

Severe anaphylactic choc revealing vascular rupture of a liver hydatid cyst observed in an emergency unit in Tunis (Tunisia)

Choc anaphylactique grave révélant une rupture intravasculaire d'un kyste hydatique du foie observé dans une unité d'urgence à Tunis (Tunisie)

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Abstract Anaphylactic shock is an exceptional mode of revelation of a liver hydatid cyst and it is in almost all cases secondary to an intraperitoneal rupture. The spread of hydatid cyst content into the bloodstream is even more exceptional. We report the case of a 36 years-old woman who presented a severe anaphylactic shock preceded by abdominal pain. Abdominal CT showed a liver hydatid cyst with a vascular communication. Operative findings confirmed the imaging data. The spread of hydatid cyst content into the bloodstream poses a double challenge as regards the management of the anaphylactic shock, and for the perioperative precautions.

Keywords Hydatid cyst · Anaphylaxis · Bloodstream · Hospital · Tunis · Tunisia · Maghreb · Northern Africa

Résumé Le choc anaphylactique est un mode de révélation exceptionnel d'un kyste hydatique du foie et il est presque dans la quasi-totalité des cas secondaire à une rupture intrapéritonéale. L'essaimage vasculaire du contenu hydatique est encore plus exceptionnel. Nous rapportons le cas d'une patiente de 36 ans qui s'est présentée en urgence dans un tableau de choc anaphylactique sévère précédé par des douleurs abdominales. La tomographie abdominale avait mis en évidence un kyste hydatique du foie avec une communication vasculaire. Les constatations opératoires avaient confirmé les données d'imagerie. L'essaimage du contenu du kyste hydatique dans la circulation sanguine pose un double défi thérapeutique, le premier est celui de la gestion du choc anaphylactique, le second est celui des précautions péri opératoires.

Mots clés Kyste hydatique · Anaphylaxie · Circulation sanguine · Hôpital · Tunis · Tunisie · Maghreb · Afrique du Nord

Introduction

Hydatid disease is a worldwide zoonosis and is localized in the liver in most cases. It is characterized by its large clinical polymorphism and the gravity of its complications which may be life threatening.

These complications can reveal the disease in 32 to 40% of cases [5,10] and are dominated by the rupture in the biliary tract. The rupture of hydatid cyst in the blood vessels is an extremely rare complication and leads to death by the spread of cyst content into the bloodstream which explains the frequency of its discovery on autopsy series [2,5].

In few survival cases, presentation can be a hydatid pulmonary embolism or an anaphylactic shock as in our case [3,7,9].

Case report

A 36 year-old woman presented to the emergency unit complaining of sudden onset of abdominal pain with pruritus, dyspnea, cough and diarrhea. The patient did not report any recent abdominal trauma.

Her past medical history included vague abdominal pain from right upper abdomen for nearly three months.

On physical examination, the patient was dyspneic with a wheezing, having periorbital edema and a diffuse maculopapular rash on the whole body, mainly on the face and the forearms. The patient presented a circulatory collapse, blood pressure was 50/30 mmHg, pulse was thready and heart rate was 150/min.

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Blood laboratory results showed leucocytosis at 19100/ml with an eosinophil count of 11% (0.5-10%) and a thrombopenia at 83 G/l.

The patient received oxygen and fluid resuscitation was started. Hypotension did not respond to fluids so, hydrocortisone sodium succinate (200 mg) and adrenaline were administered intravenously. Resuscitation maneuvers lasted several hours to counteract anaphylaxis.

Two days later, there was a significant improvement in symptoms.

Thoraco-abdominal computed tomography (CT) was performed showing a 65 x 58 x 40 mm unilocular hydatid cyst located in the segments VII and VIII of the liver. The wall of the cyst was thin and showed a discontinuity at a point, from which fluid leaked to outside in the liver parenchyma without an evident communication with a great hepatic vessel (Fig. 1). The capsule of the liver was intact.

The patient was operated. Abdominal exploration demonstrated a hydatid liver cyst in the segment VII. Pericyst was thin and intact.

Puncture and aspiration of the cyst fluid brought a bloody liquid. The cyst was opened; its content was haematic (Fig. 2A). Hydatid membranes of red blood color (Fig. 2B), were removed.

