

BRIEF REPORTS

Endoscopic resection of a choledochocele

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Choledochoceles or type III choledochal cysts are cystic dilations of the intraduodenal portion of the common bile duct. Most choledochoceles are proximal to the ampullary orifice and can be treated with endoscopic sphincterotomy.¹ Diverticular choledochoceles communicate with the bile duct via a narrow channel in the ampulla, and most of the cyst may extend into the duodenum distal to the ampullary orifice; such lesions are usually resected surgically. We present a method for endoscopic resection of a diverticular choledochocele.

CASE REPORT

The patient, a 35-year-old woman, had a long history of postprandial epigastric pain and pressure. She had not had jaundice or overt pancreatitis but did have occasional fever. The patient underwent laparoscopic cholecystectomy a year previously for similar pain associated with nausea and vomiting. Although some of her symptoms abated, the pain persisted and a subsequent endoscopy and CT showed a duodenal mass. CT also showed extensive fatty replacement of the pancreas. She was referred to our institution for EUS.

Endoscopy disclosed a large pendulous mass arising anterior to the ampullary orifice and hanging down into the distal second duodenum (Fig. 1). It appeared to be covered by normal mucosa. EUS showed that the mass was a large, thin-walled cyst, with its proximal end merging with the ampulla. The entire lesion was superficial to the duodenal submucosa, which appeared normal (Fig. 2). The cyst wall was composed of three layers. A communication from the cyst to the intra-ampullary ducts could not be demonstrated sonographically, although the intra-ampullary ducts coursed close to the cyst. A diverticular choledochocele was suspected because of the lesion's proximity to the ampulla.

Because diverticular choledochoceles join the intra-ampullary ducts via a narrow diverticular neck (Fig. 3), endoscopic sphincterotomy would prevent further filling of the cyst but was unlikely to adequately drain the lesion. Endoscopic therapy was planned with the goal of resecting the cyst and marsupializing its neck.

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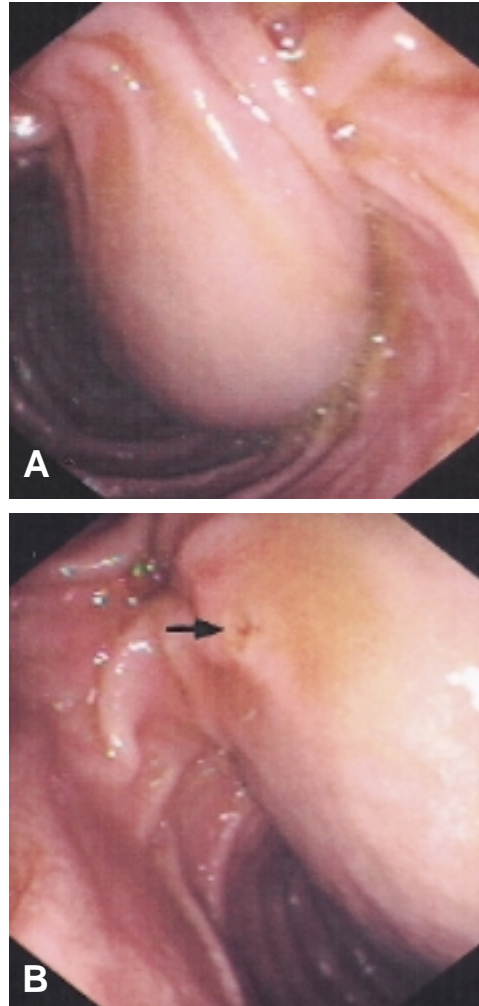


Figure 1. A, Endoscopic view of pendulous mass in second duodenum. B, Arrow points to ampullary orifice.

