

## DIAGNOSIS AND SURGICAL TREATMENT OF BILIARY-CYSTIC FISTULA CAUSED BY HEPATIC HYDATID CYST

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### Abstract

*Echinococcosis is a severe parasitic disease which is caused mostly by three species of Echinococcus (E. Granulosus, E. Multilocularis, E. Oligartus). The hydatid cysts are created in 50-80% cases in the liver (right lobe) and less in the lungs in only 10-40% cases. Patients with this disease may develop severe complications, one of the most common being the biliary-cystic fistula. Our interest focuses on two major questions; 1) if it is possible to detect those communications pre-operatively, 2) if we can predict the occurrence of fistulae post-operatively. Based on the Kjossev and Lasanoff classification, we first of all evaluate the cyst according to its topography, natural history, recurrence and possible complications. We also examine the risk factors such as age, sex, type and duration of symptoms, and alternations of the liver function test. Taking these parameters into consideration and also the predictors for existent communications, we continue with some investigations for the evidence of diagnosis. Following the above procedure, the diagnosis of biliary-cystic fistula resulted, so we continued with further investigations for the detection of the fistula type (true or occult) and the flow rhythm. For the successful resolution we chose a surgical treatment which could be performed laparoscopically or as an open surgery procedure. As a conclusion, we notice that complications can be pre-operatively and intra-operatively detected. Unfortunately there is no specific surgical treatment for the biliary-cystic fistula and for this reason we follow the conduction of a good drainage of the biliary tree and the principles. Finally as a means of prevention and a possible future recurrence of the cyst, postoperative radiological tests are recommended.*

**Keywords:** *Hepatic hydatid cyst, biliary-cystic fistula, diagnosis, surgical treatment*

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### Introduction

Echinococcus is a cestode parasite that can cause the hydatid infections [1].

The hydatid disease is caused by the ingestion of eggs of echinococcal species and especially of *Echinococcus granulosus*, with definitive hosts being sheep and dogs [2-5].

Rather for the *Echinococcus multilocularis* foxes are the most important definitive hosts [6-8]. Humans are infected accidentally by ingestion of food contaminated with eggs shed by dogs or foxes. These eggs pass through the duodenum and then invade the liver, lungs and other organs. The diagnosis of the disease is based on laboratory and imagistic investigations.

The most common biologic tests are the eosinophil count, the Imaz-Lorentz complement fixation test, Elisa test and Casoni's cutaneous test (old method) [9-11]. The imagistic investigations are in general: Ecography, CT scan (figure 1), MRI (figure 2), simple Radiography test, cholangiography and according to the location of the cyst some more specific tests can be performed [11-14].

