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Case Report

Complete resection for a giant recurrent biliary cystadenoma: A surgical case report and review of literature

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ABSTRACT

Background: Biliary cystadenoma is a rare cystic neoplasm of the liver. The clinical signs and symptoms are nonspecific, and treatment strategy is variable.

Case presentation: In this study, we presented a case of a 32-year-old female with multilocular biliary cystadenoma. The patient underwent partial removal of the hepatic cyst two times in two different hospitals for two years and that the histopathological results were biliary cystic adenoma but was successfully treated by radical resection after the second recurrence. The patient underwent a J-shaped laparotomy. The giant cystic mass measuring 20 cm × 15 cm was below the position of the right anterior segment. This lesion pushed the liver parenchyma to both sides and compressed the hepatic hilum, causing dilatation of the intrahepatic bile ducts. The patient underwent complete resection of cystic mass. During the dissection, a 0.5mm-diameter fistula of left hepatic duct with the cyst was found. It was sutured using absorbable polydioxanone (PDS 6.0) and the cystic duct tube (C tube) (6 Fr) was inserted via the cystic duct into the left hepatic duct due to drain the bile fluid.

Discussion: A biliary cystadenoma (BCA) primary origin is occasionally rare. Although imaging modalities such as ultrasound, computed tomography and magnetic resonance imaging could be suggestive, however, the definitive diagnosis is depended on the histological examination. Despite of being a benign tumor, it has a high risk of recurrence after conservative treatment. The potential risk for malignant is also present. Therefore, complete resection of the tumors is the treatment of choice.

Conclusion: We herein present a report of a rare case with had a giant biliary cystadenoma (BCA) primary origin. This report aims to improve the understanding of the diagnosis and management of this uncommon disease.

1. Introduction

Biliary cystadenoma (BCA) is a rare, benign tumor of the liver. It occurs for less than 5% of simple hepatic cysts. The risk of malignant transformation of BCA to biliary cystadenocarcinoma (BCAC) can be up to 20% [1]. Most BCA are asymptomatic, but some patients might present with abdominal pain, distension at the right upper quadrant or epigastric area as well as palpable abdominal mass. It can be found in women with oral estrogen therapy which is supposed to make the lesion grow rapidly [2–4]. Other symptoms included hemorrhage, cyst rupture [5–7]. The histopathological result is the gold standard for diagnosis. BCA is divided into 2 clinicopathological groups based on the presence or absence of ovarian mesenchymal stroma (OMS) and luminal communication of the bile duct. The recurrence rate after partial resection could be as high as 80%–90% [4,8,9]. So radical resection with negative margins is recommended (liver resection or enucleation). We report a case of biliary cystadenoma that was radically operated after two times recurrence to emphasize the importance of total resection of such tumor. All our work has been reported in line with the SCARE criteria and guidelines [10].

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2. Case presentation

A 32-year-old female patient was diagnosed with hepatitis B 7 years ago and have treated with tenofovir 300mg per day. Periodical medical examination showed a huge hepatic cyst, which gradually increased in size causing mildly painful at the right upper quadrant. The patient underwent partial removal of the hepatic cyst two times in two different hospitals for two years and the histopathological results were biliary cystic adenoma. However, the cyst persisted after the 2nd surgery then a palpable mass in the right upper quadrant with abdominal pain emerged shortly, leading to her admission to our hospital. Abdominal ultrasound confirmed a 17 × 15cm cystic mass in the center of the liver. Magnetic resonance imaging (MRI) demonstrated the giant cystic mass in the right anterior section of the liver with multiple internal septations, wall thickening and no contrast-enhancing capsule (Fig. 1).

Cyst content was heterogeneous. The lesion compressed the bifurcation of the hepatic ducts and caused dilatation of the intrahepatic biliary tree. Laboratory studies showed slightly elevated serum levels of bilirubin (9.9μmol/L), serum glutamic oxaloacetic transaminase (SGOT) (44U/L), serum glutamic pyruvic transaminase (SGPT) (41 U/L). There was also elevation of carbohydrate antigen 19-9 (CA 19-9) up to 1000 U/ml. Meanwhile, other biological tests were within normal range.