At ERCP, the lesion filled via a narrow neck that communicated with the common channel in the tip of the ampulla (Fig. 4), thus confirming the diagnosis of a diverticular choledochocele. The pancreatic duct was prominent. During the same endoscopic procedure, the cyst was aspirated using a 23-gauge sclerotherapy needle and 25 mL of bilious-appearing fluid was removed. Subsequent analysis of the fluid showed a bilirubin level of 16 mg/dL and an amylase level of 33,900 U/dL. Aspiration deflated the cyst enough to allow placement of a large polypectomy snare around it. The snare was closed around the neck of the cyst, taking care to come across the lesion below the ampullary orifice (Fig. 5). The cyst was then removed with snare cautery, leaving an opening in the ampulla anterior to the ampullary orifice (Fig. 6).

Histologic examination of the resected specimen showed a benign cyst. The wall was composed of three layers: a thin muscularis mucosa lined with small intestinal (duodenal) mucosa externally and internally.

The patient felt well after the cyst resection and was discharged home the same day. She continued to do well

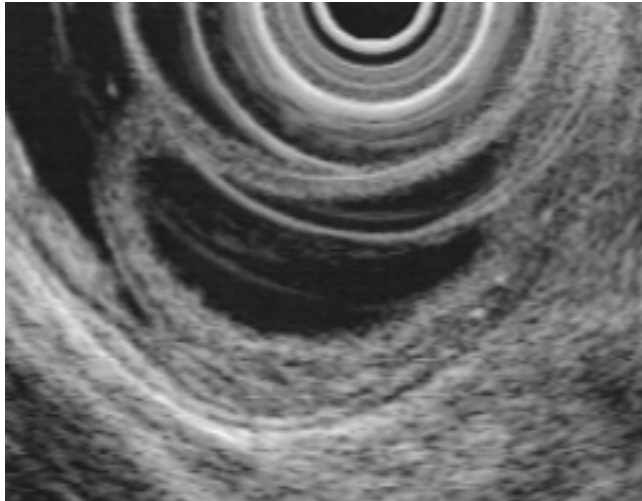


Figure 2. The proximal end of the cyst as seen by EUS. The lesion is superficial to the duodenal submucosa.

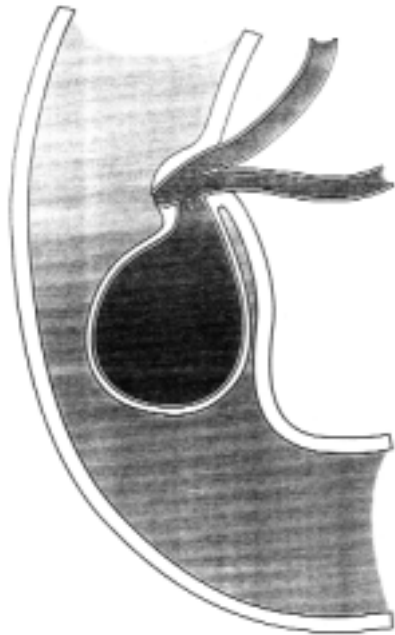


Figure 3. Schematic diagram of a diverticular choledochoceles.

1½ years later, with complete resolution of all her previous symptoms except for mild symptoms of gastroesophageal reflux. A follow-up CT of the abdomen showed no residual abnormality in the duodenum.

DISCUSSION

Choledochoceles are cystic dilations of the intraduodenal portion of the common bile duct and are the third subtype of biliary cysts in the modified classification of Alonso-Lej et al.² Choledochoceles are further subdivided into two major types. In type A, the bile duct opens into the choledochoceles, which in turn communicates with the duodenum via the

