



Unilocular macrocystic serous cystadenoma of the pancreas—atypical features: a case report

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ABSTRACT

The authors report and discuss an exceedingly rare case of a unilocular macrocystic serous cystadenomas diagnosed in a 63-year-old female patient, which was preoperatively misdiagnosed as a mucinous cystic neoplasm, due to the atypical magnetic resonance (MR) imaging features shown at presentation and the misleading results obtained through cystic fluid analysis. This manuscript overviews the typical and atypical manifestations of this entity and highlights the advantages, potential limitations and pitfalls of both MR imaging and cystic fluid analysis.

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1. Case report

A 63-year-old female patient with chief complaint of non specific recurrent abdominal pain for 1 year was referred to our institution. She reported, on one occasion, a mild elevation of amylase and C-reactive protein (CRP), identified in another institution, but without clinical evidence of acute pancreatitis.

The physical examination of the abdomen was unremarkable and complete laboratory evaluation was normal.

The patient reported no personal or family history of pancreatic disease.

The abdominal magnetic resonance (MR) imaging detected a 5 cm pancreatic unilocular cyst with lobulated contours in the uncinate process (Fig. 1A,B). The intracystic fluid presented a fluid–fluid level and had most likely a proteinaceous/hemorrhagic content, based on the high signal intensity observed on both T2- and fat-suppressed

T1-weighted MR images (Fig. 1C,D). The main pancreatic duct presented a normal diameter and no obvious communication with the cystic lesion (Fig. 1A). Pseudocyst, mucinous cystic neoplasms (MCN), lymphoepithelial cyst, and unilocular macrocystic SCA were considered in the differential diagnosis. In addition, two other subcentimetric unilocular cystic lesions were detected on the pancreatic tail (Fig. 1A; arrow). The endoscopic ultrasound-guided fine-needle aspiration (EUS-FNA) of the cystic fluid revealed elevated levels of carcinoembryonic antigen (CEA) (922 ng/ml) and a high fluid viscosity (1,78), but low levels of amylase (112 U/L) and lipase (3490 U/L). Cytology was not contributive.

The following MR examination, performed 3 months later (Fig. 2A–D), detected a change in the cystic fluid composition. The intracystic signal presented a simple fluid appearance, showing low signal intensity on fat-suppressed T1-weighted MR sequences (Fig. 2D) and high signal intensity on T2-weighted MR sequences (Fig. 2A, B, C). The subsequent cystic fluid analysis obtained by EUS-FNA revealed a striking rise of the CEA level (1900 ng/ml), low levels of amylase (35 U/L) and lipase (82 U/L), viscosity at 1.32 and mild to moderate nuclear atypia in small clusters of glandular epithelial cells, suggesting the diagnosis of MCN (Fig. 3).

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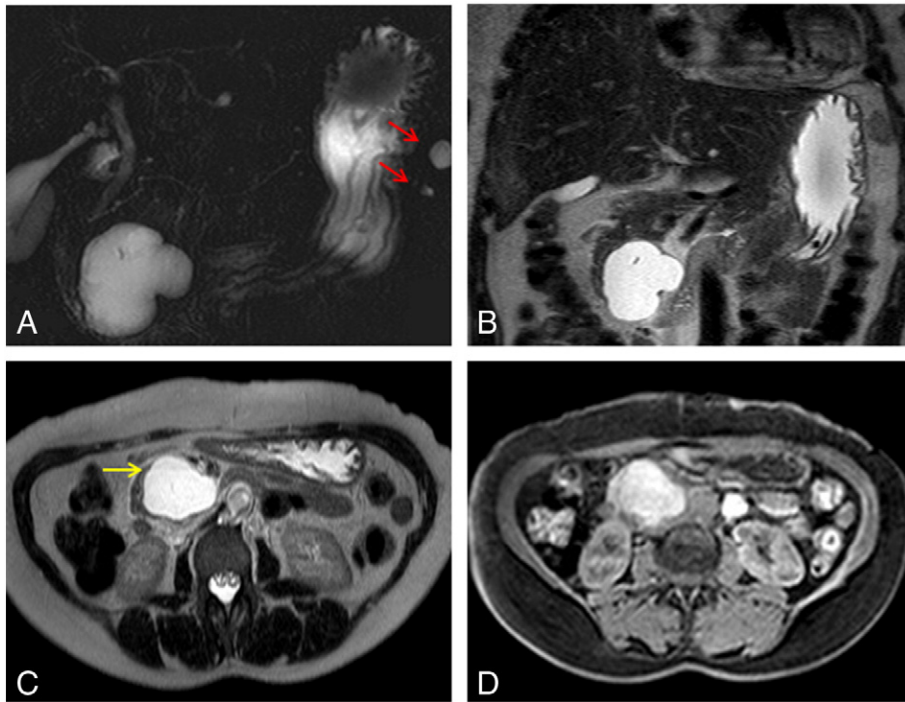