The patient underwent a J-shaped laparotomy. The giant cystic mass measuring 20 cm × 15 cm was below the position of the right anterior section. This lesion pushed the liver parenchyma to both sides and compressed the hepatic hilum, causing dilatation of the intrahepatic bile ducts (Fig. 2).

The cyst was found to contain the colored bile fluid, with multi septa. Bilirubin level in the cystic fluid was 2.4 μmoL/l. The cystic wall was sent for frozen section that showed negative for malignancy. The patient underwent complete resection of cystic mass. During the dissection, a 0.5mm-diameter fistula of left hepatic duct with the cyst was found. It was sutured using absorbable polydioxanone (PDS 6.0) and the cystic duct tube (C tube) (6 Fr) was inserted via the cystic duct into the left hepatic duct due to drain the bile fluid. Gross examination showed a multiple cystic lesion with a thick, tan-white cyst wall (Fig. 3 A, B). The histologic specimen showed that the cyst wall was lined by a single layer of cuboidal to columnar epithelial cells resembling biliary epithelium, with a stroma resembling ovarian mesenchymal stroma underneath. The cell morphology was bland without nuclear atypia or mitosis. The definitive diagnosis was a biliary mucinous cystadenoma without malignant transformation (Fig. 3C, D). The patient was discharged on the 8th-day post-surgery with no complication. Following up imaging the liver of the patient after 3 months, 6 months and 12 months showed no recurrent cystic mass.

3. Discussion

In the most recent WHO classification in 2019, BCAs or mucus-producing bile duct tumors were classified into Mucinous cyst neoplasm of the liver (MCN-L) and intraductal papillary neoplasm of the bile duct (IPNB), which was dependent on with or without communication with the bile ducts [11,12]. In pathological aspect, BCA or MCN-L was characterized with mucus-secreting cuboidal or columnar epithelium resembling biliary epithelium, with a stroma resembling ovarian mesenchymal stroma underneath [11]. To distinguish with hepatic simple (solitary) or benign cyst (BHC), BCA or MCN-L was characterized with enhanced low-attenuated multiloculated cyst with thickened irregular wall and papillary wall nodules in computed tomography (CT) [13]. By machine learning, Hardie AD. et al. have summarized three imaging features that distinguish between BCA and BHC: solid enhancing nodule, all septations arising from an external macro-lobulation and whether solitary or one of multiple cystic liver lesions [14].

BCAs are often asymptomatic and discovered incidentally. Some patients may present with palpable abdominal mass (approximately 60% of reported symptoms), distension as well as abdominal pain at the right upper quadrant or epigastric area [2,15,16]. Rarely, patients have nausea, dyspnea and weight loss [17,18]. The acute presentations are usually pain, fever due to intra-cystic bleeding, cyst rupture or infection [19]. Biliary obstruction may be due to tumor compression of the bile ducts or mucus-producing tumors causing obstruction of the biliary tract. These may include jaundice, hepatic colic, fever, chills and steatorrhea; which are often presented in cystic tumors originating from the extrahepatic bile duct [20]. Our patient was asymptomatic and incidentally diagnosed during the follow-up of hepatitis. However, after the second surgery, the cyst grew rapidly and presented as a palpable mass, causing abdominal pain.

