

A Case of Human Pancreatic Eurytremiasis.

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## Abstract

This study provides the first comprehensive imaging report of human pancreatic eurytrema. A 43-year-old man with obstructive jaundice and a pancreatic tumor was referred for diagnosis and treatment. Serum aspartate aminotransferase (AST), alanine aminotransferase (ALT), and gamma-glutamyl transpeptidase ( $\gamma$ GTP) were elevated. Computed tomography (CT) revealed a multilocular cystic lesion with delayed enhanced area in the pancreas head. On magnetic resonance imaging (MRI), the tumor was hyperintense on diffusion weighted image (DWI), and the apparent diffusion coefficient (ADC) value of the tumor was lower than that of the normal pancreatic parenchyma. Positron emission tomography with 2-deoxy-2-[fluorine-18]fluoro-D-glucose integrated with computed tomography ( $^{18}\text{F}$ -FDG PET/CT) revealed abnormally increased uptake of  $^{18}\text{F}$ -FDG in the tumor. A subtotal stomach-preserving pancreaticoduodenectomy (SSPPD) was performed on the preoperative diagnosis of pancreatic carcinoma accompanied by branch duct type intraductal papillary mucinous neoplasm (IPMN). Multifocal granulomatous lesions with necrotic areas including many parasite eggs were seen on the histology. The final diagnosis was pancreatic eurytrema.

## Key Words

Human eurytrema, Eurytrema pancreaticum, Pancreatic fluke, Pancreatic parasite infection

Conflict of interest

Yasuo Takehara is an endowed chair of the department supported by a private company.

## A Case of Human Pancreatic Eurytreemiasis

Human eurytreemiasis is one of the rarest parasite infections caused by *Eurytrema pancreaticum* (*E. pancreaticum*). Several cases of human eurytreemiasis have been reported but detailed radiological imaging features such as computed tomography (CT), magnetic resonance imaging (MRI), and positron emission tomography with 2-deoxy-2-[fluorine-18]fluoro- D-glucose integrated with computed tomography (<sup>18</sup>F-FDG PET/CT) were not included. We present a case of a human eurytreemiasis of the pancreas with special reference to the radiological and pathological manifestations.

### Case

A 43-year-old male visited a local hospital with itching and nausea and was originally diagnosed with obstructive jaundice and a pancreatic head tumor. After endoscopic retrograde biliary drainage (ERBD), he was referred to our hospital for diagnosis and treatment.

His past history was unremarkable. At laboratory examination, aspartate aminotransferase (AST) and alanine aminotransferase (ALT) levels were 39 U/L (normal range 13–33 U/L) and 72 U/L (normal range 6–30 U/L), respectively. Lactate dehydrogenase (LDH), alkaline phosphatase (ALP), and gamma-glutamyl transpeptidase ( $\gamma$ GTP) levels were 196 U/L (normal range 119–229 U/L), 308 U/L (normal range 115–359 U/L), and 156 U/L (normal range 10–47

U/L), respectively. Total bilirubin and direct bilirubin levels were 0.6 mg/dL (normal range 0.3–1.2 mg/dL) and 0.1 mg/dL (normal range 0–0.2 mg/dL), respectively. Amylase level was 72 U/L (normal range 37–125 U/L). Carbohydrate antigen 19-9 (CA19-9) level was 15 U/mL (normal range < 37 U/mL). The white blood cell count was 4500/ $\mu$ L (normal range 3800–8500 / $\mu$ L), the eosinophil level was 5.1 % (normal range 1–6 %), the C-reactive protein (CRP) level was 0.06 mg/dL (normal range < 0.3 mg/dL).

The dorsal portion of the pancreatic head was swollen with inhomogeneous densities or intensities with weak contrast enhancements, in which ill-defined multilocular cystic lesions with individual cyst size varying from 1 mm to 14 mm with non-uniformly thickened wall were distributed (Fig. 1, 2 and 4).

