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Case Report

Biliary Cystadenoma with High Dysplasia Detected Incidentally in a Young Patient Admitted for Percutaneous Abscess Drainage

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Abstract

Biliary cystadenomas are uncommon lesions with clinical and radiological characteristics that overlap with other cystic liver lesions. Here, we intended to discuss a biliary cystadenoma found in a 37-year-old female patient who had been treated for a liver abscess and had been sent to our clinic with a long-term hydatid cyst diagnosis.

Keywords: Abscess, biliary cystadenocarcinoma, biliary cystadenoma, computed tomography, excision, ultrasonography

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Biliary cystadenoma (BCA) is a rare benign cystic neoplasm of the liver originating from the biliary epithelium.[1] In about 85% of cases, it originates from the intrahepatic bile ducts. There are two subtypes of BCA that have been defined, including serous and mucinous types. Mucinous BCAs, which are uncommon, are more likely to develop into cancer.[2,3] BCAs may receive a diagnosis by chance or when a complication arises. The principal preoperative diagnostic tool is contrast-enhanced computed tomography (CT).[1]

Here, we present the treatment of an incidentally discovered BCA in a patient who was referred to our clinic for percutaneous liver abscess drainage and had previously been diagnosed with a hydatid cyst.

Case Report

A 37-year-old female patient, who applied to the surgery clinic with complaints of abdominal pain, weakness, high fever, and chills. She had tenderness in the right upper quadrant and was in bad condition; leukocytes: 5500 UL, CRP: 140 mg/dl, GGT: 243 IU/L, ALP: 210 IU/L, tumor markers and echinococcal indirect hemagglutination (IHA) test results were negative. The patient was diagnosed with a "hydatid cyst" based on an eccentric abdominal CT done about a year earlier (Fig. 1), and she was treated with albendazole. Due to the location and size of the lesion, surgery was not possible. The histology of the surgical sample was unspecific, and the patient was medically followed up. Abdominal ultrasonography (US) depicted a multiloculated cystic lesion

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measuring 11x12x15 cm in size, with irregular thick walls and septations, dense content, occasionally leveling, and a tendency to merge with other lesions, was discovered in the liver segment IV-VIII. Under the guidance of US and CT, the Seldinger procedure was used to insert a 10F diameter percutaneous drainage catheter into the lesion, which was identified as an abscess (Fig. 2). A microscopic analysis of the drained purulent material indicated numerous leukocytes and Corynebacterium striatum in its culture. Clinical and laboratory results for the patient significantly improved within the first week of drainage. Despite the fluid flowing from the catheter seeming less purulent and being present in smaller amounts, the drainage continued. The internal septations showed notable thickening and contrast enhancement despite reduction in the lesion size. Similar results were observed with CT and the magnetic resonance cholangiopancreatography (MRCP) examination, which was carried out to assess the link between the pancreas and the biliary system (Fig. 3). The abscess content of the lesion entirely vanished after about two months of percutaneous drainage therapy and periodic catheter modifications. Due to the patient's persistent yellowish catheter drainage and the lesion's persistent radiological appearance, particularly the contrast-enhancing internal thick septa, we hypothesized that our patient might have BCA-biliary cystadenocarcinoma (BCAC). Because the pre-operative diagnosis was BCAC and the clinical state was satisfactory, no culture was performed on the patient. The patient was then referred to a hepatobiliary surgery center. Despite having undergone two consecutive trucut biopsies of the lesion in this case, a histopathological diagnosis was not possible. The patient was not given Frozen. Laboratory results from the patient before surgery showed

