Case Series

Biliary cystadenoma in young females with favorable surgical outcomes: A case series

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Abstract

Background: Biliary cystadenoma is a rare cystic neoplasm of the liver with potential for malignant transformation, often presenting with nonspecific clinical features. Preoperative diagnosis can be challenging due to overlapping imaging characteristics with other cystic hepatic lesions and variable tumor marker levels. We present a case series of two female patients with biliary cystadenoma who exhibited distinct clinical and radiological presentations. The first case involved a young female presenting with acute symptoms, obstructive jaundice, and markedly elevated CA 19-9 levels, mimicking a malignant lesion. Imaging revealed a large multiloculated cystic lesion with biliary compression, and intraoperative findings confirmed biliary communication, making surgical excision technically demanding. The second case presented with a more indolent course, mild symptoms, and typical imaging features without biliary involvement, allowing for straightforward surgical management. Both patients underwent complete surgical enucleation, and histopathological examination confirmed biliary cystadenoma without evidence of malignancy. The postoperative course was uneventful in both cases, with favorable outcomes. This case series highlights the varied clinical spectrum of biliary cystadenoma, the limitations of tumor markers such as CA 19-9 in differentiating benign from malignant lesions, and the importance of complete surgical enucleation for optimal outcomes.

Keywords: Biliary cystadenoma; Hepatic cystic lesion; CA 19-9; Liver tumor; Cystic liver neoplasm; Surgical excision

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1. Introduction

Biliary cystadenoma is an uncommon benign cystic neoplasm of the liver, predominantly affecting middle-aged women. It is characterized by multiloculated cystic architecture, internal septations, and the presence of ovarian-type stroma on histopathology. Although benign, it carries a risk of malignant transformation into cystadenocarcinoma, making early diagnosis and complete surgical excision essential.

Clinically, biliary cystadenoma often presents with nonspecific symptoms such as abdominal pain, fullness, or a palpable mass, or may remain asymptomatic for long periods. In some cases, especially when the lesion compresses adjacent biliary structures, patients may present with obstructive jaundice. Radiologically, these lesions typically appear as well-defined, multiloculated cystic masses with thin septations on contrast-enhanced computed tomography (CECT) and magnetic resonance imaging (MRI). However, differentiation from other cystic liver lesions such as simple hepatic cysts, hydatid cysts, and cystic neoplasms can be challenging [1,2].

Tumor markers such as carbohydrate antigen 19-9 (CA 19-9) may be elevated in biliary cystadenoma, sometimes to markedly high levels, mimicking malignant hepatobiliary tumors. This overlap can create a diagnostic dilemma, particularly when combined with atypical clinical features [3].

Definitive diagnosis is established by histopathological examination following surgical enucleation. Complete resection is the treatment of choice, as incomplete removal is associated with recurrence and potential malignant transformation [4].

In this case series, we present two patients with biliary cystadenoma demonstrating varied clinical presentations, radiological findings, and surgical complexities. This highlights the diagnostic challenges and emphasizes the importance of appropriate surgical management for favorable outcomes.

2. Case Presentation

2.1 Case 1

A 34-year-old female presented with complaints of abdominal pain for 20 days and vomiting for 7 days. The vomiting was non-bilious and non-blood stained. The patient also reported yellowish discoloration of urine. The abdominal pain was progressive and localized to the right upper quadrant.

On examination, the patient was icteric. Per abdominal examination revealed a soft mass measuring approximately 10 × 7 cm in the right hypochondrium. The mass was mobile with respiration and was tender on palpation. Bowel sounds were present.

Laboratory investigations revealed hemoglobin of 10 g/dL, total leukocyte count of 7,700 cells/mm^3, and platelet count of 1.82 lakh/mm^3. Liver function tests showed elevated total bilirubin of 5.4 mg/dL, with direct bilirubin of 1.9 mg/dL and indirect bilirubin of 1.2 mg/dL. Transaminases were elevated with AST of 328 IU/L and ALT of 188 IU/L. Alkaline phosphatase was 263 IU/L. Serum albumin was 5.8 g/dL. Renal function was normal with serum creatinine of 0.5 mg/dL. Coagulation profile showed an INR of 1.3. Tumor marker CA 19-9 was markedly elevated (>120,000 U/mL).

