

# A rare case of pancreatic endometriosis in a postmenopausal woman and review of the literature

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## Abstract

Pancreatic endometriosis is very rare with only a few cases reported in the literature. The imaging features are non-specific and the definitive diagnosis is usually only established after surgery. We report on a 68-year-old woman with left upper quadrant pain who demonstrated a mass in the pancreatic tail on imaging. Laboratory results showed only mildly elevated liver enzymes, tumor markers were within the normal range. A left pancreatectomy was performed, frozen section suggesting a benign lesion, and final histopathology confirmed endometriotic cysts. A research of the literature found only eight reported cases of endometriotic cysts of the pancreas, with the majority affecting premenopausal women. Preoperative diagnosis is challenging and most patients undergo resection because of suspected neoplasm. Thorough diagnostic workup may help in avoiding extensive surgery and reduce postoperative complications.

## Keywords

Endometriosis, pancreas, benign, postmenopausal, computed tomography (CT), magnetic resonance imaging (MRI)

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## Introduction

Endometriosis is a fairly common condition, affecting about 5–10% of women in the reproductive age group. Extragenital manifestations of endometriosis occur in up to 40% of patients with pelvic endometriosis, with intestinal (mostly rectum and sigma), urinary tract, abdominal wall/peritoneal, and thoracic manifestations being the most common. Endometriotic cysts have been described in multiple organs such as the lungs, liver, or brain, but there are only a few reports in the literature regarding involvement of the pancreas (1–4). A search on PubMed found eight case reports, with most of the patients within the pre-menopausal or perimenopausal age range. There are some more cases reported concerning hepatic endometriosis, with some of the patients being postmenopausal women (5–15). Patients with pancreatic endometriosis presented with a history of abdominal pain, acute pancreatitis, weight loss, or acute abdomen. The imaging features demonstrated cystic lesions of variable sizes and a varying degree of

hemorrhagic content, with a nodular component described in one case (11). Preliminary diagnoses included cystic neoplasms and pancreatic pseudocysts.

## Case report

A 72-year-old woman with a preliminary diagnosis of pancreatic cancer was referred for further diagnostic workup and assessment regarding surgery. There was a history of increasing abdominal pain in the upper left quadrant; the medical history included an umbilical hernia, hypertension, previous cholecystectomy, appendectomy, and hysterectomy as well as a

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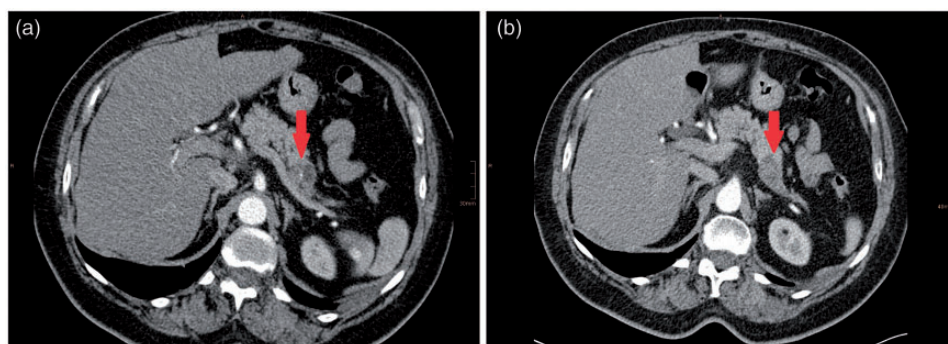


hernia repair. There was no known history of pancreatitis or endometriosis.