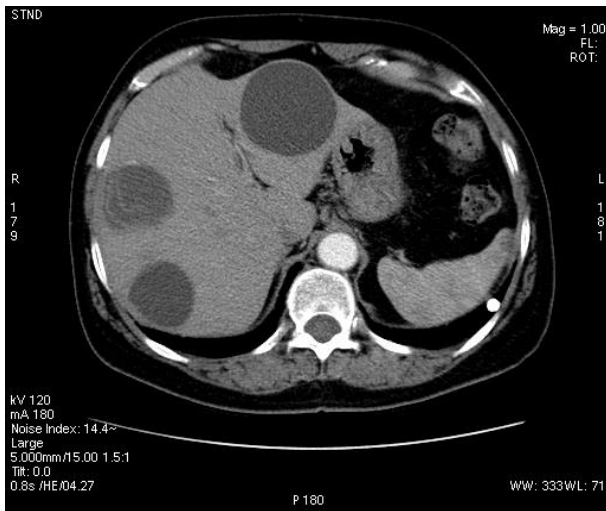


Figure 1 - CT scan showing the cyst

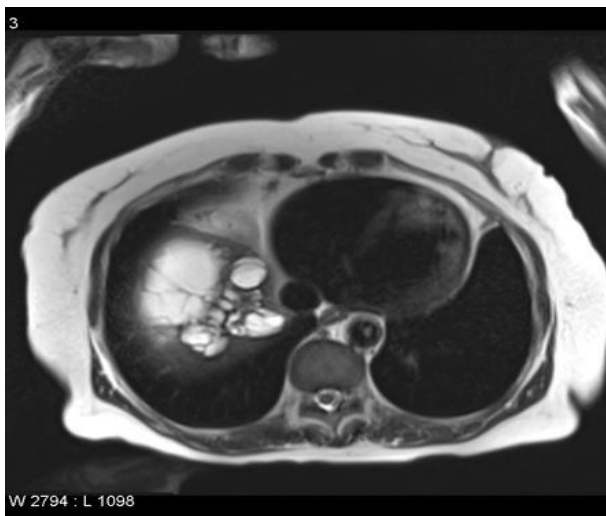


Figure 2 - MRI showing the cyst

**Material and Methods**

If the cyst is located in the liver, we have to evaluate the cyst, based on the Kjossev and Lasanoff classification that has 4 parameters: i) the topographic location, ii) the nature of the liver hydatid cyst, iii) the recurrence and iv) the complications.

i)The topography (T) can be described according to the coinauds segments that are 8 in

number. Additionally the exact pre-operative cyst location influences the laparotomy choice. (Figure 3)

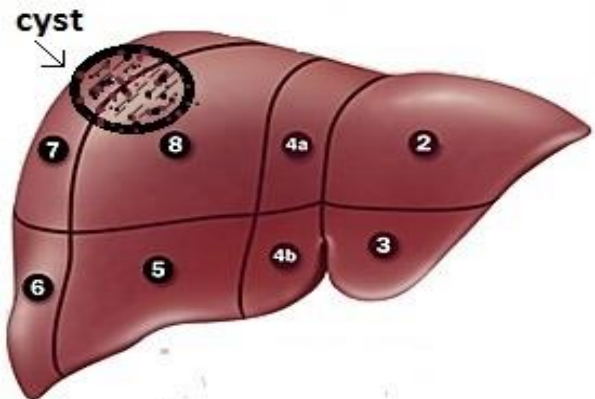


Figure 3 – Location of the cyst

ii) The second parameter is the Nature of the liver hydatid cyst that is symbolized with N and is numbered from N1 to N5. This describes the morphology and some changes of the cyst wall but also some developing structures. (Figure 4)

More details:

N1: simple cyst with no internal architecture except sand

N2: cyst with detached endocyst secondary to rupture

N3: cyst with daughter cyst or matrix

N4: densely calcified cysts, 30 % of liver hydatid cysts have calcifications of different magnitude

N5: residual cavity/ hepatic lesion after surgery including post-operative fistula

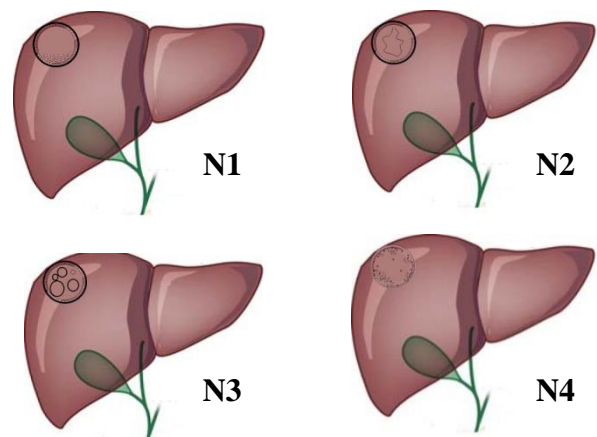


Figure 4 - Nature of the liver cyst

iii) The 3<sup>rd</sup> parameter is the recurrent cyst that has a rate of 8 to 15 % and can be univesicular or multivesicular, heterogenous and infected.

iv) Regarding the 4<sup>th</sup> parameter, the complications of the hepatic hydatid cyst, are symbolized with C and classified from 0 to 6.

