

Imaging Diagnosis of Tubular Adenoma of Common Bile Duct: Case Report and Review of the Literature

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Received April 15, 2018; revision requested July 28, 2018; revision received August 1; accepted August 2.

Abstract: Tubular adenoma of common bile duct (CBD) is an extremely rare benign extrahepatic bile duct adenoma. We report a case of surgery and histopathological proved CBD tubular adenoma. A 69-year-old man was admitted to the hospital for yellow urine for one month. Clinical, radiological and laboratory results were analyzed. Medical literature in PubMed pertaining to similar cases was reviewed. A solid mass was detected in the distant part of CBD, with dilation of the extrahepatic and intrahepatic bile duct by ultrasound, contrast enhanced ultrasound (CEUS), computer tomography (CT) and magnetic resonance imaging (MRI). It showed heterogeneous enhancement both in contrast-enhanced CT, MRI and CEUS. After operation, pathological findings indicated a tubular adenoma of CBD. Our case highlighted the fact that tubular adenoma of CBD was demonstrated as a homogeneous mass in the distant part of CBD.

Key words: Tubular adenoma; Common bile duct; Imaging; Benign; Diagnosis

Advanced Ultrasound in Diagnosis and Therapy 2018;02:139–42

DOI: 10.37015/AUDT.2018.180813

Bile duct adenoma is a benign, indolent lesion. It is most frequently encountered incidentally as a solitary nodule in medical imaging. Histologically, it is a well-circumscribed, disorderly collection of irregular ductules lying within a connective tissue stroma with various degrees of sclerosis and chronic inflammation [1]. Although bile duct adenoma are generally regarded as having no malignant potential, it rarely gives rise to adenocarcinoma.

Up till now, limited cases of imaging character of bile duct adenoma have been reported globally. The diagnostic importance lying on its frequent confusion with cholangiocellular or metastatic adenocarcinoma [2]. Herein, we discussed a case of a tubular adenoma of common bile duct (CBD), which was preoperatively diagnosed by ultrasound, contrast-enhanced ultrasound (CEUS), computer tomography (CT) and magnetic resonance imaging (MRI).

Case report

A 69-year-old man was referred to our hospital complaining of yellow urine for one month. Mild increased serum alkaline phosphatase and serum γ -glutamyl trans peptidase were observed. No increase in serum tumor markers, such as carcinoembryonic antigen or carbohydrate antigen 19-9 (Table 1). No clinical evidence of hepatitis. Gray scale ultrasound showed intrahepatic biliary tree was slightly dilated and the diameter of CBD was 9mm. A homogeneously hypo-echoic mass of 20mm×13mm was detected in the distant part of CBD, with clear boundary and no acoustic shadowing (Fig. 1). The lesion was suspicious for tumor or sludge in CBD according to the ultrasound findings. CEUS was performed by LOGIC E9 (GE Healthcare, Milwaukee, WI, USA) ultrasound system and with 1.5–5.0MHz convex array probes. After injection of 2.0 mL SonoVue® (Bracco Imaging Spa, Milan, Italy)

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suspension, the mass at the distant part of CBD was synchronously and heterogeneously enhanced. The enhancement began at 15 s, reached to peak at 20 s, decreased from 31 s and became hypo-enhanced at 78 s (Fig. 2). As the lesion was obviously enhanced after contrast agent injection, the possibility of sludge was excluded. Also, because the enhancement of the lesion decreased earlier than its surrounding normal pancreas parenchyma, it was suspected to be a benign tumor by CEUS.

Table 1 Laboratory data at initial hospitalization

| Clinical characteristics | Value |
|------------------------------------|-------------------------|
| White blood cell | $14.13 \times 10^9/L$ |
| Red blood cell | $4.65 \times 10^{12}/L$ |
| Carbohydrate antigen 19-9 | 13.5 U/mL |
| Carcinoembryonic antigen | 1.01 ng/mL |
| Alpha-fetoprotein | 1.8 ng/mL |
| Carbohydrate antigen 125 | 7.3 IU/L |
| Alkaline phosphatase | 187 U/L |
| Alanine aminotransferase | 44 IU/L |
| γ -Glutamyl trans peptidase | 89 U/L |

Unenhanced abdominal CT showed a tumor of $10\text{mm} \times 10\text{mm}$ in the duodenal papilla with dilation of the extrahepatic and intrahepatic bile duct. While dynamic contrast-enhanced CT revealed heterogeneous enhancement in arterial and the venous phase (Fig. 3). The tumor was suspected to be benign CBD tumor.

