Tubulopapillary adenoma of the common bile duct presenting with jaundice

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ABSTRACT

In this report, an adult patient with tubulopapillary adenoma of the common bile duct that manifested with jaundice is presented. Diagnostic challenges were analyzed. Although adenomas of the common bile duct are rare, they should be kept in mind in the differentiation of lesions of this region. It should be remembered that these lesions radiologically could mimic carcinoma and choledocholithiasis. Endoscopic resection should be considered as the primary method for treatment. Histopathology is the gold standard in diagnosis.

Keywords: Adenoma, tubulopapillary, common bile duct, histopathology, endoscopic resection

INTRODUCTION

Villous/tubulovillous adenomas are benign epithelial tumors usually detected in the colon. They often present by polyps elevated from the mucosa. These, mostly sporadic detected lesions can sometimes be a component of diseases such as familial adenomatous polyposis, Gardner's syndrome and Peutz-Jegher syndrome (1, 2). Villous/tubulovillous adenomas are rare in the common bile duct, and similar lesions of this localization are called papillary/tubulopapillary adenoma. General symptoms are bile duct obstruction and jaundice. They may be confused with common bile duct stones and malignant tumors during pre-operative evaluation (3, 4). Thus, unnecessary extensive surgical procedures can be applied in some cases. The definite diagnosis is only made by postoperative histopathologic examination of the surgical specimen. Herein, an adult male patient who presented with jaundice and had common bile duct tubulopapillary adenoma was presented with emphasis on diagnostic challenges.

CASE PRESENTATION

A 51-year-old male patient was admitted to the clinic with persistent jaundice and pain in the right hypochondriac region for the past week. On physical examination, the skin and mucosal surfaces were icteric. The right hypochondriac region was mildly tender on palpation. The laboratory examinations were characteristic for obstructive jaundice. The magnetic resonance cholangiopancreatography (MRCPG) showed intra- and extra-hepatic bile duct dilatation and a mass in the distal portion of the common bile duct causing obstruction that was compatible with a stone. The patient was prepared for endoscopic retrograde cholangiopancreatography (ERCPG). Informed consent was obtained from the patient and his relatives prior to the procedure. A polypoid lesion was observed in the ampulla of Vater during duodenoscopy. The lesion was extracted with snare polypectomy method after 1: 10000 diluted adrenaline injection to its base. The common bile duct was then cannulated by the ERCPG catheter and the contrast material was injected. The widest portion of the common bile duct dilatation was observed to be 2.0 cm in diameter and a filling defect was detected in the distal portion. A balloon catheter was inserted to the common bile duct and the lumen was cleared by expanding the proximal portion with air. Meanwhile, a short-pediced, dark pink polypoid lesion that was 1.5 x 1.0 cm in size was seen to move towards the intestinal lumen and was removed by applying snare polypectomy again (Figure 1). After the procedure, the patient's symptoms quickly regressed. Three days after the operation the patient's biochemistry tests were within normal limits. On histopathologic examination, the lesion was composed of villous and tubular structures consisting of a stroma of spindle cells lined with dysplastic single-layer columnar epithelium (Figure 2). Based on these findings, the patient was diagnosed as tubulopapillary adenoma of the common bile duct with mild dysplasia. Any recurrence or complication was not observed during the six-month follow-up period. Magnetic resonance cholangiopancreatography obtained four months after the procedure was normal. On follow-up duodenoscopy, ampulla of Vater appeared normal.

DISCUSSION

Adenomas are benign tumors composed of epithelial tissue, and are the most common lesions of the digestive tract. Generally, they appear as single, well-defined polypoid lesions. According to the World Health Organization classification there are five types of adenoma in the gallbladder and extrahepatic bile ducts: tubular, papillary, tubulopapillary, biliary cystadenoma, and papillomatosis (adenomatosis).

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Figure 1. Polypoid lesion and its resection within the common bile duct on endoscopic evaluation

Figure 2. Microscopic view of tubulovillous adenoma in the common bile duct (hematoxyline eosin; x40)

Adenomas are more common in the gallbladder than in the common bile duct. Adenomas are detected in 0.3-0.5% of cholecystectomy materials performed for chronic cholecystitis and cholelithiasis. Tubular adenomas are more common in the digestive system, while papillary/tubulopapillary adenomas are less frequent (5). These have the same morphologic characteristics as intestinal villous/tubulovillous adenomas, and have a high risk of malignancy (6, 7). The first papillary (villous) adenoma of the common bile duct in the English literature has been reported by Saxe et al. (9) and a total of 27 cases have been reported so far (3, 8).

Adenomas of the gallbladder and extrahepatic bile ducts are more common in women. Conversely, approximately 70% of common bile duct papillary/tubulopapillary adenomas are detected in men. The age range of cases presented in the English literature is 27-84, with an average age of 63.6 years (3). Adenomas usually present with jaundice, abdominal-especially right upper quadrant- pain, dyspepsia, nausea and vomiting symptoms similar to other lesions that cause common bile duct obstruction. Although the majority of adenomas in the ampullary region are found sporadically, they can sometimes manifest as a component of polyposis syndromes (1, 2).

The preoperative diagnosis of common bile duct adenomas is very difficult. The radiologic suspicion of malignancy in adenomas has been reported several times (4). Sometimes, as in our case, the adenoma could be perceived as a common bile duct stone in radiologic evaluations. Likewise, endoscopic examinations are insufficient in terms of ruling out malignancy. Malignancies can be correctly evaluated only in procedures performed by very experienced biliary endoscopists. In such cases, what makes endoscopic examination superior to radiologic evaluations is the possibility of obtaining biopsy for histopathologic examination. Histopathologic evaluation is the most reliable method in the diagnosis of adenoma. Histopathologically bile duct adenomas are composed of a dysplastic epithelium and stroma consisting of connective tissue such as their gastrointestinal tract counterparts. In situ carcinoma component was also determined in some biliary adenomas (3).

There is no consensus on the optimal treatment method of common bile duct adenomas of the ampullary region and the distal portion of the common bile duct (3). The implementation of endoscopic resection in patients with common bile duct adenoma with high-risk of malignancy has been first proposed in 1992 by Sturgis et al. (10). However, it was emphasized that the recurrence risk was high.

Similarly, local endoscopic resection (papillectomy) of ampullary region adenomas can be successfully carried out. The same method can be applied with sphincterotomy in distal intra-ductal adenomas, particularly in patients with suspected malignancy (11). Ariche et al suggested common bile duct resection along with hepatoduodenal ligament lymph node dissection in suspicious lesions of the middle portion of the common bile duct (7).

The prognosis of common bile duct adenomas is good. Nevertheless, the anatomical structure of this region restricts surgical procedures, which in turn leads to insufficient resection and recurrence in some cases. Careful histopathologic examination of the resection material is very important in terms of predicting prognosis.

CONCLUSION

Although common bile duct adenomas are rare, they must be kept in mind in the differential diagnosis of lesions of this region. It should be considered that they resemble carcinoma or common bile duct stones on radiological examinations in many cases. Endoscopic local resection should be considered as the main treatment method, and histopathologic evaluation is the gold standard for diagnosis.

Informed Consent: Written informed consent was obtained from patient's parents and patient who participated in this case.

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