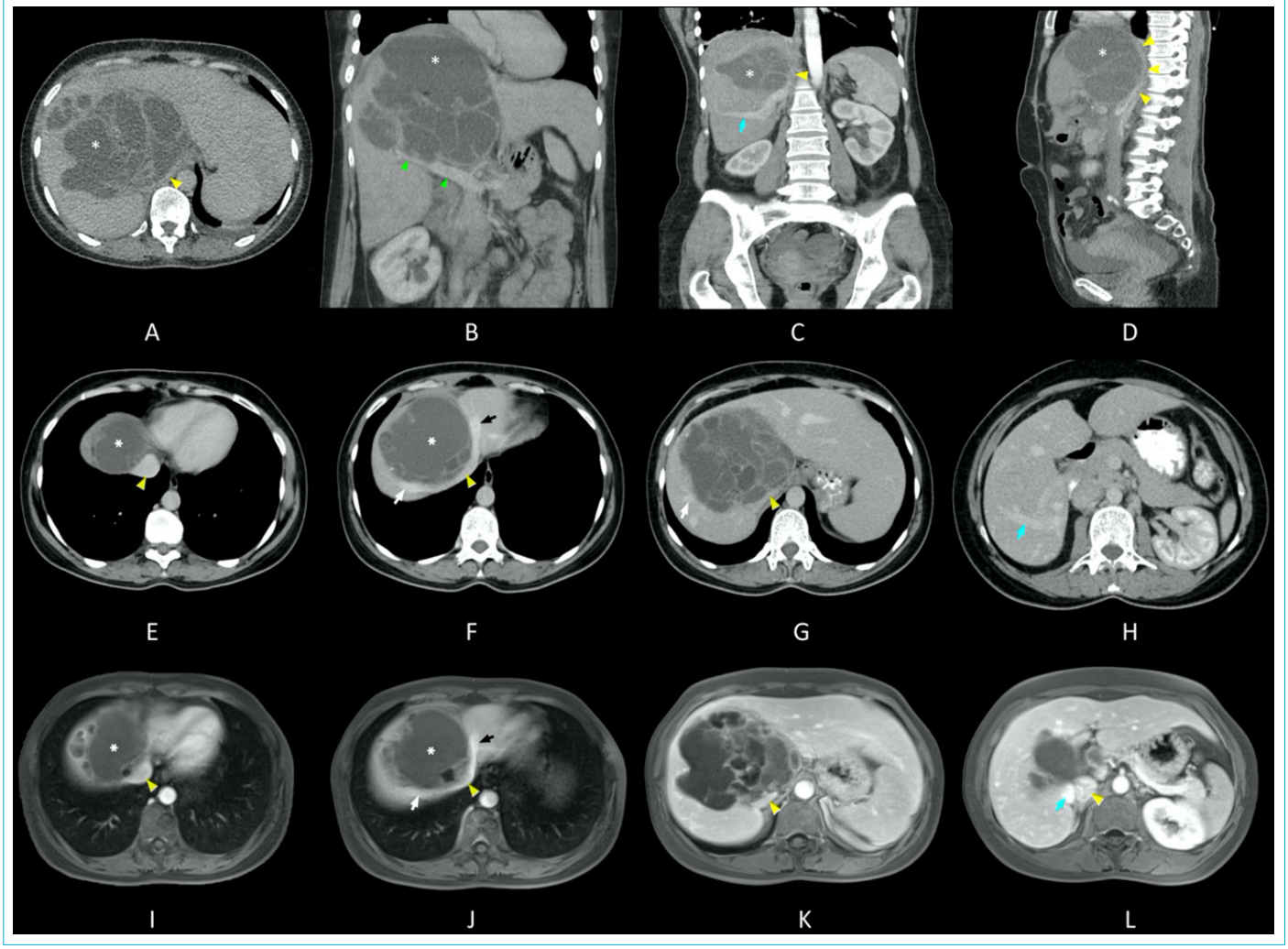

Figure 1. Imaging prior to the treatment. On axial CT **(a-h)** and MRI **(i-l)** images with intravenous contrast, the BCAC (asterisk), inferior vena cava (IVC; yellow arrowhead), left hepatic vein (black arrow), middle hepatic vein (white arrow), right hepatic vein (blue arrow) and portal vein (green arrowhead) are shown. Because of the mass effect, there is a deviation in the middle and left hepatic veins as well as IVC compression at the hepatic level (yellow arrowhead).