On contrast enhanced CT (Fig. 1), there were weak enhancements of the tissue around each cystic lesion in the portal phases. Delayed enhancements were also marginal for the tissue. There was no calcification in the pancreas on precontrast CT. On T2-weighted MRI performed before ERBD at the local hospital, the dorsal portion of the pancreatic head was replaced by inhomogeneously hyper- and hypo- intense lesions with partially ill-defined cystic components (Fig. 2A). There were inhomogeneously high intensities on diffusion weighted image (DWI) (Fig. 2C), and the apparent diffusion coefficient (ADC) value of the lesion was lower than that of normal pancreatic parenchyma (Fig. 2D). On magnetic resonance cholangiopancreatography

(MRCP), diffuse dilatation of the bile duct was seen (Fig. 2B), which was considered to be caused by stenosis of the lower portion of the common bile duct (CBD) due to the compression by the swollen pancreatic head. However, the main pancreatic duct (MPD) was not dilated (Fig. 2B) because the lesion affected mainly the dorsal portion of the pancreatic head. On  $^{18}\text{F}$ -FDG PET/CT, inhomogeneously increased uptake of  $^{18}\text{F}$ -FDG was seen in the dorsal portion of the pancreatic head with a maximum standard uptake value (SUVmax) of, 4.45 except for the surrounding area of the ERBD tube. Otherwise, there was no abnormally increased uptake of  $^{18}\text{F}$ -FDG on maximum intensity projection (MIP) (Fig. 3).

Based on the above imaging findings, although the imaging findings of T2-weighted images and MRCP were not entirely typical, the preoperative diagnosis for the pancreatic mass lesion was adenocarcinoma accompanied by the branch duct type IPMN considering negative eosinophilia. The atypical pattern for IPMN was considered to be caused by neoplastic growth of the cystic portion of the IPMN, or accompanied pancreatic ductal cancer invasion to preexisting branch duct type IPMN. Subsequently, the patient underwent subtotal stomach-preserving pancreaticoduodenectomy (SSPPD). On the resected specimen, yellowish-white round lesions were seen in the head of the pancreas. The MPD was normal because the lesion affected mainly the dorsal portion of the pancreatic head (Fig. 4). Microscopic examination revealed multifocal granulomatous lesions with necrotic areas surrounded by histiocytes including multinucleated

giant cells. The largest lesion was approximately 1.4cm in diameter. In the necrotic area, numerous parasite eggs were seen (Fig. 5). Although no worm was detected, eurytremaiasis was diagnosed on the specific shape of the eggs.

## Discussion

*E. pancreaticum* is a parasite commonly found in the pancreatic and biliary passages of cattle, hogs, sheep, goats, and water buffaloes mainly in Asia and South America [1]. In Japan, the parasite is mainly found in the pancreas of pasture cattle in Kyushu, the southern area of Shikoku, the Izu islands, and Western and Southern areas of mainland Japan.

Regarding human eurytremaiasis, its transmission likely occurs by the ingestion of grasshoppers (second intermediate host of the parasite) harboring metacercariae stages of the parasite [2]. The metacercariae can migrate into the pancreatic duct, where they develop to be adult worms.

Clinical symptoms include abdominal distress, dyspepsia, vomiting, diarrhea, and occasionally jaundice, and hepatomegaly with tenderness. No therapeutic trials have been conducted [3].

Detailed medical investigation after surgery, revealed that the patient lived in the vicinity of the estuary of a river in Shikoku. In addition, he had past history of anisakis infection linked to eating raw fish two and five years ago. Furthermore, he had the habit of eating deer and wild boars on a daily basis. Although the route of infection was undetermined, it seemed to be related

to his eating habits. The patient was discharged after surgery receiving guidance regarding his eating habits.