In most cases of BCA, the lesions are diagnosed incidentally by ultrasound (US) or computed tomography [21]. US is initial screening with a sensitivity of 90% for the patients with suspected BCA [22]. Contrast-enhanced computed tomography allows the differential diagnosis of BCA or MSN-L from solitary bile duct cysts [21]. On MRI, these tumors are usually described as multiple septations, thick wall, homogeneous low intensity on T1 weighted images, high intensity on T2 weighted images [23,24]. Cystic fluid signal intensity depends on the protein content. Moreover, MRI demonstrates anatomic relationships between the cyst and the biliary tree which can aid in surgical planning [24,25]. Our patient, MRI showed a large multiseptated cystic lesion in the right anterior hepatic with wall thickening. Cystic fluid has heterogeneous signal on T1, T2 weighted images. Intrahepatic duct was dilated due to cystic compression of the bifurcation of the hepatic duct. Echinococcal cyst and liver abscesses can be difficult to distinguish from BCA [26]. Echinococcal cysts usually present as cyst degeneration, collapsed membranes and daughter cyst inside the cyst [26,27]. Liver abscesses may have multiple septations with wall thickness and thicker than that seen with BCA [25,26]. In a middle-aged woman with a multilocular cystic liver mass, the primary diagnostic consideration should be BCA [28]. The study of Choi et al. consisting of 31 patients demonstrated that the presence of a septum and septal thickening as

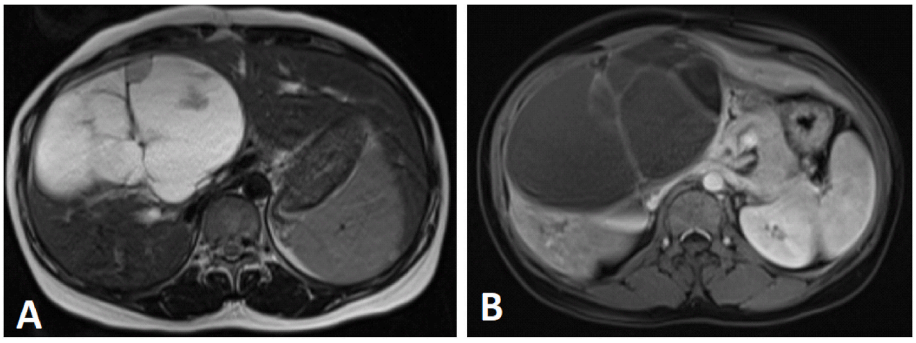

Fig. 1. Magnetic resonant imaging of the liver shows a multilocular cystic. (A) T2-weighted image shows a multiple septation cystic lesion. (B) T1 weighted image shows multiple thick internal separations with enhancement, there was no mural nodules.

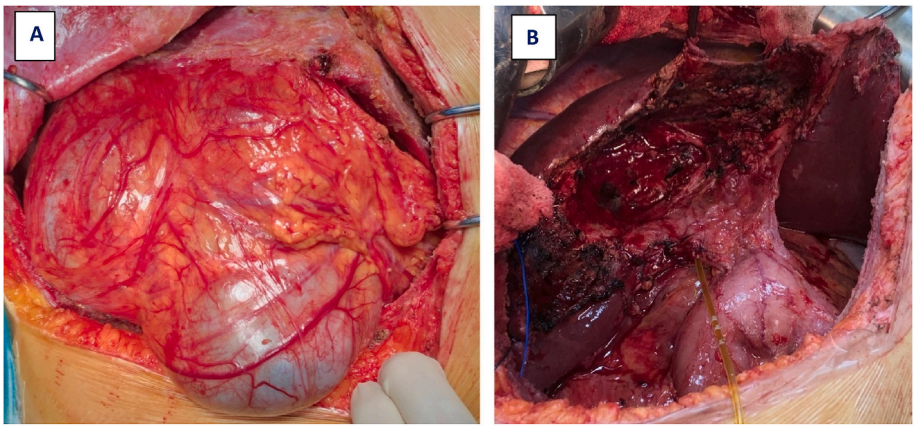

Fig. 2. Intraoperative imaging. A) Cystic mass below the right anterior and 4 segment. B) The site of cystic mass post-enucleation.

