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Santorinicele - A Case Report

Dr. Anagha Rajeev Joshi[1], Dr. Sukhada Vilas Kulkarni[2]

[1]MBBS, MD, DMRE, Department of Radiology, LTMG hospital, Sion, Mumbai, Maharashtra, India

[2]MBBS, MD, Department of Radiology, LTMG hospital, Sion, Mumbai, Maharashtra, India

Abstract: ***We present a rare case of santorinicele, diagnosed by ultrasonography (USG) and confirmed on multidetector computed tomography (MDCT) scan of abdomen. We present the clinical details and imaging findings, followed by discussion of the etiology, pathogenesis, and imaging of this condition.***

Keywords: Santorinicele, Pancreas divisum, Santorini, Wirsung, Wirsungocele

1. Introduction

Santorinicele refers to the cystic dilatation of the dorsal pancreatic duct (duct of Santorini), just proximal to the minor papilla [1]. It has been suggested to be a possible cause of a relative stenosis at the minor papilla causing high intraductal pressure and resultant pancreatitis. It is commonly associated with pancreas divisum or other congenital anomalies. We describe a case of a santorinicele along with wirsungocele without pancreas divisum or other congenital pancreatic anomalies that was identified incidentally by multidetector computed tomography.

2. Case Report

A 45 yr old female was admitted with complaints of mild generalized abdominal pain. She did not have any other significant medical or surgical history. She was non-smoker and non-drinker with no family history of gastrointestinal disease. The physical examination as well as the laboratory investigations, including a complete blood count, liver function tests, serum amylase level were within normal limits.

USG of abdomen revealed atrophic pancreas with dilated main pancreatic duct almost mimicking a pancreatic pseudocyst. The accessory duct was also dilated (Figures 1A, 1B).

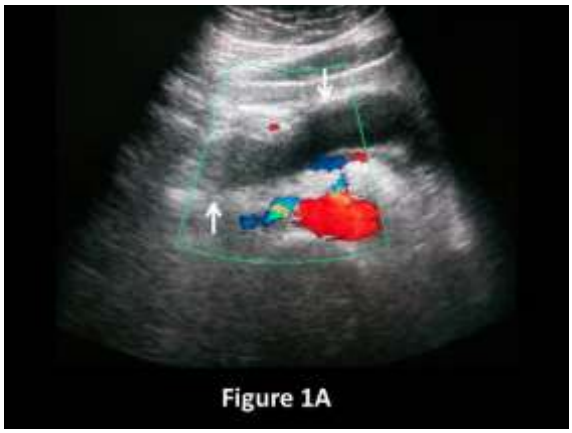

Figure 1A: Colour Doppler ultrasonography of abdomen, showing dilated main pancreatic duct i.e. duct of Santorini (white arrow) showing no colour uptake.

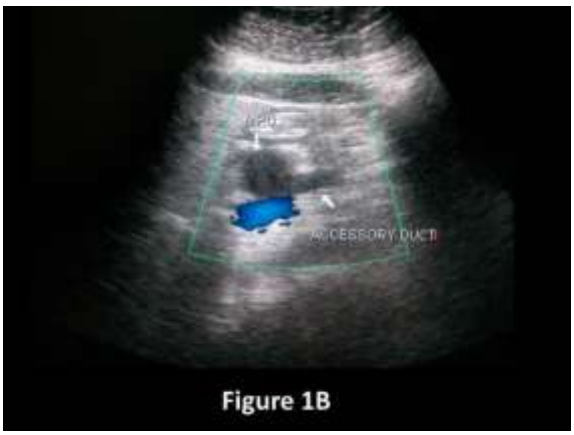

Figure 1B: Colour Doppler ultrasonography of abdomen, showing dilated main pancreatic duct (duct of Santorini) and dilated accessory duct i.e. duct of Wirsung (white arrows) showing no colour uptake

MDCT scan, done later revealed dilation of main pancreatic duct measuring 1.8 cm along with dilatation of the accessory pancreatic duct (Figures 2A, 2B, 2C, 2D).The dorsal pancreatic duct was equal or slightly more prominent than the ventral pancreatic duct.