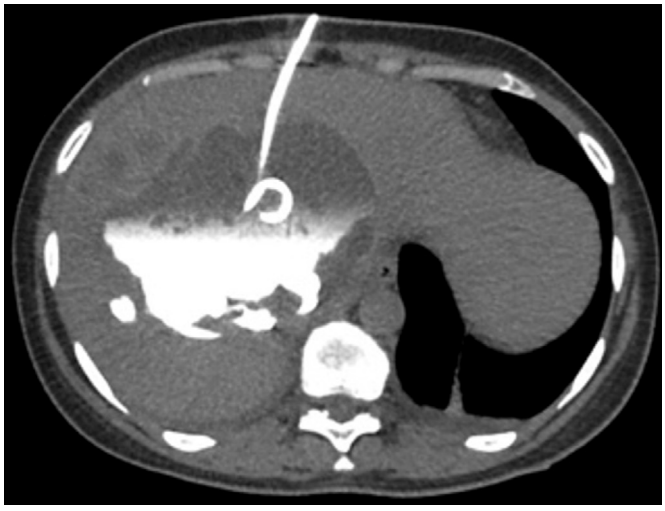

Figure 2. CT-guided drainage of the lesion with a catheter.

ALP=260 IU/L, ALT=722 IU/L, AST=838 IU/L, Albumin=27 g/dL, total bilirubin=1.74 mg/dL, direct bilirubin=0.85 mg/dL, GGT=639 IU/L, LDH=960 IU/L, and CRP=3.13 mg/L. Surgical intervention was performed with a Makuuchi J incision. A mass reaching approximately 20 cm in diameter in the central sector of the liver and compressing the left medial, caudate and right lobe was revealed with the help of intraoperative US. A previously placed drainage catheter area, located between segments 4-8, was observed. Central hepatectomy and isolated caudate lobectomy were performed six times with the help of Pringle maneuver, preserving the right posterior segments (Fig. 4). The lesion was reported histopathologically as BCA displaying high-grade dysplasia.

Discussion

The clinical features of BCAs are highly variable and often nonspecific.[4] Due to the possibility of conversion to BCAC, diagnosing BCAs is crucial.[5]

BCAs are most frequently seen intrahepatic (85%) and infrequently in the gallbladder.[1,6] The female to male ratio is 4:1, and they are most frequently observed in middle-aged women. Pregnancy and the usage of oral contraceptives have been linked to their increased occurrence in women.[7,8] There was no such history in our case. Because of their silent nature, BCAs can grow to be as large as 35 cm in size.[8] Right upper quadrant pain, distention, nausea, vomiting, and a palpable mass are the most typical symptoms when present. Rarely, patients may exhibit complications like bleeding, rupture, infection, obstructive jaundice, and venous obstruction. The malignant change of BCAs to BCAC, which has been observed to occur up to 20% of the time,