Parasitic granulomas should have been included in the differential diagnosis in case with a cluster of ill defined low density lesions surrounded by weak enhancement on contrast-enhanced CT. However, because the patient was afebrile and the white blood cell count (including eosinophil) and the CRP level was not elevated, we couldn't reach the diagnosis of infectious disease preoperatively. Although absence of MPD dilatation was an atypical finding for invasive ductal cancer, we considered it could be adenocarcinoma accompanied by the branch duct type IPMN affected mainly the dorsal portion of the pancreatic head.

There are nine case reports concerning human eurytremiasis in Japan. Seven were diagnosed by the presence of eggs, and two were diagnosed by the presence of adult worms [1]. Of those, Matsunaga et al. [4] reported that they identified the filling defect created by adult worms in endoscopic retrograde pancreatography (ERP). However, CT, MRI, and <sup>18</sup>F-FDG PET/CT findings were not reported. In this case, since there was no lesion in other organs on images, it was unlikely that the parasite moved into the pancreas by penetrating the gastrointestinal tract or through vessels. It is likely that the parasite invaded deep into the pancreatic duct through the duodenal papilla. Furthermore, we considered the multifocal lesion formed as a result of cystic dilation of the branch pancreatic duct because microscopic examination revealed numerous

parasite eggs in the lesion. We believe that the invasion of the parasite into the pancreatic ductal branches and subsequent occlusions of the ductal orifice formed multilocular cystic lesions.

In conclusion, radiologists should be aware that human eurytreemiasis may be detected as a cluster of ill-defined multilocular cystic lesion in the swollen pancreas on images.

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## Figure legends

Fig. 1. Coronal reformatted images of multiphase contrast enhanced CT (A: arterial-portal venous phase; B: late portal venous phase, C; equilibrium phase). Several ill-defined multilocular cystic lesions are seen in the swollen pancreatic head measuring 4 cm in diameter (circle). Arrowhead: ERBD tube; asterisk: portal vein. There are weak enhancements of the tissue around each cystic lesion in the portal phases. Delayed enhancements are also marginal for the tissue.

Fig. 2. MRI performed before ERBD.

The mass in the pancreatic head (circle) consisted of inhomogeneous hyper- and hypo- intense areas on T2 weighted images (A). On MRCP (B), dilatation of the bile duct (arrowhead) is seen, but MPD is not dilated. The mass is hyperintense on diffusion weighted image (DWI) (C), and low signal on corresponding ADC-map (D), which suggests lower ADC value of the lesion compared to normal pancreatic parenchyma.

Fig. 3. <sup>18</sup>F-FDG PET/CT (A, B: axial fusion image; C: maximum intensity projection image).

Abnormally increased uptake of <sup>18</sup>F-FDG is seen in the pancreatic head mass (circle). The SUVmax is 4.45 except for the portion of the common bile duct in which the ERBD tube is placed (arrowhead). There is otherwise no abnormally increased uptake of <sup>18</sup>F-FDG in the body (C).

Fig. 4. Resected specimen.

On macroscopic view before formaldehyde fixation, yellowish-white round lesions (arrows) are seen in the swollen pancreas head. Duo: duodenum; CBD: common bile duct; and MPD: main pancreatic duct. A ruler (one scale is 1mm) is on the lower left side.

Fig. 5. Histological specimen (H&E stain).

A: On low power (x12.5) image, multifocal granulomatous lesions with necrotic areas (asterisk) are seen in the pancreatic head. B, C: on magnified images (x 100 and x 400), many *E. pancreaticum* eggs are identified by their specific morphological features (arrows).

Fig. 1.

