

A Case of Santorinicele without Pancreas Divisum: Diagnosis with Multi-detector Row Computed Tomography

A santorinicele is defined as a focal cystic dilatation of the terminal portion of the dorsal pancreatic duct at the minor papilla. Most cases reported previously were associated with pancreas divisum and a santorinicele without pancreas divisum is known to be rare. We recently experienced a typical case of a santorinicele without pancreas divisum in a 67-yr-old woman with abdominal pain and hematochezia, subsequently proven to be the result of an ischemic colitis. The santorinicele was diagnosed incidentally with multi-detector row computed tomography using a minimum intensity projection technique, which clearly showed a cystic dilatation of the terminal portion of the dorsal pancreatic duct and a communication between the ventral and dorsal pancreatic ducts. This finding was also confirmed by a magnetic resonance cholangiopancreatography.

Key Words : Pancreas; Santorinicele; Tomography, Spiral Computed

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INTRODUCTION

A santorinicele is a focal cystic dilatation of the terminal portion of the dorsal pancreatic duct at the minor papilla, and has been reported in patients with pancreas divisum (1). Clinically this rare anomaly has been suggested to be a possible cause of a relative stenosis of the minor papilla, which in association with pancreas divisum, results in a high intraductal pressure and resultant pancreatitis (1-7). We recently encountered a patient with a santorinicele without pancreas divisum. Although there is only one report of a santorinicele without pancreas divisum, it was combined with bifid pancreas, which is a congenital anomaly where the primitive pancreatic channels fail to fuse into a single main duct during the development of the pancreas (8). We describe a typical case of a santorinicele without pancreas divisum or other congenital pancreatic anomalies that was identified incidentally by multi-detector row computed tomography (MDCT).

CASE REPORT

A 67-yr-old woman was admitted complaining of param-

bilical and lower left abdominal pain with intermittent hematochezia. She had been previously treated for diabetes mellitus and hypertension with an oral hypoglycemic agent and a calcium channel blocker for the previous 10 yr. She was a non-smoker, and non-drinker with no family history of gastrointestinal disease. The physical examination was negative except for the paraumbilical and left lower abdominal tenderness. The laboratory studies, including a complete blood count, liver function tests, serum amylase level, were within the normal range. The colonoscopic findings were diffuse erythema, edema, ulcer, and friability of the mucosa through the proximal descending and distal transverse colon, which was consistent with ischemic colitis. MDCT (LightSpeed Pro; GE Medical Systems, Milwaukee, WI, U.S.A.) was performed after the colonoscopy. The CT scan protocol was as follows: 150 mL of iopromide (Ultravist 370; Schering, Berlin, Germany) was injected through an 18-gauge angiographic catheter that was inserted into a forearm vein at a flow rate of 3 mL/sec by using a mechanical injector. Portal venous phase images were acquired following a scanning delay of 70 sec from the time of initiation of contrast material injection, and the delayed phase images followed after a scanning delay of 3 min. The CT parameters included a detector configuration

of 1.25 mm \times 16, a gantry rotation speed of 0.5 sec, and a table feed of 20 mm per gantry rotation. CT showed mild and diffuse wall thickening from the distal transverse colon to the descending colon. It also showed a small round low-density lesion at the distal dorsal pancreatic duct, which was adjacent to the duodenal wall (Fig. 1A). The 1.25-mm transverse CT scans were reconstructed at 0.625-mm intervals on a dedicated workstation (Advantage Window 4.2; GE Medical Systems). Reformation was performed using a minimum intensity projection technique. The coronal reformatted image using a minimum intensity projection technique clearly showed a cystic dilatation of the distal dorsal pancreatic duct and a communication between the ventral and dorsal pancreatic ducts (Fig. 1B). These findings were compatible with a santorinicele without pancreas divisum. The dorsal pancreatic duct was equal to or slightly more prominent than the ventral pancreatic duct. The subsequent magnetic resonance cholangiopancreatography (MRCP) confirmed the cystic dilatation of the distal dorsal pancreatic duct. There was no evidence of pancreas divisum or any other congenital anomaly of pancreas (Fig. 1C). Based on the findings of imaging studies, the diagnosis of a santorinicele without pancreatic divisum was made.

The patient did well after supportive cares including intravenous fluids and antibiotics for ischemic colitis, and then was discharged on the 9th day of admission without any problems.