Contrast-enhanced computed tomography (CECT) of the abdomen revealed a well-defined exophytic cystic lesion arising from segment IV of the liver, measuring approximately 10 × 9 × 12.5 cm. The lesion showed a thin capsule measuring 1–2 mm in thickness with internal septations. No solid component, nodularity, calcification, or post- contrast enhancement was noted in the arterial, portal venous, or delayed phases. The lesion extended towards the porta hepatis with possible mild compression of the biliary confluence.

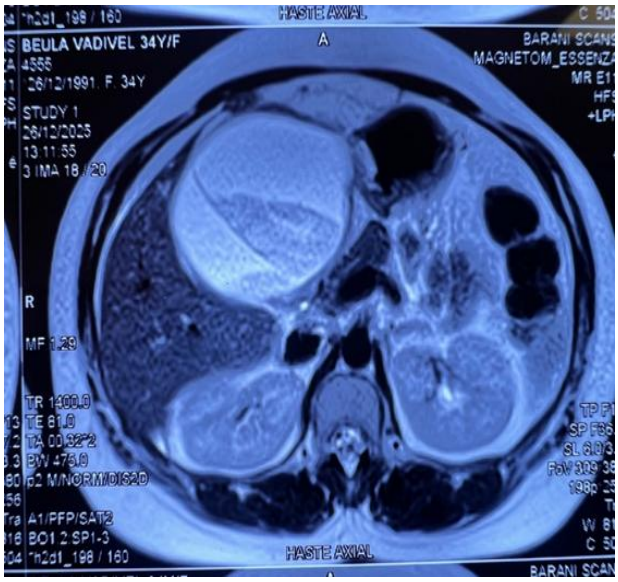

Fig (1): MRI Abdomen (axial section) showing a well-defined cystic lesion in segment IV of the liver with internal septations and hyperintense signal

Magnetic resonance imaging (MRI) of the abdomen demonstrated a large exophytic cystic lesion involving segments IV and IVa of the left lobe of the liver, measuring approximately 12 × 10 × 9 cm. The lesion showed internal septations measuring 1–2 mm in thickness with areas of internal debris. No solid component or nodular enhancement was noted. The lesion appeared hyperintense on T1-weighted imaging.

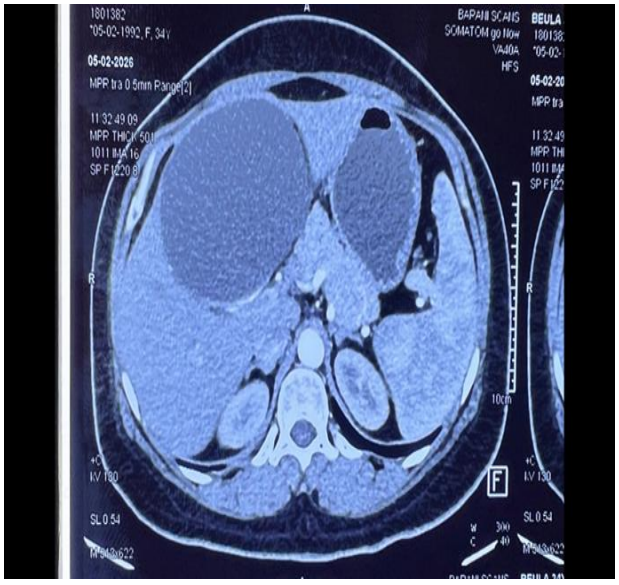

Fig (2): Contrast-enhanced CT scan showing a large well-defined cystic lesion in segment IV of the liver with internal septations and mass effect