Fig. 1. MR imaging firstly performed during the diagnostic workup of the pancreatic cystic lesion. Coronal Magnetic Resonance cholangiopancreatography (MRCP; A) and coronal T2-weighted MR images (B) depict the presence of a unilocular cystic lesion with lobulated contours, located in the pancreatic head. Two smaller lesions are visible in the pancreatic tail (red arrows). Axial T2-weighted (C) and axial unenhanced fat-suppressed T1-weighted (D) MR images. The high signal intensity of the cystic fluid on T1-weighted MR image suggests a proteinaceous/hemorrhagic component. An intracystic fluid-fluid level is observed on T2-weighted MR images (yellow arrow).

Considering the high suspicion of a potentially malignant cystic lesion, a surgical resection was proposed and a pylorus-preserving duodenopancreatectomy was performed. On gross examination, the mass presented as a thin-walled unilocular cyst and was filled with

serous-fluid (Fig. 4A,B). Microscopy revealed a single-layered epithelium composed of cuboidal cells, with round, hyperchromatic nuclei, no atypia and high intracytoplasmic levels of glycoproteins (Fig. 4C). Neither intracystic mucin content nor solid mural nodules were

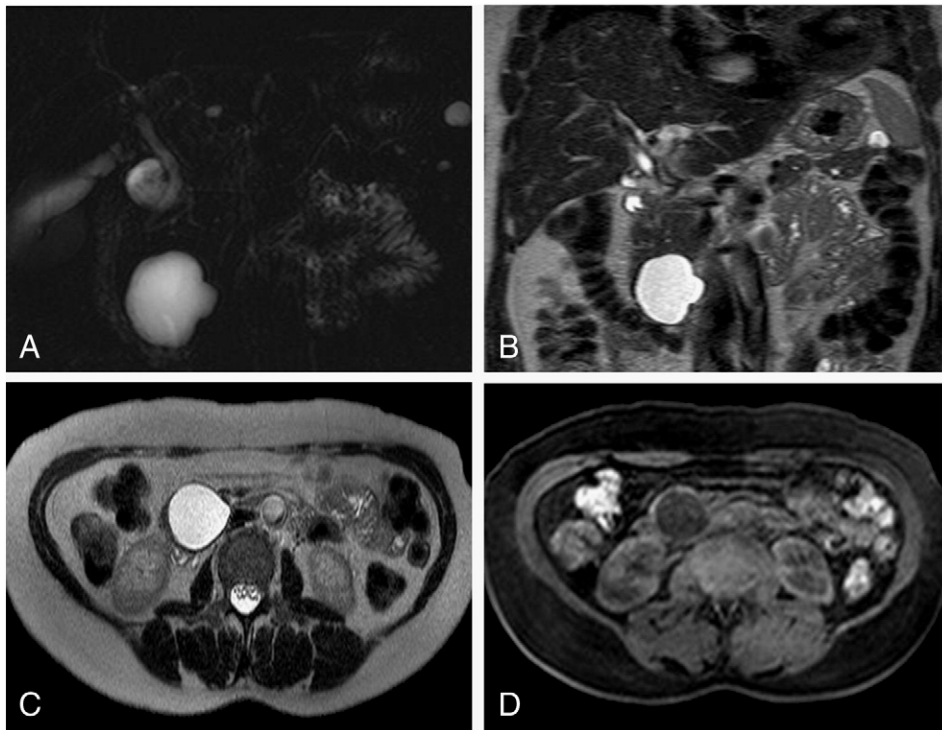


Fig. 2. MR imaging performed three months later demonstrated size stability of pancreatic cystic lesions. Coronal magnetic resonance cholangiopancreatography (MRCP; A), coronal (B) and axial (C) T2-weighted MR images and axial unenhanced fat-suppressed T1-weighted (D) MR images. Despite maintaining high signal intensity on T2-weighted MR sequences, the cystic fluid had become hypointense on T1-weighted MR sequences suggesting a simple fluid appearance.

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