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Case Report

Intraductal papillary mucinous neoplasm complicated by a gastropancreatic fistula

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ABSTRACT

One of the rare complications of low-grade pancreatic neoplasms is fistulization into nearby structures. This often does not present clinically, but is incidentally identified in patients who have been imaged serially to monitor the progression of the disease. In this report, we present an uncommon complication of an intraductal papillary mucinous neoplasm, which developed a spontaneous gastropancreatic fistula in a patient who was conservatively managed. The clinical course, imaging features, and management of this case are discussed.

Introduction

Intraductal papillary mucinous neoplasm (IPMN) of the pancreas are a spectrum of benign to malignant epithelial neoplasms that are characterized by papillary proliferation, duct dilation, and cyst formation. The incidence of IPMN has been increasing as improvements have been made at identifying cystic pancreatic lesions by imaging [1]. Anatomically, IPMNs are classified as main pancreatic duct (MPD) type or branch pancreatic duct type, based on the ductal involvement of the tumor. The neoplasms originate from stem cells in the pancreatic duct with progression to dysplasia and eventually to invasive carcinoma. The risk of malignant transformation is lower in the branch pancreatic duct type [2]. As IPMN may not progress to carcinoma and majority of the patients are asymptomatic, management differs based on clinical risk factors. Surgery is warranted for neoplasms with high-risk features including MPD $\geq$ 1 cm, enhancing mural nodularity, or biliary obstruction. Worrisome features include cyst size $\geq$ 3 cm, MPD dilatation between 5-9 mm, nonenhancing mural nodularity, and abrupt change in main duct caliber and distal pancreatic atrophy. Imaging findings can be confirmed with endoscopic ultrasound which shows mural nodularity or continuity with the MPD. These neoplastic processes can be followed with CT or MRI based on size. Current recommendations suggest

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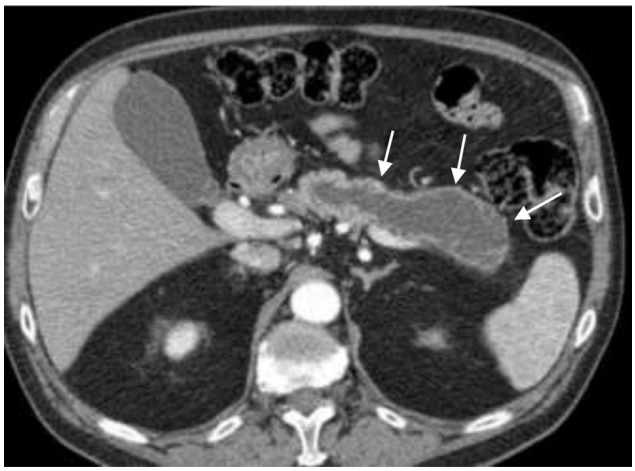

Fig. 1 – Axial contrast-enhanced CT from initial presentation in October 2011 shows a low attenuation mass in the pancreatic body and tail (arrows).

follow-up intervals at 2-3 years for cysts less than 1 cm; annual follow-up for cysts 1-2 cm for a total duration of 2 years, and 3-6 months for cysts greater than 2 cm. Any symptomatic patient is a candidate for surgery based on cyst location and extent of MPD involvement [3].

Tumors may cause complications such as pain, weight loss, pancreatitis-like symptoms, pancreatic insufficiency, and fistula forming to adjacent organs. While clinical presentations may indicate growth and complications of the IPMN, imaging plays an important role in monitoring the size and identifying complications. We present an interesting case of a spontaneous gastropancreatic fistula formation secondary to an IPMN in a patient being followed with conservative management.

Case report

An 87-year-old male presented to our institution's minimally invasive and robotic surgery clinic with recurrent episodes of pancreatitis over the past 7 years. Although the patient was asymptomatic at the visit, it was noted that the patient experienced an episode of pancreatitis a month ago.

Initial CT examination of the abdomen in October 2011 showed diffuse cystic dilatation of the body and tail of the MPD measuring 7.1 × 2.7 cm and appeared contiguous with the MPD distally, suspicious for main duct type IPMN (Figs. 1 and 2). Subsequent MR examination of the abdomen confirmed the fusiform dilation of the MPD occupying the body and tail, measuring 8.4 × 3.3 cm (Fig. 3). Endoscopic ultrasound with fine needle biopsy was performed with cytology demonstrating benign ductal cell with focal mucinous changes, confirming IPMN. Histologically, ductal dilation with adjacent pancreatic fibrosis, atrophy, and inflammation support diagnosis of IPMN (Fig. 4).

