CASE REPORT

A rare cause of dilated bile duct incidentally detected on imaging

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SUMMARY

Adenomas involving the extrahepatic bile ducts are rare benign tumours of the biliary tract. Given the asymptomatic nature of these tumours and the natural progression of biliary adenoma to carcinoma including cholangiocarcinoma, early detection is challenging. We present a case of an octogenarian woman with a remote history of gallstones who initially presented to the gastroenterology office for an incidental finding of biliary dilation, and whose subsequent imaging and biopsy suggested common bile adenoma. However, the patient was deemed a poor surgical candidate for pancreaticoduodenectomy, and unfortunately represented 5 months later with findings suggesting cholangiocarcinoma. This case provides a unique opportunity to study the natural progression of biliary adenoma to cholangiocarcinoma.

BACKGROUND

Adenomas involving the extrahepatic bile ducts are rare benign tumours of the biliary tract.^{1 2} Given the asymptomatic nature of these tumours and the natural progression of biliary adenoma to carcinoma including cholangiocarcinoma, early detection is challenging. We present a case of incidentally detected common bile duct (CBD) adenoma subsequently progressed to cholangiocarcinoma.

CASE PRESENTATION

An octogenarian woman with a remote history of gallstones presented to the gastroenterology office for an incidental finding of biliary dilation up to 19 mm detected on chest CT performed for evaluation of lung nodules. The patient denied abdominal pain, nausea, vomiting, jaundice or weight loss. Her medical history was significant for abdominal aortic aneurysm, coronary artery disease, peripheral vascular disease and chronic kidney disease.

INVESTIGATIONS

The bilirubin, alkaline phosphatase and transaminases were normal. Endoscopic ultrasound revealed a small soft tissue non-shadowing lesion in the distal CBD without evidence of a pancreatic head lesion (figure 1). Endoscopic retrograde cholangiopancreatography (ERCP) showed diffuse dilation of the biliary tree with a fixed filling defect without focal stricture (figure 2). Forcep biopsies obtained from the distal CBD revealed adenomatous epithelium with high-grade dysplasia (figure 3). A biliary stent was not placed due to normal liver tests.

Figure 1 Endoscopic ultrasound showed a small non-shadowing lesion in the distal bile duct (arrow). CBD, common bile duct; MPD, main pancreatic duct; HOP, head of pancreas.

TREATMENT

The patient was deemed a poor surgical candidate for pancreaticoduodenectomy and so was monitored clinically.

OUTCOME AND FOLLOW-UP

Five months after initial presentation the patient presented with jaundice, upper extremity bruising and steatorrhea. Her alkaline phosphatase was

Figure 2 Cholangiogram revealed diffuse biliary dilation with a small fixed filling defect just proximal to the ampulla without evidence of dominant stricture (arrow).

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Figure 3 Forcep biopsies revealed adenomatous epithelium with high-grade dysplasia.

elevated to 491 units/L and total bilirubin was 6 mg/dL. Repeat ERCP revealed a 15 mm occlusive stricture in distal CBD with diffuse proximal biliary dilation. Brush cytology showed atypical ductal cells suspicious for adenocarcinoma (figure 4). Repeat imaging did not reveal any enlargement of pulmonary nodules.

DISCUSSION

Biliary tract tumours are rare entities, more common in women than in men; they can occur in gallbladders or extrahepatic ducts.^{3 4} They can be classified according to the morphology or histology types. According to WHO classification, they are divided into five different types: tubular, papillary, tubulopapillary, biliary cystadenoma and papillomatosis.⁵ In a study that included 201 gallbladder adenomas, the most commonly found histology were pyloric, intestinal (tubular, papillary and tubulopapillary), foveolar and biliary.⁶ Meanwhile, adenomas of the extrahepatic biliary tree are incredibly sparse and intestinal histology is the commonest.⁵ Morphologically they are divided into papilloma, tubular adenoma, villous adenoma or tubulovillous adenoma.^{2 7} Rarely there can be fibromas and neuromas.¹ The presence of tubular histology confers a higher risk for malignant conversion.²

Adenomas in gallbladders are usually detected incidentally through cholecystectomies. Patients with extrabiliary adenomas, on the other hand, have symptoms similar to cholangiocarcinoma with obstruction of biliary trees causing jaundice, fever, abdominal pain, biliary colic and pruritus.² Our patient,

Figure 4 Cytology demonstrated normal ductal cells adjacent to atypical ductal cells.

however, was asymptomatic and the adenoma was detected incidentally. It has been postulated that biliary adenomas may follow the adenoma-to-carcinoma sequence, but given the rarity this is not firmly established.² Nevertheless in a recent study comparing 45 patients with intraductal papillary neoplasms of the bile duct to cholangiocarcinoma, it was found that the process of bile duct carcinogenesis correlates to stepwise activation of common oncogenic pathways.⁸

