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Santorinicele Containing a Pancreatic Duct Stone in a Patient with Incomplete Pancreas Divisum

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Santorinicele, a focal cystic dilatation of the distal duct of Santorini, has been suggested as a possible cause of the relative stenosis of the accessory papilla, is associated with complete pancreas divisum, which results in acute episodes of pancreatitis or pain. This report describes a case of a santorinicele, which was initially detected by upper gastrointestinal endoscopy as a polypoid mass, in a patient with recurrent abdominal pain. The mass was subsequently proved to be a santorinicele containing a pancreatic duct stone associated with incomplete pancreas divisum on endoscopic retrograde pancreatography. To the best of our knowledge this is believed to be the first description of a santorinicele associated with these characteristic findings.

Key Words: Santorinicele, pancreas divisum, incomplete, pancreatic duct, calculus

INTRODUCTION

A santorinicele is defined as a focal cystic dilatation of the end of the dorsal pancreatic duct at the minor papilla.[1] This anomaly is believed to result from a combination, either acquired or congenital, of a relative obstruction and a weakness of the distal ductal wall. Furthermore, a santorinicele has been suggested to be a possible cause of the relative stenosis of the minor papilla, which in association with pancreas divisum (PD) results, with a high intraductal pressure, is responsible for recurrent episodes of acute pancreatitis. Herein, a patient with a longstanding history of abdominal pain, who was found to have a santorinicele containing a pancreatic duct stone, is reported. Although there are several reports of santorinicele, to the best of our knowledge, this is the first description of a santorinicele associated with a pancreatic duct stone, particularly in a patient with an incomplete PD.

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CASE REPORT

A 40-year-old man presented with epigastric pain, which was related to heavy meals. The patient had experienced similar intermittent episodes for several years, but the causes for his complaint were not previously found. A physical examination was noncontributory. The laboratory studies, including a complete blood count, liver function tests, serum amylase and lipase, were within the normal ranges. The abdominal ultrasonography was unremarkable. On upper gastrointestinal endoscopy (GIF-XQ 240, Olympus, Tokyo, Japan), the stomach revealed a multiple patchy hyperemia, with a mild intestinal metaplasia on the antrum and lower body. The duodenum was normal, with the exception of a round mass-like lesion at the proximal descending portion. Lateral-viewing duodenoscopy (TJF-240, Olympus, Tokyo, Japan) showed the mass to be an enlarged minor papilla, which was protruding into the duodenal lumen. The orifice of the minor papilla could not be identified (Fig. 1). On palpation with a biopsy forceps the lesion was slightly movable, but very firm and hard. Therefore, an unexposed type of papillary tumor was initially suspected. Endoscopic retrograde

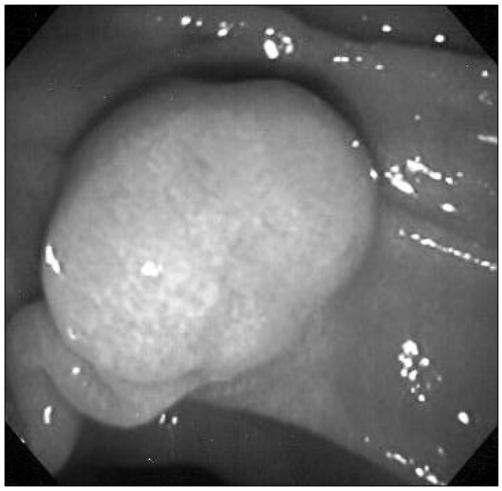

Fig. 1. Endoscopic image of the minor papilla showing a protruding polypoid configuration, which subsequently proved to be a santorinicele containing a pancreatic duct stone.

cholangiopancreatography (TJF-240, Olympus, Tokyo, Japan) was performed, the following day, for a precise evaluation of the minor papillary lesion and the relationship between the lesion and the pancreatic duct. Pancreatography, via a major papilla, disclosed a small broom-like arborization of the ventral pancreatic duct and a slightly dilated dorsal pancreatic duct (5.5 mm at the head, 3.5 mm at the body). In addition, both ventral and dorsal pancreatic ducts were communicated by another channel, suggesting an incomplete PD, even though the communicating channel was wider than usual. In particular, a peculiar configuration, i.e. a cystic dilatation (6 × 5 mm) with a central round filling defect, was observed at the termination of the dorsal pancreatic duct at the minor papilla (Fig. 2). This finding was most consistent with a solitary pancreatic duct stone in a santorinicele. The cholangiography was normal. A biliary sphincterotomy, with a needle knife papillotome (MTW, Wesel, Germany), was performed on the protruding santorinicele. Just after the sphincterotomy, a whitish ovoid stone was easily exposed and removed spontaneously from the santorinicele (Fig. 3). There were no complications related to the procedure.

