Ampullectomy of an unusual lesion developing 20 years after endoscopic treatment of a type III choledochocele

A type III choledochocele is a rare congenital cystic dilation of the intraduodenal portion of the main bile duct, representing 5% of all choledochal cysts according to the Toldani classification [1]. Choledochoceles may cause biliary obstruction and carcinoma development with a reported incidence of 2.5%. To date, endoscopic treatment remains the less invasive technique, which consists of cystic mass removal associated with sphincterotomy [2,3]. However, the risk of carcinoma development may persist even after treatment and reports of long-term outcomes are lacking [4].

Herein we report the original case of a 78-year-old woman presenting with a previous history of sphincterotomy for a type III choledochocele 20 years ago (> Video 1). She presented with a 12-mm low-grade dysplasia adenoma arising from the sphincterotomy scar (> Fig. 1), mimicking an ampulloma. To the best of our knowledge, such a lesion has yet to be reported. Imaging modalities (endoscopic ultrasound and magnetic resonance imaging) revealed a 9-mm moderate dilation of the main bile duct upstream of a distal stenosis (> Fig. 2).

A new endoscopic procedure was performed. The patient was placed in a dorsal position under general anesthesia. Similar to an ampullectomy, a duodenoscope under CO2 insufflation was used. An en bloc resection was performed using a 15-mm braided snare (Lariat; Life Partners Europe, Bagnolet, France) and the specimen was immediately retrieved (Fig.3). Cholangiography revealed a delayed clearance of the contrast due to the distal stenosis (▶ Fig.4). Complementary intraductal biopsies of the stenosis were performed using a pediatric biopsy forceps. Prevention of post-ampullectomy pancreatitis was ensured by intrarectal indometacine ad-





▶ Video 1 Ampullectomy of an unusual lesion that developed 20 years after endoscopic treatment of a type III choledochocele.



► Fig. 1 Endoscopic view of the ampulloma-like lesion involving the biliary tract and the whole ampulloma mass.

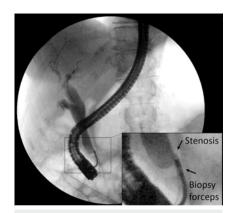


► Fig. 2 Endoscopic aspect of the duodenal wall after resection.

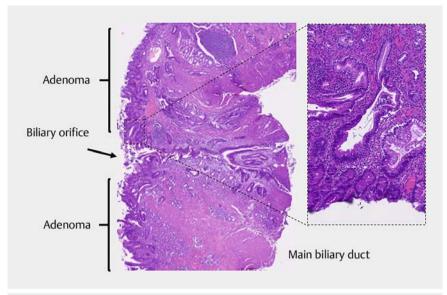
ministration, pancreatic stenting with a 5-Fr×5-cm plastic stent, and ringer lactate hyperhydration. The patient was discharged after 24h without complications.

Final histology confirmed the R0 resection of a low-grade dysplasia adenoma. Biopsies of the stenosis revealed only fibrotic tissue. The 4-month follow-up duodenoscopy confirmed the absence of local recurrence.

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▶ Fig. 3 Additional biopsies of the stenosis were guided by cholangiography. Cholangiography revealed a distal stenosis of the main bile duct. Biopsy sampling of the stenosis was performed using pediatric biopsy forceps.



▶ Fig. 4 Histological features of the ampulloma-like resection. Hematoxylin and eosin staining revealed a tubulous adenoma with low-grade dysplasia arising and surrounding the biliary orifice.

Competing interests

The authors declare that they have no conflict of interest.

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References

- [1] Todani T, Watanabe Y, Narusue M et al. Congenital bile duct cysts: classification, operative procedures, and review of thirtyseven cases including cancer arising from choledochal cyst. Am J Surg 1977; 134: 263–269
- [2] Culetto A, Miranda-Garcia P, Tellechea JI et al. Endoscopic treatment of a choledochocele. Endoscopy 2016; 48: E286
- [3] Lobeck IN, Dupree P, Falcone RA et al. The presentation and management of choledochocele (type III choledochal cyst): a 40-year systematic review of the literature. J Pediatr Surg 2017; 52: 644–649
- [4] Kim TH, Park JS, Lee SS et al. Carcinoma arising in choledochocele: is choledochocele innocent bystander or culprit? Endoscopy 2002; 34: 675–676

Bibliography

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