At the time of diagnosis, the patient decided on conservative management. Over the next year, the patient became

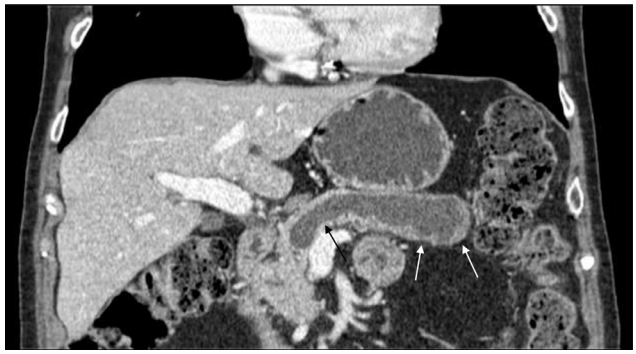

Fig. 2 – Coronal reconstructed contrast-enhanced CT of the abdomen obtained in October 2011 shows a low attenuation mass in the pancreatic body and tail (arrows) contiguous with the main pancreatic duct which is markedly dilated (black arrow).

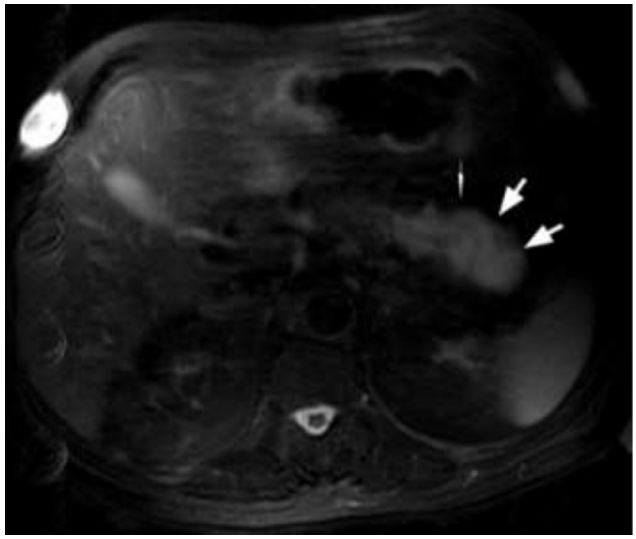

Fig. 3 – Axial T2 weighted fat saturated MR image obtained in June 2011 shows a T2 hyperintense mass causing fusiform dilation of the main pancreatic duct in the body and tail (arrows), confirming the CT findings.

symptomatic with back and abdominal pain. Repeat CT examination of the abdomen approximately 1 year after initial presentation showed progressive diffuse ductal dilatation with an increase in size of the overall cystic mass measuring up to 11.3 cm in greatest axial dimension. Further serial follow-up CT imaging was completed at 6 and 12-month intervals. In June 2017, 6 years after the initial CT, there was progressive enlargement of the cystic mass which measured 12.3 × 5.6 × 5.5 cm occupying the entire pancreatic body and tail and extending to the head (Fig. 5). There was increased bulbous distention of the pancreatic body with atrophic residual pancreatic parenchyma. Given the patient's age and clinical condition, the patient agreed on serial CT examinations at 6-month intervals rather than surgery. In June 2018, nearly 7 years after the initial presentation, a contrast-enhanced CT examination of the abdomen demonstrated a communication between the

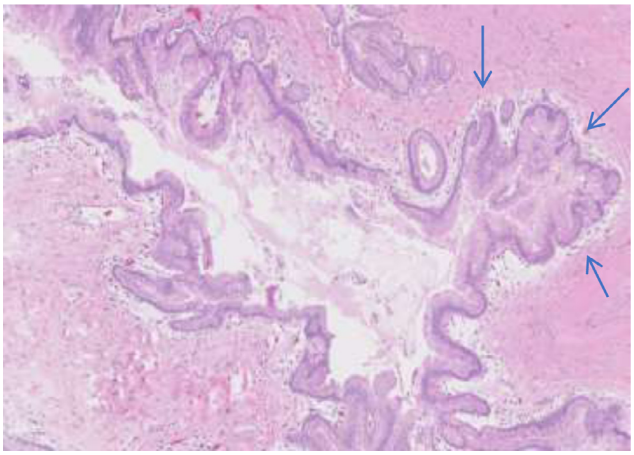

Fig. 4 – High power H&E stain from a similar patient at our institution showing the epithelium composed of mucinous metaplasia forming papillary projections into the duct lumen (arrows). Pools of mucin are found throughout the lumen of ducts. Adjacent fibrosis and atypical glandular focus <0.5 mm are suspicious for early invasive focus. These are characteristic findings for IPMN.

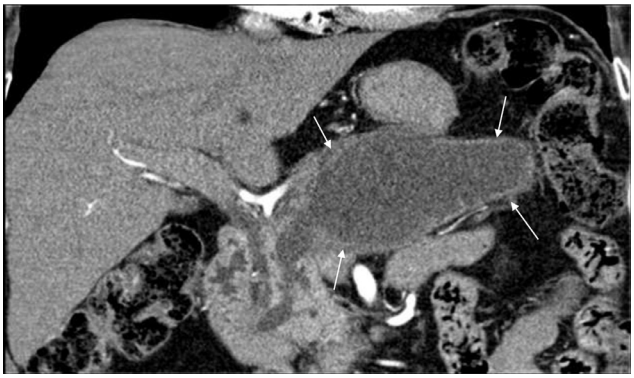

Fig. 5 – Coronal reconstructed contrast enhanced CT of the abdomen in June 2017 shows further enlargement of the cystic pancreatic mass occupying the majority of the pancreatic body and tail (arrows).

