of cases, splenic cysts are hydatid in origin. Hydatid disease is an anthropozoonosis caused by the dog tapworm *Echinococcus granulosus*. It is cosmopolitan, rampant in the Mediterranean basin of North Africa, in Latin America and in Eurasia [1]. Kenya is the main source of human hydatid disease (cystic echinococcosis) in SubSaharan Africa [1,2]. Few cases have been reported in Central Africa. Cases of spontaneous or post traumatic intraperitoneal rupture of hydatid cysts have been reported. We are presenting the case history of a 40 years old Cameroonian living in a rural area and owner of hunting dogs. He was admitted to the casualty unit of the university teaching hospital, Yaounde, complaining of acute abdominal pain following physical stress complicated by a hypovolemic shock secondary to intraperitoneal bleeding caused by a splenic hydatid cyst. Total splenectomy was performed and post surgery monitoring was uneventful.

2. CASE HISTORY

A 40 years old Cameroonian man, living in a rural area and owner of many hunting dogs was admitted to the Yaounde University Teaching Hospital for acute abdominal pain which had been evolving for over 7 days following physical activity and accompanied by vomiting, fever and general body weakness. His past medical history indicated the presence of a splenic cyst diagnosed a year earlier (**Figure 1**), for which he was given medical treatment (which unfortunately was not documented); hydatid serology at the time was positive.

Physical examination revealed pallor, weakness, fever, a blood pressure of 100/50 mmHg, a pulse at 98 p/min and a tender mass at the left hypochondrium (**Figure 2**). Rectal examination was unremarkable. A full blood count revealed severe anemia with 4 g/dl hemoglobin and 1500/mm^3 eosinophils but other features were normal. Abdominal ultrasound and CT revealed a large splenic cyst whose content was cloudy (**Figure 3**). There was no other associated visceral lesion on abdominal imaging, cardiac ultrasound or chest X-ray. On the basis of the acute abdominal pain, hemodynamic instability and severe anemia, a total splenectomy was carried out 48 hours later. He was given Albendazole (Zentel®) 400 mg twice daily from the day before surgery to day-15 after surgery. Post-operatory monitoring was uneventful. Histopathologic analysis of the cystic tissue revealed a univesicular cyst whose wall stained positive on PAS.

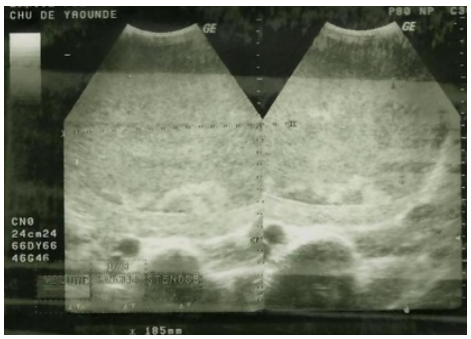


Figure 1. Abdominal US undertaken a year earlier showing the hyper echoic wall of a cyst whose content is hyper echoic.

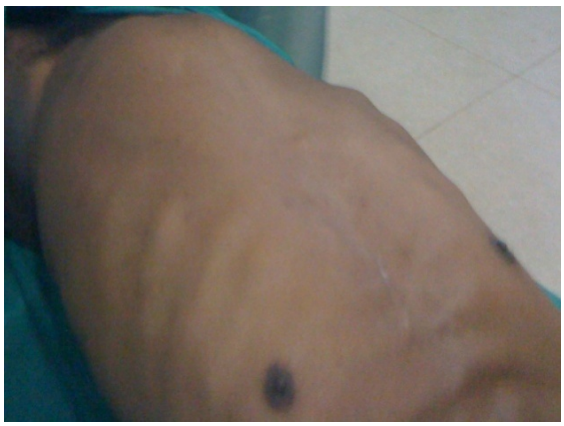
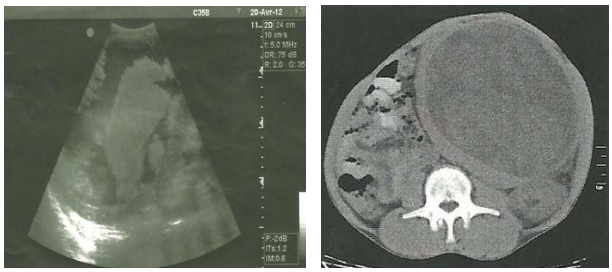


Figure 2. Image of a left hypochondriac swelling, with the patient lying on his back.