DISCUSSION

A santorinicele is believed to be analogous to a dilatation of the most distal common bile duct, which is commonly known as a choledochocele. It was first described in 1994 by Eisen et al. (1), who reported 4 patients with pancreatitis and pancreas divisum accompanied by a focal cystic dilatation of the terminal portion of the dorsal pancreatic duct by endoscopic retrograde cholangiopancreatography (ERCP). Since the first description, santoriniceles have been reported in patients with pancreas divisum, either complete or incomplete (2-7). Most reports of santoriniceles have common features such as pancreas divisum and pancreatitis. The santorinicele has been suggested to be a possible cause of the relative stenosis of the accessory papilla, which in association with unfused dorsal and ventral ducts results in the high intraductal pressure responsible for the recurrent episodes of acute pancreatitis. Therefore, a sphincterotomy or balloon dilatation is believed to be effective in patients with a santorinicele and pancreatitis (3-5, 7).

The prevalence of this rare anomaly is unknown, and it is unclear if it is congenital in origin or is an acquired lesion secondary to a stenosis of the dorsal duct orifice. Because most santoriniceles have been reported in elderly patients, it has been assumed that a santorinicele is most probably an acquired rather than a congenital condition. Some santoriniceles

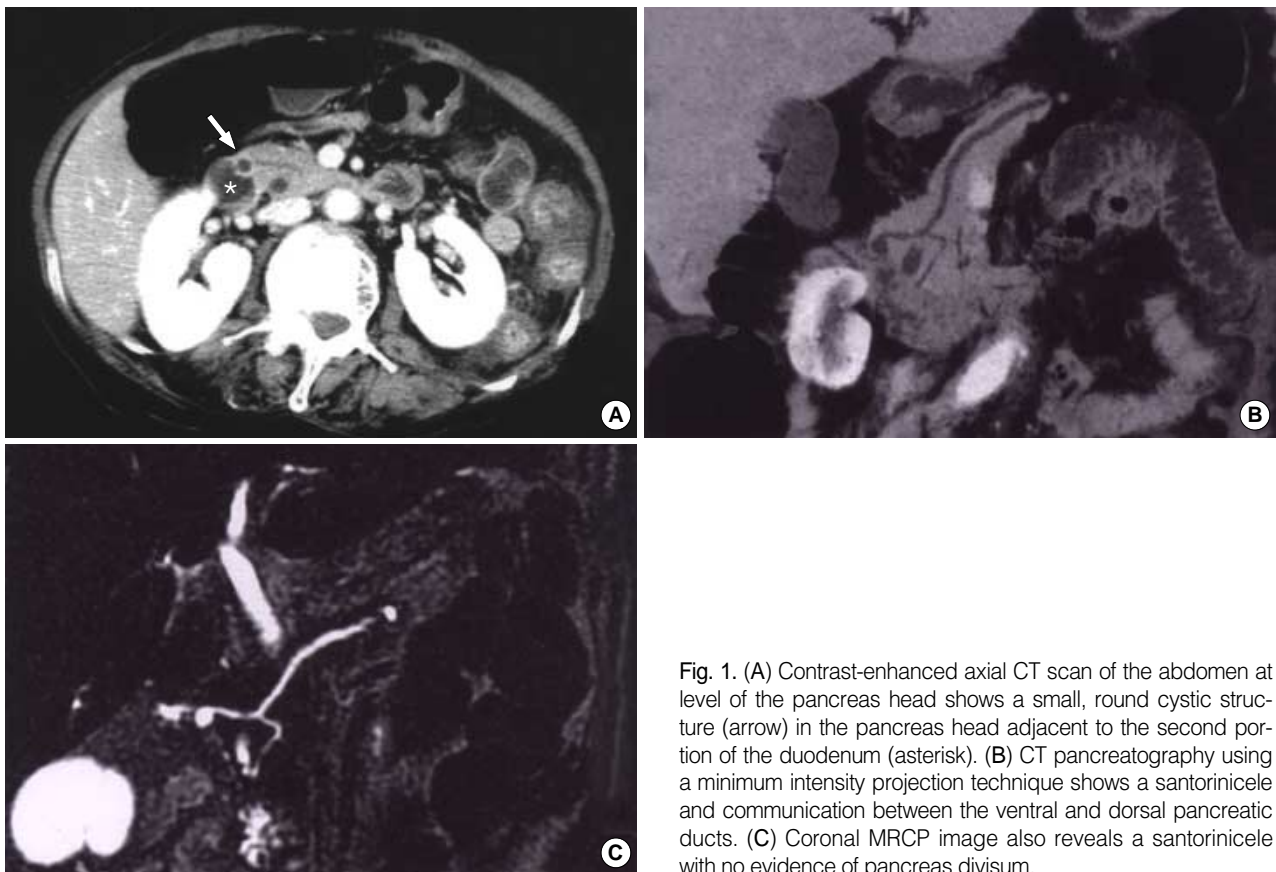


Fig. 1. (A) Contrast-enhanced axial CT scan of the abdomen at level of the pancreas head shows a small, round cystic structure (arrow) in the pancreas head adjacent to the second portion of the duodenum (asterisk). (B) CT pancreatography using a minimum intensity projection technique shows a santorinicele and communication between the ventral and dorsal pancreatic ducts. (C) Coronal MRCP image also reveals a santorinicele with no evidence of pancreas divisum.

are associated with an adjacent duodenal diverticulum. Structural changes might contribute to the acquired mucosal weakness, thereby facilitating the formation of a santorinicele. The association with a duodenal diverticulum may further support the acquired etiology. However, a report of a santorinicele in a pediatric patient without a duodenal diverticulum suggests that the pathogenesis is congenital in some cases (4).