Therefore, the transformation of her adenoma to cholangiocarcinoma provides a unique opportunity to study the natural progression of biliary adenoma to cholangiocarcinoma. Although it could be argued that the initial biopsies may have missed an underlying malignancy, the patient's clinical course suggests benign disease at the time of initial imaging, followed by rapid progression to overt malignancy. Despite treatment, cholangiocarcinoma has a very poor prognosis, therefore surgery should be performed if CBD adenomas are detected.^{9 10} In cases where surgery is not an option, endoscopic resection has been described.¹¹ Radiofrequency ablation or photodynamic therapy have been shown to be safe in the treatment of malignant biliary obstruction and may be reasonable alternatives to surgery. However, this should be further evaluated in clinical trials.^{12 13} In our patient these options were not pursued due to her unwillingness to undergo multiple procedures. Instead she underwent placement of a fully covered, self-expanding, metal biliary stent for palliation.

Learning points

- ▶ Evaluation of postcholecystectomy bile duct dilation over 10 mm should include liver tests, endoscopic ultrasound and endoscopic retrograde cholangiopancreatography, if possible.
- ▶ If bile duct adenomas are detected, surgery should be considered given the poor prognosis of cholangiocarcinoma.
- ▶ If the patient is not a surgical candidate, radiofrequency ablation or photodynamic therapy may be some alternatives which need further investigation.

Contributors KPL analysed the data, reviewed the literature and wrote the manuscript. DN analysed the data, developed the pathological images and reviewed the manuscript. DD designed, revised and developed radiological images and critically reviewed the manuscript for important intellectual content. VM coordinated, designed, revised and developed radiological images and critically reviewed the manuscript for important intellectual content.

Competing interests None.

Patient consent Obtained.

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REFERENCES

- 1 Fletcher ND, Wise PE, Sharp KW. Common bile duct papillary adenoma causing obstructive jaundice: case report and review of the literature. *Am Surg* 2004;70:448–52.
- 2 Kim B, Joo S, Joo K. Carcinoma in situ arising in a tubulovillous adenoma of the distal common bile duct: a case report. *World J Gastroenterol* 2008;14:4705–8.
- 3 Sang X, Sun Y, Mao Y, et al. Hepatobiliary cystadenomas and cystadenocarcinomas: a report of 33 cases. *Liver Int* 2011;31:1337–44.
- 4 Meng X, Li J, Zhang W, et al. [Intrahepatic biliary cystadenoma: experience with 10 consecutive cases at a single center]. *Nan Fang Yi Ke Da Xue Xue Bao* 2011;31:1733–6.
- 5 Hamilton SR, Aaltonen LA. Pathology and genetics of tumours of the digestive system. In: *WHO classification of tumors. Pathology and genetics of tumours of digestive system*. Hamilton SR, Aaltonen LA, eds. Lyon, France: IARC Press, 2000:204.

- 6 Albores-Saavedra J, Chablé-Montero F, González-Romo MA, *et al.* Adenomas of the gallbladder. Morphologic features, expression of gastric and intestinal mucins, and incidence of high-grade dysplasia/carcinoma in situ and invasive carcinoma. *Hum Pathol* 2012;43:1506–13.
- 7 Lee SH, Lee DS, You IY, *et al.* [Histopathologic analysis of adenoma and adenoma-related lesions of the gallbladder]. *Korean J Gastroenterol* 2010;55:119–26.
- 8 Schlitter AM, Born D, Bettstetter M, *et al.* Intraductal papillary neoplasms of the bile duct: stepwise progression to carcinoma involves common molecular pathways. *Mod Pathol* 2014;27:73–86.
- 9 Wirasorn K, Ngamprasertchai T, Chindapasirt J, *et al.* Prognostic factors in resectable cholangiocarcinoma patients: carcinoembryonic antigen, lymph node, surgical margin and chemotherapy. *World J Gastrointest Oncol* 2013;5:81–7.
- 10 Farley DR, Weaver AL, Nagorney DM. 'Natural history' of unresected cholangiocarcinoma: patient outcome after noncurative intervention. *Mayo Clin Proc* 1995;70:425–9.
- 11 Sturgis TM, Fromkes JJ, Marsh W Jr. Adenoma of the common bile duct: endoscopic diagnosis and resection. *Gastrointest Endosc* 1992;38:504–6.
- 12 Steel AW, Postgate AJ, Khorsandi S, *et al.* Endoscopically applied radiofrequency ablation appears to be safe in the treatment of malignant biliary obstruction. *Gastrointest Endosc* 2011;73:149–53.
- 13 Pereira SP, Aithal GP, Ragunath K, *et al.* Safety and long term efficacy of porfimer sodium photodynamic therapy in locally advanced biliary tract carcinoma. *Photodiagnosis Photodyn Ther* 2012;9:287–92.

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