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A Case of Ampullary Adenoma that Developed to Cancer 7 Years After Initial Diagnosis

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Conflict of interest: None declared

Patient: Male, 81
Final Diagnosis: Ampullary cancer
Symptoms: Jaundice
Medication: —
Clinical Procedure: Endoscopy
Specialty: Gastroenterology and Hepatology

Objective: Unusual clinical course





Background: Although ampullary adenomas have been reported to be considered as precancerous lesions, there have been very few reports of cases in which cancer occurred after long-term follow-up. We herein report a case of ampullary adenoma that developed to cancer after long-term observation.

Case Report: An 81-year-old man was referred to our hospital due to a tumor at the ampulla of Vater. Histological examination revealed a tubular adenoma. Because the patient refused treatment, follow-up by duodenoscopy, EUS, MRCP, and forceps biopsy was planned. There was no change in the tumor for 6 years. Seven years after the initial diagnosis, he developed from jaundice. Duodenoscopy showed an easy-bleeding, reddish, uneven surface area of the tumor and NBI demonstrated an irregular, non-structured surface pattern. EUS demonstrated invasion of the duodenal muscularis and infiltration into the bile duct. Histological examination revealed a well-differentiated adenocarcinoma.

Conclusions: The clinical course of this case provides evidence of the adenoma-carcinoma sequence.

MeSH Keywords: Adenocarcinoma • Adenoma • Ampulla of Vater • Carcinogenesis • Narrow Band Imaging

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Background

Although ampullary adenomas have been reported to be considered as precancerous lesions [1,2], there have been very few reports of cases in which cancer occurred after long-term follow-up. In cases of familial adenomatous polyposis (FAP), long-term observation is usually performed in accordance with the endoscopic surveillance program. Recently, the usefulness of magnified endoscopic examination, such as narrow-band imaging (NBI), for ampullary adenoma has been reported [3,4]. However, there have been very few reports of

ampullary adenoma in which carcinogenesis could be confirmed by long-term observation with NBI. We herein report a rare case of ampullary adenoma that developed to cancer after long-term observation, in which the change of endoscopic findings was confirmed by NBI.

Case Report

An 81-year-old man was referred to our hospital due to a tumor at the ampulla of Vater. Duodenoscopy revealed a discolored and partially reddish-colored, lobular, protruding tumor at the ampulla of Vater (Figure 1A). NBI demonstrated an oval-shaped, pinecone/leaf-shaped villi pattern around the ampullary