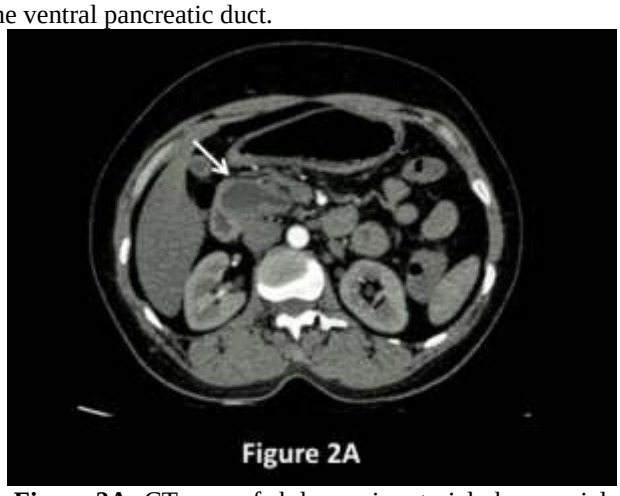

Figure 2A: CT scan of abdomen in arterial phase, axial section showing dilated duct of Santorini (white arrow).

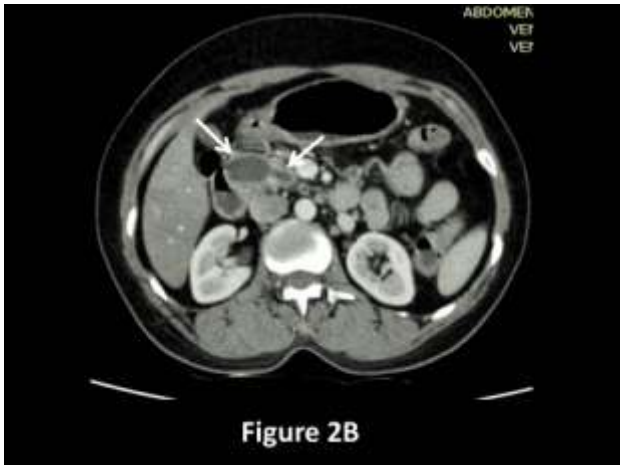

Figure 2B: CT scan of abdomen in venous phase, axial section showing dilated duct of Santorini and duct of Wirsung (white arrows)

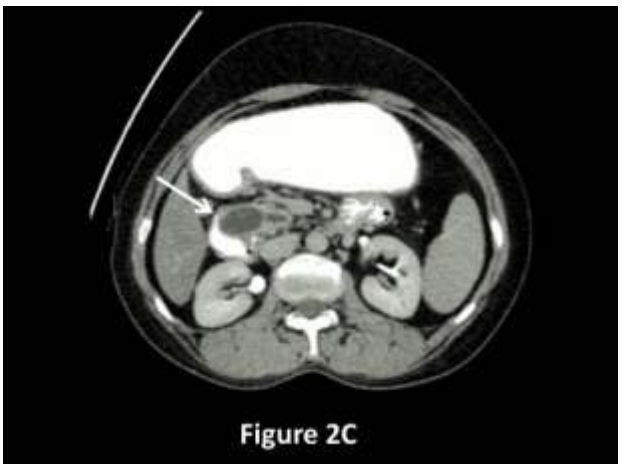

Figure 2C: CT scan of abdomen in delayed phase with patient in right decubitus, after oral contrast administration, axial section showing dilated duct of Santorini bulging into the duodenum (white arrow).

Figure 2D: CT scan of abdomen in venous phase, coronal reconstruction showing dilated duct of Santorini and duct of Wirsung (white arrows)

Magnetic Resonance Cholangiopancreatography (MRCP) was performed which confirmed the findings of santorinicele and wirsungocele (Figure 3).

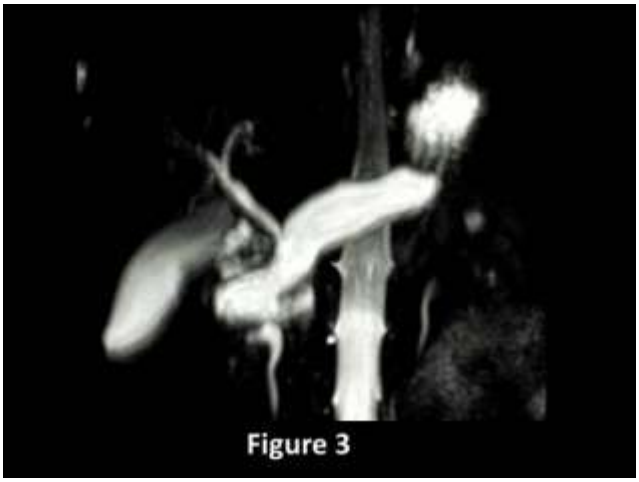

Figure 3: MRCP sequence, showing dilated duct of Santorini and duct of Wirsung (white arrows)

When abdominal MDCT is performed for purposes other than evaluation of pancreatic disease, as in our patient, the finding of an atypical lesion should be correlated with the clinical history and findings on MRI / MRCP or Endoscopic Retrograde Cholangiopancreatography (ERCP) [2] .

