Peroral cholangioscopy of programmed cell death-1 inhibitor-related sclerosing cholangitis: three case reports

Programmed cell death-1 (PD-1) inhibitor therapy is indicated for many types of malignancies, but can lead to immune-related adverse events [1, 2]. Recently, PD-1 inhibitor-related cholangitis and its clinical features, including peroral cholangioscopy, were reported [3–5]. However, the diagnostic criteria of PD-1 inhibitor-related cholangitis are unclear. We herein report peroral cholangiography for three patients with suspected pembrolizumab-related sclerosing cholangitis.

Case #1 was a 61-year-old man with bladder cancer who had been treated with pembrolizumab as third-line chemotherapy. After his fifth course of pembrolizumab, he was admitted to our hospital complaining of pyrexia. Case #2 was an 89-year-old man with bladder cancer. After his fourth course of pembrolizumab as second-line chemotherapy, he was admitted to our hospital for examination of liver dysfunction. Case #3 was a 63-year-old man with lung cancer. After his seventh course of pembrolizumab as first-line chemotherapy, he was admitted to our hospital because of liver dysfunction.

Laboratory data from all three patients revealed increased liver and biliary enzymes. Computed tomography and endoscopic ultrasonography consistently showed diffuse symmetric wall thickening of extrahepatic bile ducts without any obstruction (Fig. 1a, b). Endoscopic retrograde cholangiopancreatography revealed an irregular bile duct wall in all three patients (Fig. 1c). Peroral cholangioscopy revealed band-like narrowing of the wall of the biliary tract in all three patients and diverticulum-like outpouching in patients #1 and #3 (Fig. 1d; Video 1).

A biopsy of the lesions of the extrahepatic bile duct showed inflammatory cells in the bile duct epithelium (Fig. 2). All patients received internal treatment with ursodeoxycholic acid. Patient #1 was also treated with oral prednisolone. Liver and biliary enzymes improved gradually in all three patients, although normalization of enzyme activities was not achieved.

Endoscopy_UCTN_Code_CCL_1AZ_2AJ
Video 1 Peroral cholangioscopy showing multiple areas of band-like narrowing of the wall of the biliary tract, which was present in all patients, and a diverticulum-like outpouching, which was present in patients #1 and #3.

Fig. 2 Histopathological appearance showing an infiltration of inflammatory cells (arrows), including neutrophils and lymphocytes, in the epithelium of the extrahepatic bile duct (arrowheads).

Competing interests
None

The authors
Takumi Onoyama, Yohei Takeda, Masayuki Kato, Mirai Edano, Ryohsei Tarumoto, Kazuya Matsumoto, Hajime Isomoto
Division of Medicine and Clinical Science, Department of Multidisciplinary Internal Medicine, Tottori University, Yonago, Japan

Corresponding author
Takumi Onoyama, MD, PhD
Division of Medicine and Clinical Science, Department of Multidisciplinary Internal Medicine, Faculty of Medicine, Tottori University, 36-1 Nishi-cho, Yonago 683-8504, Japan
golf4to@yahoo.co.jp

References


Bibliography
DOI https://doi.org/10.1055/a-0948-1271
Published online: 2019
Endoscopy
© Georg Thieme Verlag KG
Stuttgart · New York
ISSN 0013-726X

ENDOSCOPY E-VIDEOS
https://eref.thieme.de/e-videos

Endoscopy E-Videos is a free access online section, reporting on interesting cases and new techniques in gastroenterological endoscopy. All papers include a high quality video and all contributions are freely accessible online.

This section has its own submission website at https://mc.manuscriptcentral.com/e-videos

Onoyama Takumi et al. POCS of PD-1 inhibitor-related sclerosing cholangitis... Endoscopy