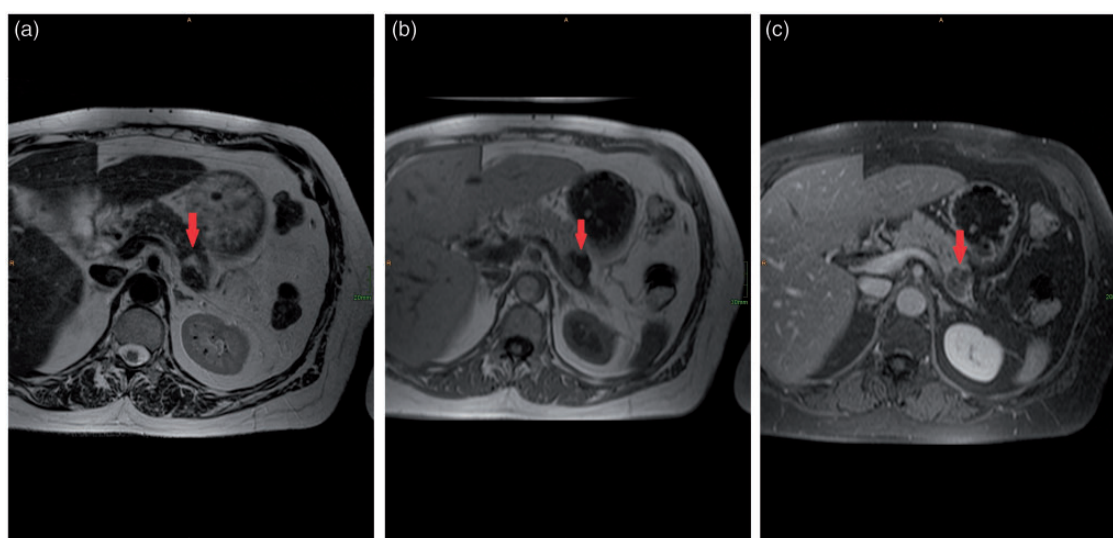
On physical examination the abdomen appeared distended and tender, most pronounced in the left upper quadrant. Blood tests showed an elevated level of gamma glutamyltransferase (GGT) ( $2.21 \mu\text{mol}/(\text{s} \cdot \text{L})$ , normal range  $<0.70$ ), alanine transaminase (ALAT) ( $0.69 \mu\text{mol}/(\text{s} \cdot \text{L})$ , normal range  $<0.60$ ), and C-reactive protein (CRP) ( $5.6 \text{ mg/L}$ , normal range  $<5$ ). Tumor markers<sup>®</sup> (carcinoembryonic antigen [CEA], cancer antigen 19-9 [CA 19-9], cancer antigen 125 [CA 125], neuron-specific enolase [NSE], and chromogranin A) were all within the normal range.

Cross-sectional imaging of the abdomen revealed a pancreatic mass which was considered suspicious for malignancy. A computed tomography (CT) examination (Siemens Sensation 16, Siemens Healthcare

GmbH, Erlangen, Germany; 80 mL Solustrast 300<sup>®</sup> i.v., Bracco Imaging SpA, Milan, Italy; delay 35 s) showed a partly cystic, partly solid lesion ( $3 \times 2.4 \text{ cm}$ ) in the pancreatic tail with a small calcification; there was no evidence of tumor invasion into adjacent structures or metastases (Fig. 1a and b). Magnetic resonance imaging (MRI) (Siemens Sonata, 1.5 T, T2 TSE tra, T1 fl2D in/opposed phase tra, T2 TSE 3D MRCP, T1 fl2D dynamic, and T1 fl3D, Testbolus, T1fl3D after injection of 48 mL Magnevist<sup>®</sup> i.v., Bayer Healthcare, Leverkusen, Germany) showed a partially cystic, partially hemorrhagic lesion with subtle rim enhancement and hypotrophy of the pancreatic tail distally to the lesion. The main pancreatic duct was not dilated and no communication between the lesion and the pancreatic duct system was visible (Fig. 2). Considering patient age



