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Case Report

Proximal Common Bile Duct Adenomyoma, - A Case Report

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ABSTRACT

Proximal common bile duct adenomyoma is a rare benign tumor. It is more common in female and elderly. It produces obstructive jaundice mimic to cholangiocarcinoma. It is usually evaluated by liver function test, tumor marker (CA19-9), Magnetic resonance cholangiopancreatogphy (MRI-MRCP), and endoscopic retrograde cholangiopancreatography (ERCP) with biopsy. Confirmatory diagnosis is almost impossible before surgery. Confirmation is only possible after surgery and histopathological examination. Recently we have encountered a 31 years old man who presented with obstructive jaundice. He was evaluated as usual and diagnosed as a case of hilar cholangiocarcinoma (Bismuth type I). Extrahepatic excision along with pericholedochal lymph node, lymph node around common hepatic artery and retro-duodenal lymph nodes were removed en-block and the operation was completed by Roux-en-Y reconstruction. Histopathological examination confirms the lesion as bile duct adenomyoma. The patient is symptom free without any recurrence at 3 years follow up after surgery. We are going to report the case as bile duct adenomyoma in a young male which is a very rare tumor for understanding its presentation, diagnosis and treatment in light of published literature.

Keyword: Proximal common bile duct, adenomyoma,

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Introduction

The adenomyoma is a localized non-neoplastic mass lesion and it is composed of bland dilated glandular elements interspersed among thick bundles of smooth muscle. It occurs most commonly in uterus and Gynecologist frequently deals this problem. Adenomyoma is also found in gallbladder sometimes and surgeon confuses frequently with gallbladder carcinoma. Incidence of benign bile duct tumors is 0.1% of all biliary tract surgery.[1] Among them adenomyoma in bile duct is extremely a rare condition. It is most commonly occurred in female. The ratio between male and female is 1:3. The mean age of biliary adenomyoma is 60 years.[2] It may remain asymptomatic but it usually produces obstructive jaundice if it presents at bile duct or ampulla.

Pre-operative diagnosis is challenging except ampullary adenoma where endoscopic biopsy can be done. Treatment of the adenomyoma depends on its site. We report here a young male with proximal common bile duct (CBD) adenomyoma (very rare) who was successfully treated by resection of supraduodenal part of CBD and Roux-en-Y hepaticojejunostomy. It was successfully treated by resection of supra-duodenal part of CBD followed by Roux en Y hepaticojejunostomy

Case Report

A 31 years old male presented with progressive jaundice for 2 months with intense itching, anorexia and weight loss. He also complained of recurrent attacks of high fever with chills and rigors which was associated with upper abdominal pain. He was deeply icteric and malnourished. Scratch marks were found all over the body. Gallbladder was palpable. There was no ascites and other organomegaly. Laboratory data showed; hemoglobin: 11.1 g/dl, ESR: 10 mm in 1st hour, WBC: 7500/mm3, serum total bilirubin: 34.0 mg/dl, alkaline phosphatase: 304 U/L, SGPT: 24 U/L, serum albumin: 26 g/L and CA19-9 level: 24 IU/ml, Ultrasonography of abdomen showed a hyper-echoic area at the proximal common bile duct (diameter of 9 mm). MRCP (Fig. 1) revealed a signal void area at the junction of common hepatic duct, cystic duct & proximal common bile duct. Intra-hepatic duct was grossly dilated and common bile duct was normal. Gallbladder was elongated and distended. On the basis of clinical, laboratory and imaging data he was diagnosed as cholangiocarcinoma (Bismuth-Corlette Type I) involving the proximal common bile duct. Per operative findings was a nodular soft tissue growth at the proximal common bile duct 2 cm below the confluence (Fig. 2). Tumor was free from surrounding structures. Lymph nodes along the hepatoduodenal ligament, common hepatic artery and retroduodenal area were found enlarged. We excised the supra-duodenal portion of common bile duct, gallbladder, the tumor, common hepatic duct about 1cm above the growth with enlarge lymph nodes as an en-block specimen. Biliary reconstruction was done in the form of Roux en Y hepatico-jejunostomy. Gross histological examination of the specimen showed a 0.7 mm thick growth at the junction of the common hepatic duct, cystic duct and proximal common bile duct. Microscopic examination showed the growth composed of hyperplastic proliferation of the glandular element and muscles with no evidence of malignant changes in cellular component. No malignant changes were noted in lymph nodes. On the basis of histopathological data the tumor was labeled as adenomyoma of upper bile duct (Figure: 3a-b). Patient was symptom free without any recurrence at 3 year follow up after operation. His liver function and tumor marker level was within normal limit.