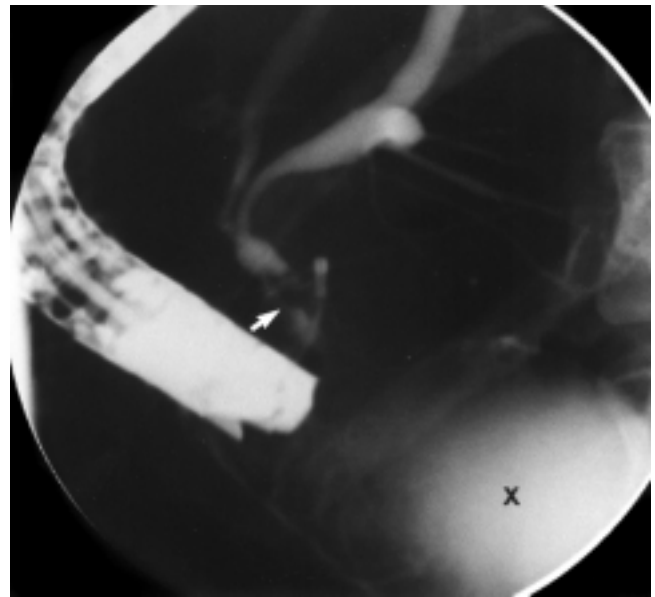


Figure 4. Retrograde cholangiogram showing a diverticular choledochoceles; arrow points to the neck of cyst, and x indicates the contrast-filled cyst.

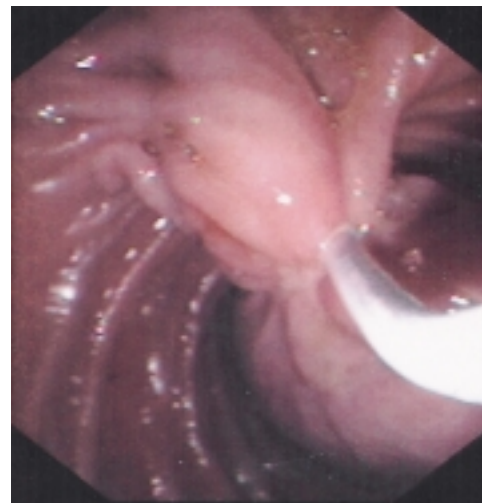


Figure 5. Endoscopic view of snare tightening around the neck of the cyst.

ampullary orifice or another small opening. In type B, the bile duct opens directly into the duodenum, and the choledochoceles communicates with the distal common bile duct via a small neck in a diverticular fashion.³ Our patient had the IIIB or diverticular type of choledochoceles. This lesion is sometimes referred to as a duodenal duplication because it is lined by duodenal-type mucosa; we favor the term diverticular choledochoceles because the lesion communicates with the ducts and is filled with bile and pancreatic juice, not duodenal secretions.

Choledochoceles can usually be treated with endoscopic sphincterotomy, which unroofs the

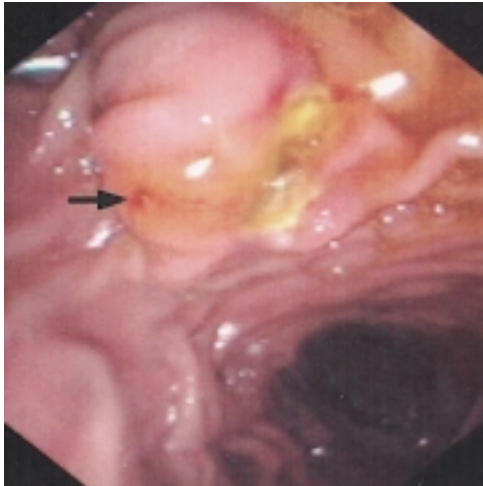


Figure 6. Postresection endoscopic view of the ampulla; arrow points to the ampullary orifice.

cyst.^{1,4-9} Surgical sphincteroplasty or excision are also well described.^{2,9,10} Large diverticular choledochoceles are generally treated surgically, particularly when the cyst extends below the ampullary orifice, as in this case. Sphincterotomy might not drain this type of cyst and would not reduce the risk of lithiasis in the cyst^{2,6} or rarely carcinoma^{11,12} in the undrained lesion. Although endoscopic incision and drainage of type IIIB cysts have also been reported,¹ we chose to resect this choledochocoele, more closely mimicking the surgical approach to this lesion. Snare resection was only possible after partial deflation of the cyst with a sclerotherapy needle.