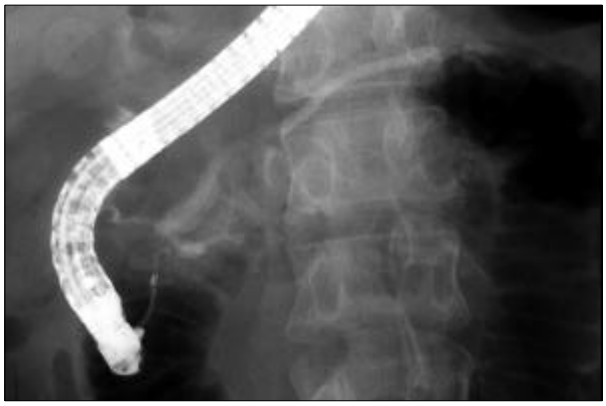

Fig. 2. Endoscopic retrograde pancreatogram demonstrating a cystic dilatation, with a central filling defect at the terminal dorsal duct and an incomplete pancreas divisum.

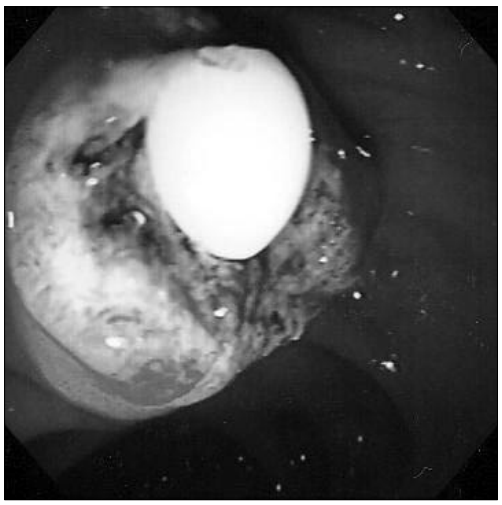

Fig. 3. Endoscopic image of the santorinicele showing an ovoid pancreatic duct stone, which was exposed immediately after a sphincterotomy.

DISCUSSION

Santoriniceles are believed to be analogous to a similar dilatation of the most distal common bile duct, commonly known as a choledochocele. It has been suggested to represent a congenital or acquired weakness of the mucosal wall in the setting of the obstruction.[1] Most santoriniceles have been reported in elderly patients,[1-5] suggesting they are most probably acquired rather than congenital. In addition, some santoriniceles are associated with adjacent duodenal diverti-

cula.[1] Structural changes may contribute to the acquired mucosal weakness, thereby facilitating santorinicele formation. However, reports of santorinicele, including a pediatric patient, suggest that some cases may be secondary to a congenital weakness.[6,7]

The present case had two characteristic findings; firstly an incomplete PD. Since all santorinicles reported, which are believed to be of an acquired origin, have been detected in patients with a complete PD, it is unclear whether a santorinicele may be present in patients without this anomaly. However, the relative obstruction to the pancreatic flow through the communicating branch in an incomplete PD may lead to dorsal duct hypertension,[8,9] which is sufficient to trigger an acute episode of pancreatitis or pain, or weaken the duodenal wall at the minor papilla. Furthermore, a stenosis of the minor papilla in an incomplete PD may lead to the drainage of the dorsal pancreatic outflow to the ventral duct system through the communicating branch. In the long term, the communicating branch may adapt to the impeded pancreatic outflow, secondary to the stenosis of the minor papilla, and finally become wider. It is conceivable that the wide communicating branch provides comparatively adequate drainage of the dorsal duct. Therefore, the patient had no serious symptoms or complications, even though he complained of intermittent abdominal pain, which developed only after eating heavy meals. In addition to an incomplete PD, the second characteristic finding was the presence of a pancreatic duct stone in the santorinicele. A stone in the santorinicele is extremely rare, and to our knowledge, only one previous case has been reported.[6] The calculus in the santorinicele indicates the degree of long-standing stasis, and might be one of the causes of the orifice obstruction.

Magnetic resonance pancreatography (MRP) is a noninvasive method for evaluating the pancreatobiliary system. Several groups have reported the excellent reliability of MRP in demonstrating pancreatic duct abnormalities, such as cysts, strictures, dilatations, and PD, as well as santorinicles.[3-5,10,11] In particular, secretin-stimulating MRP, by increasing the exocrine pancreatic outflow, not only improves the conventional MR imaging of the pancreatic duct anatomy, but may also be useful for diagnosing a pancreatic papillary stenosis or a dysfunction and for detecting a reduced pancreatic exocrine reserve.[3-5,12] Therefore, it appears reasonable that the vast majority of santoriniceles reported to date had been diagnosed with secretin-stimulating MRP. Fortunately, in this case, the santorinicele was easily detected using upper gastrointestinal endoscopy because the minor papilla showed the constant polypoid configuration, which may be caused by a pancreatic duct stone in the santorinicele.

In conclusion, our case is believed to be the first description of a santorinicele associated with a pancreatic duct stone and an incomplete PD. However, it is difficult to explain exactly whether the santorinicele was the result of a stenosis of the minor papilla, due to a stone, or an increased dorsal ductal pressure associated with an incomplete PD, or the cause of stone formation due to the chronic stasis of pancreatic secretion. To clarify these problems, further identification of more such cases is warranted.

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