CASE REPORT

Triple communicating complicated hepatic hydatid cyst: an unusual presentation and laparoendoscopic management

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SUMMARY

Echinococcal disease in humans had been widely reported in the literature for its unusual presentation and location. We discuss a rare case of suppurated left hepatic hydatid cyst with contaminant transdiaphragmatic rupture communicating into the right pleural cavity along with fistulisation of cyst into the left hepatic duct and stomach which is very rare according to our knowledge. A 65-year-old man presented to us with features of cholangitis and sepsis. Initial radiological investigations revealed multiloculated cystic mass in the left lobe of cirrhotic liver communicating with the left hepatic duct and extending to the right pleural cavity with dilated common bile duct. Endoscopic retrograde cholangiopancreatography confirmed the diagnosis yielding hydatid membranes with presence of transgastric fistula in the left hepatic duct and provided postoperatively continuous internal drainage. Old age, complicated cyst, compromised respiratory status, sepsis and cirrhotic liver precluded us to plan for conservative surgical approach (laparoscopic drainage of mediastinal contents) with successful outcome.

BACKGROUND

Echinococcal disease in humans had been widely reported in the literature for its unusual presentation and location. We discuss an unusual case of suppurated left hepatic hydatid cyst with contaminant transdiaphragmatic rupture communicating into the right pleural cavity along with fistulisation of cyst into the left hepatic duct and stomach. These types of cases lead to diagnostic as well as therapeutic challenges. The cyst presented with combination of most common as well as rare complication of hepatic hydatid cyst. Therapeutic options should be tailored according to the patient's need from the various options available in management.

CASE PRESENTATION

A 65-year-old man presented with a 3-month history of right upper quadrant pain and dyspepsia along with difficulty in breathing from past 1 month. On clinical assessment, the patient was febrile, icteric, with decreased air entry on the right side of the chest and a palpable gallbladder. Haematological parameters revealed raised leucocyte count ($19\,700\text{ cells/mm}^3$), serum bilirubin (3.2 mg/dL) with direct bilirubin (2.5 mg/dL) and INR (1.5) at admission.

INVESTIGATIONS

Radiological imaging (CT/Ultrasonography) revealed a multiloculated cystic mass in the left lobe of cirrhotic liver communicating with the left hepatic duct and extending to the right pleural cavity, dilated common bile duct (1.2 cm) and intrahepatic biliary radicals (IHBR) up to head of pancreas, enlarged lymph nodes around peripancreatic, coeliac and portal area, right pleural effusion and consolidated right lower lobe lung. Ca19-9 levels were high (1140 IU/mL) and α -fetoprotein levels and lactic dehydrogenase levels were within normal limits. Serology for echinococcal antigen was positive. Owing to element of high suspicion of complicated hepatic hydatid cyst along with features of cholangitis, an endoscopic retrograde cholangiopancreatography (ERCP) was planned.

ENDOSCOPIC MANAGEMENT

ERCP was performed under sedation and guide wire was cannulated followed by wide sphincterotomy of the ampulla using standard sphincterotome. Balloon sweep yielded infected bile and membranes typical of hydatid cyst (figure 1). Repeated balloon sweep were performed along with copious saline irrigation and cholangiogram demonstrated dilated left-sided biliary radicals. A 7 Fr double pigtail stent was deployed. During withdrawal of scope, fistulous opening was identified in the antrum of stomach closer to lesser curvature and guide wire was cannulated with instillation of dye through it.

To our surprise, it was found delineating the IHBR and extravasation of contrast into the right

Figure 1 Endoscopic retrograde cholangiopancreatography and removal of hydatid membranes.

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Figure 2 Fluoroscopic view of common bile duct stent and cholangiogram obtained via gastric fistulous opening.

pleural cavity (figure 2). The 7 Fr double pigtail stent was deployed over the guide wire through the antrum for providing adequate internal drainage of hydatid cyst (figure 3). Postprocedural contrast-enhanced CT of the abdomen and thorax was obtained which confirmed the above findings (figure 4). Diagnosis of transdiaphragmatic rupture of suppurated hydatid cyst communicating into the right pleural cavity along with contaminant fistulisation to the left hepatic duct and stomach was made.

OPERATIVE MANAGEMENT

In view of wider transdiaphragmatic communication of cyst with the pleural cavity along with cirrhosis of liver, laparoscopic drainage of mediastinal collection was planned after 3 days of ERCP. The anterior wall of the stomach was found densely adherent to the left lobe of the liver which in turn was plastered to diaphragm (figure 5). Mediastinal extensions of cystic contents were approached by creating an opening in the diaphragm transabdominally (figure 6). Infected contents were aspirated using a 10 mm suction cannula followed by irrigation of the cavity with scolicedal agents. The other end of transgastric stent was found in the mediastinal cavity (figure 7). Twenty-eight Fr drain were placed in the mediastinal cavity and Morrison's space.

