

CASE REPORT

Solitary True Cyst of the Pancreas in Adults. A Report of Two Cases

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ABSTRACT

Context Solitary true cysts of the pancreas in adults are extremely rare and only few cases have been reported in the literature. The etiology and natural history of these lesions remain unknown and treatment is not standardized. We describe two additional resected cases. **Cases reports** The first patient was a young woman with an incidental 3 cm cyst located in the pancreatic head who underwent enucleation. The second patient was a young woman with a large 8 cm symptomatic cyst located in the pancreatic tail who underwent a laparoscopic spleen-preserving distal pancreatectomy. Histological examination revealed fibrous walls lined by a monolayer of cuboidal epithelium in both cases. **Conclusions** A preoperative work-up alone does not always allow an accurate diagnosis, but it is useful in determining lesion characteristics and guiding therapeutic decision making. When surgery is indicated, a limited resection is warranted in most cases.

INTRODUCTION

Solitary true cysts of the pancreas in adults are extremely rare and only a few cases have been reported in the English literature [1, 2, 3, 4, 5, 6, 7, 8, 9]. These cysts are typically unilocular and lined by cuboidal epithelium. The Etiology and natural history of these lesions remain unknown and treatment is not standardized. Even though imaging techniques continue to improve, diagnostic accuracy is sometimes limited. We herein describe two additional resected cases.

CASE REPORTS

Case 1

The first patient was a 37-year-old woman admitted to our department for a left acute urolithiasis. She had no history of alcohol abuse, gallstone disease, pancreatitis and abdominal trauma. Abdominal ultrasound revealed an incidental anechogenic 3 cm cystic lesion in the pancreatic head. Serum tumor marker levels (CEA, CA 19-9) were within the normal range, as well as biochemical markers such as amylase and lipase. A Helical computed tomography (CT) scan first showed a 3 cm thin-walled, non contrast-enhancing, unilocular lesion in the pancreatic head (Figure 1). Magnetic

resonance imaging (MRI) showed a unilocular lesion hyperintense on T2-weighted image without internal septations and a normal pancreatic and common bile duct. Endoscopic ultrasonography (EUS) was not performed since the cystic lesion was 3 cm in diameter and the patient was young. Surgical resection was scheduled. At laparotomy, the cyst was located in the pancreatic head, well circumscribed and distinct from the main pancreatic duct, justifying cyst enucleation. Frozen section evaluation was negative for malignancy. Macroscopically, the cystic lesion was 3 cm in diameter, unilocular, thin-walled, and contained a clear fluid. Analysis of the cystic fluid revealed amylase and lipase levels similar to those of normal serum and tumor markers levels within the normal range. Histologically, the cyst had thin fibrous walls lined by a cuboidal epithelium and normal adjacent pancreatic



Figure 1. Computed tomography scan showing a 3 cm thin-walled, non contrast-enhancing, unilocular cystic lesion in the pancreatic head.

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parenchyma (Figure 2). Immunohistochemical analysis was negative for CA 19-9 staining. The Post-operative course was uneventful and the patient was discharged 8 days after surgery. At the 2-year follow-up, she is alive without evidence of recurrence.

Case 2

The second patient was a 21-year-old woman admitted to our department for recurrent episodes of epigastric pain, nausea and dyspepsia. The Past history was again unremarkable. Physical examination revealed a marked tenderness in the upper left abdomen. Serum tumor marker levels and biochemical markers were within the normal range. Helical CT scan showed an 8 cm thin-walled, non contrast-enhancing, unilocular lesion in the pancreatic tail with minimal dilatation of the pancreatic duct (Figure 3). At laparoscopy, the large cyst was located in the pancreatic tail without evidence of liver or peritoneal involvement and a laparoscopic spleen-preserving distal pancreatectomy was carried out. Frozen section evaluation was negative for malignancy. Macroscopically, the cystic lesion was 8 cm in diameter, unilocular thin-walled, containing clear fluid. Fluid analysis was negative for enzyme and tumor marker levels and the cyst had the same histological features as in the first case. Immunohistochemical analysis was also negative for CA 19-9 staining. During the postoperative course a pancreatic fistula was conservatively resolved and the patient was discharged 21 days after the operation. At the 6-month follow-up, she is alive without evidence of recurrence.