(a) (b)

Figure 3. Abdominal US (a) and a contrast-enhanced CT of the abdomen (b) showing a large splenic mass with well demarcated borders and a cloudy content.

Also present were *E. granulosus* hooklets and a granulomatous inflammatory reaction with epitheloid and giant cells.

3. DISCUSSION

Hydatid disease is a helminthiasis caused by the development in humans of the larval form of *E. granulosus*. In its classical life cycle, the domestic dog is the final or definitive host while domestic ungulates such as sheep are intermediate hosts. Man occasionally enters the cycle by ingesting eggs of *E. granulosus* [2]. The spleen is the most common site of infection following hepatic (50% - 70%) and pulmonary (25% - 40%) localizations [2]. It can either occur in isolation or in association with an extrasplenic localization, most often hepatic [2,3]. So far, to the best of our knowledge, intracystic bleeding has never been reported as a complication of hydatid disease. Spontaneous or post traumatic intraperitoneal rupture of the hydatid cyst generally characterizes the natural history of splenic hydatid cysts. Rupture into the peritoneal cavity is rare but remains a life threatening complication [4,5]. Small cysts can remain asymptomatic for several years. The clinical features of hydatid disease depend on the affected organ, the diameter of the cyst, its position, its effect on both the infected organ and on neighboring organs, and the eventual presence of complications such as rupture or infection [6]. Acute abdominal pain is the main clinical feature in case of complication [2,4]. Risk factors for rupture of hydatid cysts include trauma, the dimension of the cyst, a superficial localization and the patient's young age [6-8]. In addition to bleeding disorders, these same factors could equally account for intracystic hemorrhage, as is the case with physical stress, a feature of this patient.

Diagnosis is considered on the basis of clinical features, positive serology and imaging tests (particularly abdominal ultrasound and CT—with or without contrast enhancement) [9-12]. In the case of active bleeding, abdominal CT (without contrast enhancement) shows a hyperdense lesion within the cyst. Abdominal ultrasound coupled to a Doppler can rule out an aneurysm or pseudo aneurysm of the splenic artery [9]. Selective arteriography has both diagnostic and therapeutic value in case of massive bleeding. Pseudocysts which follow blunt splenic trauma or splenic infarction must be ruled out [13]. Diagnosis is confirmed by pathologic analysis [2].

Surgery remains the mainstay in the treatment of hydatid cysts of the spleen, especially in case of bleeding [2, 14], although it does not prevent recurrence [15]. It is best in case of large cysts, infection, localization in a vital organ, or in case of complication [6,15]. Total splenectomy is indicated in case of large cysts. It also has the advantage of preventing recurrences [2]. Other surgical

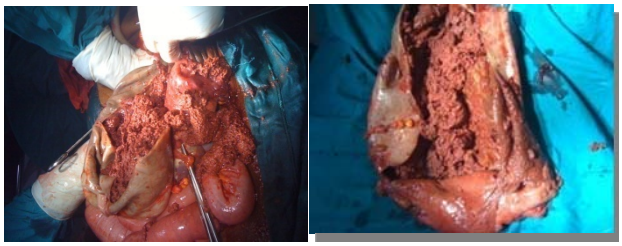


Figure 4. Image showing the hemorrhagic content of the splenic mass (after surgery).

options exist [6]. Percutaneous treatment (including puncture, aspiration, injection and reaspiration) has been practiced since the 1980s [6,15] but a potential complication is the occurrence of anaphylactic shock which can be minimized by administering the benzimidazole drugs, albendazole or mebendazole [16]. The use of this approach is greatly limited by the type of the presenting lesion, notably types II and IIIb multilocular cysts of the WHO-IWGE (Informal Working Group on Echinococcosis) classification (**Figure 5**) [17]. Deciding on which therapeutic option to use remains challenging [18]. In the absence of complications, the benzimidazoles remain a good alternative to invasive surgery and percutaneous treatment. Albendazole at 10 mg/kg twice daily (generally 400 mg) remains the preferred regimen (**Table 1**) [6,19].