Since all santoriniceles reported were detected in patients with pancreatic divisum, it was unclear if a santorinicele might be present in patients without this anomaly. In 2003, a santorinicele without pancreatic divisum was reported by Byeon et al. (8). This is the only report of a santorinicele not associated with pancreas divisum. However, instead of pancreas divisum, the patient had another congenital anomaly, bifid pancreas, which is an anatomical variant where the main pancreatic duct is bifurcated along its length. On the other hand, our case was characteristic in two ways. First, the patient did not have pancreas divisum and other congenital pancreatic anomalies. Second, CT detected the santorinicele incidentally. Since all santorinicele reported were diagnosed by MRCP or ERCP, this is the first case reported of a santorinicele without pancreas divisum detected by CT.

In conclusion, this case demonstrates that a santorinicele can exist without pancreas divisum. Furthermore, MDCT is believed to be a very useful diagnostic technique for delicate lesions like as the present case.

REFERENCES

1. Eisen G, Schutz S, Metzler D, Baillie J, Cotton PB. *Santorinicele: new evidence for obstruction in pancreas divisum. Gastrointest Endosc 1994; 40: 73-6.*
2. Peterson MS, Slivka A. *Santorinicele in pancreas divisum: diagnosis with secretin-stimulated magnetic resonance pancreatography. Abdom Imaging 2001; 26: 260-3.*
3. Costamagna G, Ingrosso M, Tringali A, Mutignani M, Manfredi R. *Santorinicele and recurrent acute pancreatitis in pancreas divisum: diagnosis with dynamic secretin-stimulated magnetic resonance pancreatography and endoscopic treatment. Gastrointest Endosc 2000; 52: 262-7.*
4. Tang H, Kay CL, Devonshire DA, Tagge E, Cotton PB. *Recurrent pancreatitis in a child with pancreas divisum. Endoscopic therapy of a santorinicele. Surg Endosc 1999; 13: 1040-3.*
5. Seibert DG, Matulis SR. *Santorinicele as a cause of chronic pancreatic pain. Am J Gastroenterol 1995; 90: 121-3.*
6. Manfredi R, Costamagna G, Brizi MG, Spina S, Maresca G, Vecchioli A, Mutignani M, Marano P. *Pancreas divisum and "santorinicele": diagnosis with dynamic MR cholangiopancreatography with secretin stimulation. Radiology 2000; 217: 403-8.*
7. Joo KR, Bang SJ, Shin JW, Kim DH, Park NH. *Santorinicele containing a pancreatic duct stone in a patient with incomplete pancreas divisum. Yonsei Med J 2004; 45: 952-5.*
8. Byeon JS, Kim MH, Lee SK, Yang DH, Bae JS, Kim HJ, Lee SS, Seo DW, Min YI. *Santorinicele without pancreas divisum. Gastrointest Endosc 2003; 58: 800-3.*