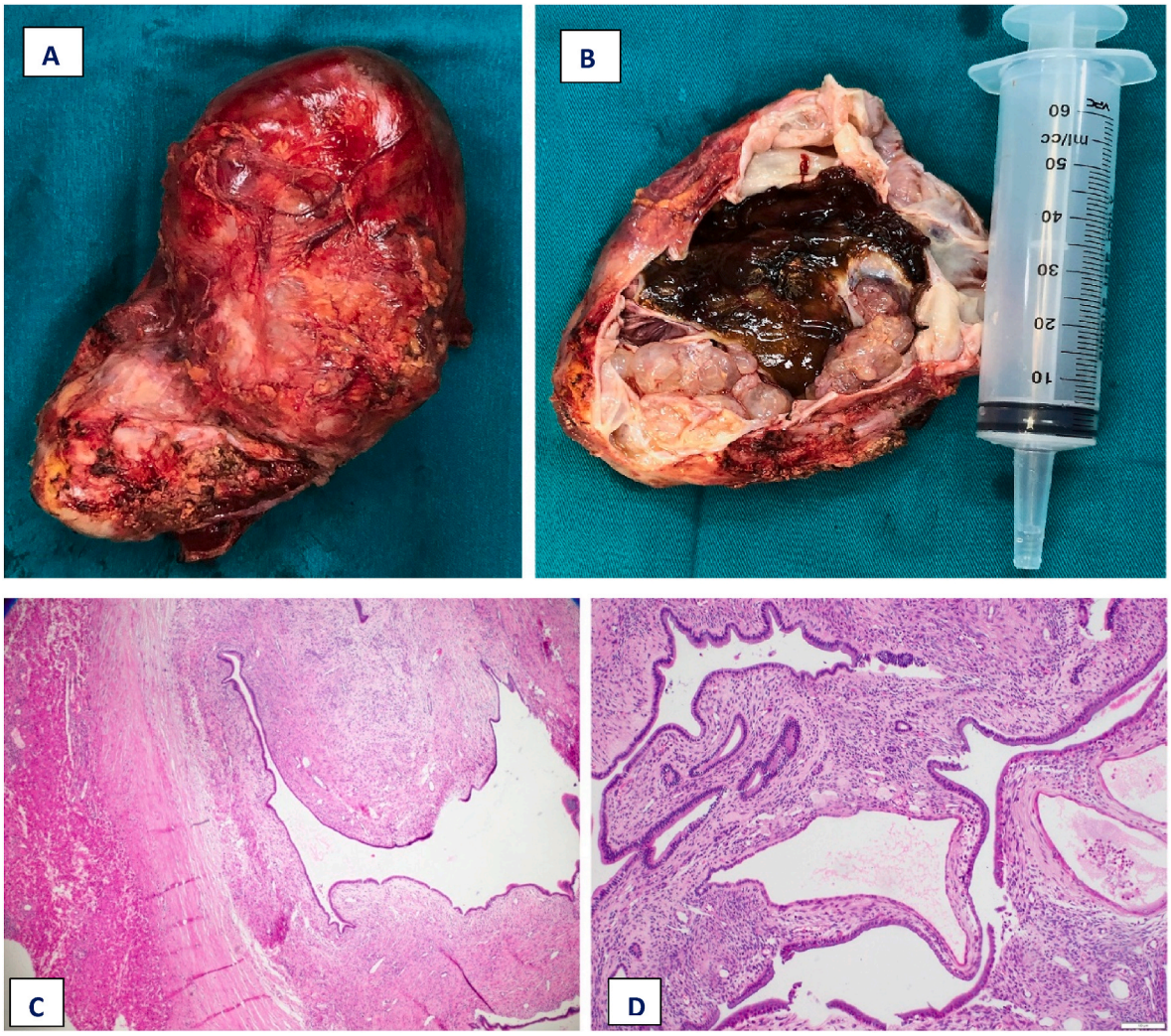

Fig. 3. Macroscopic appearance. A) Cystic mass post enucleation. B) Cystic mass with multilocular insides. Histological pictures showing the cyst wall consisted of epithelial cells with ovarian mesenchymal stroma. C) Cyst is lined by cuboidal epithelium resembling biliary epithelium with surrounding stroma. (H&E, 40X). D) A multiloculated cyst is lined by single layer of columnar - cuboidal epithelium with basally placed nuclei, which is absent of multilayering, nuclear atypia or mitosis. (H&E, 100 X).

significant predictors of BCA versus simple hepatic cysts [29].