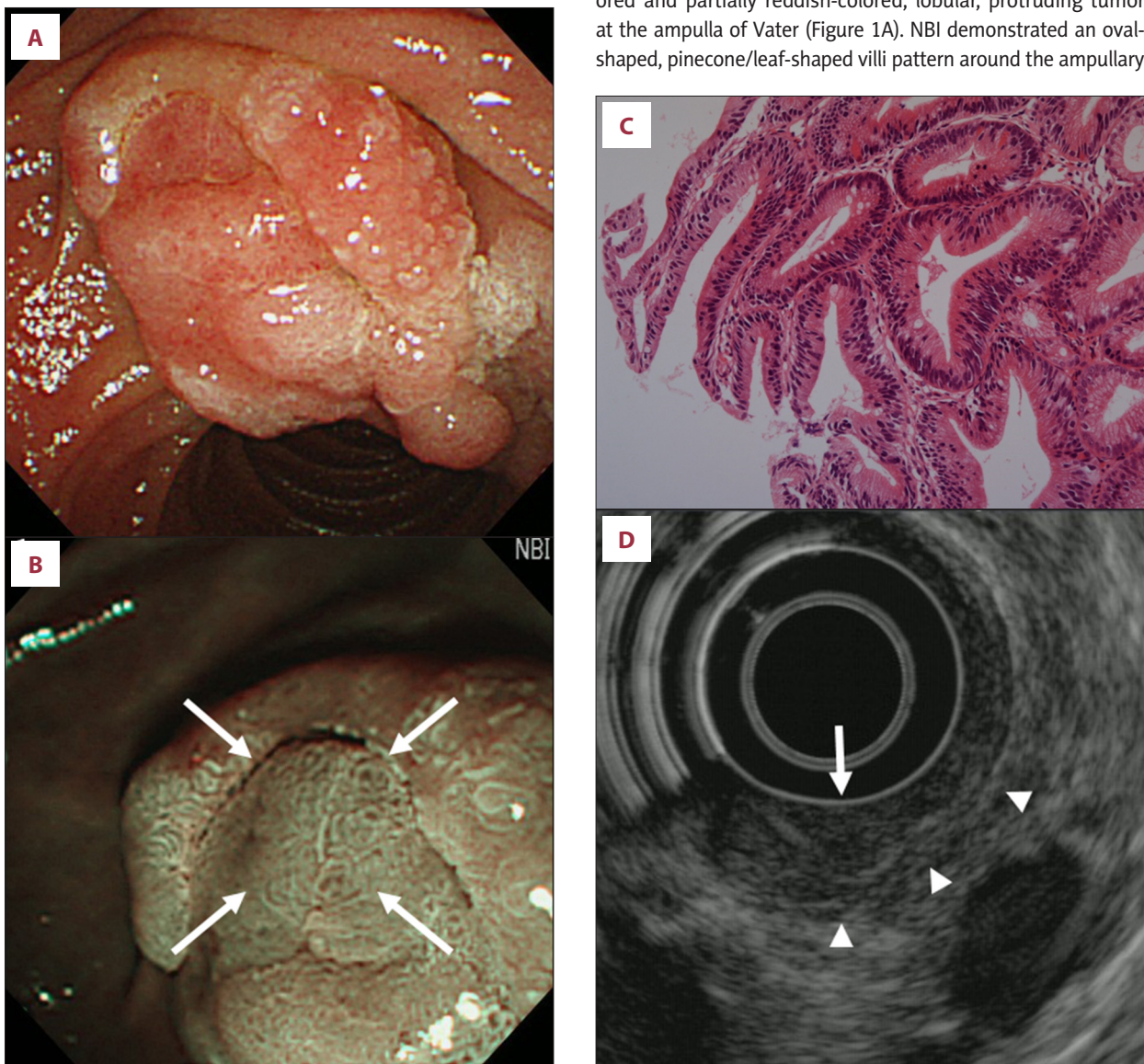


Figure 1. (A) Duodenoscopy: a discolored and partially reddish, lobular, protruding tumor at the ampulla of Vater. (B) Narrow-band imaging (NBI) revealed an oval-shaped, pinecone/leaf-shaped villi pattern (arrow). (C) Histopathological findings of the biopsy specimen from both the surface and ampullary orifice showed tubular adenoma. (D) Endoscopic ultrasonography visualized a hypoechoic mass (arrow) limited to the ampulla of Vater without invasion of the duodenal muscularis propria layer (arrowhead).

orifice (Figure 1B). Histological evaluation by forceps biopsy from both the surface and ampullary orifice showed a tubular adenoma (Figure 1C). Magnetic resonance cholangiopancreatography (MRCP) showed no dilation of the bile duct (BD) or of the pancreatic duct (PD). Endoscopic ultrasonography (EUS) did not detect ductal infiltration by the tumor into the BD or the PD (Figure 1D). He had no history of familial adenomatous polyposis (FAP). Since the patient refused treatments, such as endoscopic papillectomy and pancreato-duodenectomy, follow-up was planned. EUS, duodenoscopy, and forceps biopsy demonstrated no change for 2 years after the initial diagnosis. Only MRCP was performed due to advanced age, showing no change of the tumor for a further 4 years.

He developed jaundice 7 years after the initial diagnosis. Duodenoscopy revealed an easy-bleeding, reddish, uneven surface area of the tumor (Figure 2A). NBI demonstrated an irregular, non-structured surface pattern of the reddish area (Figure 2B). Forceps biopsy from the ampullary orifice showed well-differentiated adenocarcinoma (Figure 2C). EUS demonstrated a hypoechoic mass at the ampulla of Vater with invasion of the duodenal muscularis and the BD (Figure 2D). CT and MRI revealed no distant metastasis. Since he once again refused surgical treatment, endoscopic biliary stenting was performed.

Discussion

Ampullary adenoma associated with familial adenomatous polyposis (FAP) has been reported to have a low risk for the