The inner surface of the cyst cavity was examined gently and during that meticulous examination a blood clot was discovered (Fig. 2C). It was removed and a wall defect was found through which we found a continuous bleeding from a vessel nearby the cyst wall (Fig. 2D). The wall defect corresponded to the ruptured area of the cyst viewed on the scan. This vessel was then ligated. The parasite was sterilized, partial cystectomy and omentopexy were performed. Post-

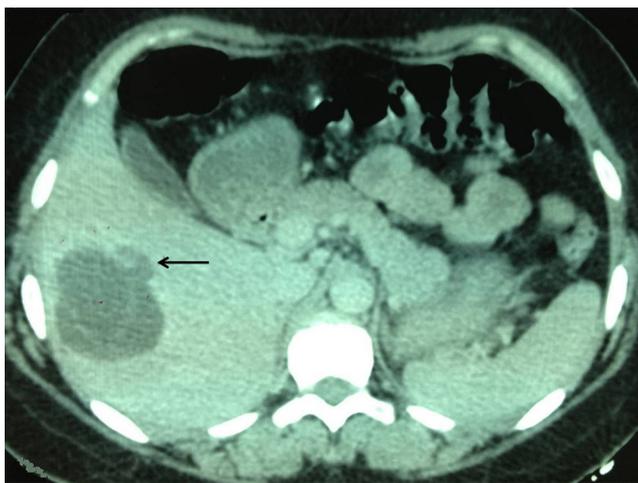


Fig. 1 On abdominal CT: Disruption of the cystic wall and a fluid leakage to outside in the liver parenchyma / *TDM abdominale : rupture de la paroi kystique et extravasation du contenu kystique dans le parenchyme hépatique*

operative course was uneventful. The patient was discharged four days later.

The serodiagnosis by enzyme-linked immunosorbent assay (ELISA) was done in post operative period; it was positive and remained positive 18 months after surgery. Albendazole treatment, 800 mg daily, was prescribed for six months, then was stopped because the patient was willing to pregnancy.

Discussion

Hepatic hydatid cyst results from the development in the liver parenchyma of a larva of *Echinococcus granulosus*. The cyst has three layers: the outer layer, called pericyst, is a fibrous tissue resulting from the inflammatory response of the host against the parasite; the middle laminated membrane is an acellular layer which allows the passage of nutrients, water and electrolytes from the host and the inner membrane, known as the endocyst, is a germinal layer that's the origin of the formation of the scolexes. These scolexes are the larval stage and infective form of the parasite. They are discharged in the cyst [6].

The content of the cyst is essentially water, scolexes and a few proteins. This content is strongly antigenic.

Spillage of cyst material into circulation occurs spontaneously or after trauma. It releases a high antigenic fluid in the circulation which causes IgE-mediated anaphylactic reactions [4,6].

On experimental studies, administration of this content to animals leads to systemic manifestations such as cardiopulmonary changes including a "triple response" of the systemic blood pressure ending in prolonged hypotension and pulmonary arterial hypertension.

An immediate, severe neutropenia is followed by a rebound neutrophilia and eosinophilia. Red cell volume and plasma protein concentration are increased, and coagulation time is prolonged. Pathological changes, confined to the lungs, consists of congestion, periarteriolar and peribronchiolar hemorrhage, edema, and sequestration of polymorphonuclear cells in the alveolar capillaries [7,9].

The mechanism of these reactions is complex. In some cases, it is typically a hypersensitivity reaction type I associated with immunoglobulin E in response to the high plasma concentration of *Ag. Echinococcus*. Anaphylactic reaction may also be secondary to complement activation with the release of anaphylatoxins. In vitro, the hydatid fluid activates alternative complement pathway with the formation of C3a [7,9].

In human practice, leakage of the cystic contents into the blood circulation is a very rare event because of the compliance of the vessel walls that are frequently laminated or completely occluded by the cystic mass effect. Local