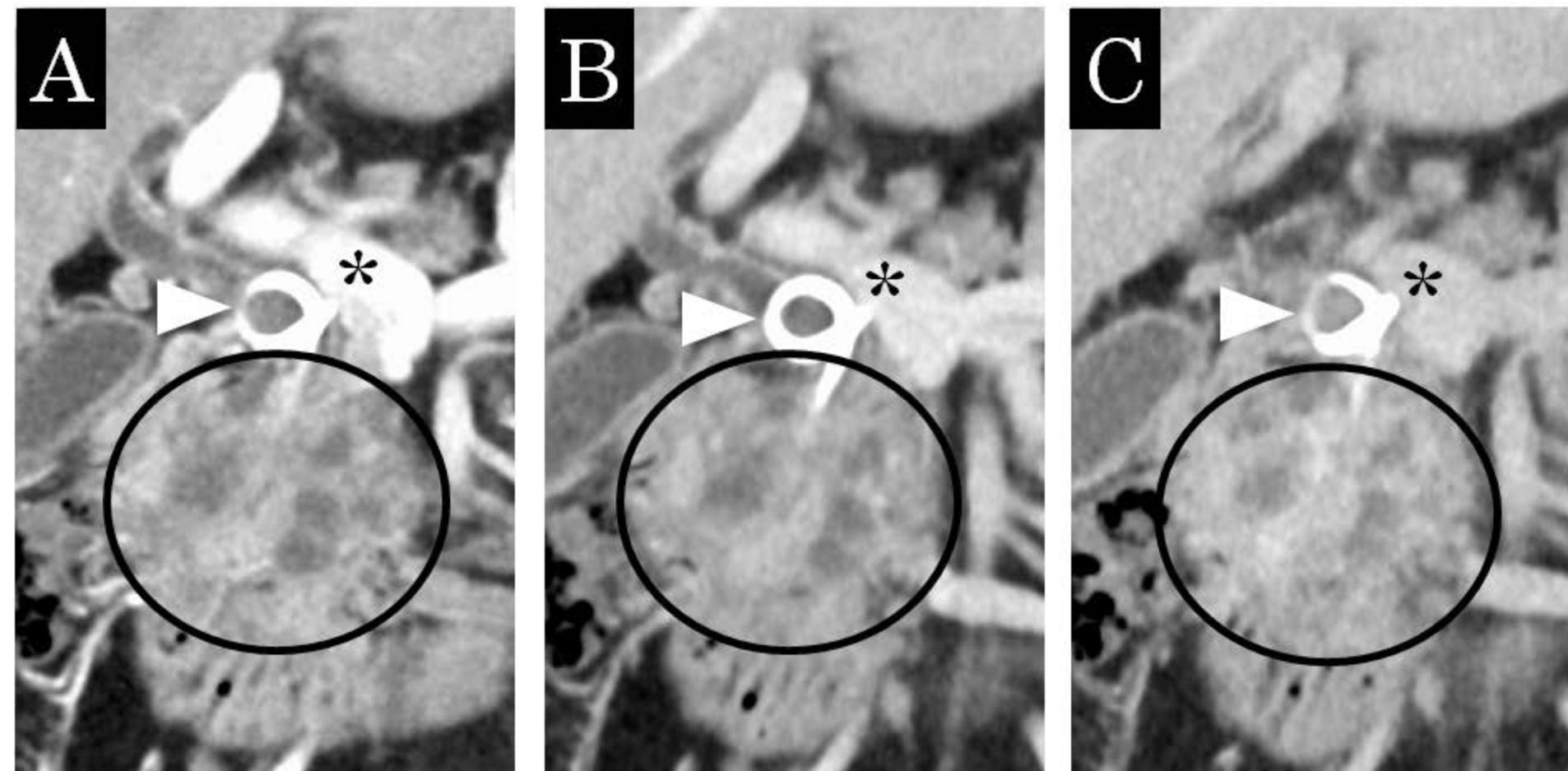


Fig. 2.