OUTCOME AND FOLLOW-UP

Postoperative course was uneventful and the patient was discharged on 10th postoperative day after laparoscopic drainage

Figure 4 CT image—postendoscopic retrograde cholangiopancreatography.

with a total stay of 17 days in the hospital. Albendazole therapy was administered for 3 months. Follow-up was performed on 6th week, 12th week and 6 months after surgery. X-ray of the chest and ultrasound of the abdomen performed during the follow-up period showed resolution of right basal lung consolidation and decrease in the size of dilated biliary radicals in 12 weeks. After 1 year of follow-up there was no recurrence.

DISCUSSION

Long asymptomatic course of hydatid cyst is well renowned but the exact incidence is not known. Liver involvement (50–75%) is most common followed by lungs (20–25%) and other organs (10–15%).^{1–2} Rupture into biliary tree (5–45%) is reported to be the most common complication of hepatic hydatid cyst, followed by suppuration of the cyst, communication to the pleural, pericardial and peritoneal cavity.^{3–5} However, few unusual presentations of hepatic hydatid cyst reported in the literature are concomitant rupture into the pleural and biliary tree,^{6–7} rupture into the duodenum,⁸ spontaneous intraperitoneal rupture⁹ and anaphylactic shock due to spontaneous rupture into the peritoneal cavity.¹⁰

The spectra of transbiliary rupture of hydatid cyst are occult communication (10–37%) and frank intrabiliary rupture (3–17%).¹¹ Occult rupture is usually silent and may be accompanied by suppuration or can evolve towards a frank rupture.¹² In frank rupture, daughter vesicles and fragmented membranes escape into the biliary tree causing obstruction, cholangitis or

Figure 3 Placement of transgastric stent via gastric fistula.

Figure 5 Intraoperative findings.

Figure 6 Creation of opening in the diaphragm to approach mediastinal extension.

septicaemia as in the above mentioned case.¹³ Suppuration of bile (23%) occurs most frequently due to cystobiliary communication.⁵ Concomitant pleural and biliary rupture is very unusual and considered to be a difficult condition to manage due to high morbidity and mortality.¹⁴ Moreover, there are very few case reports which describe fistulisation of hydatid cyst into the stomach.^{15 16}

High values of Ca19-9 levels in liver hydatid cyst as in the above case had been reported in few cases in the literature.^{17 18} The possibility for such high levels are chronic inflammation, hyperbilirubinaemia due to obstructive jaundice, acute cholangitis due to frank rupture of hydatid cyst in the biliary tract and substances produced by parasites that may closely resemble the Lewis X antigen or closed related structures which are recognised by anti-Ca19-9 antibodies leading to the elevation of Ca19-9 levels.¹⁹

The role of ERCP is now well established in managing biliary complications of hydatid cyst.^{20 21} In the preoperative period, it allows confirmation of diagnosis and managing biliary obstruction. During postoperative period, stenting establishes continuous internal drainage and decreases the chances of biliary fistula formation and aids in healing of biliary fistula.^{20–22} ERCP becomes modality of choice in the management of such complicated cysts as is in our case. Manouchehr *et al*²² reported the successful endoscopic management alone, and along with medical therapy in cases of transbiliary rupture.

Although thoracic approach is recommended in case of rupture of cyst into the pleural cavity due to higher incidence of

lung abscess and involvement of bronchioles, a transabdominal approach is preferred in case of concurrent abdominal pathology. Kilani *et al*²³ mentioned about posterolateral thoracotomy approach in 40 cases of transdiaphragmatic rupture into pleural cavity that reported significant morbidity and mortality in their series.

The successful outcome of laparoscopic management of hydatid cyst had already been described by Palanivelu *et al*.²⁴ Laparoscopic approach along with its inherent benefits provided the access to difficult areas along with controlled drainage without adding to the pulmonary morbidity in this case.

Old age, complicated cyst, compromised respiratory status, sepsis and cirrhotic liver precluded us to plan for conservative surgical approach (laparoscopic drainage) with endoscopic stenting (biliary and transgastric) with successful outcome. To the best of our knowledge, triple communicating hydatid cyst (communication of cyst with left hepatic duct, stomach and pleural cavity) managed laparoendoscopically is the first reported case in the literature.

Learning points

- ▶ High index of suspicion should be maintained for diagnosing such an unusual case of hydatid cyst.
- ▶ Multimodal management should be a preferred modality for treating complicated hydatid disease.
- ▶ Among the various surgical options available, the need of type of surgery should be tailored according to individual patient.

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Figure 7 Drainage of mediastinal collection with placement of drain.

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