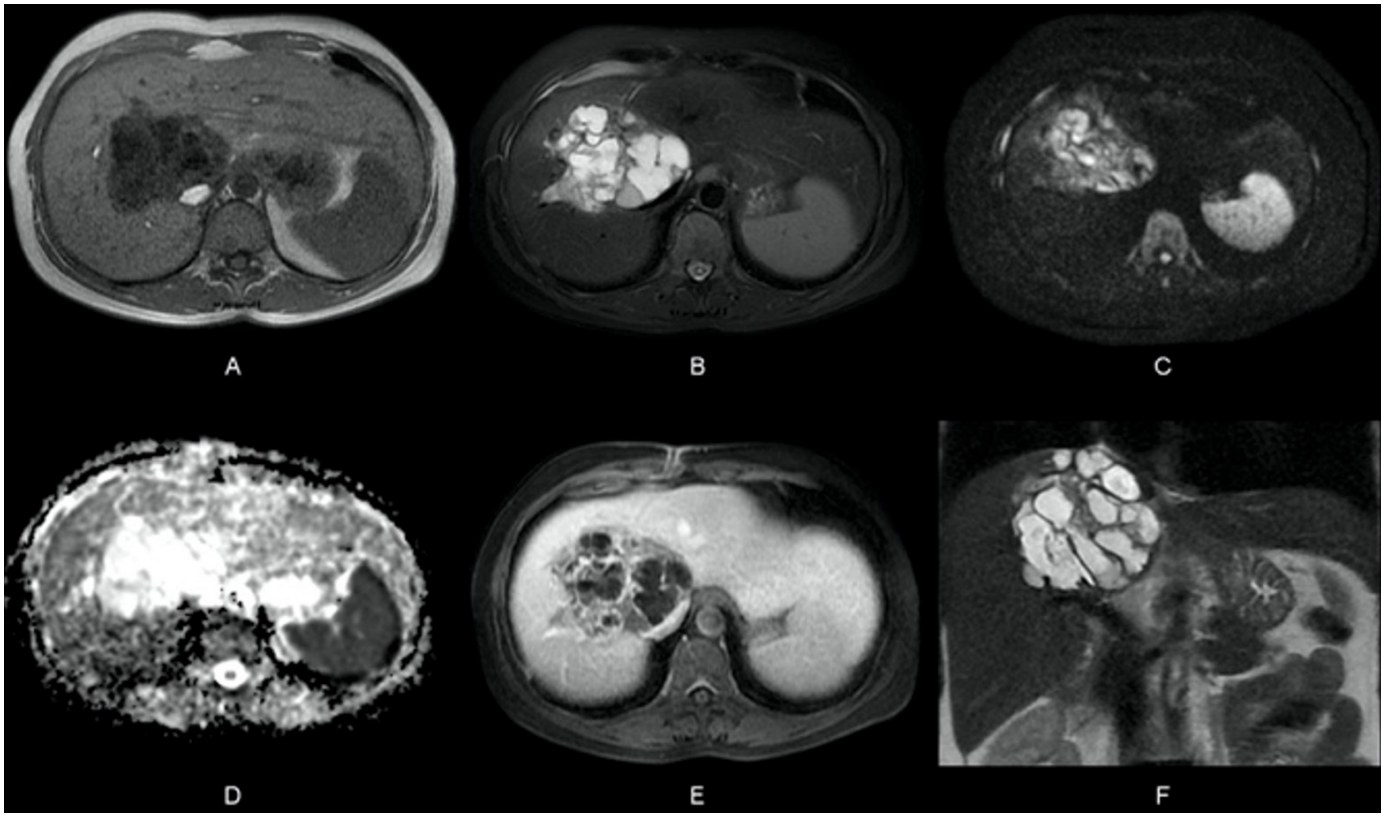

Figure 3. Post-treatment magnetic resonance imaging shows the multiseptal and multiloculated cystic lesion. **(a)** axial T1 weighted imaging (WI), **(b)** T2-fat saturated image, **(c)** diffusion WI, **(d)** apparent diffusion coefficient, **(e)** contrast-enhanced axial plan, **(f)** coronal plan T2-WI.