Discussion

Although adenomyoma is relatively common in gallbladder, it is rare in bile ducts and it is extremely rare in proximal bile duct. In 2000, Beazley and Blumgart[3], reviewed 115 patients, of them the anatomic distribution of adenomyoma in bile duct were: 47% in ampulla or its close proximity, 27% in common bile duct, 15% in common hepatic duct, 8% in right and left hepatic duct, and 3% in cystic duct (Fig. 4).

Bedirli and his colleagues[4] reviewed literature from 1964 until 2000 and found 49 articles of 59 biliary adenomyoma. Among 59 biliary adenomas; 35 were found in gallbladder, 11 in extrahepatic biliary tree, 9 in ampulla of the Vater, and 4 in duodenum. Alejandro J. Pérez-Alonso and colleagues[5] had ten years study of adenomyoma of biliary tract. They found 24 patients with adenomyomatosis out of 5141 surgical specimens from 2000 to 2010 periods. Among the 24 patients of adenomyoma, 20 were located in fundus of the gallbladder, 2 in cystic duct and 2 in distal common bile duct. Therefore, recent and past published data shows that biliary adenomyoma is a rare tumor, particularly proximal common bile duct. As a rare one the patient is reported here to understand the overall management of adenomyoma of proximal bile duct.

Symptoms of adenomyoma vary depending on location of tumor. Patients with adenomyoma in the gallbladder are usually asymptomatic or have mild to moderate epigastric pain[6]. Patients with adenomyoma in ampulla of the Vater and biliary tree are usually present with obstructive jaundice and cholangitis[7]. Our case is presented with deep jaundice, intense inching, anorexia and weight loss which is similar with other report[7-8]. Preoperative diagnosis of biliary tract adenomyoma is a challenging job as the symptoms are almost similar with malignant mass. Even biochemical, radiological and imaging findings are also similar with malignant mass. However, if the adenomyoma is at the ampulla of vater, there is an option of

endoscopic biopsy[9]. Proximal common bile duct adenoma is really difficult to diagnosis prior to surgery. Intraoperative frozen section biopsy from the mass may differentiate whether the lesion is benign or malignant[2,10]. Sometimes preoperative endoscopic and radiologic evaluation and intraoperative frozen section biopsy can all prove but insufficient for differentiating between adenomyoma and a malignant tumor [4]. The adenomas are usually soft, hardly palpable and there is a little or no resistance during passing ductal probes, for that reason sometimes it is difficult to detect during the operation [11]. Imai et al.[12] states that histological examination is required for confirmation of adenomyoma. In histological point of view, most reported cases of adenomyoma revealed a lobular configuration of ducts and ductules without dysplastic change. The differentiating point between adenomyoma and adenocarcinoma are the glands with basally located monotonous nuclei and a lobular arrangement of small ducts without desmoplastic reaction may represent adenomyoma and with desmoplastic reaction may produce adenocarcinoma[13]. The treatment depends on the location and symptoms of the adenomyoma. Complete excision of the tumor and lymph node is recommended for excluding malignancy [12, 14]. It is safe to do total excision of the tumor with its surroundings are to have tumor free margin and biliary drainage procedure. If any lymph adenopathy is present, it should be excised and histopathological examination to find out any malignant foci [9]. Because of preoperative lack of confirmation of the diagnosis, benign tumor is treated aggressively as like as malignant one which are suspected to be malignant [11]

Conclusion

Adenomyoma of the bile duct is a rare disease. It is rarer in young and male patient. The proximally located adenomyoma is very rare. As our patient covers the all rare facts of adenomyoma of bile, diagnosis and treatment of the present case is discussed in this article. Complete excision of the bile duct adenomyoma and biliary reconstruction has an excellent outcome.