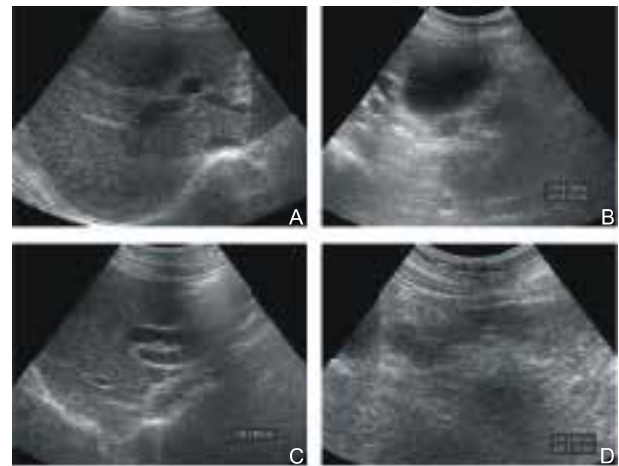


Figure 1 Gray scale ultrasound scan showed dilated inner-hepatic bile ducts (A). Gallbladder was slightly enlarged (B) and common bile duct (CBD) dilated (C). A suspected tumor of $20\text{ mm} \times 13\text{mm}$ in the distant part of dilated CBD (D).

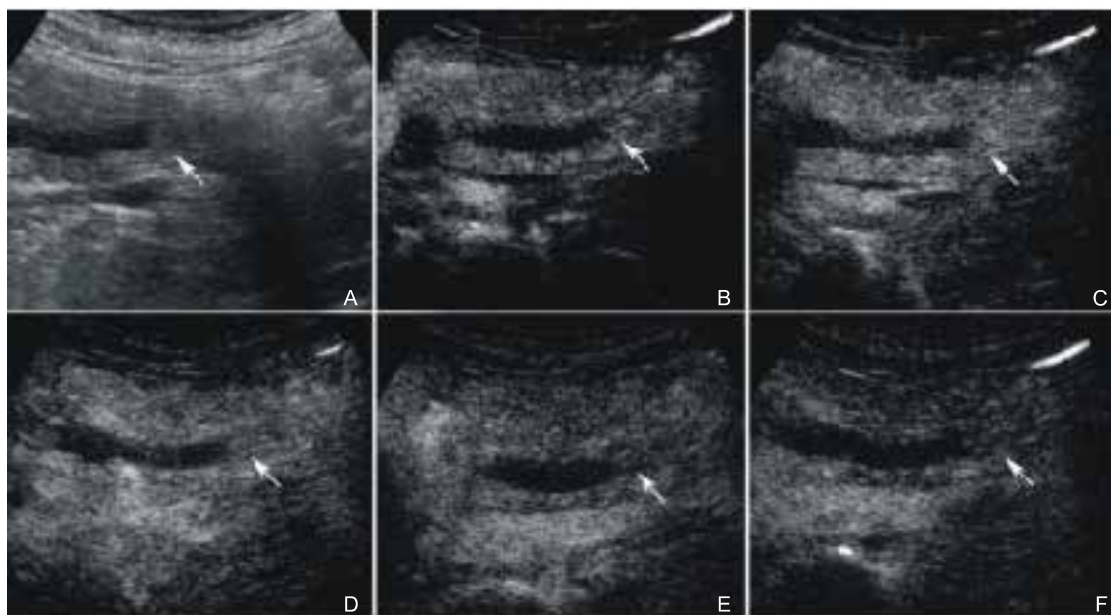


Figure 2 Contrast-enhanced ultrasound (CEUS) scan showed a $20\text{ mm} \times 13\text{ mm}$ sized non-shadowing mass (arrow) in the lower portion of the common bile duct on B mode ultrasound (A). The lesion (arrow) showed heterogeneous iso-enhancement 15 s after injection of contrast agent on CEUS (B). The enhancement reached to peak enhancement at 20s after injection of contrast agent on CEUS (C) and became iso-enhanced from 26 s (D). The enhancement of the lesion (arrow) decreased from 31s (E) and became hypo-enhanced at 78s after contrast agent injection (F).