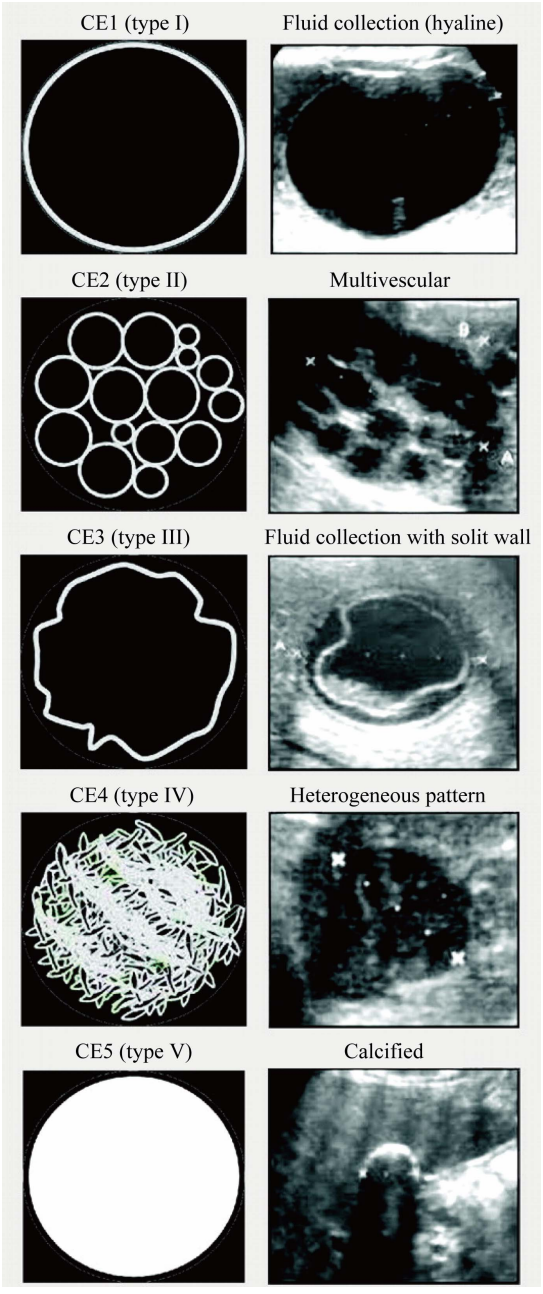


Figure 5. WHO-IWGE Ultrasound classification of CE cysts. With permission from Ref. [6].

Table 1. Suggested stage specific approach to treatment of uncomplicated cystic echinococcosis of the liver. Adapted from Brunetti and colleagues (Elsevier) (Ref. [6]).

WHO classification	Surgery	Percutaneous treatment	Drug treatment	Suggested treatment	Resource setting
CE1	No	Yes	Yes	<5 cm: albendazole	Optimal
				<5 cm: PAIR	Minimal
				>5 cm: PAIR + albendazole	Optimal
				>5 cm: PAIR	Minimal
CE2	Yes	Yes	Yes	Other PT + albendazole	Optimal
				Other PT	Minimal
CE3a	No	Yes	Yes	<5 cm: albendazole	Optimal
				<5 cm: PAIR	Minimal
				>5 cm: PAIR + albendazole	Optimal
				>5 cm: PAIR	Minimal
CE3b	Yes	Yes	Yes	Non-PAIR PT + albendazole	Optimal
				Non-PAIR PT	Minimal
CE4				Watch and wait	Optimal*
CE5				Watch and wait	Optimal*

*Minimal may not be applicable here because in low resourced remote endemic areas it may be impossible or too expensive to travel to the nearest hospital just to obtain a diagnosis. PAIR = puncture, aspiration, injection, reaspiration; PT = percutaneous treatment.

In conclusion, intracystic bleeding of a splenic hydatid cyst caused by *E. granulosus* is a rare clinical entity. It should be considered in the setting of an acute abdomen in every patient carrying a hydatid cyst, especially in highly echinococcosis-endemic zones.

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