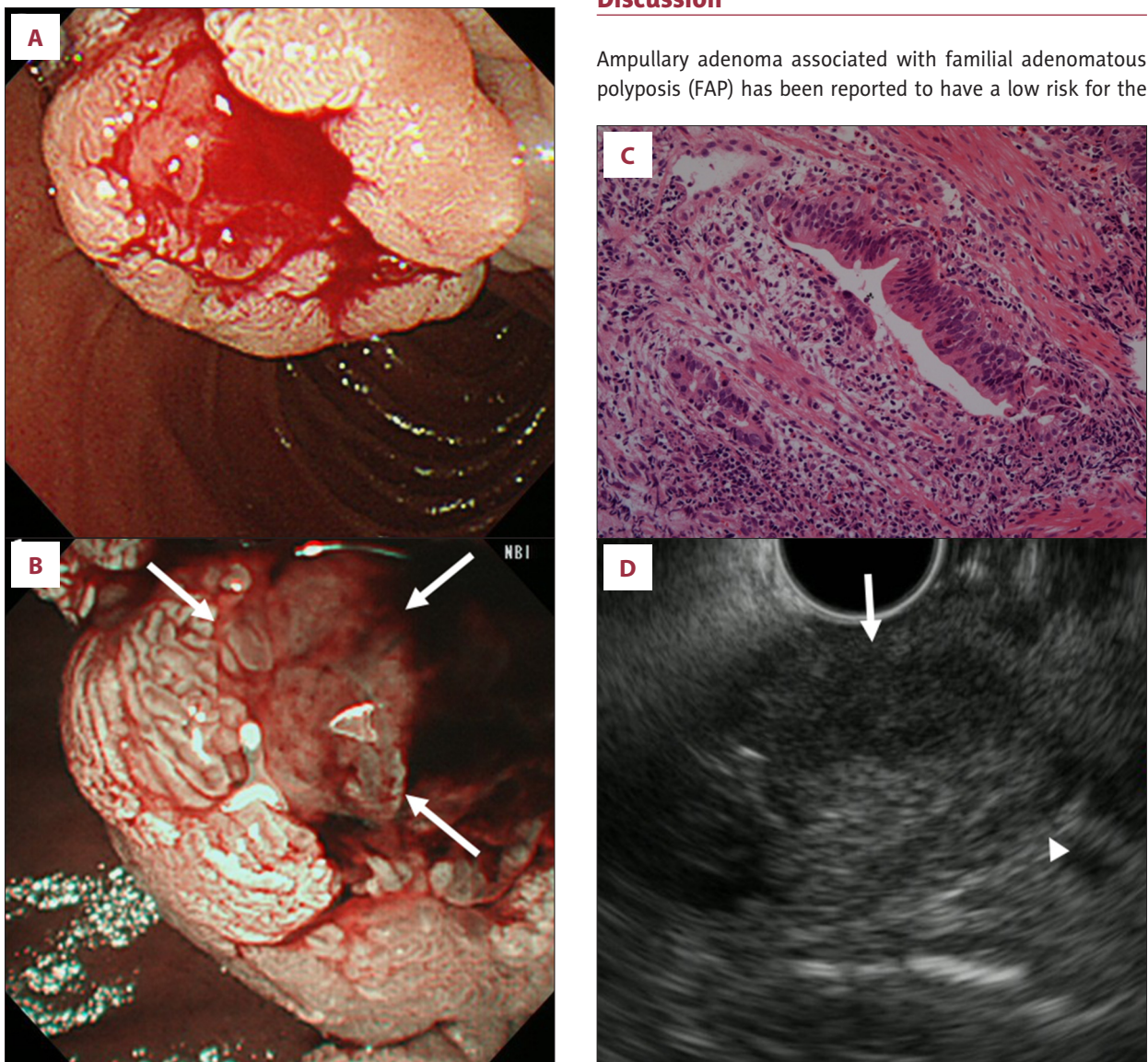


Figure 2. (A) Duodenoscopy: an easy-bleeding, reddish, uneven surface area was observed around the BD orifice. (B) NBI revealed an irregular and non-structured surface pattern (arrow). (C) Histopathological findings of the biopsy specimen from the ampullary orifice showed well-differentiated adenocarcinoma. (D) Endoscopic ultrasonography (EUS) visualized a hypoechoic mass (arrow) with invasion of the duodenal muscularis propria layer (arrowhead).

development of ampullary cancer [5–7]. In contrast, the natural history of sporadic ampullary adenoma remains unclear. Because of the high percentage of coexisting adenoma structures in ampullary adenocarcinomas, the adenoma-carcinoma sequence applies to the ampulla of Vater [2]. However, reports regarding carcinogenesis of ampullary adenoma after long-term follow-up are extremely rare. Based on the PubMed database and Japanese literature database 'Igakuchuou-Zasshi' with the key words 'ampullary adenoma' and 'cancer', only 1 case of sporadic ampullary adenoma that developed to cancer after long-term observation was reported from 1983 to 2014 [8]. Misawa et al. [8] reported a case of ampullary adenocarcinoma diagnosed by biopsy as having developed tubular adenoma 3 years before the diagnosis. In this case, the diagnosis of development to cancer from adenoma was based on the appearance of jaundice in the same way as in the present case. Carcinogenesis was suggested to have developed from the ampullary common duct (Ac) and to have invaded the ampulla Vater and sphincter of Oddi, with subsequent infiltration of the muscularis propria layer of the

duodenum. There are few reports of NBI findings for ampullary tumors. Uchiyama et al. [3] classified the surface structures of the ampullary lesions using magnifying endoscopy with NBI into 3 groups: 1) oval-shaped villi, 2) pinecone/leaf-shaped villi, and 3) irregular/non-structured. All cases of adenoma and adenocarcinoma had type II/III surface structures. In the present case, at the initial diagnosis, NBI demonstrated an oval-shaped, pinecone/leaf-shaped villi pattern (type I). Seven years later, the non-structured surface pattern (type III) around the ampullary orifice was shown by NBI and the biopsy specimen confirmed adenocarcinoma. NBI findings were thought to correspond to histological findings.

Conclusions

In our present case, the development to cancer from adenoma was demonstrated by the endoscopic appearance with NBI after long-term follow-up. The clinical course of this case provides evidence of the adenoma-carcinoma sequence.

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