

CASE REPORT

Recurrent Acute Pancreatitis and Wirsungocele. A Case Report and Review of Literature

Rajesh Gupta, Sandeep Lakhtakia, Manu Tandan, Darisetty Santosh,
Guduru Venkat Rao, Duvvuru Nageshwar Reddy

Asian Institute of Gastroenterology. Hyderabad, India

ABSTRACT

Context The association of Santorinicele with pancreas divisum has been well described. There is an increased risk of recurrent acute pancreatitis in patients with pancreas divisum who also have Santorinicele. Focal saccular dilation of the terminal part of the main pancreatic duct has been described as an incidental finding and termed, 'Wirsungocele'.

Case report We report a case of a 39-year-old male who had recurrent episodes of acute pancreatitis. Laboratory tests, US of the abdomen and CECT of the abdomen confirmed acute pancreatitis. MRCP showed focal saccular dilation of the terminal part of the main pancreatic duct suggestive of Wirsungocele. An ERCP confirmed MRCP findings. An endoscopic pancreatic sphincterotomy was performed and a 5 Fr single pigtail pancreatic stent was placed. The pancreatic stent was removed after 4 weeks. At the 12-week follow-up, the patient was asymptomatic.

Conclusion This case report describes the association of Wirsungocele with recurrent acute pancreatitis.

INTRODUCTION

Cystic dilatation of the intramural portion of the dorsal pancreatic duct, i.e. Santorinicele,

has been described in association with pancreas divisum [1, 2]. It has also been shown that patients with pancreas divisum who have Santorinicele have an increased risk of recurrent acute pancreatitis and are more likely to benefit from pancreatic endotherapy [3, 4, 5]. Focal saccular dilatation of the terminal part of the main pancreatic duct has been aptly termed, Wirsungocele by Baron *et al.* [6]. However, it was reported as an incidental finding. We report a patient who had recurrent acute pancreatitis and a Wirsungocele.

CASE REPORT

A 39-year-old male presented to our institute with a history of recurrent episodes of pancreatitis during the previous year. His symptoms had started one year earlier when the first attack of pancreatitis occurred. The results of his tests showed elevated serum lipase (3,046 IU/L; reference range: 5.6-51.3) and amylase (3,156 IU/L; reference range: 0-220 IU/L).

Other examinations, which included a complete blood count, liver function test, kidney function test, lipid profile and transabdominal ultrasound, were normal. He improved with conservative treatment. A second attack of pancreatitis recurred after 3 months and this also subsided with conservative treatment. The patient remained asymptomatic for the next 6 months. In the past 3 months, the patient had two more



Figure 1. MRCP showing focal saccular dilatation of the terminal part of the main pancreatic duct.

episodes of documented pancreatitis which also subsided with conservative treatment. Contrast-enhanced CT of the abdomen was also done which revealed a bulky pancreas.

The patient was referred to our institute for evaluation and further management. The examination included a complete blood picture, liver function tests, kidney function tests, lipid profile, serum amylase, lipase and IGg4, and transabdominal ultrasound: all were normal. An EUS showed a heterogeneous pancreas. MRCP showed focal saccular dilatation of the terminal part of the main pancreatic duct measuring 11x7 mm suggestive of Wirsungocoele (Figure 1). An ERCP confirmed the MRCP finding (Figure 2). An endoscopic pancreatic sphincterotomy was performed and a 5 Fr single pigtail pancreatic stent was placed. The pancreatic stent was removed after 4 weeks. The patient was asymptomatic at week 12 of the follow-up.

DISCUSSION

Santorinicele (a cystic dilatation at the orifice of the minor papilla) has been well documented among patients with recurrent attacks of acute pancreatitis and pancreas divisum [1, 2, 3]. It has been postulated that relative stenosis at the minor papilla in patients with pancreas divisum leads to an increase in intraductal pressure. This increased intraductal pressure, in addition to congenital or acquired weakness in the duct wall, leads to the formation of a Santorinicele which, in turn, predisposes further obstruction

at the minor papilla causing recurrent attacks of pancreatitis [4, 5].

Until now, an association between Wirsungocoele and recurrent acute pancreatitis has not been reported. Baron *et al.* described Wirsungocoele as an incidental finding in their case report. To our knowledge, this is the first case of recurrent acute pancreatitis in association with a Wirsungocoele. Though endoscopic US did not show any focal cystic dilatation of the main pancreatic duct, both MRCP and ERCP confirmed focal saccular dilatation of the terminal part of the main pancreatic duct consistent with a diagnosis of a Wirsungocoele. It is possible that EUS could not visualize the focal cystic dilatation due to compression of the duodenal wall by the echoendoscope. Several theories have been proposed to explain the etiology and pathophysiology of terminal cystic ductal dilatations. It has been postulated that decreased autonomic innervation of the sphincter of Oddi leads to non-coordination



Figure 2. Endoscopic retrograde pancreatogram showing focal cystic dilatation of the terminal part of the main pancreatic duct.

of the sphincter and functional obstruction at the papillary orifice [7]. The anomalous pancreatico-biliary junction with a long common channel (more than 5 mm) has also been proposed to be an important factor in the formation of choledochoceles [8]. Some studies have shown a focal dilation of the main pancreatic duct in the head region with increase of age [9].

There was no demonstrable peri-ampullary diverticulum or anomalous pancreatico-biliary junction to predispose the ductal wall to weakness in our patient. However, the possibility of functional obstruction at the papillary orifice cannot be ruled out. The pathophysiological mechanism for Wirsung-occele formation is unclear. Whether the association of recurrent acute pancreatitis and Wirsung-occele is causative or incidental remains to be established. Similarly, the role of pancreatic endotherapy is also unsubstantiated.

In conclusion, this case report clearly shows the presence of Wirsung-occele in patients with recurrent acute pancreatitis.

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Correspondence

Rajesh Gupta
Asian Institute of Gastroenterology
6-3-661, Somajiguda, Hyderabad
500082 India
Phone: +91-40.23.378.888
Fax: +91-40.23.324.255
E-mail: rajeshdrgupta@msn.com

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