

CLINICAL CASE

HEPATIC HYDATID CYST RUPTURED INTRABILIARY IN A 28-YEAR-OLD HIV POSITIVE PATIENT**Dan Nicolae Păduraru^{1,2}, Georgiana Radu¹, Daniel Ion^{1,2}, Alina Tinca^{1,2}, Bianca Smarandache³, Erika Oprea², Gabriel Constantinescu^{1,4}, Octavian Andronic^{1,2}**¹“Carol Davila” University of Medicine and Pharmacy, Bucharest, Romania²Bucharest University Emergency Hospital, Bucharest, Romania³“Dr. Ion Cantacuzino” Clinical Hospital, Bucharest, Romania⁴Bucharest Emergency Hospital, RomaniaCorresponding author: Dan Nicolae Păduraru
E-mail: dan.paduraru.nicolae@gmail.com**Abstract**

Echinococcosis is an endemic disease in countries from the Mediterranean area, Eastern Europe, the Middle and the Far East, Australia, South America and Eastern Africa. However, it has a worldwide distribution because the dog, a definitive host, is a ubiquitous animal. Another important factor that led to a worldwide spread of the disease is population migration, as the number of cases in non-endemic regions has been increasing. We present the case of a 28-year-old HIV positive patient suffering from a liver hydatid cyst that had ruptured inside the biliary pathways. US and CT were the imaging tools used to establish a preoperative diagnosis and the main therapeutic approach was neutralizing the parasite using 90° alcohol, Lagot deroofting, contents' evacuation through open surgery and appropriate drainage afterward. ERCP was very useful in unblocking the main biliary pathways after the surgery as well as installing a stent to ensure its further permeability.

Keywords: *hydatid disease, hepatic hydatid cyst, complicated hydatid disease, ruptured hydatid cyst*

Introduction

Echinococcosis is an endemic disease in countries from the Mediterranean area, Eastern Europe, the Middle and the Far East, Australia, South America and Eastern Africa. However, it has a worldwide distribution because the dog, a definitive host, is a ubiquitous animal. Another important factor that led to a worldwide spread of the disease is population migration, as the number of cases in non-endemic regions has been increasing [1].

Biologically, *E. Granulosus* is a cestode species of the genus *Echinococcus* with only three stages of development. Its life cycle

involves two mammalian hosts. The adult cestode will inhabit the intestine of a carnivore (considered a definitive host) while producing eggs that contain infective oncospheres. Through the intestines of the definitive host, either cestode segments containing eggs or free eggs will be released into the environment. These eggs are found and ingested by an intermediate host animal. The eggs open inside the digestive tract of the intermediate host, thus releasing the metacestode, a larval stage. Typically, the mature metacestode can produce numerous protoscoleces. Each one of these protoscoleces has the potential to develop into an adult parasite

after being ingested again by another definitive host [2].

The hydatid disease is a potentially lethal zoonosis. In humans, only the following species can cause infection: *E. Granulosus*, *E. Multilocularis* and *E. Vogeli* [2]. Humans are intermediate hosts; thus, the occurrence of a hydatid cyst is accidental, through the ingestion of *E. Granulosum* eggs directly by contact with canines or ovines, or indirectly by food, water and contaminated objects or surfaces. The larvae develop inside the digestive tract of the patient, are absorbed into the portal vein and then travel all over the body [3]. This is why the liver is the most commonplace for hydatid cysts (more than 2/3 of all patients). Moreover, most patients have only one affected organ. Sometimes the cysts grow (with an average 1-30 mm per year) without any noticeable change throughout the years; other times, the cysts may completely disappear without external intervention. One of the most common problems caused by a hydatid cyst is compression, whether it is compression of surrounding hepatocytes and fibrosis or compression and displacement of biliary ducts. Spontaneous intrabiliary rupture may occur because of the high intracystic pressure. When a cyst ruptures inside the biliary tract, an overt passage of hydatid material takes place. This scenario was classified as frank perforation. Another scenario where only signs of suppuration are present and the leakage of hydatid material is occult may take place [1].

Classical therapeutic management of hepatic hydatid cysts consists of medical treatment, open surgery, endoscopic treatment and the newer minimally invasive methods [3].

Case presentation

A 28-year-old patient came to the Emergency Department with the following symptoms: high fever (41 degrees Celsius), diarrhea, jaundice and pain in the upper right quadrant for about 48 hours before the admission. He is HIV positive, has B and C hepatitis, has been a Heroin addict 10 years prior and has hyperthyroidism.

Clinical exam reveals a distended abdomen, painful both spontaneously and when palpated, more accentuated in the right upper quadrant. There were no signs of muscle defense.

Initial blood tests revealed high total bilirubin (13.62 mg/dL) and high direct bilirubin (10.30 mg/dL). No inflammatory syndrome was present (leucocytes count was 8.28 m/mm³).