DISCUSSION

Pancreatic cystic lesions are increasingly being detected due to a heightened awareness and the widespread use of cross-sectional imaging [9, 10, 11, 12]. Solitary true cysts of the pancreas are typically diagnosed in childhood, predominantly in infants, suggesting a congenital nature [1, 13]. They are single or multiple, usually small and can be found in association with von Hippel-Lindau syndrome or polycystic disease. Descriptions in adults are extremely

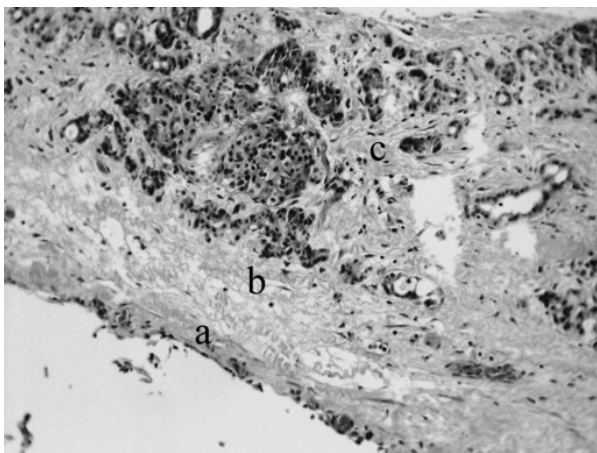


Figure 2. Histopathologic examination showed a fibrous cyst wall (b) lined by a monolayer of cuboidal epithelium (a) with adjacent pancreatic parenchyma (c) (H&E x100).

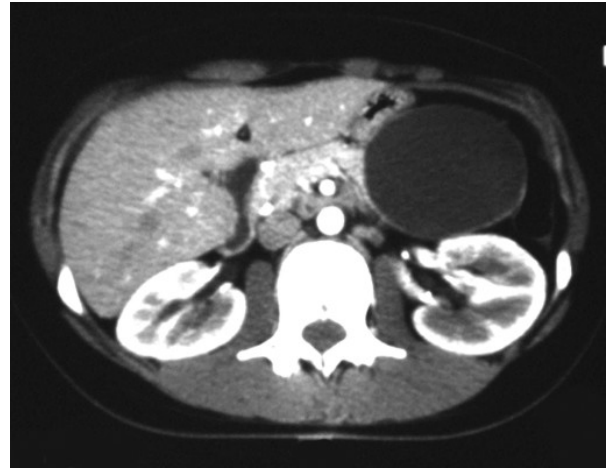


Figure 3. Computed tomography scan showing a large 8 cm thin-walled, non contrast-enhancing, unilocular cystic lesion in the pancreatic tail.

rare. The mean age of patients at diagnosis is 45 years with a predominantly female prevalence and most cysts are located in the tail of the pancreas. Symptoms are related to the size and location of the lesion, but some patients may be asymptomatic [1, 2, 3, 4, 5, 6, 7, 8, 9]. Modern imaging techniques usually show a well-defined unilocular cyst with a thin wall, no mural nodularity and calcifications on enhancement with contrast. However, even with the combined use of laboratory data, clinical information and modern diagnostic imaging (CT scan, MRI and EUS) it may be difficult to accurately differentiate between some solitary true cysts and cystic neoplasms of the pancreas, particularly macrocystic serous cystadenoma or a small unilocular mucinous tumor [1, 2, 3, 4, 5, 6, 7, 8, 9, 14, 15, 16, 17]. Cyst fluid analysis may be helpful, but it is not always a sensitive and specific test, since both normal and elevated tumor marker levels have been reported [1, 2, 3, 4, 5, 6, 7, 8, 9, 14, 15, 16, 17]. Solitary true pancreatic cysts are often large and always unilocular, without communication with the ductal system or internal septa. They are lined by a single layer of cuboidal epithelium without morphological alterations, usually containing a clear fluid with normal amylase and lipase concentrations. The epithelial lining cells may be positive to CA 19-9 antibodies by immunohistochemically staining [1, 2, 3, 4].

Excluding symptomatic cases, surgical resection is mostly indicated for two reasons: the possibility of growth of the lesion, especially in young patients and the inability to exclude a cystic neoplasm definitely. According to most authors, cystic lesions of 3 cm in diameter should always be resected, especially in young patients as in our first case [9, 10, 11, 12, 18, 19]. Treatment varies depending on cyst size and location but a conservative approach is advisable. Enucleation is a safe and effective procedure for small lesions, especially if located in the pancreatic head, but the benign nature of the cyst should be confirmed by frozen section evaluation intraoperatively. A major

procedure such as a spleen-preserving distal pancreatectomy may otherwise be necessary [1, 2, 3, 4, 5, 6, 7, 8, 9, 10, 20, 21, 22]. Laparoscopic surgery may represent a sound and effective approach in selected lesions, as was the case in our second patient [5, 10, 23, 24].

In conclusion, solitary true cysts of the pancreas in adults are extremely rare. Excluding symptomatic cases, surgical resection is mainly indicated when a differential diagnosis with cystic neoplasms is impossible. A limited resection is warranted in most cases.

Conflict of interest The authors have no potential conflicts of interest

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