Isolated Ampullary Adenoma Causing Biliary Obstruction

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Abstract

This is an interesting case of an isolated ampullary adenoma causing biliary obstruction that required surgical excision. We describe a patient who presented with a six month history of recurrent attacks of typical biliary pain radiating from the right upper quadrant of the abdomen to the back, nausea and vomiting, which we attributed to a large pedunculated tubulovillous adenoma. Abdominal ultrasound and endoscopic ultrasonography provided useful information in the diagnostic assessment of ampullary adenoma. Sporadic duodenal adenomas are an increasingly recognized condition in those with familial adenomatous polyposis syndromes as well as sporadic cases.

Keywords

Sporadic duodenal adenoma – intestinal ultrasound – endoscopic ultrasonography (EUS) – radical surgery.

Introduction

Adenomas of the major duodenal papilla, also known as ampullary adenomas, can occur sporadically or in the context of genetic syndromes such as familial adenomatous polyposis (FAP). These lesions have the potential to undergo malignant transformation to ampullary cancer \cite{1}.

The identification of duodenal and ampullary adenomas occurs in the setting of incidental esophagogastroduodenoscopic findings, surveillance of FAP, and in evaluation of symptomatic ductal obstruction. Because of the risk for malignant progression, similar to the transformation of colonic adenomas into adenocarcinomas, it is important to perform biopsy, resect, and survey these lesions. Sporadic duodenal adenomas, histologically mostly villous or tubulovillous, are an increasingly recognized condition in those with FAP syndromes as well as sporadic cases.

Adenomas of the major duodenal papilla account for 5\% of gastrointestinal neoplasms \cite{2} but are being identified more frequently with increasing use of upper endoscopy and ERCP.

ERCP and endoscopic ultrasonography (EUS) provide useful information in the assessment of ampullary adenomas. EUS and intraductal US (IDUS) have emerged as useful techniques to assess the depth of involvement in patients with ampullary neoplasms. These modalities allow the assessment of intraductal extension and extension beyond the muscularis propria and can allow evaluation of periampullary lymph nodes in those patients suspected of having cancer. EUS or IDUS of lesions that appear suspicious for harbouring cancer may help to select which patients can be considered candidates for endoscopic versus surgical therapy and in the guiding of the surgical therapy. EUS has been shown to be superior to CT, magnetic resonance imaging, or transabdominal ultrasonography for tumor staging \cite{3}.

Adenoma of the major duodenal papilla can be treated either endoscopically or surgically. Endoscopic treatment consists of endoscopic resection and thermal ablation.

The surgical options include transduodenal local excision (ampullectomy) and radical pancreatoduodenectomy. Surgical resection has been the mainstay for resection of adenomas of the major duodenal papilla \cite{4-6}.

Case report

A 66-year-old man presented 5 years after laparoscopic cholecystectomy with obstructive jaundice and a six month history of recurrent attacks of typical biliary pain radiating from the right upper quadrant of the abdomen to the back, nausea and vomiting. During these episodes he was found to have fluctuating abnormal biochemical liver test results, particularly for alkaline phosphatase (ALP). Between the attacks the liver tests returned to normal. Ultrasound scan evidenced only a mildly dilated bile duct, a feature commonly seen following cholecystectomy. Moreover, abdominal ultrasound demonstrated a large polyloid lesion...
of the second portion of duodenum with fluid collection anteriorly to it, accompanied by luminal substenosis (Fig. 1). Radial scanning of the same lesion with a 12-MHz probe revealed a voluminous “mulberry-form” polypoid lesion, 35 mm in diameter, with a heterogeneous hypoechogenic pattern, that protruded widely at the level of papilla of Vater from the mucosal surface remaining limited to this layer (Fig. 2). An ERCP showed no biliary calculi and confirmed mild bile duct dilatation.

Side-viewing upper endoscopy and biopsy were used to assess the polyp of the duodenal papilla. The histopathological examination showed a tubulovillous adenoma with low grade dysplasia. Diagnosis of adenoma was made by endoscopic findings, benign histologic findings, and EUS.

Although polypectomy during endoscopy has been used successfully to remove small intestinal polyps, this patient underwent surgical resection because of the large size of the polyp. Therefore, a transduodenal resection of ampulla and reconstruction were performed (Fig. 3). Postoperative course was without complications. The patient has not experienced biliary pain up to now.

**Fig 1.** US appearance with a convex probe of a large polypoid lesion of the second portion of duodenum with collection of surrounding fluid (arrows).