The first imaging technique used in the management of this case was an abdominal ultrasound. The results were as follows: a moderate dilation of the biliary ducts in the left hepatic lobe, echodense material inside the main biliary duct and a hydatid cyst adjacent to the hilum and bed of the cholecyst.

Following the information acquired from the abdominal ultrasound, an abdominal CT scan was mandatory. In our patient's case, this investigation showed an expansive process resembling a young hydatid cyst with multiple daughter vesicles, measuring 87/79 mm in length and 89 mm in height, projected in the VIIIth and Vth hepatic segments. Moreover, it had projections throughout the hepatic hilum, underneath it, a wide contact with the cholecyst, a small contact with the duodenum on its second portion and was touching vascular hepatic and portal vessels posteriorly. The cyst's irregular contour, the important dilation of the intrahepatic biliary canals of the left hepatic and VIIIth segments, the dilation of the main biliary duct to approximately 20 mm as well as 2 punctiform pneumatoceles projected inside the left biliary duct raised the suspicion of a ruptured cyst (Figure 1).

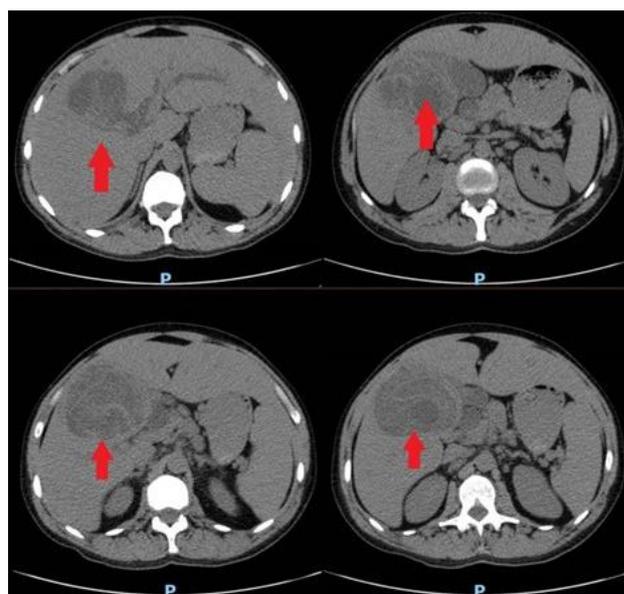


Figure 1 – Abdominal CT scan. Transversal plane. The red arrow points towards the hydatid cyst

The patient was scheduled for emergency surgery under general anesthesia. Intraoperative, a cystic formation measuring about 100/90 mm was found on the visceral side of the right hepatic lobe (Figure 2). In order to neutralize the contents of the cyst, we used 90 degrees alcohol. Afterward, a Lagrot unroofing was performed in order to evacuate the daughter vesicles as well as the prolifere membrane – approximately 500 mL of liquid in total. Also, a cholecystectomy was performed. To ensure the adequate drainage of bile, 2 drainage tubes were placed inside the remaining cavity alongside with a transcystic one.



Figure 2 – Intraoperative aspect showing the cyst after the cholecystectomy, before the drainage

Next, a cholangiogram was scheduled in order to see whether any remaining residue was still inside the biliary ducts. After 40 mL of contrast was introduced through the transcystic duct, we could see the following: a partial opacification of the left hepatic duct, a full opacification of the right hepatic duct and a weak opacification of the main hepatic duct before its union with the cystic duct. Also, some contrast went into the restant cavity. There was zero opacification of the choledochal duct (Figure 3).

As the cholangiogram showed intense blockage in the biliary ducts, an ERCP (Endoscopic Retrograde Cholangio-Pancreatography) was scheduled the next day. It showed multiple daughter vesicles and other residues inside the canals, blocking the passage of bile (Figure 4). After the biliary ducts were unplugged, a stent was inserted in order to keep a good flow of bile through them and to prevent another blockage.

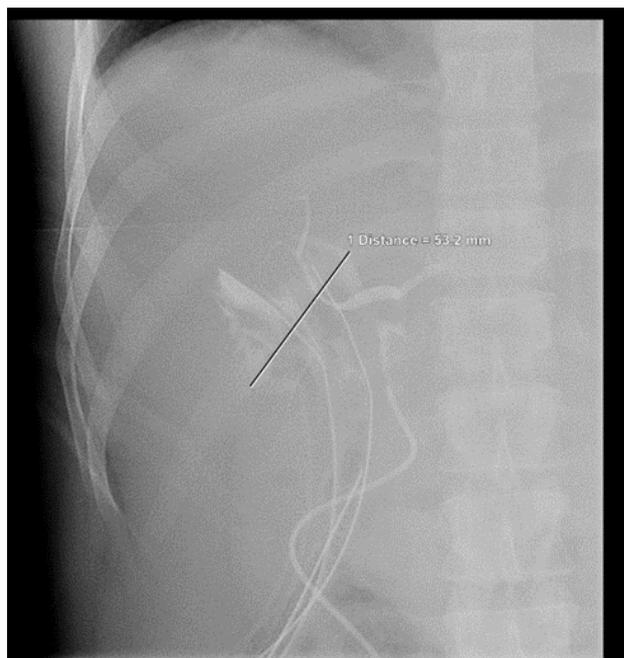


Figure 3 – First cholangiogram



Figure 4 – ERCP. Daughter vesicles and prolifere membrane in the biliary tract

A second cholangiogram was performed to ensure that the biliary tract was permeable. This time, contrast was able to pass inside the whole extrahepatic biliary tract (Figure 5).