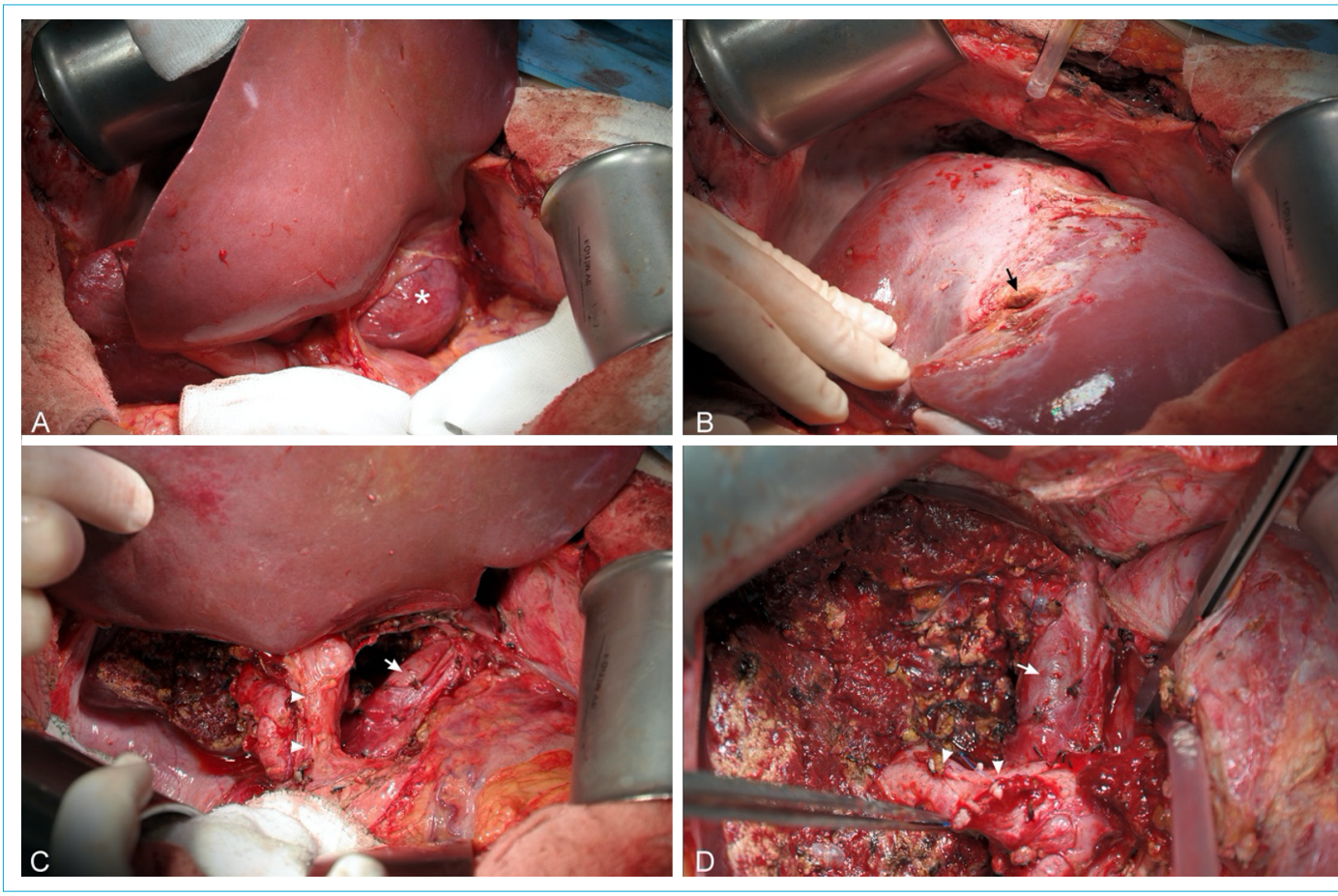

Figure 4. Intraoperative images show **(a)** Biliary cyst adenoma (asterisk) compressing the right and left lobes and the caudate lobe in the central sector, **(b)** the area of previously placed drainage catheter at the junction of segment 4-8 (black arrow), **(c)** Inferior vena cava (long arrow) with portal vein (arrows head) isolated in the central sector, **(d)** Inferior view of the vena cava (long arrow) after resection.

is the most feared side effect of BCAs. However, there were no reported physical examination findings that were associated with malignant potential.[4,6,9,10]