**Fig 2.** EUS finding of adenoma of the major duodenal papilla: a voluminous “mulberry-form” polypoid lesion originating from the mucosal surface remaining limited to this layer. The proper muscle layer is intact (arrows).

**Fig 3.** Appearance of the duodenal tubulovillous adenoma at laparotomy.
Discussion

The case is important, given the increased recognition of such lesions outside the context of polyposis syndromes. Sporadic duodenal adenoma is considered an uncommon finding. In a large series from Germany, only 6.9% of 378 duodenal polyps found at 25,000 upper gastrointestinal endoscopies were adenomatous [7]. Most adenomas were found incidentally at endoscopy but occasionally caused bleeding or obstruction of the duodenum or ampulla of Vater.

The distribution of small bowel adenomas shows a predominance at the ampulla and periampullary region, with decreased numbers proximally in the duodenum and distally in the small bowel, with a small peak in the distal ileum.

Duodenal adenomatous polyps are more common at gastroduodenal junction and have 3%-5% risk of developing adenocarcinoma in life time [8]. Tumors of the major duodenal papilla, also known as ampullary adenomas, are histologically mostly villous or tubulovillous adenomas. Brunner gland adenomas are very rare tumors and less than 150 cases have been reported in the English literature [9]. Size of the tumor is important in differentiating adenoma from Brunner gland hyperplasia. The size less than 1 cm is referred to as Brunner gland hyperplasia. Adenocarcinoma of the duodenum not originating from the region of ampulla is an uncommon neoplasm.

There is no consensus on which ampullary adenomas should be kept under surveillance and which lesions should be removed endoscopically or surgically but it is known that these lesions have the potential to undergo malignant transformation to ampullary cancer [10].

An incidental, small ampullary adenoma may not require further evaluation or therapy, depending on the clinical context; instead, lesions with high-grade dysplasia often warrant therapy because they may harbour malignancy missed at biopsy and to prevent progression to malignancy.

Treatment options for patients with advanced adenomas include endoscopic modalities and surgery [11]. Endoscopic papillotomy in FAP for papillary adenomas has been reported with long-term success rates in up to 67% of patients, and complications (including pancreatitis, bleeding, cholangitis, and, rarely, death) have been reported in up to 25% of patients [12]. Options besides snare polypectomy include endoscopic ablative techniques including multipolar electrocoagulation, laser, and argon plasma coagulation.

The high risk of recurrence of duodenal and periampullary polyposis with local therapy makes it an unattractive option for control of disease, and often it is followed by surgical therapy for definitive treatment [13].

Ampullary adenomas have historically been treated surgically. Surgical options include pancreaticoduodenectomy (Whipple’s procedure) or transduodenal ampullectomy.

We believe that because of high recurrence rates and a distinct frequency of malignant transformation of duodenal adenomas, a submucosal resection should only be performed in a high-risk patient. In these patients, the resected specimen has to be examined as accurately as possible to rule out a submucosal microinvasion. Radical surgery should be performed in all normal risk patients with juxtapapillary adenomas.

It is also necessary to give adequate emphasis to a clinically important association between sporadic duodenal adenoma and colorectal neoplasia.

The frequency with which a duodenal adenoma is associated with colorectal neoplasia is not well described. A previous clinicopathological study described 21 cases of duodenal adenomas, of which 11 underwent colonoscopy. Four of these 11 cases were classified as having FAP. Of the remaining 7 cases of sporadic duodenal adenomas, four (57%) were found to have colorectal neoplasia [14].

Another study involving the highest number of sporadic duodenal adenomas found that comparing the findings between patients with sporadic duodenal adenoma and a control group of patients presenting for endoscopies, colorectal neoplasias were significantly more common in the duodenal adenoma group [15].

A possible explanation for the association between duodenal adenoma and colonic neoplasia may be that they share common pathogenetic pathways. This may include genetic and/or environmental factors. The data on this problem are currently unclear. For instance, analyses of genetic mutational steps in duodenal and colorectal carcinogenesis show differences in the frequency and site of APC gene mutations while showing some positive correlations for late events, such as p53 mutations [16].

In conclusion, ampullary and duodenal adenomas have the potential for malignant transformation and require appropriate diagnostic evaluation. Moreover, patients with sporadic ampullary or duodenal adenomas are at increased risk for colon polyps and should be offered screening colonoscopy.

References