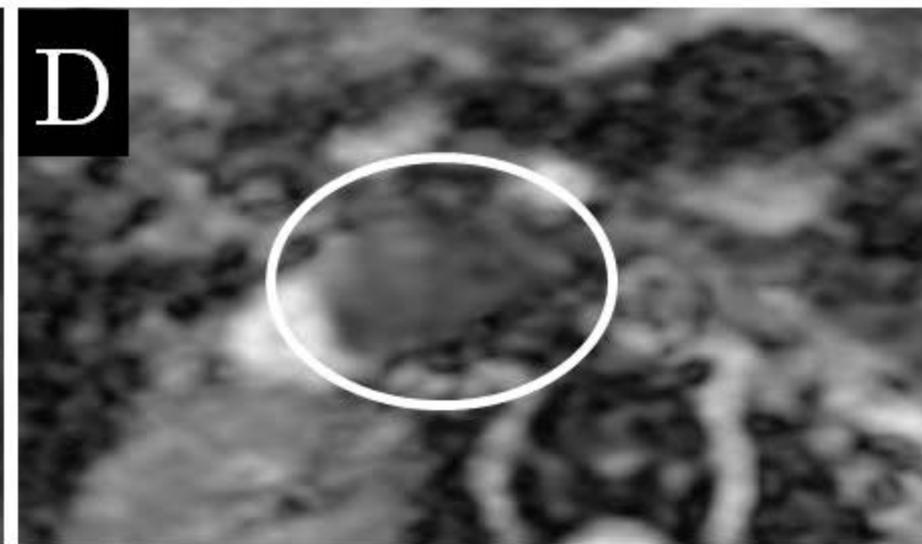
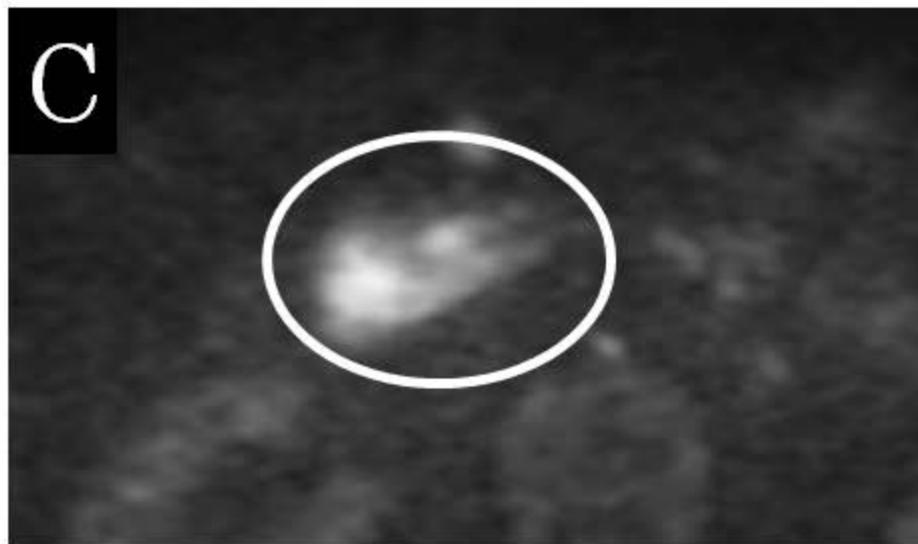
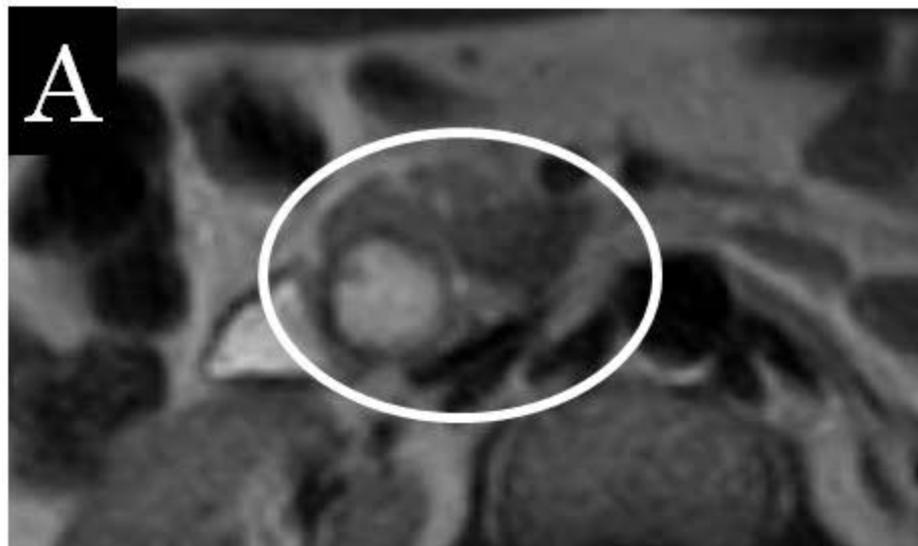


Fig. 3.