Endoscopic ultrasound (EUS) was performed which revealed dilated main as well as accessory pancreatic duct. As the patient had persistent symptoms, she underwent lateral pancreatico-jejunostomy with Roux-en-Y anastomosis. The patient had uneventful post-operative course in the ward with resolution of abdominal pain and was discharged with no new complaints.

3. Discussion

Pancreas develops from two outpouchings, i.e. dorsal and ventral pancreatic buds of the endodermal lining of the foregut. The dorsal pancreas develops faster than the ventral pancreas. In addition, the ventral pancreas rotates toward the dorsal pancreas. Finally, the ventral and dorsal pancreas join and the ductal systems fuse so that secretions from the ventral pancreas enter the shared ductal system of the ventral pancreas (duct of Wirsung) and common bile duct. In the final anatomic arrangement, the head of the pancreas originates from both the dorsal pancreas and the ventral pancreas, the body and tail of the pancreas originate from the dorsal pancreas, whereas the ventral pancreas portion is called the uncinate process.In most cases, the pancreatic duct from the dorsal pancreas fuses with the pancreatic duct from the ventral pancreas to form the main pancreatic duct. After fusion, the pancreatic secretions from the entire pancreas and biliary secretions gain access to the duodenum by way of the ventral pancreatic duct [3].

Though congenital anomalies of the pancreas and pancreatic duct are relatively uncommon and are often discovered as an incidental finding in asymptomatic patients, some of these anomalies may lead to various clinical symptoms such as recurrent abdominal pain, nausea and vomiting. [4]

Anatomic variations and developmental anomalies of the pancreas and pancreatic duct include variations of the course of the pancreatic duct (descending, sigmoid, vertical and

loop shaped course), variation of the configuration of the pancreatic duct (bifid configuration with dominant duct of Wirsung, dominant duct of Santorini without divisum, absent duct of Santorini and ansa pancreatica), duplication anomalies, anomalous pancreaticobiliary ductal junction, cystic dilatations of terminal portions of the ducts of Wirsung and Santorini termed Wirsungocele and Santorinicele, pancreas divisum, annular pancreas, ectopic pancreas, pancreatic agenesis and hypoplasia of the dorsal pancreas and accessory pancreatic lobe[4].

Santorinicele refers to the cystic dilatation of the dorsal pancreatic duct (duct of Santorini), just proximal to the minor papilla. 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. Santorinicele is the dilatation involving the duct of Santorini, whereas Wirsungocele involves the duct of Wirsung.

Santorinicele has been most commonly associated with pancreatic divisum, i.e. santorinicele with pancreas divisum (SWPD). It has also been recorded without pancreatic divisum i.e. Santorinicele without pancreas divisum pathophysiology (SWOPP), however many of these are accompanied by a dominant dorsal pancreatic duct, in which the caliber of the dorsal pancreatic duct was wider than that of the ventral duct, which is similar pathophysiology to that of pancreas divisum. SWOPP is a relatively rare condition found mostly in patients suffering chronic pancreatitis, and considered an acquired condition[1].

The santorinicele has been suggested to be a possible cause of the relative stenosis of the accessory papilla, which if associated with unfused dorsal and ventral ducts results in the high intraductal pressure responsible for the recurrent episodes of acute pancreatitis.

The prevalence of this lesion 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. As most santoriniceles have been reported in elderly patients, it has been assumed that a santorinicele is most probably an acquired condition. Structural changes might contribute to the acquired mucosal weakness, facilitating the formation of santorinicele. The association with duodenal diverticulum in some cases may further support the acquired etiology[5]. However, a report of santorinicele in a pediatric patient without a duodenal diverticulum suggests that the pathogenesis is congenital in some cases[6].

4. Conclusion

Congenital anomalies of pancreas and pancreatic duct, though not commonly encountered in the radiological evaluation, radiologists must be aware of these anomalies. MDCT may allow diagnosis of these anomalies; however MRCP is most accurate in depicting the details. Recognition of developmental anomalies of the pancreas and pancreatic duct is important because these anomalies may be a surgically correctable cause of recurrent pancreatitis, help in surgical planning and prevention of inadvertent ductal injury.

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Author Profile

Dr. Anagha Rajeev Joshi, MBBS, MD, DMRE, Department of Radiology, LTMG hospital, Sion, Mumbai, Maharashtra, India

Dr. Sukhada Vilas Kulkarni, MBBS, MD, Department of Radiology, LTMG hospital, Sion, Mumbai, Maharashtra, India