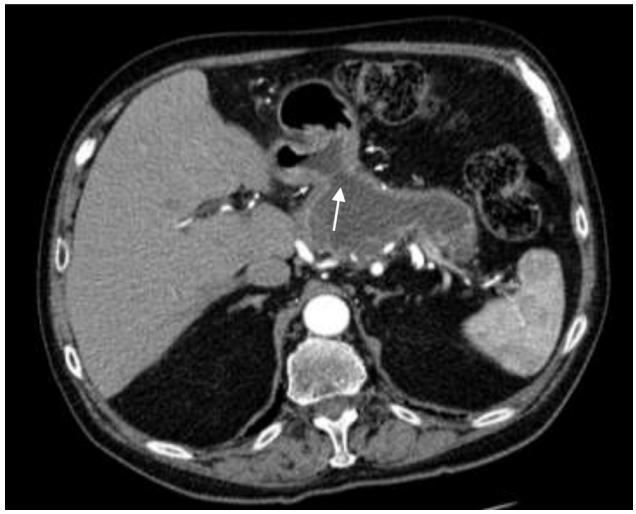

Fig. 6 – Axial contrast-enhanced CT of the abdomen in June 2018 demonstrates a gastropancreatic fistula (arrow) with low attenuation mucinous secretions in the gastric lumen.

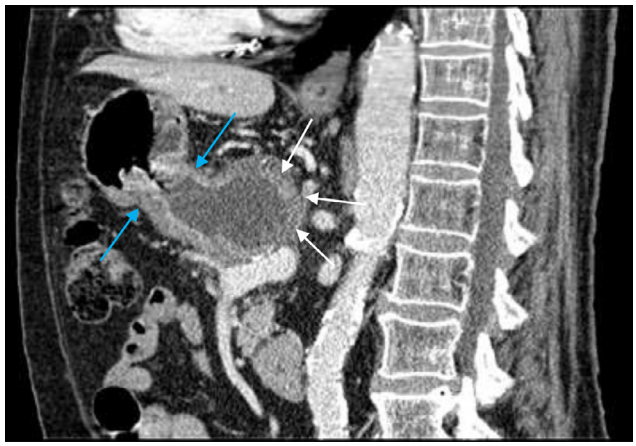

Fig. 7 – Sagittal reconstructed contrast-enhanced CT of the abdomen in June 2018 shows a thick walled pancreatic mass (white arrows) communicating with the gastric lumen forming a gastropancreatic fistula (blue arrows). Color version of figure is available online.

ventral surface of the pancreas and lesser curvature of the stomach with low-density fluid in the gastric lumen, indicating a gastropancreatic fistula (Figs. 6 and 7). The patient remained asymptomatic at this time and no intervention has been performed to this date.

Discussion

Given an increase in the incidence of IPMN without an increase in IPMN-related mortality, the long-term sequela and complications will frequently manifest and require medical attention [1]. The low incidence of pancreatic cancer in IPMN indicates that a majority of IPMNs do not progress to malignancy, and patients may be managed conservatively with imaging follow-up. Therefore, tumor complications and growth must be routinely monitored so appropriate treatment can be initiated.

Fistula is not a common complication of IPMN, with reported incidence near 7%. The pathophysiology of fistulization is thought to be a result of mechanical penetration but the neoplasm can also spread to adjacent organs via invasion and create a fistula communication [4]. A majority of the fistulas are complications of main duct IPMNs; however, fistulas are not specific to that subtype [5].

IPMN fistulization has multiple clinical consequences due to the different organs the neoplasm penetrates. In a large retrospective study with 423 patients, a majority of fistulas involved the duodenum, followed by the stomach, common bile

duct, and colon [5]. Some patients developed multiple fistulas, highlighting the importance of close follow-up with known IPMN. Small, uncomplicated pancreatic fistulas can spontaneously close with somatostatin. Complicated pancreatic fistulas are managed endoscopically via pancreatic stent placement and/or pancreatic sphincterotomy.

A fistula may be asymptomatic incidental finding on imaging. Once documented, this fistula must be monitored for further complications. It has been reported that tumor can also develop in the fistula [6,7]. Although, given the low incidence of this finding, long-term consequences of a gastropancreatic fistula are not well-established and require continual research to determine ideal intervals for imaging follow-up. In our case, the patient remained asymptomatic despite the fistula without intervention.

Conclusion

In conclusion, we present an infrequently encountered complication of gastropancreatic fistula of IPMN, in a patient who underwent conservative management. Imaging features and important clinical issues, including management of the gastropancreatic fistula are reviewed with an emphasis on long-term follow-up and appropriate management in low-risk patients.

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