Abdom Imaging 26:260–263 (2001)
DOI: 10.1007/s002610000156

Abdominal Imaging

Santorinicele in pancreas divisum: diagnosis with secretin-stimulated magnetic resonance pancreatography

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Received: 13 July 2000/Accepted: 23 August 2000

Abstract
A Santorinicele, or cystic dilatation of the dorsal pancreatic duct at the minor papilla, is seen in a small number of patients with pancreas divisum and may indicate obstruction at the minor papilla, a risk factor for pancreatitis. We present a case of a Santorinicele that was diagnosed with secretin-stimulated magnetic resonance pancreatography and treated with minor papillotomy.

Key words: Pancreatic ducts, MR—Pancreatic ducts—Pancreas divisum—Santorinicele—Secretin.

Pancreas divisum is the most common congenital anomaly of the pancreatic ductal system and has been reported to occur in up to 10% of the general population in an autopsy series [1]. The embryologic etiology for pancreas divisum is the failure of complete fusion of the dorsal and ventral pancreatic anlagen. The duct of Santorini drains the dorsal pancreas to the minor (or accessory) papilla, whereas the duct of Wirsung drains the ventral pancreas to the major papilla. Although usually asymptomatic, patients with pancreas divisum may develop pancreatitis, believed to be due to an inability of the minor papilla to accommodate the exocrine secretions of the dorsal pancreas.

Recent reports in the gastroenterology literature have described a "Santorinicele," or unusual cystic dilatation of the termination of the dorsal pancreatic duct at the minor papilla, in a small number of patients with pancreas divisum (Fig. 1) [2–4]. Noninvasive imaging with secretin-stimulated magnetic resonance pancreatography (MRP) of pancreatic function has also been recently described in the radiology literature [5, 6]. We present a case of pancreas divisum with a Santorinicele that was diagnosed with a secretin-stimulated MRP study and subsequently treated endoscopically with minor papillotomy.

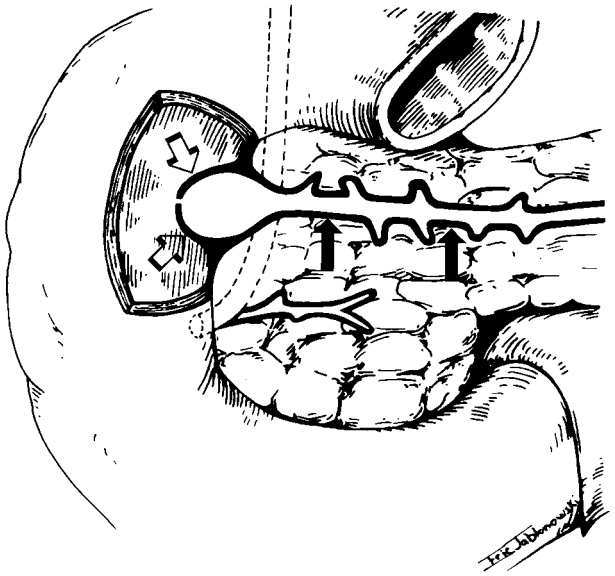

Fig. 1. Line drawing with exposed view of the region of the minor papilla depicts typical pancreatic duct anatomy in pancreas divisum, with the dorsal pancreatic duct (*arrows*) terminating in a Santorinicele (*open arrows*) at the minor papilla. The common bile duct (*dashed lines*) and ventral pancreatic duct terminate normally at the major papilla.

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Case report

A 71-year-old woman was referred for further evaluation of multiple episodes of acute recurrent pancreatitis. She had undergone an open cholecystectomy 16 years previously and had her most recent bout of pancreatitis 3 months before presentation. She had undergone three prior endoscopic retrograde cholangiography (ERC) examinations; the last examination was performed 5 years previously at an outside hospital. Prior endoscopic therapy included a biliary sphincterotomy for treatment of stenosis of the major papilla. No endoscopic retrograde pancreaticogram (ERP) was obtained at the time of the previous endoscopic examinations. Prior abdominal computed tomographic and ultrasound examinations performed 6 years previously showed only mild dilatation of the pancreatic duct in the pancreatic body and tail.

At the time of her recent referral, the patient's serum amylase, lipase, total bilirubin, and transaminase levels were normal. The serum triglyceride level was elevated to 275 ng/dL (normal = 83–141 ng/dL). Physical examination of the abdomen was normal. A repeat endoscopy examination showed changes of a prior biliary sphincterotomy at the major papilla. Repeat ERC showed a dilated biliary tree, post cholecystectomy, with no choledocholiths, and rapid drainage of contrast through the major papilla to the duodenum. Attempts to enter or locate the pancreatic duct orifice from the major papilla for an ERP were unsuccessful. Attempted cannulation of the minor papilla was also unsuccessful.

