

CASE REPORT

Recurrent Acute Pancreatitis in a Patient with Wirsungocele and Neuroendocrine Tumor of Ampulla of Vater

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ABSTRACT

Context Wirsungocele has recently been shown to be associated with acute recurrent, severe necrotizing pancreatitis and chronic pancreatitis or chronic pain in abdomen. Till to date there is no report on association of wirsungocele with an ampullary neuroendocrine tumor, and recurrent pancreatitis. **Case report** We report a first ever case of wirsungocele diagnosed on EUS, its association with neuroendocrine tumor of ampulla and recurrent acute pancreatitis. **Conclusion** This case report highlights the diagnostic utility of EUS in diagnosing small ampullary pathology like wirsungocele and neuroendocrine tumor.

INTRODUCTION

Recurrent acute pancreatitis can rarely be due to an obstructive pathology at the ampulla of Vater. EUS is an excellent modality for detecting small ampullary pathology. Recently wirsungocele has been described as an etiological factor for recurrent pancreatitis. We encountered a patient who had recurrent pancreatitis due to a wirsungocele and a small neuroendocrine tumor at the ampulla of Vater, both diagnosed by endosonography.

CASE REPORT

A 63-year-old male had two episodes of acute pancreatitis by biochemistry and abdominal ultrasonography. EUS findings showed an ampullary mass and cystic dilatation of terminal portion of duct of Wirsung suggestive of wirsungocele (Figures 1 and 2). EUS guided FNA from the ampulla suggested a neuroendocrine tumor. MRCP confirmed EUS findings of wirsungocele and ampullary mass (Figure 3). ERCP (Figure 4) and endoscopic ampullectomy was then performed and patient made an uneventful recovery. The histopathological examination and immunohistochemistry showed neuroendocrine tumor (pathological classification: G1) with cells expressing synaptophysin and chromogranin (Figure 5). The Mib proliferation index was less than 2%. The pancreatic duct was seen well dilated within the tumor tissue.

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Key word Endosonography; Neuroendocrine Tumors; Pancreatitis

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DISCUSSION

Wirsungocele is the term coined for cystic dilation of the terminal part of the duct of Wirsung. We found a total of eight cases of wirsungocele described in five papers indexed in PubMed [1, 2, 3, 4, 5]. Abu-Hamda *et al.* first described wirsungocele as an incidental finding on ERCP [1]. Four of these cases (50.0%) of wirsungoceles are associated with acute recurrent pancreatitis [1, 2, 3, 4, 5]. Five reported cases (62.5%) are males and age of presentation is after 4th decade [1, 2, 3, 4, 5]. Endoscopic pancreatic sphincterotomy and stenting appears very effective and durable [2, 3, 4].



Figure 1. EUS of the ampulla of Vater showing cystic dilation of terminal pancreatic duct.

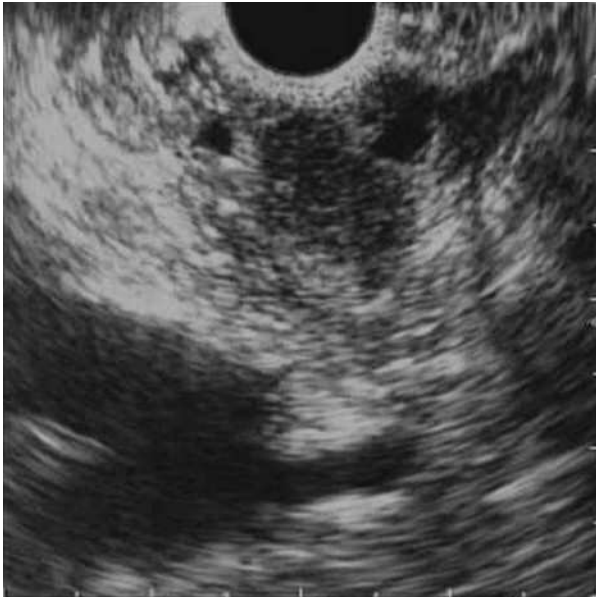


Figure 2. EUS of the ampullary region showing hypoechoic lesion at the ampulla. The wirsungocele is also seen.

Our patient also had good symptom relief following endotherapy. EUS as a diagnostic tool has not been used adequately so far. For the first time we could demonstrate wirsungocele on EUS and its association with neuroendocrine tumor.