After the patient was hospitalized, gadopentetate dimeglumine (Magnevist®, Bayer Schering Pharma AG, Berlin, Germany) enhanced abdominal MRI was performed. The tumor showed low intensity on T1-weighted images (Fig. 4A) and high intensity on T2-

weighted images (Fig. 4B). Contrast-enhanced MRI revealed that this tumor had heterogeneous enhancement on arterial phase (Fig. 4C). Local stenosis was detected in the distant part of common bile duct in coronal plane (Fig. 4D). The tumor was suspected to be malignant by MRI.



Figure 3 Unenhanced and enhanced computed tomography (CT) scan showed a tumor of 10 mm ×10mm in the duodenal papilla (A). In addition, tumor showed heterogeneous enhancement in arterial (B) and the venous phase (C).

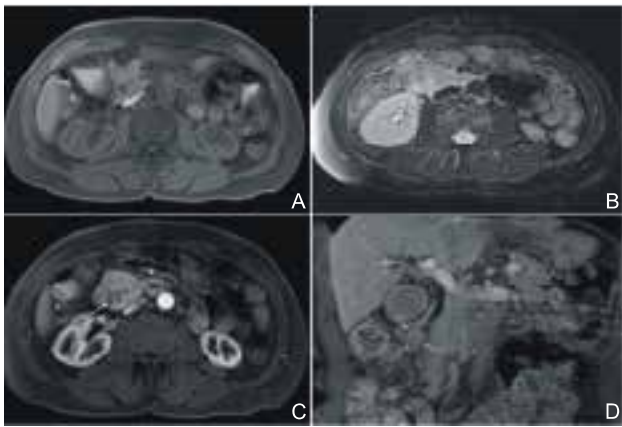


Figure 4 Gadopentetate dimeglumine (Magnevist®, Bayer Schering Pharma AG, Berlin, Germany) enhanced abdominal magnetic resonance image scan showed the lesion on T1- weighted image (A), T2- weighted image (B), arterial phase (C) and venous phase (D).

Therefore, the patient accepted operation. During the operation, surgeons detected a 20 mm × 16 mm polypoid mass at the junction between duodenal papilla and CBD. The CBD was partial obstructed. Histology showed the tumor consisted of small heterogeneous tubular ducts with fibrous tissues, without cell atypia or mitotic activity. It is well-circumscribed, with disorderly collection of irregular ductules lying within a connective tissue stroma, associated with varying degrees of sclerosis and chronic inflammation (Fig. 5). There was no lymph node metastasis. Thus, pathological diagnosis was confirmed as tubular adenoma of CBD, with moderate proliferative of glandular epithelium.

Discussion

CBD adenoma is an extremely rare benign tumor of extrahepatic bile duct. Although the actual incidence of adenoma of CBD is unknown, it is benign and asymptomatic tumor and is always detected incidentally [3]. Histologically, adenomas arise from the epithelial lining of duct, may be either tubular, papillary, or tubulopapillary [4]. Pathologically, tubular common bile duct adenoma is a well-demarcated, small sized lesion (<1

cm). It is characterized by well-circumscribed, disorderly collection of irregular ductules lying within a connective tissue stroma that shows varying degrees of sclerosis and chronic inflammation [1,5]. A variable proportion of CBD adenomas are still confused with malignant well-differentiated adenocarcinoma preoperatively [6]. The clinical manifestations of a CBD adenoma include progressive and intermittent jaundice, right upper quadrant abdominal pain, dyspepsia, nausea and vomiting which were similar to ampullary tumors [7]. It usually takes few weeks to months to make confidential diagnosis after appearance of clinical symptoms. As CBD adenomas may produce obstructive jaundice, which can be easily confused with a malignant neoplasm or stone [8]. Nowadays, in clinical practices, these tumors were known to be progressed to carcinoma, and in order to relief symptoms associated with ampulla obstruction, surgical resection is usually performed [1,3,7].