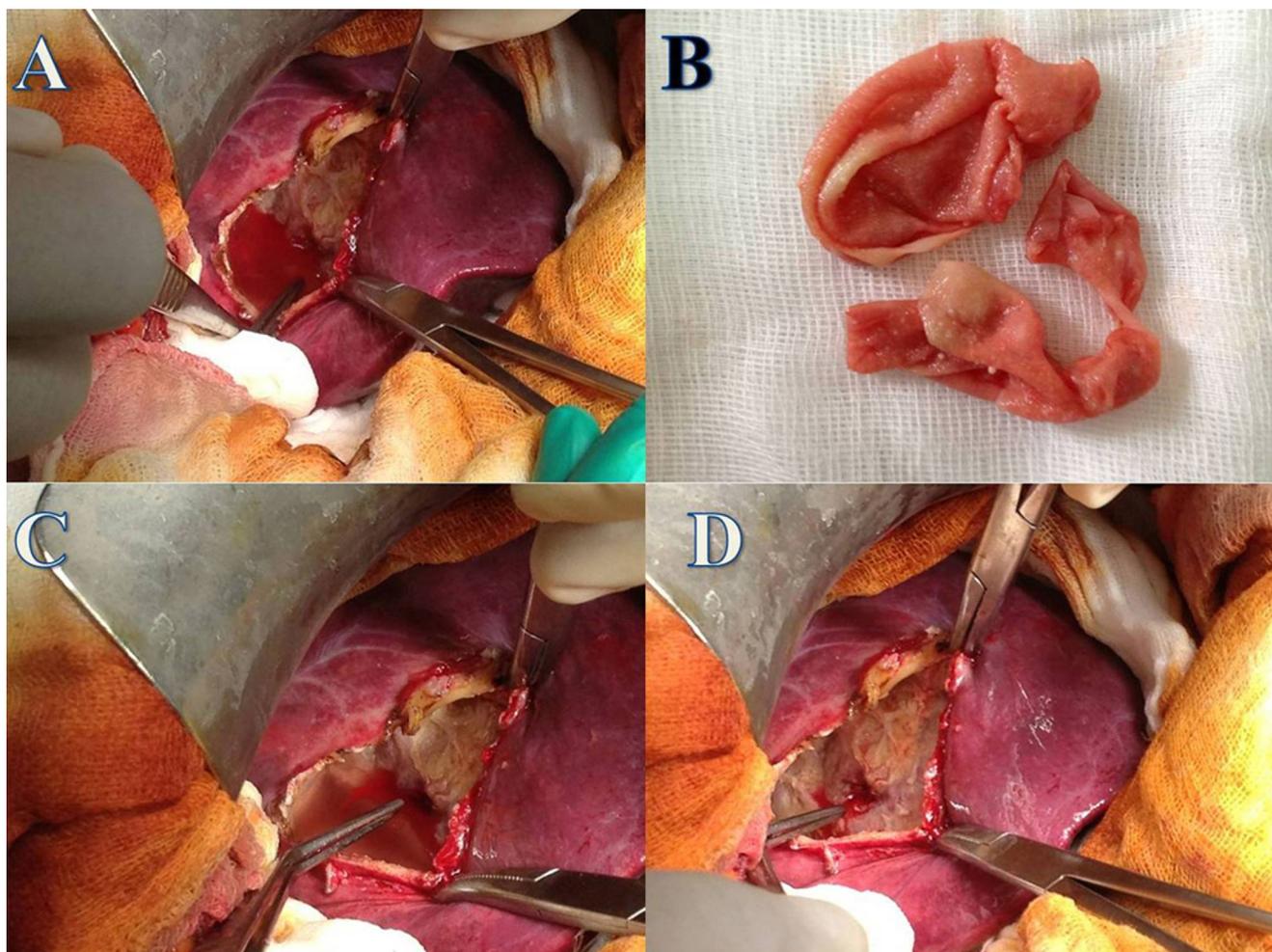


Fig. 2 A. Cystic content was haematic. B: Hydatid membranes have the color red blood. C: A meticulous examination of the inner layer discovered a blood clot. D: Removing of the clot reveals a wall defect through which a continuous bleeding was found / *A. Le contenu kystique était hématique. B: Les membranes hydatiques sont au couleur du sang. C: Un examen minutieux de la paroi kystique découvre un caillot de sang. D: Le retrait du caillot révèle une brèche au niveau de la paroi kystique qui est le siège d'un saignement actif*

thrombosis theoretically prevents hydatid intravascular spread. Second, this complication is exceptionally discovered in vivo since this incident causes the migration of daughter vesicles or hydatid membrane into bloodstream leading to lethal pulmonary embolism or to an anaphylactic shock also frequently fatal [1-3, 8,9]. Sometimes, allergic reaction is paucisymptomatic and often unreported.

This complication is habitually described as an intraoperatively surprise in totally asymptomatic patients. The discovery of the cysto-vascular communication is often made during the aspiration of cyst contents.

According to Zaouche's multicentric study about 2013 cases of hydatid cyst of the liver, only four cases (0.2%) of vascular rupture were found [10].

The review of these observations and other reports [2,7,10] reveals that the majority of these cysts sits on the hepatic dome at segments VII and VIII similar to our case or

on the retro-hepatic segment I. The main presentation is massive hydatid emboli. An anaphylactic shock is rarely described.

The treatment of the anaphylactic reaction has no particularities. However, some anesthetic and surgical precautions should be taken [4] to prevent a recurrent anaphylactic reaction.

Fluid resuscitation and prophylactic preoperative intravenous administration of HSHC will be administered during the intervention in the infusion intravenously since induction is required. These precautions have been taken into consideration in our patient.

Some authors advocated the combination of preoperative histamine H1 and H2 receptor blockers that has been shown to lessen the hemodynamic changes during the operation [4,8].

Adrenaline, which is the sole effective treatment of the cardiovascular collapse, should be nearby. Anesthesia of these patients must avoid Suxamethonium Chloride and morphine which may promote histamine release.

Some surgical precautions are mandatory like blood supply because blood transfusion may be necessary, anticipate a second vacuum if massive bleeding occurs and to get ready for vascular exclusion of the liver before the puncture of the cyst if the communication is large.

Conflict of interest : The authors declare not have any conflict of interest.

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