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PORTAL CAVERNOMA COMPLICATING A PERSISTENT POSTOPERATIVE BILE LEAK AFTER LIVER HYDATID CYST SURGERY: -A REPORT CASE-

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ABSTRACT

Introduction: Hydatid cyst is a parasitic infection caused by the larval form of Echinococcus granulosus. Portal hypertension is an unusual postoperative complication after a liver hydatid cyst surgery.
Case presentation: To illustrate this rare condition, we present the case of a 32-year-old patient operated for a liver hydatid cyst with late onset postoperative complications. The patient suffered from secondary digestive bleeding due to portal hypertension and was therefore treated with a distal splenorenal shunt (Warren).
Conclusion: Surgery of hydatid cyst may cause severe complications like portal hypertension and in certain cases, distal splenorenal anastomosis might represent an important treatment option that needs to be performed in specialised centres.

Keywords: Hydatid; Liver cyst; Portal hypertension; Splenorenal shunt.

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INTRODUCTION

Hydatid cyst is a parasitic infection caused by the larval form of Echinococcus granulosus. It arises in human's liver in 72%-93% of cases [1]. To treat, different attitudes are feasible: a conservative approach (no touch), pharmacotherapy with benzimidazole compounds, endoscopical approach, radiofrequency thermal ablation, percutaneous needle aspiration, and surgery as invasive treatment option. There has not yet been reached a consensus on the optimal therapeutic approach, however, surgery represents the standard treatment for liver hydatid cyst [2, 3]. Surgical treatment may cause certain complications like deep bleeding or hematoma, deep infection and bile leakage, which occur in 10%-28% of the cases [3, 4]. One of the unusual complications is the occurrence of a portal hypertension.
The present paper reports the case of a patient operated for a liver hydatid cyst with late onset of portal hypertension treated with a distal splenorenal shunt (Warren, DSRS).

CASE REPORT

A 32-year-old male was operated for a hydatid cyst of the liver segment 1. He underwent a simple unroofing of the protruding dome of the cyst with a drain placed in the residual cavity. At postoperative day 3 (POD3), a bile leak externalized by the drain was identified and managed conservatively (drainage on contact, monitoring and quantification until complete closure of the fistula). The patient was discharged from hospital at POD 9. Spontaneous closure of the fistula occurred after 11 months of progressive regression of the bile output.
We note that no signs of portal hypertension have been reported during surgery, including oesophageal varices, collateral vein circulation enlarged spleen or ascites.
The patient was missed for 4 years until he experienced 2 episodes of massive bleeding from esophageal varices that were initially managed with endoscopic varices ligation and blood transfusion. Abdominal computed tomography (CT) with angiography showed a portal cavernoma with a venous thrombosis spreading to the portal vein and the superior mesenteric vein and a splenomegaly. There was no evidence of chronic liver disease or

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recurrence of liver hydatid cyst. Viral hepatitis B and C serologies were negatives. Thrombophilia assessment was negative (including protein S, protein C, protein C-resistance activity, factor V and VIII, factor-II-mutation, Jak-2-gene-mutation and homocystein). These data were consistent with the hypothesis that the persistent bile leak and subsequent lasting infection caused portal thrombosis, cavernoma and hypertension in our patient.

Three months after the last bleeding episode, the patient was referred to our center for a recurrence that was out of endoscopic control. As thrombosis spreads to other veins of the portal venous system such as the superior splenic and mesenteric vein, a splenectomy could not be effective and the patient underwent a salvage surgery that consisted of splenic vein and left renal vein shunting (Warren procedure) (fig.1).

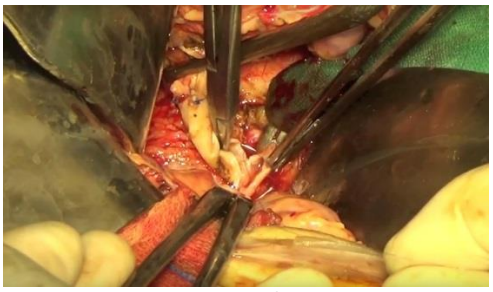

Fig.1: Operating view of splenorenal shunt

The postoperative courses were uneventful and the patient was discharged from the hospital at POD 11. He is alive without any bleeding recurrence with a patent shunt (fig.2) 24 months after the operation.

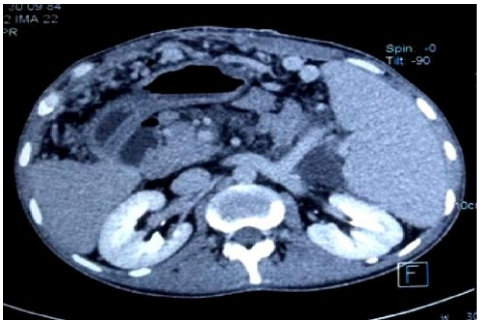

Fig.2: CT scan image showing the splenorenal shunt

DISCUSSION:

A portal vein thrombosis can occur in the portal vein, its branches or also with extension into the splenic, superior mesenteric or inferior mesenteric vein [5]. Portal thrombosis can be associated with surgery, especially hepato-pancreato-biliary surgery. Reports on liver transplantation and duodenopancreatectomy show an incidence rate of 5-16% and 1-2%, respectively [6]. The etiology is based on the Virchow's triad consisting of venous stasis, hypercoaguable state as well as endothelial lesions. Endothelial lesions may be caused by intra-abdominal infection, inflammation or direct manipulation of the portal vein in case of splenectomy, shunt surgery, liver transplantation or other abdominal surgery. In our case the conservative treatment of the biliary fistula with a permanent drain and the subsequent inflammatory syndrome represent probable reasons for the portal thrombosis.

Liver hydatid cyst surgery can cause postoperative biliary fistula with an incidence rate of 8.2-26% [7,8]. Usually this complication can be handled well by using distinct and systematic surgical approaches as described by our team permitting good results even in most complex cases.[9]

Portal vein thromboses show symptoms like abdominal pain, nausea or fever but may also be asymptomatic. They can be diagnosed by color duplex sonography or CT with angiography with high sensibility and specificity.[10] The treatment include anticoagulation, thrombolysis or surgical thrombectomy. Not sufficiently treated, the thrombosis can lead to portal hypertension, development of portal cavernoma and esophageal varices, all of which increase the postoperative morbidity and mortality. The bleeding of esophageal varices has a high mortality rate of 40% and high incidence of early rebleeding [11]. Primary treatment options are vasoconstrictors and endoscopic varices ligation, however, up to 20-30% of these procedures are not successful [11]. A transjugular intrahepatic portosystemic shunt (TIPS) offers a therapeutic option in this situation permitting control of the bleeding in 69% [12]. The implantation of the shunt might be difficult or even impossible if the thrombosis is spread into other veins of the portal venous system such as the splenic or superior mesenteric vein. As this was the case in our patient, a surgical shunt was indicated and performed.

CONCLUSION:

Portal cavernoma is unusual complication of hydatid cyst surgery; its treatment is mainly surgical. The distal splenorenal shunt is the most preferable surgical procedure despite a considerable morbidity, mortality and increased risk for thrombosis of the anastomosis. Therefore, this procedure should be

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performed in specialized centers for hepatobiliary surgery by experienced surgeons.

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