Biliary intraductal papillary mucinous neoplasm visualized intralesionally through a fistula with the duodenal bulb

A 53-year-old man was admitted to our hospital with abdominal pain. He had undergone surgery for a pancreatic pseudocyst related to chronic alcoholic pancreatitis 7 years previously at another hospital. Magnetic resonance imaging (MRI) 3 years later had shown a cystic lesion, 4 cm in diameter, at the hepatic hilum, with two small mural nodules. The patient was then lost to follow-up for 3 years.

Following admission to our hospital, MRI showed a 6 × 6.5-cm, inhomogeneous solid lesion between the liver, pancreas, and duodenum, with compression of the duodenal bulb (Fig. 1 a) and invasion of the left biliary duct, common hepatic duct (Fig. 1 b), hepatic artery, and portal vein. Gastroscopy showed the presence of a compression on the anterior wall of the duodenal bulb. The compression had two fistulous orifices with villous tissue at the edges, which were exuding whitish mucus (Fig. 2 a). A diagnostic gastroscope was inserted through one of the fistulous orifices revealing that the internal structure of the neoplastic lesion that was forming a fistula to the duodenal bulb was a multiloculated cavity covered with a whitish, villous tissue (Fig. 2 b–d; Video 1). A biopsy of the villous tissue at the edge of the fistula revealed fragments of epithelial neoplasia with papillary structures and focal adenocarcinomatous transformation (Fig. 3 a).

The patient underwent surgery, and a neoplastic lesion that was infiltrating the biliary tract and the vascular structure at the hepatic hilum, as well as fistulizing to the duodenal bulb, was seen. The pancreas was normal. The surgeon decided to perform a gastroduodenal anastomosis. Histology of the surgical sample showed an adenocarcinoma, consistent with a biliary intraductal papillary mucinous neoplasm (IPMN-B), the papillary structure of the neoplastic lesion that was forming a fistula to the duodenal bulb was a multiloculated cavity covered with a whitish, villous tissue.

Video 1

Endoscopic examination of the internal structure of the neoplastic lesion that was forming a fistula to the duodenal bulb, showing a multiloculated cavity covered by a whitish, villous tissue.
features of which were fistulizing to the duo-
denal bulb (Fig. 3b).

IPMN-B is a type of bile duct tumor with
certain unique features, which include in-
traductal papillary growth and mucin
production. Recent studies have revealed
pathological similarities between the
IPMN-B and pancreatic IPMN (IPMN-P),
and some even consider the two neo-
plasms to be counterparts [1, 2]. There
are several reports of IPMN-P fistulizing
to surrounding organs [3–5]; however, to
the best of our knowledge, this is the first
report of an IPMN-B forming a fistula with
the duodenal bulb. This occurrence ap-
pears to confirm a similarity in behavior
between IPMN-B and IPMN-P.

Fig. 3 Histological appearances of the neoplastic lesion from: a endoscopic biopsy fragments showing
epithelial neoplasia with papillary structures and focal adenocarcinomatous transformation; b the sur-
gical resection showing adenocarcinoma, compatible with biliary intraductal papillary mucinous neo-
plasm (IPMN-B), with papillary structures that are fistulizing to the duodenal bulb.

Competing interests: None

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