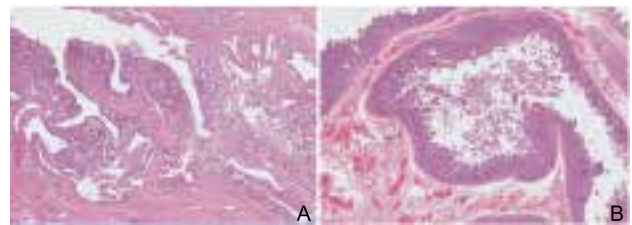


Figure 5 Microscopic features of the tubular adenoma (Hematoxylineosin stain) showed tumor consisted of small heterogeneous tubular ducts with fibrous tissues (A, × 100). There was no cell atypia or mitotic activity (B, × 40).

It has been reported that preoperatively differential diagnosis between CBD adenoma and malignant tumor is very difficult by current imaging methods [9]. Due to recent advances in diagnostic techniques, various imaging approaches have been applied to make early diagnosis of CBD adenomas, which were often detected as high-grade dysplasia or carcinomas in situ. Conventional B mode ultrasound is the first-line imaging investigation for biliary diseases. It can make convenient and accurate diagnoses of intrahepatic and extrahepatic bile duct dilations. However, it is lack of the ability

to depict the microcirculation of the suspected lesion. Sludge, non-shadowing stones, blood clots, and other benign tumors, could not be ruled out by conventional B mode ultrasound [10]. Recently, with the use of contrast-enhanced agents and the development of CEUS technology, it is possible to visualize disease features in small structures, promising results have been achieved in the field of bile duct disease [11]. CEUS was able to make differentiation diagnosis between CBD cancer and sludge or stone without acoustic shadowing [12]. As the blood supply of the common bile duct is entirely arterial, defining the enhancement process of the lesions in CBD is complicated and quite different from that of the liver [12–13]. It is recommended that the process is classified as early phase (10-30 s after contrast injection) and late phase (31-180 s after contrast injection) [13]. In our case, slightly CBD dilation with a homogeneously hypo-echoic mass in the distant part was a characteristic finding on conventional ultrasound. However, conventional ultrasound can hardly differentiate tumor in the CBD from sludge since they have similar echogenicity. Real time CEUS was performed preoperative to provide more details about the suspected tumor in CBD. CEUS allows real-time visualization of the enhancement of the lesion in the distant part of CBD conveniently and accurately. It is an effective approach for different diagnose of tumors in CBD [14]. On CEUS, the mass was identified as fast enhancement after injection of contrast agent. Early wash out than its surrounding pancreas parenchyma could be observed. Thus confirming the neoplastic nature of the lesion, meanwhile sludge or stone in the distant part of CBD can be easily excluded. As the enhancement decreased gradually so that the tumor became hypo-enhanced during the late phase in CEUS, it was suggested to have relationship with the fibrous stroma within the tumor [1,3].

In our case, this benign tumor showed heterogeneous iso-enhancement both in contrast-enhanced CT and MRI. CT and MRI had distinguished advantages to demonstrate the accurate location and shape of the tumor, to get some information about whether the tumor was invasive or not [14–15].

Currently, as few cases of imaging features of CBD tubular adenomas have been described, it is clinically challenging to diagnose before operation. Radiologic imaging techniques, especially contrast enhanced CT, MRI and CEUS are useful for detecting and characterization tumor in the distant part of CBD [16].

Conclusion

Comparing to CT and MRI, CEUS has advantages such as no radiation, real time observation and more easily to be performed. However, the detection of lesion

in distal part of CBD is more operator dependent. When a homogeneous mass is detected in the distant part of CBD and shows heterogeneous enhancement both in contrast-enhanced CT, MRI and CEUS, CBD tubular adenomas should be included in the differential diagnosis.

Conflict of Interest

This work was supported by the National Natural Science Foundation of China (Grant No. 81571676 and 81501471). and supported by Program for Shanghai Outstanding Medical Academic Leader

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