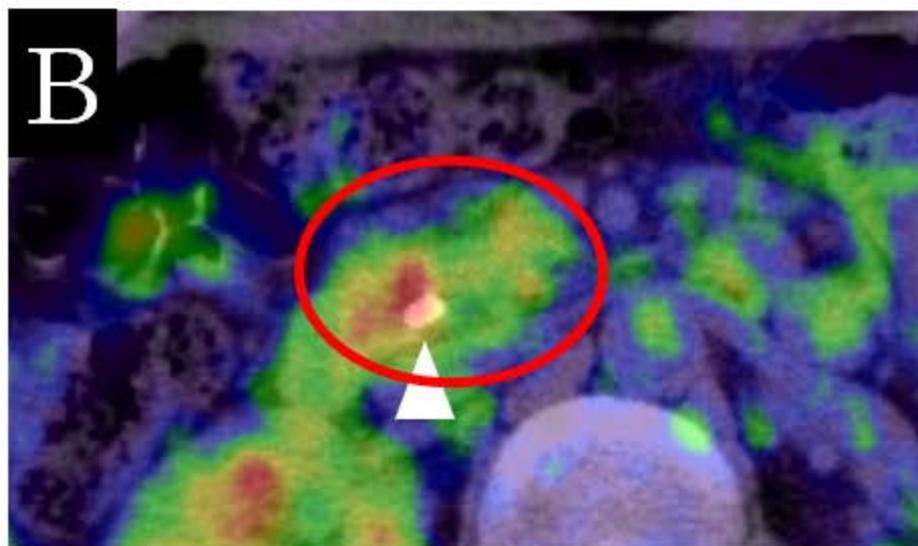
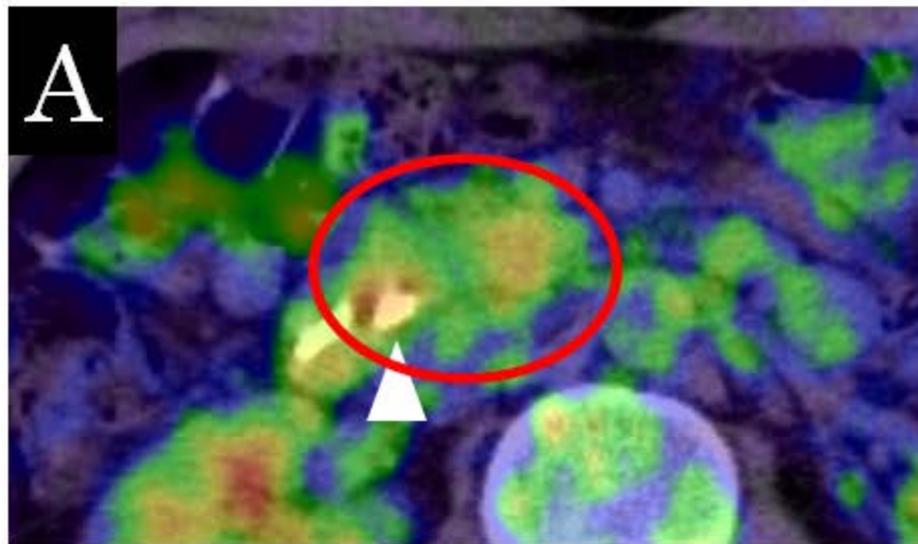


Fig. 4.