Ampullary neuroendocrine tumor is a rare entity, and constitutes less than 1% of all gastrointestinal neuroendocrine tumors [5]. In our case possibility of wirsungocele formation due to long standing obstruction by the neuroendocrine tumor is unlikely. A complete upstream ductal dilatation is more likely to occur due to a tumor. The episodes of acute recurrent pancreatitis were more likely to be due to wirsungocele as the duct is not compressed by the tumor on histology.

CONCLUSION

This case report highlights the diagnostic utility of EUS in diagnosing small ampullary pathology like

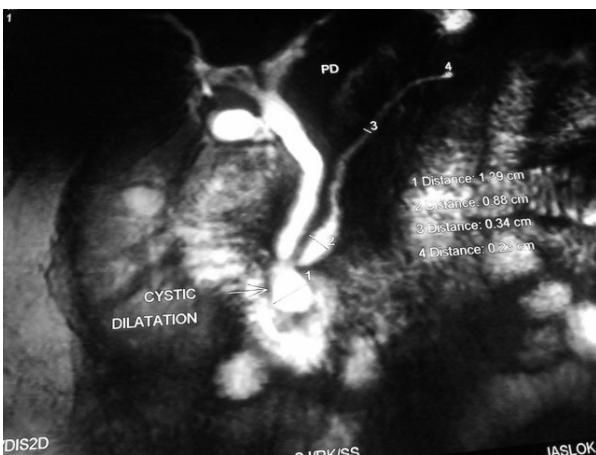


Figure 3. MRCP showing cystic dilation of terminal part of main pancreatic duct.

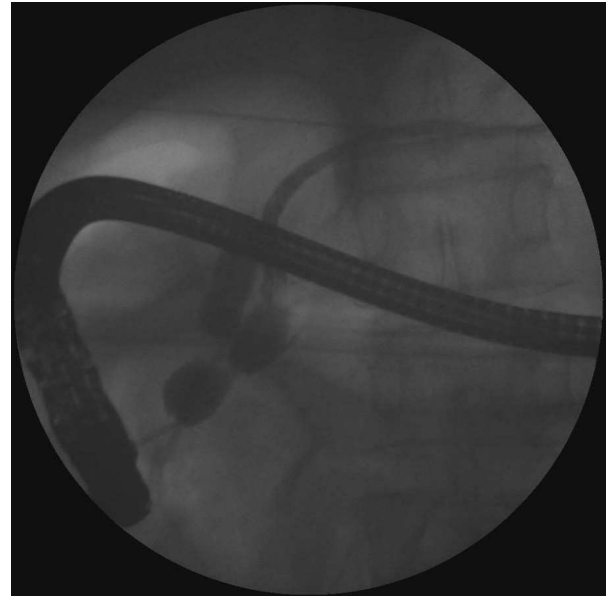


Figure 4. ERCP showing dilation of terminal part of pancreatic duct.

wirsungocele and neuroendocrine tumor. The association of wirsungocele with neuroendocrine tumor of ampulla and recurrent pancreatitis has also been reported for the first time.

Conflict interests The authors have no potential conflict of interest

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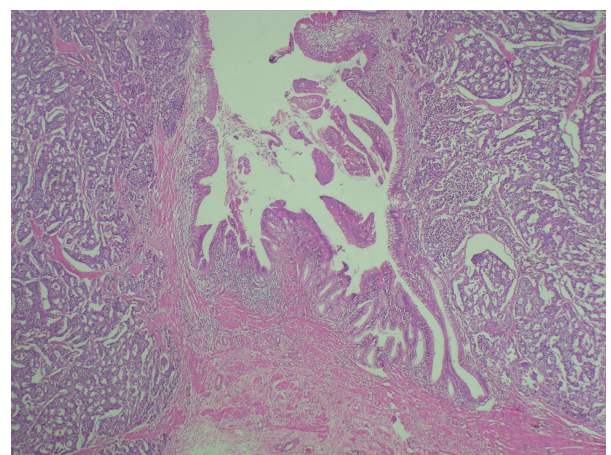


Figure 5. Microphotograph showing neuroendocrine tumor at the ampulla. The pancreatic duct is also seen. (Magnification, x40).

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