EUS has been previously used to diagnose and guide therapy of choledochoceles,^{6,13-15} and it played an important role in the endoscopic treatment of our patient. EUS demonstrated the cystic nature of this lesion and also showed that it was superficial to the duodenal wall. Although the back wall of the cyst was not visible endoscopically, EUS visualized the entire cyst, thereby allowing us to proceed with endoscopic resection.

Endoscopic aspiration and snare resection are a novel approach in the treatment of diverticular choledochoceles. The combination of EUS and ERCP permits a rational diagnostic and therapeutic approach, avoiding resection of a lesion that involves deeper

layers of the duodenal wall. It appears that, when favorable anatomy is documented, these rare lesions can be successfully managed by endoscopic methods.

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REFERENCES

1. Dunham F, Engelholm L, Tossaint J, Deltenre M, Lambilliotte JP, Cremer M. Investigation des dilatations Kystiques idiopathiques du choledoque par cholangio-pancreatographie. *Acta Gastroenterol Belg* 1981;44:274-83.
2. Alonso-Lej F, Rever W Jr, Pessagno D. Congenital choledochal cyst with a report of 2 and an analysis of 94 cases. *Int Abstr Surg* 1959;108:1-30.
3. Chi-loo Lu S, Kaplowitz N. Diseases of the biliary tree. In: Yamada T, editor. *Textbook of gastroenterology*. Philadelphia: JB Lippincott; 1995.
4. Siegel J, Harding G, Chateau F. Endoscopic incision of choledochal cysts (choledochocoele). *Endoscopy* 1982;13:200-2.
5. Schmidt H, Bauer J, Wiessner V, Schonekas H. Endoscopic aspects of choledochocoeles. *Hepatogastroenterology* 1996;43:143-6.
6. Tanno S, Maguchi H, Obara T, Fujii T, Santos S, Itoh N, et al. Endoscopic treatment of gallstone-impacted choledochocoele precisely diagnosed by endoscopic ultrasonography. *Endoscopy* 1996;28:470-1.
7. Venu R, Geenen J, Hogan W, Dodds W, Wilson S, Stewart E, et al. Role of endoscopic retrograde cholangiopancreatography in the diagnosis and treatment of choledochocoele. *Gastroenterology* 1984;87:1144-9.
8. Gerritsen J, Janssens A, Kroon H. Choledochocoele: treatment by endoscopic sphincterotomy. *Br J Surg* 1988;75:495-6.
9. Martin RF, Biber BP, Bosco JJ, Howell DA. Symptomatic choledochocoeles in adults: endoscopic retrograde cholangiopancreatography recognition and management. *Arch Surg* 1992;127:536-9.
10. Sarris GE, Tsang D. Choledochocoele: case report, literature review and proposed classification. *Surgery* 1989;105:408-14.
11. Donald JJ, Coral A, Lees WR. Choledochocoele complicated by carcinoma. *Clin Radiol* 1989;40:101-3.
12. Ozawa K, Yamada T, Matumoto Y, Tobe R. Carcinoma arising in a choledochocoele. *Cancer* 1980;45:195-7.
13. Tio T, Rohde P, Sie L, Tytgat G. Endosonography in the pre-operative diagnosis of choledochocoele. *Gastrointest Endosc* 1992;38:381-3.
14. Meyenberger C, Bertschinger P, Wirth H, Marincek B, Bischof T, Ammann R. Dilatation of the common bile duct: what does endoscopic sonography contribute? *Schweiz Med Wochenschr* 1994;124:642-8.
15. Avunduk C, Weiss R, Hampf F, Navab F. Obstructing choledochocoele: diagnosis by endoscopic ultrasound. *Abdom Imaging* 1995;20:72-4.