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A rare case of gastric fistulization of a main-duct IPMN: a case report

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ABSTRACT

We report a rare case of gastric fistulization in a patient with main-duct (MD) intraductal papillary mucinous neoplasm (IPMN). Fistulization to adjacent organs is a rare complication of this disease, mainly affecting the duodenum; the fact that the stomach was the only organ involved makes this case even more peculiar. The diagnosis of the fistula was made after examination with Endoscopic Ultrasound (EUS), since a previous computed tomography (CT) scan wasn't able to detect it.

Key words: Neoplasm; Endoscopy; Ultrasound; Fistula; Pancreas

TEXT

Introduction

IPMNs belong to the category of cystic pancreatic tumors; their incidence is reported to vary from 0.31 to 4.35/100000.¹ IPMNs can be classified into 3 groups, based on imaging features: MD-IPMN, characterized by diffuse or segmental dilation of the main pancreatic duct, branch duct (BD)-IPMN, characterized by pancreatic cysts in communication with a normal main pancreatic duct (MPD) and mixed type IPMN, when characteristics of both types are present.^{2,3} Their main histological feature is the proliferation of mucin producing epithelial cells, with excessive production of mucus leading to dilation of the ducts. IPMNs can also be classified in benign, "borderline" and malignant according to the grade of the dysplasia of the proliferating cells.⁴

IPMNs can show malignant transformation as a complication and, among all the other ones that may occur (i.e. hemorrhage, acute pancreatitis, perforation), fistulization to adjacent organs is the rarest.⁵ The duodenum seems to be the most common site of fistulization, followed by the stomach, the common bile duct and the colon.

Here we present the case of a MD-IPMN fistulizing in the stomach, with no involvement of the duodenum.

Case report

In 2015, a 72-year-old patient, with benign prostatic hyperplasia and a recent onset of diabetes mellitus, presented with abdominal mesogastric pain started some weeks before along with a 5 kg loss during the last months. Blood analysis showed inflammation with WBC 10470/mm³ and C reactive protein (CRP) 82 mg/dL. Abdominal US showed an inhomogeneous mass in the mesogastric-epigastric area, with some anechoic areas inside it. Subsequent CT scans demonstrated an inhomogeneous enlargement of the pancreatic head and neck, dilation of the MPD and cystic dilation of many collateral ducts.

The patient was then referred to our Endoscopy Unit to undergo EUS. Endoscopically a 3 cm sub-epithelial yellowish mass was seen in the gastric antrum (Fig. 1) and, in the duodenum, a patulous ampulla of Vater with extruding mucus (the so-called fish-mouth sign) was noted (Fig. 2). EUS demonstrated a diffuse dilation of the MPD up to 17 mm, mucus filling the duct and cystic spaces containing contrast-enhanced papillae (SonoVue® 5 ml iv) (Fig. 3). Moreover, the nature of the gastric mass was discovered: fistulization of a side branch of the MPD into the gastric wall was revealed by EUS (Fig. 4).

A diagnosis of MD-IPMN was made and the patient was sent to a surgical consultation but he refused surgery.

Discussion

We report a rare case of MD-IPMN with a fistula involving the stomach in a 72-year old male presenting with abdominal pain. IPMNs are a rare entity among pancreatic tumors, and fistulization is one of their rarest complications. Incidence of fistulas in IPMNs varies in percentage according to different studies. We performed an analysis of the recent literature and, to our knowledge, the largest series of patients were described by Ravaud *et al*⁶ in 2015 (423 patients) and Kobayashi *et al*⁷ in 2008 (274 patients): the percentage of fistulization ranged from 1.9% in the French study to 6.6% in the Japanese one. This difference can be explained by the diagnostic methods that were used in the two groups: MRI and/or CT in the former, EUS and/or endoscopic retrograde cholangiopancreatography (ERCP) in the latter. Furthermore in the French study, although 75% of the patients had more than one organ involved, the duodenum was affected in every single patient⁶.

Conclusions

The rarity of this case is explained by the fact that, in order of frequency, fistulas occur primarily to the duodenum and then to other organs, whereas in this case the stomach was the only organ involved.

It should be kept in mind that fistulization does not strictly adhere to the criteria of malignancy since the mechanism involved includes either high mechanical pressure in the pancreatic ducts filled with mucin or inflammatory stimulation with direct invasion⁸⁻¹⁰: it is possible to hypothesize that both mechanisms were here involved.

It should be noted that, in our case, the fistula was eventually discovered by EUS examination, as not previously detected by CT.

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NOTES

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Authors' contributions.—

De Angelis C and Saracco GM critically revised the manuscript; Bruno M and Gaia S performed the endoscopy and acquired the data; Cortegoso Valdivia P and Bruno M equally contributed to analysis and interpretation of the data and wrote the manuscript. The manuscript have been read and approved by all the authors; all the requirements needed have been met. No data from the study has been or will be published separately

TITLES OF FIGURES

Figure 1.— a yellowish sub-epithelial mass was noted in the gastric antrum

Figure 2.— thick mucus extruded from a patulous ampulla of Vater

Figure 3.— contrast enhanced papillary projections after infusion of SonoVue[®] 5 ml i.v.

Figure 4.— a dilated pancreatic duct (arrow) fistulizing in the gastric submucosa (asterisk)