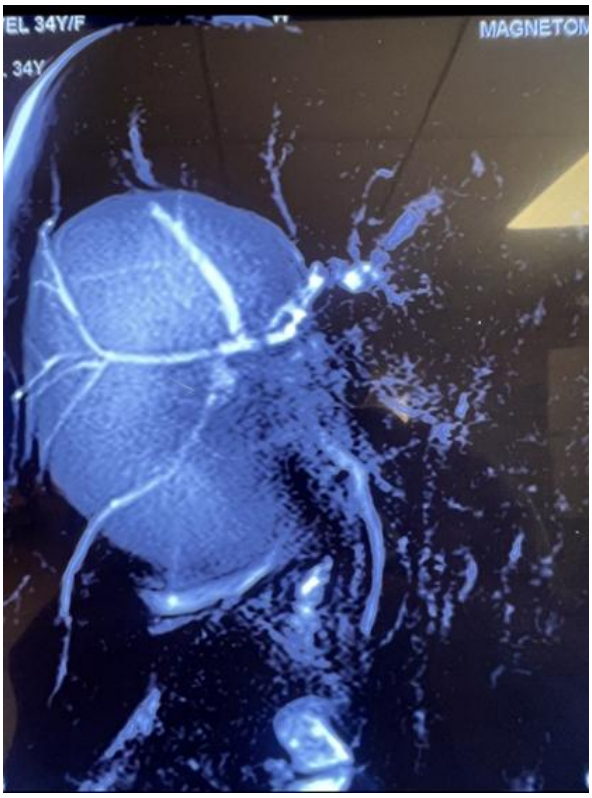

Fig (3): MRCP image showing a multiloculated cystic lesion in segment IV of the liver with internal septations and communication with the biliary tree

Based on the clinical, radiological, and laboratory findings, a provisional diagnosis of a cystic hepatic neoplasm, likely biliary cystadenoma, was made.

The patient was taken up for surgery under general anesthesia with epidural analgesia in the supine position. A right subcostal incision was made. Intraoperatively, a large cystic lesion was found occupying segment IV of the liver and extending into the hilar region. Approximately 300 mL of bile-stained fluid was aspirated from the cyst. The lesion was dissected from the liver surface using meticulous hand dissection along with hydro dissection techniques. The cyst wall was adherent to the gallbladder, and cholecystectomy with complete cyst excision was performed.

Intraoperative saline injection revealed a small rent in the left hepatic duct, which was repaired using polydioxanone (PDS) sutures. A 7 Fr pigtail catheter was inserted through the cystic duct into the common bile duct for drainage. Hemostasis was achieved, and a subhepatic drain was placed.

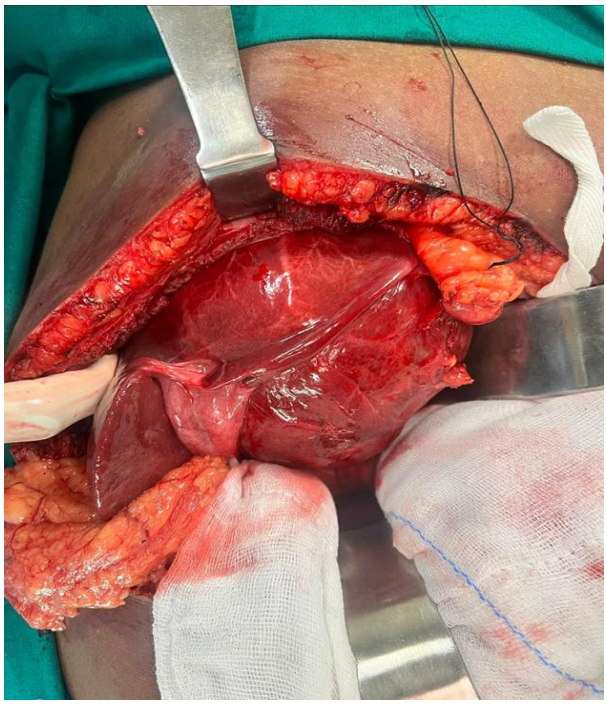

Fig (4): Intraoperative photograph showing a large exophytic cystic lesion arising from segment IV of the liver with hilar extension

Histopathological examination confirmed the diagnosis of Biliary cystadenoma, with no evidence of malignancy.

The postoperative course was uneventful. The patient was mobilized on postoperative day 1 and started on a liquid diet, which gradually advanced to a soft diet. The subhepatic drain was removed on postoperative day 6. The patient had a favorable recovery and remained asymptomatic on follow-up.

2.2 Case 2

A 48-year-old female presented with complaints of dull aching pain in the right upper quadrant of the abdomen for 3 months, associated with early satiety and intermittent nausea. There was no history of fever, vomiting, or jaundice.

On examination, the patient was not icteric. Per abdominal examination revealed a well-defined, non-tender mass measuring approximately 8 × 6 cm in the right hypochondrium, which showed slight movement with respiration. Bowel sounds were present.