The patient then underwent an abdominal MR examination, including a secretin-stimulated MRP, performed with a 1.5-T scanner (Signa Horizon, GE Medical System, Milwaukee, WI, USA) using a torso phased array coil. After the pancreatic duct was optimally localized with half-Fourier acquisition single-shot fast spin-echo images (repetition time = infinite, echo time = 1055 ms, field of view = 20 cm × 20 cm, matrix = 256 × 256, flip angle = 90 degrees, section thickness = 40 mm), sequential oblique coronal images were repeated every 2 min through 16 min after the intravenous injection (1 clinical unit/kg) of secretin (Ferring Pharmaceuticals, Suffern, NY, USA). The MRP examination (Fig. 2) confirmed pancreas divisum and showed an abnormal response to secretin, with prolonged dilatation of the dorsal pancreatic duct. With time, the sequential MRP images also showed an approximately 8- × 10-mm focal dilatation of the distal dorsal pancreatic duct at the level of the minor papilla, consistent with a "Santorinicele" (Fig. 2).

The patient subsequently underwent repeat endoscopy, which was notable for a visible prominent bulge at the minor papilla. With successful selective cannulation of the dorsal pancreatic duct through the minor papilla, ERP imaging confirmed dilatation of the dorsal pancreatic duct to approximately 6 mm. A minor papilla sphincterotomy was then performed with a papillotome, with observation of an immediate gush of secretions from the pancreatic duct. A temporary pancreatic duct stent was placed to reduce the risk of postprocedural pancreatitis. Follow-up fluoroscopy performed 2 weeks later showed that the stent had migrated from the pancreatic duct and likely had passed in the stool. The patient has remained well, with no recurrence of pancreatitis in the 20-month follow-up interval since the procedure.

Discussion

Although the clinical significance of pancreas divisum has been controversial, some series have found an increased prevalence of pancreatitis [7], attributed to relative dorsal pancreatic duct obstruction at a stenotic minor papilla. Relative obstruction to exocrine flow may lead to dorsal duct hypertension, which is thought to cause pain. In support of this theory is the observation of a Santorinicele in patients with pancreatitis and pancreas divisum [2–4]. The Santorinicele, or focal cystic dilatation of the distal duct at the minor papilla, has been considered analogous to a similar dilatation of the most distal common bile duct, termed a choledochocele or type III choledochal cyst [3]. Santoriniceles have been postulated to represent congenital or acquired weakness of the mucosal wall in the setting of obstruction [4]. Most Santoriniceles have been reported in the elderly population [2], often associated with adjacent duodenal diverticula. The diverticula may contribute to acquired mucosal weakness, thereby facilitating Santorinicele formation. Observation of a Santorinicele in a pediatric patient [4], however, suggests that some cases may be secondary to congenital weakness. In the pediatric case reported, calculi were also observed in the Santorinicele, indicating chronic stasis of pancreatic secretions and providing further evidence of an obstructive process.

Secretin stimulation of pancreatic exocrine secretion has been reported to be a useful adjunct to sonographic evaluation of the pancreatic duct [8] and has also been recently used with MRP [5, 6]. Whereas both secretin-stimulation ultrasonography and MRP assess the physiologic response to secretin by measure of a normally transient increase in pancreatic ductal caliber, only MRP provides a complete depiction of the entire pancreatic ductal anatomy. Although the dorsal duct may be successfully followed by ultrasound to the minor papilla in some cases, in others it may be obscured distally by bowel gas. Ultrasonography also rarely depicts the ventral duct system. With an increase in pancreatic duct caliber, the secretin-stimulation MRP improves MR imaging of pancreatic duct anatomy, facilitating and often confirming the diagnosis of pancreas divisum.