The recurrence rates of BCA after fenestration, aspiration, sclerosis, internal drainage, marsupialization, or partial resection are up to 80–90% (8,9). Rapidly recurring cyst after aspiration may denote misdiagnosed BCA [30]. Therefore, formal surgical resection with negative margins is recommended [31,32]. Liver resection or enucleation depends on the individual patients, anatomical relationships of the cyst and surgeon experience (25–27). Cystic lesions localized to one side

of the liver should be performed hepatectomy, centrally located lesions that consist hepatic, portal vein and biliary tract should be enucleated [33].

There are two important issues while making a diagnosis of BCA. One is misdiagnosed as a simple hepatic cyst or a hydatid cyst, so that partial resection leads to postoperative recurrence and the second is difficult to distinguish from BCAC either before and during surgery [2,34]. Therefore, the recommendation is complete resection of any suspected BCA to decrease postoperative recurrence and to avoid misdiagnosis of malignant neoplasm [2,34]. We should not leave remaining BCA based on no evidence of malignancy from intraoperative frozen section. Intra-operative frozen section is often insensitive due to small focus of malignant that can be missed [31,32,35]. Our patient was operated on two different times at two different hospitals with the same operating procedure that was fenestration and had a rapidly postoperative recurrence. So, this patient was misdiagnosed as a simple hepatic cyst in the previous surgery.

At the third time of surgery, we removed completely the tumor and found intraoperative bile leak from the left hepatic duct. The C tube was inserted via the cystic duct into the left hepatic duct and was relieved on the 21st day postoperative without complication.

4. Conclusion

Biliary cystadenoma or mucinous cyst neoplasm of the liver is a rare type of hepatic cyst. Imaging diagnoses are non-specific, with histology and immunohistochemistry analysis are gold standards. If there is a multilocular and thick-walled cystic on liver imaging modalities, BCA should be suspected. Complete resection of the tumors (enucleation or liver resection) should be performed to avoid recurrence and to prevent the malignant degeneration of Cyst adenoma to Cyst adenocarcinoma postoperatively. Our report emphasizes the difficulty of diagnosis and management for this rare type of hepatic neoplasms.

Provenance and peer review

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Ethical approval

The study was approved by the Research Ethics Committee of Hanoi Medical University Hospital. The procedures used in this study adhere to the tenets of the Declarations of Helsinki.

Consent

The patients have consented to the submission of the case report for submission to the journal.

Author contribution

Hoang NGUYEN conceived and wrote the manuscript.

Ngoc Cuong NGUYEN edited the manuscript, provided imaging diagnosis as well as illustrated figures.

Thi Tra My THIEU analyzed the data and followed up the patient.

Tuan Hiep LUONG edited the manuscript.

Ngoc Minh TRAN provided histological imaging diagnosis as well as illustrated figures to the article.

Dang Hung NGUYEN and An Khang NGUYEN performed the operation.

All authors contributed to the interpretation of the results, discussed the results. All authors read and approved the final manuscript to submit. All authors contributed to the interpretation of the results, discussed the results. All authors read and approved the final manuscript to submit.

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Declaration of competing interest

The authors declare that they have no conflicts of interests.

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Abbreviations

BCA	Biliary cystadenoma
BCAC	Biliary cystadenocarcinoma
CT	Computed tomography
EGD	Esophagogastroduodenoscopy
GI	Gastrointestinal
MRI	Magnetic resonance imaging
OMS	Ovarian mesenchymal stroma
SGOT	serum glutamic oxaloacetic transaminase
SGPT	Serum glutamic pyruvic transaminase

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2022.103785.

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