Imaging techniques are essential for diagnosing BCAs. They are single cystic tumors that are mainly found in the fourth segment of the liver.[4] In our case, the lesion was rather extensive and was placed between the fourth and eighth segments. Fine internal septations and calcifications, as well as multiloculated lesions with an echogenic capsule, are features of BCA in US. A critical finding in Doppler US for the differential diagnosis of septal hemorrhage. This discovery is evidenced by contrast enhancement in the septa on CT and magnetic resonance imaging (MRI). In the preoperative phase, CT outperforms US. MRI may reveal distinct signal characteristics for the cysts.[11,12] BCAs are not communicating with the biliary system. Examinations following contrast reveal only minor septal and capsular enlargement.[13] The laboratory is typically normal in BCA patients, with the exception of elevated liver enzyme and bilirubin levels in a small number of recorded cases. High levels of CA19-9 and CEA may indicate the onset of cancer.[4]

Due to the possibility of cell spread and the emergence of peritoneal carcinomatosis, fine-needle aspiration of the cyst fluid is not advised for diagnosis. In BCA, percutaneous tru-cut biopsy does not produce data for histological confirmation.[4,14] In cases with suspected BCA, radical excision is advised due to the possibility of malignancy and the high risk of recurrence.[4] This procedure is also indicated as the primary treatment and most conclusive diagnostic method. Two percutaneous biopsies were done on our patient, but the lump had to be completely removed in order to provide a conclusive diagnosis. Another therapeutic option is percutaneous ablation, which has a recurrence rate of 80% reported.[5] This rate is 5-10% after total resection.[4]

Simple hepatic cysts are the first lesions that come to mind in the differential diagnosis. The lack of internal septation and papillary protrusions set them apart from BCA. Hemorrhagic cysts are difficult to differentiate with US, however with CT, they are easier to distinguish from BCAs. Hemorrhagic cysts generally show up in T1 on an MRI.[4] The differential diagnosis also includes mesenchymal hamartomas, pyogenic hepatic abscesses, hydatid cysts, and undifferen-

tiated embryonal sarcomas. The presence of internal gas, perilesional edema and the differences in segmental perfusion usually suggest an infectious etiology. Radiologically, BCAs are frequently mistaken with the hydatid cyst Gharbi type 3 subtype.[12] Hydatid cysts can also exhibit negative serological responses and, in rare cases, septal enlargement, making it difficult to diagnose them, especially in locations where they are common. However, negative serological test, the presence of septate and high-density cystic lesion, papillary protrusion within the lesion, and most importantly, the presence of enhanced septations support the diagnosis of BCA.[15] Children and young men are more likely to develop embryonal sarcomas and mesenchymal hamartomas.[5] Imaging techniques might not be able to distinguish some atypical cystic lesions from cystadenocarcinoma and BCA. Due to variations in cystic fluid density brought on by intracystic bleeding or the presence of uneven septal thickening brought on by secondary infection, differential diagnosis may be challenging. In our situation, the lesion's development into an abscess made differential identification and diagnosis challenging. In addition, our patient had a history of surgical and medical follow-up with a hydatid cyst diagnosis for a while in an external center.

The prognostic data after resection in patients with BCA are limited. Patients should be made aware of the possibility of both recurrence and malignant transformation, particularly if total resection is not carried out. The distinction between BCA and BCAC may not always be accurately made by radiological imaging techniques. If the BCA exhibits variable wall thickness, papillary ridges, septal calcifications, internal debris, bile duct dilatation or a mural nodule with contrast enhancement, the likelihood of malignant transformation should be taken into account.[4,11,12]

Conclusion

BCAs can grow significantly in the liver and manifest as life-threatening problems because of their quiet path. They need to be diagnosed quickly because they frequently appear at younger ages and always carry a risk of becoming malignant. The fact that it is frequently mistaken for the hydatid cyst lesion, which is widespread in endemic nations like our case, may play a significant role in the delay in the diagnosis and treatment of BCA. Additionally, it is crucial to separate it from other benign hepatic cysts due to its unique clinical management. Excisional surgery serves as the only means of diagnosis and treatment in BCA due to the limited diagnostic value of tru-cut biopsy.

Disclosures

Informed consent: Written informed consent was obtained from the patient for the publication of the case report and the accompanying images.

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