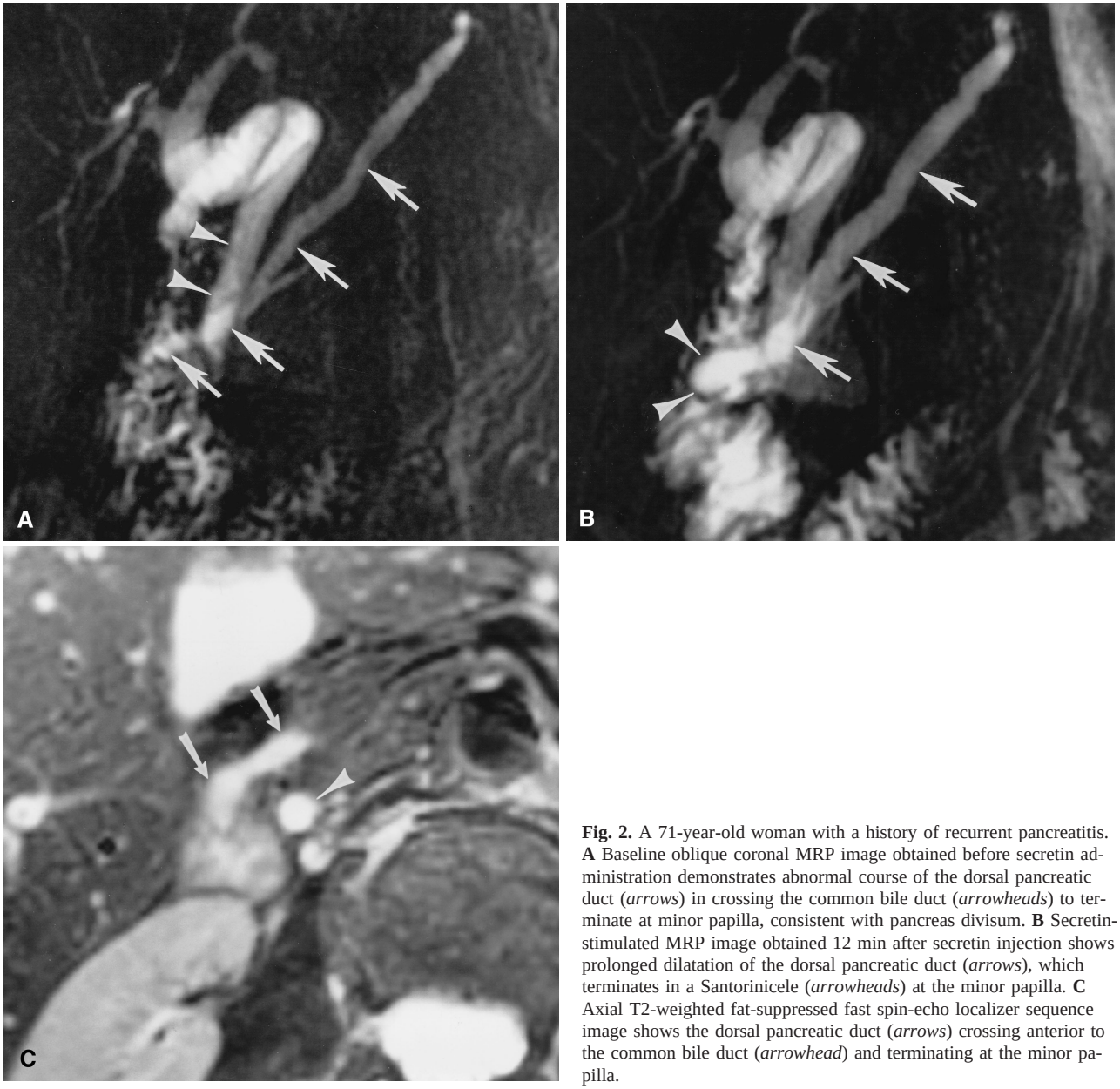

Fig. 2. A 71-year-old woman with a history of recurrent pancreatitis. **A** Baseline oblique coronal MRP image obtained before secretin administration demonstrates abnormal course of the dorsal pancreatic duct (*arrows*) in crossing the common bile duct (*arrowheads*) to terminate at minor papilla, consistent with pancreas divisum. **B** Secretin-stimulated MRP image obtained 12 min after secretin injection shows prolonged dilatation of the dorsal pancreatic duct (*arrows*), which terminates in a Santorinicele (*arrowheads*) at the minor papilla. **C** Axial T2-weighted fat-suppressed fast spin-echo localizer sequence image shows the dorsal pancreatic duct (*arrows*) crossing anterior to the common bile duct (*arrowhead*) and terminating at the minor papilla.

The secretin-stimulation MRP not only improves MR imaging of pancreatic duct anatomy but also is a physiologic challenge to assess the significance of any degree of accompanying stenosis of the minor papilla. In addition to the abnormal response of prolonged pancreatic duct dilatation, the present case also illustrates a Santorinicele at the minor papilla. Patients with a positive secretin-stimulation test for minor papilla stenosis may be more likely to benefit from an endoscopic minor papillotomy [8]. In the present case, the Santorinicele that becomes more apparent after secretin administration is a strong indication of significant papillary stenosis, adding further evidence to the positive abnormal pancreatic duct response to secretin. The noninvasive secretin-stimulation MRP diagnosis of a Santorinicele is a useful finding in patients with pancreas divisum and may help predict which patients are likely to respond to endoscopic papillotomy, although further study of the clinical response to papillotomy in these patients is warranted.

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