More specific:

C0 - without complications

C1 – adhesions to the adjacent structures

C2 - compression of the neighboring structures

C3 - biliary cyst communication

C4 - infection of liver hydatid cysts

C5 - communicating rupture of liver hydatid cyst

C6 - direct rupture of live hydatid cyst

In our case, in the biliary-cystic fistula the C3 show the biliary cystic communication.

After making the complete evaluation of the hydatid cyst, we continue with some further investigations that are predictable for possible fistula. From liver function tests we observe that the patient pre-operatively has increased GGT (gama glutamyltransferase), ALP (alkaline phosphatase) as well as modifications of Bt (total bilirubin), Bd (direct bilirubin), GOT (glutamate oxalate transaminase) and GPT (glutamic pyruvate transaminase).

It is remarkable that the topography, the volume, the number of cysts and also infection and age increase the risk for fistula.

Afterwards, when we diagnosed the biliary cystic fistula, we had to evaluate the type and the flow rhythm, so we could continue with the treatment.

There are two types of fistulae: franc (true) or occult communications. In the case of franc communication the cyst is > 10 cm, multivesicular, solitary and is located in the left lobe. Patients with prolonged history have an increased risk. Instead in the case of occult or silent (as it is called), the diameter of the cyst is >14.5, and there is a presence of biliary leakage, the fistula is multilocular, degenerated and also presented as external biliary fistula being recognized during operation. Finally it can be detected from increased levels of: ALP, GGT, Bt, Bd, Eosinophils.

There are in addition, some more specific diagnostic recommendations for the cysto-biliary communications. Examples are:

▪ ERCP (Endoscopic Retrograde CholangioPancreatography) - an important investigation for diagnosis of major biliary

communication and clearance of common bile duct.

▪ DIRECT CHOLANGIOGRAPHY - an intra-operative cholangiography that is performed through a cystic drain or a T-tube in a suspect intrabiliary rupture.

▪ PTC (Percutaneous Transhepatic Cholangiography) - access to the biliary tree in cases where ERCP has been unsuccessful

▪ MRCP (MRI & Magnetic Resonance Cholangiopancreatography) - noninvasive method for diagnosis and treatment. Reveals extensively dilated hepatic bile ducts and common bile duct filled with hydatid cyst material.

After all the above necessary investigations we should continue with the treatment, which in most cases is the operation.

## Results

Before beginning the surgical treatment of the disease, we should take into consideration that some groups of patients are inoperable. Surgery is contraindicated in elderly patients or patients with cardiac diseases, tumors or immunosuppression. On the other hand, the surgical indications include the flow rhythm. If there is *high flow* we perform operation earlier than the *low flow fistulae* which is performed after a 3 weeks waiting period. Also another indication is the cases of numerous cysts that increase the risk of multiple fistulae. Furthermore a large hydatid cyst and patients who developed mechanical jaundice or any other disease affecting the neighboring organs, are criteria for operability.

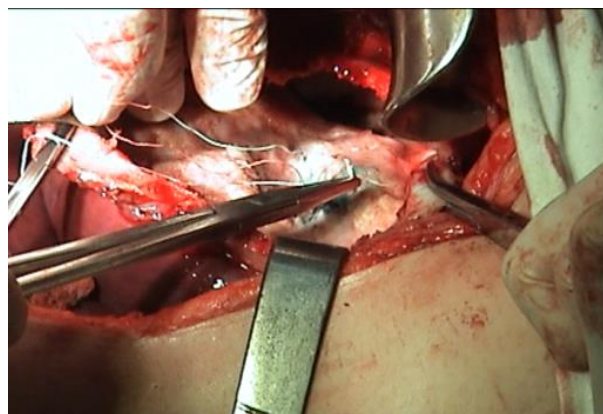


Figure 5 – The “Direct suture”