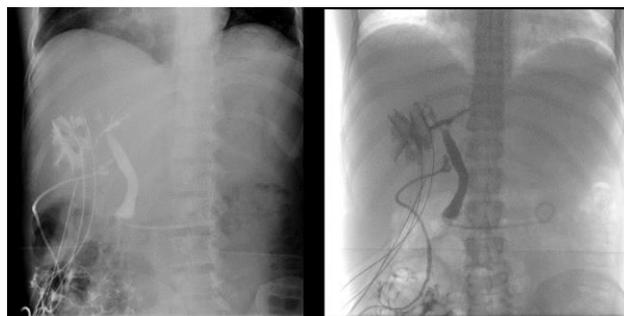


Figure 5 – Second cholangiogram showing permeable biliary ducts

The patient's evolution was favorable and the clinical symptoms disappeared progressively. The fever came to normal the following day after surgery and the jaundice disappeared completely 7 days after. Three days after surgery the total bilirubin had a value of 3.1 ml/dL and the direct

bilirubin had a value of 2.6 ml/dL. 2 days later they had normal values. The CT scan performed 6 months after the surgery, revealed the restoration of the hepatic tissue (Figure 6).

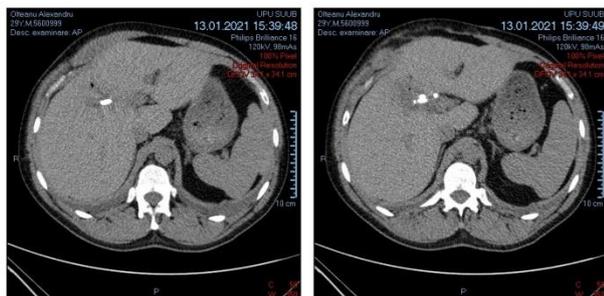


Figure 6 – Transverse section of the computed tomography performed 6 months after the surgery. Note the absence of the initial hepatic lesion and the restoration of the hepatic architecture.

Discussions and Conclusion

Hydatid disease is an endemic zoonosis in Eastern Europe, including Romania [4]. It is a difficult pathology to manage because of the various locations in which cysts may appear. It can cause general (anaphylactic shock), as well as local complications (depending on its location). The main affected site is the liver, and it is usually easy to diagnose at this level because it shows characteristic imaging findings. On the other hand, it does not produce any symptoms while the cyst is small, so the patient usually comes to the hospital only when complications arise. Some potential complications in liver hydatidosis are: intrahepatic complications, transdiaphragmatic thoracic and abdominal involvement, biliary communication, portal vein involvement, abdominal wall involvement, perforation into adjacent viscera, as well as exophytic growth. Moreover, secondary involvement may be seen in almost any anatomic location due to its hematogenous dissemination [5]. In our case, the cyst created a severe complication as it ruptured intrabiliary.

Ultrasonography (US) is very useful in the diagnosis of hydatid disease and particularly in cases involving the liver. It can easily detect cystic membranes, septa, hydatid sand as well as daughter vesicles. However, the cyst is best described using a Computed Tomography (CT) scan, as it can show cyst wall calcification, cyst infection, cyst wall defects and potential passage of contents through a defect, just like in our

patient's case. US is useful in demonstrating rupture not only in cases that involve wide communication, but also when it describes indirect signs of biliary communication such as increased echogenicity or biliary duct dilation. In the case of transdiaphragmatic disease, chest radiography, US, CT and MRI (Magnetic Resonance Imaging) are all very useful in depicting it. When peritoneal seeding may occur, CT is the most useful tool in assessing it [5].

A particularity of our case were the other infectious complications, as our patient was positive for HIV and B and C hepatitis viruses. In patients with HIV infection, various opportunistic infections have been reported. However, echinococcal cysts are not common in this situation, especially with liver involvement. The first available cases were published in 2012 by Amis Javed et al., who described 3 cases of liver hydatid cysts in HIV positive patients [6].

To conclude, we present the case of an HIV positive patient suffering from a liver hydatid cyst that had ruptured inside the biliary pathways. US and CT were the imaging tools used to establish a preoperative diagnosis and the main therapeutic approach was neutralizing the parasite using 90 degrees alcohol, Lagot deroofting, contents' evacuation through open surgery and appropriate drainage afterward. ERCP was very useful in unblocking the main biliary pathways after the surgery as well as installing a stent to ensure its further permeability. The patient made a full recovery.

References

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