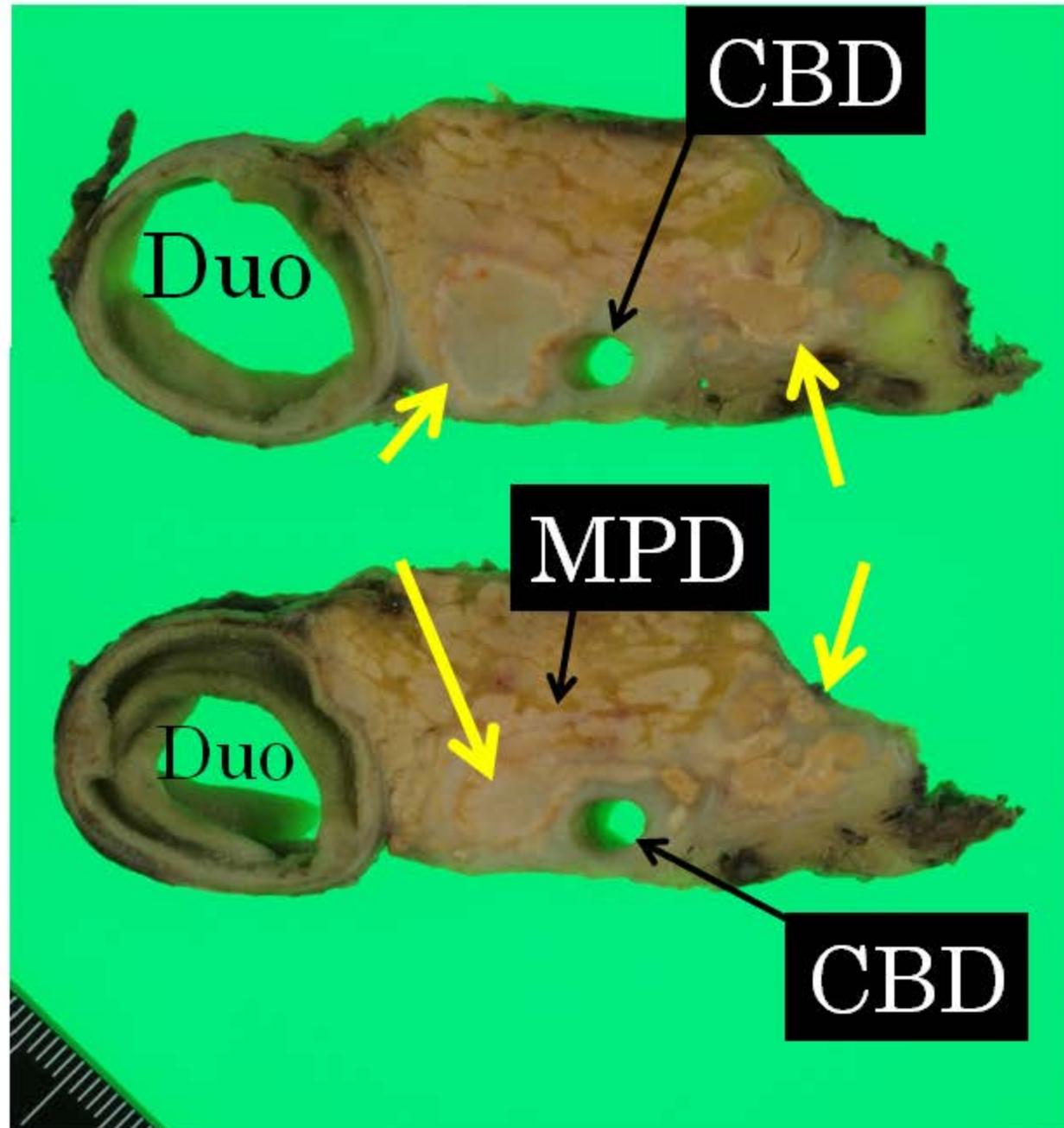


Fig. 5.