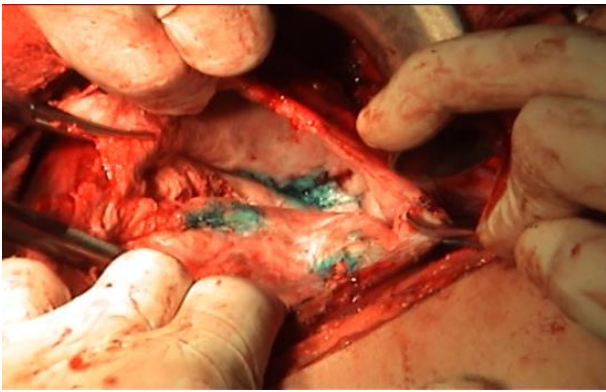


Figure 6 - The "Direct suture"

The treatment of the cysto-biliary communication is based on several techniques. The "Direct suture" (Figure 5 and 6) is used for small cysto-biliary fistula which could be sutured using a resorbable material. Next choice is the "Repair using a T-Tube" in which is allowed to restore canal continuity and to drain the hepatic territory of upstream. The T-Tube is kept 4 to 6 weeks and is withdrawn only after cholangiography. If a complete pericystectomy (Napalkoff total cystectomy, Figure 7 and 8) is not realizable under good conditions, then two other techniques of treatment of cysto-biliary communication could be performed. The one is "Transparieto-hepatic fistulization", which was described by Perdromo and we use a T-tube from which the one branch is put intra-biliary and the second one intra-cystic. The T-tube drainage is kept for 4 to 6 weeks.



Figure 7 – Extracted cyst

The second surgical choice is called "Internal drainage technique" and was invested by Goinard. It is chosen to be performed especially for central cysts with a large bilio-cystic fistula. The technique is pericystectomy by suture of pericyst and then follows the insertion of a T-

tube in the common bile duct, which will make drainage for the next 5 to 8 weeks. Finally some other surgical procedures are the ERCP, atypical hepatectomy and Pericystic – jejunal anastomoses.



Figure 8 – Extracted cyst - section

## Discussions

Echinococcosis is epidemiologically found all over the world. Specifically the specie *E. Granulosus* is endemic in America, Asia, Africa (north) and common in Europe particularly in Turkey and Greece [14]. Referring to *E. Multilocularis* the incidence is high in the northern hemisphere, like North America, Asia, central and northern Europe parts [15].

The Hepatic Hydatid Cyst disease is usually detected very late – years after the formation of the cysts, and commonly accidentally, so the incidence for complication forms of the disease is very high. Severe and many times lethal complications can occur according to the site of the cysts. [16]. In the Liver, the parasite causes decreased function of the organ which may lead to liver failure, but primarily causing cholestatic jaundice or communications through fistulae by neighboring organs like with the biliary tree and with the thorax - lungs, or even with the skin (very rare).

Even if surgery remains the gold standard treatment for the disease and its complications, sometimes the criteria are not enough to proceed in operation and so alternative therapies must be performed. Preferable is the introduction of PAIR and chemotherapy in inoperable patients. Further more a minimally invasive procedure as

laparoscopy has been proven beneficial by many surgeons due to the low morbidity outcome and to the better visual control of the cyst cavity. Unfortunately there are many criteria that must be filled and exclude the laparoscopic procedure and they are based on: fistulae formation, central localization of cyst, number of the cysts, thickened or calcified wall of the cyst [17-20].

## Conclusions

- Liver is the most common site for the hydatid cyst
- The high rate of cases of liver hydatid cyst are diagnosed in the complications stage
- The cysto-biliary fistula is one of the first complications of Echinococcosis
- Intra-hepatic and extra-hepatic communications with the biliary tree can be detected preoperatively and intraoperatively
- Non specific surgical treatment.
- Effective and good drainage of biliary tree & maintenance of the principles
- For prevention of recurrent cyst or fistula radiological tests post-operatively are often recommended

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