

Adenomyoma of ampulla: a rare cause of obstructive jaundice

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ABSTRACT

Adenomyoma is a rare benign lesion occurring commonly in the fundus of the gallbladder in the biliary tract. Ampullary adenomyoma is rarer still, presenting as obstructive jaundice due to its location at the ampulla and may mimic malignancy on clinical and radiological examination. Endoscopic biopsy may not be always diagnostic if the lesion is deep seated. Histologically these lesions show a combination of diverse benign epithelial and mesenchymal elements, which should not be confused with an invasive tumor because of its arrangement. We report three cases of adenomyoma located at the ampulla presenting as obstructive jaundice. The diagnosis was confirmed on pancreaticoduodenectomy resection specimens.

INTRODUCTION

In the hepatobiliary system adenomyoma is rare outside the fundus of gallbladder but can arise throughout the entire biliary tree including papilla of Vater.⁽¹⁾ According to WHO classification, adenomyoma and adenomyomatous hyperplasia are defined as duct like structures accompanied by hyperplasia of smooth muscle cells.⁽²⁾ Despite its benign nature, presentation of ampullary adenomyoma as biliary obstruction leads to clinical misdiagnosis as carcinoma and is usually treated by extensive surgery. We present 3 cases of adenomyoma of ampulla presenting as obstructive jaundice for which pancreaticoduodenectomy was performed.

CASE REPORT

All pancreatico-duodenectomy cases received in the Department of Pathology for 3 consecutive years at our institute were reviewed and we found 3 cases of adenomyoma. All three patients were males with ages of 58 years, 65 years and 81 years and presented with abdominal pain, obstructive jaundice and loss of appetite. Ultrasonography showed dilatation of common bile duct in all 3 cases but no obvious mass in two (58Y male, 65Y male) and a nodule at ampulla measuring 1.5x1cm in third case (81Y male). Pylorus preserving pancreatico-duodenectomy was done in all three cases. Macroscopically the ampulla was firm and showed a well defined lesion measuring 0.8cm and 1.5cm respectively in our first and third case (58Y male, 81Y male). The second case (65Y male) had firmness at ampulla but did not show any lesion. Histology showed similar morphology in all cases. The lesion was composed of proliferating benign glands of varying size lined by gastric antral epithelium and separated by bands of fibromuscular tissue. Overlying mucosa was unremarkable.

DISCUSSION

Adenomyoma is a rare benign lesion arising anywhere in the gut with sign and symptoms depending on size and location. Patients with ampullary adenomyoma usually present with features of biliary obstruction and weight loss with rare reports of acute pancreatitis.(3)) Diagnosis is confirmed by histological examination which is not always possible on endoscopic biopsies particularly if lesions are not visible at the ampulla. ERCP image can be a useful diagnostic approach as it allows inspection of periampullary region and visualization of bile duct and pancreatic duct. Definite diagnosis is not easy to confirm before resection. People have used endoscopic snare removal in polypoidal lesions however this may sometimes cause pancreatitis as a complication.(4) Histologically the epithelial component can show pancreatic, gastric, intestinal or brunner gland tissue, which can be confused with ectopic pancreatic tissue, myoepithelial hamartoma, fibroadenoma, and mesenchymal lesions. The neoplastic or malformative origin of this lesion is still subject to controversy. It is considered incomplete or abortive variant of pancreatic heterotopia. The term ectopic pancreas is used if there is normal pancreatic differentiation. If the differentiation is into various abnormally arranged pancreatic and glandular elements, terms like adenomyoma, myoepithelial hamartoma, adenomyosis and foregut choristoma are used.(5) Awareness of adenomyoma is important as it is a rare lesion mimicking carcinoma clinically as well as radiologically. Pathologist should be familiar with morphological features so that it is not mistaken for well differentiated adenocarcinoma because of smooth muscle fibers running in between glands. Confirmation of diagnosis occurs on histopathological examination. Endoscopic biopsies may be helpful in superficial lesions but are not always diagnostic. Lesions located deeper will still need extensive resection.

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