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Figure 1

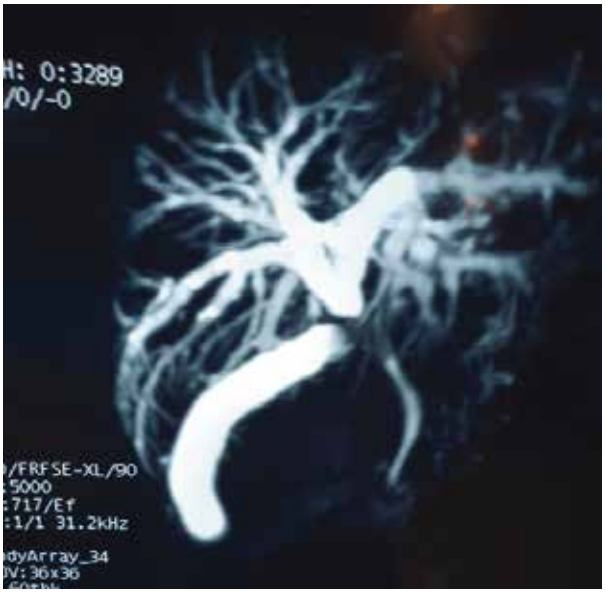


Figure-2

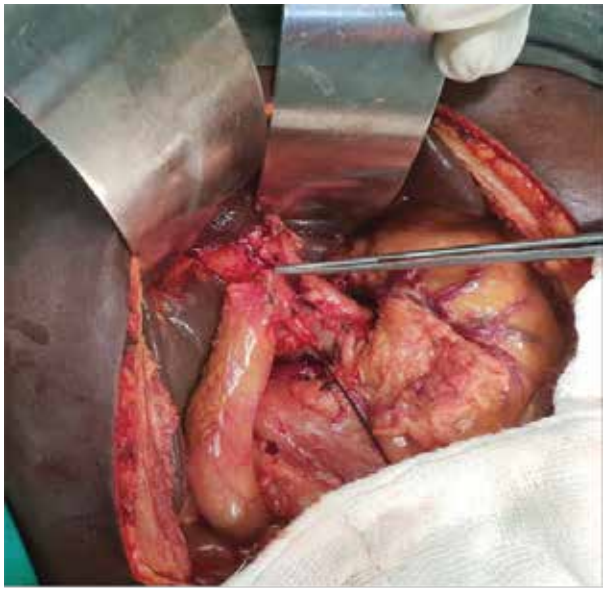
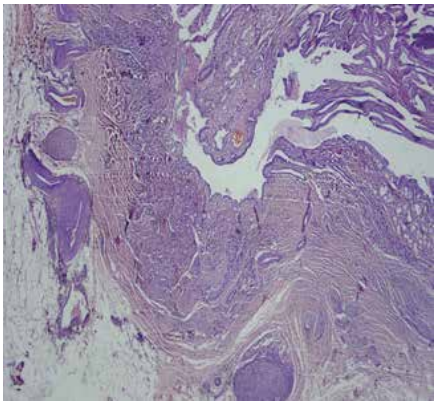


Figure-3

3a



3b

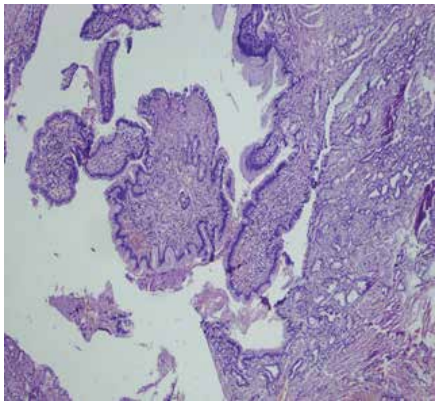


Figure-4

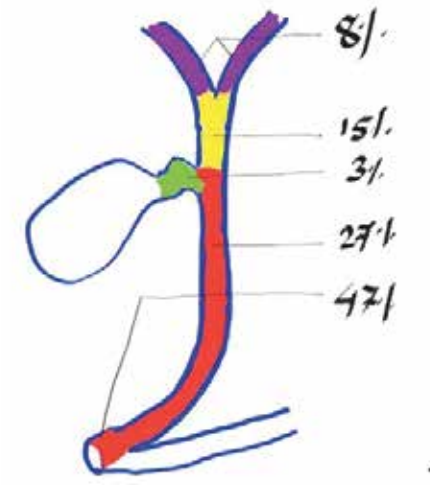


Figure Legends

Figure 1: MRCP shows signal void area at the proximal CBD and junction of cystic and hepatic duct.

Figure 2: Per-operative findings- tumor is indicated by the pointed area of the forceps. Gallbladder was elongated.

Figure 3a: Shows hyperplasia of the surface mucosa and dilated glands surrounded by hyperplastic smooth muscle and thick nerve bundle.

Figure 3b: Shows hyperplastic polyp with hyperplasia of the surface mucosa and dilated glands surrounded by hyperplastic smooth muscle and thick nerve bundle).

Figure 4: Site and percentage of bile duct adenomyoma.