Laboratory investigations revealed hemoglobin of 11.2 g/dL, total leukocyte count of 6,800 cells/mm^3, and platelet count of 2.1 lakh/mm^3. Liver function tests were within normal limits. Tumor marker CA 19-9 was mildly elevated (approximately 120 U/mL).

Contrast-enhanced computed tomography (CECT) of the abdomen demonstrated a well-defined multiloculated cystic lesion in segment VI of the liver, measuring approximately 9 × 7 × 8 cm. The lesion showed thin internal septations without any solid component, calcification, or post-contrast enhancement.

Magnetic resonance imaging (MRI) revealed a cystic lesion with low signal intensity on T1-weighted images and high signal intensity on T2-weighted images, with thin septations and no nodularity, suggestive of a benign cystic hepatic lesion.

Based on the clinical and radiological findings, a provisional diagnosis of biliary cystadenoma was made.

The patient underwent elective surgical excision of the lesion. Intraoperatively, a well-encapsulated cystic lesion was identified in segment VI of the liver without involvement of the hilar structures. The cyst was exercised completely using careful dissection, and no biliary communication was noted.

Histopathological examination confirmed the diagnosis of Biliary cystadenoma, with no evidence of malignancy.

The postoperative course was uneventful, and the patient recovered well with no complications. On follow-up, the patient remained asymptomatic.

3. Discussion

Biliary cystadenoma is a rare benign cystic tumor of the liver, accounting for a small proportion of hepatic cystic lesions. It predominantly affects women and is believed to arise from aberrant bile duct epithelium, often associated with ovarian-type stroma. Despite its benign nature, it is clinically significant due to its potential for recurrence and malignant transformation into cystadenocarcinoma if inadequately treated [5,6].

The clinical presentation of biliary cystadenoma is variable and largely depends on the size and location of the lesion. Most patients present with nonspecific symptoms such as abdominal pain, fullness, or a palpable mass. However, when the lesion is located near the hepatic hilum, it may cause biliary obstruction leading to jaundice. In the present case series, Case 1 demonstrated an acute presentation with abdominal pain and obstructive jaundice, whereas Case 2 had a more indolent course with mild symptoms and no biliary involvement. This highlights the wide clinical spectrum of this entity.

Radiological imaging plays a crucial role in diagnosis. On contrast-enhanced computed tomography (CECT) and magnetic resonance imaging (MRI), biliary cystadenomas typically appear as well-defined multiloculated cystic lesions with internal septations and a thin capsule. The absence of solid components, nodularity, or contrast enhancement generally favors a benign diagnosis.

In Case 1, imaging revealed a large exophytic cystic lesion with septations and extension towards the porta hepatis, causing compression of the biliary confluence. In contrast, Case 2 demonstrated a smaller lesion located in segment VI without hilar involvement. These differences significantly influenced the surgical complexity.

Tumor markers, particularly CA 19-9, may be elevated in biliary cystadenoma and can mimic malignant hepatobiliary neoplasms. Markedly elevated CA 19-9 levels, as seen in Case 1, can lead to diagnostic confusion and raise suspicion of malignancy. However, such elevations have been reported in benign cystic lesions due to mucin production or biliary communication. In Case 2, only mild elevation of CA 19-9 was observed, correlating with the absence of biliary obstruction or communication. This underscores the limited specificity of CA 19-9 in differentiating benign from malignant cystic liver lesions [7].

Surgical excision remains the treatment of choice for biliary cystadenoma. Complete resection is essential to prevent recurrence and eliminate the risk of malignant transformation. Incomplete procedures such as aspiration or marsupialization are associated with high recurrence rates and are therefore not recommended [4]. In the present series, both patients underwent complete surgical excision with favorable outcomes. Case 1

required meticulous dissection due to hilar involvement and intraoperative identification of a small rent in the left hepatic duct, which was successfully repaired. Case 2, on the other hand, allowed for straightforward excision due to the absence of biliary involvement.

Histopathological examination remains the gold standard for definitive diagnosis. The presence of cyst lining epithelium with underlying ovarian-type stroma confirms biliary cystadenoma and helps distinguish it from other cystic lesions of the liver.

This case series emphasizes the varied clinical presentations, imaging features, and surgical challenges associated with biliary cystadenoma. It also highlights the importance of considering this diagnosis in cystic hepatic lesions, even in the presence of markedly elevated tumor markers, and reinforces the role of complete surgical enucleation in achieving favorable outcomes.

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