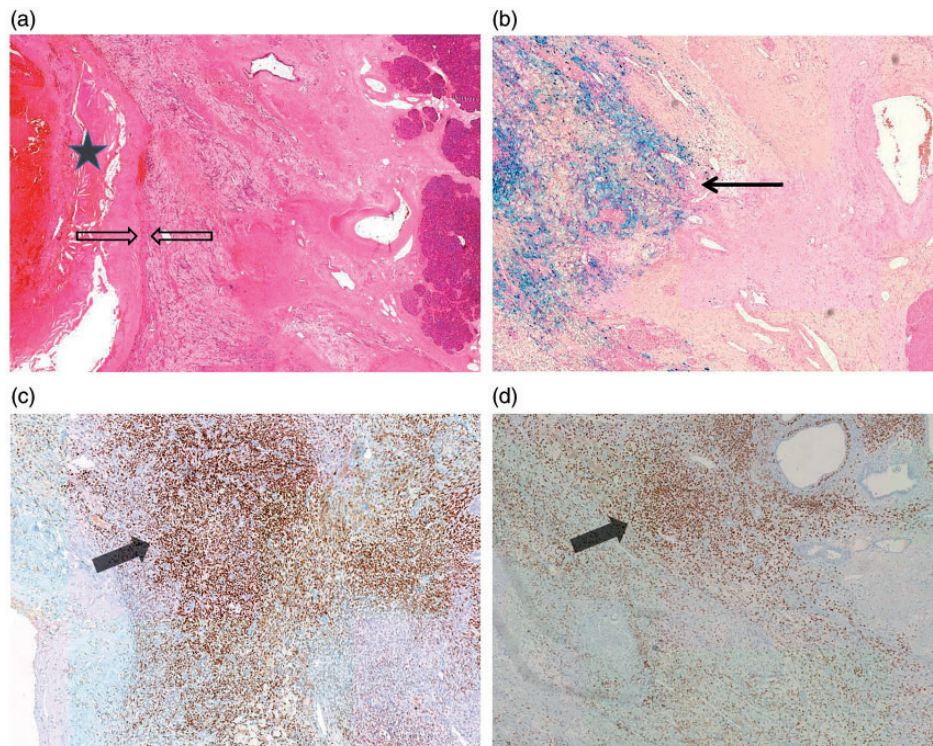
**Fig. 1.** CT of the upper abdomen (arterial phase) showing (a) a partly cystic lesion with a small calcification and (b) a more solid part, in the pancreatic tail.



**Fig. 2.** MRI of the upper abdomen. (a) T2W sequence, (b) TIW sequence, and (c) TIW sequence post contrast showing a cystic lesion with some hemorrhagic content, subtle rim enhancement and hypotrophy of the pancreatic tail distally to the lesion. The main pancreatic duct was not dilated and no communication between the lesion and the pancreatic duct system was visible.

and gender, mucinous neoplasm, serous cystic neoplasm, and pseudocyst as well as intraductal papillary mucinous neoplasm (IPMN) were possible differential diagnoses. Due to the hemorrhagic content and absence of a polycystic structure or honeycomb appearance, a serous cystic neoplasm was deemed unlikely. Since there was no communication with the pancreatic duct system, an IPMN was also excluded. A pseudocyst was considered, but the patient had no history of pancreatitis. Therefore, a preliminary diagnosis of mucinous neoplasm was made. A left hemipancrcreatectomy was performed; since the splenic vein was partly adherent to the pancreatic tail, a partial resection and ligation was performed, with venous drainage of the spleen sufficient via gastric veins so that no reconstruction was necessary. The intraoperative frozen section histology suggested a completely resected benign lesion without further differentiation possible on the frozen sections. Postoperatively the patient suffered from a short episode of respiratory problems, which settled under continuous positive airway pressure (CPAP) within 1 day, and was discharged 17 days post operation. At follow-up 5 years after surgery, the patient is asymptomatic,

showing a small cystic lesion at the resection margin with no evidence of hemorrhage, which is considered to be most likely a small postoperative residual pseudocyst. The definitive histopathology report read focal multicystic lesion with several endometriotic cysts, partially with regressive changes (Fig. 3a–d). Microscopically, there were multiple cysts lined with flat to cubic epithelium without dysplasia, partly with regressive changes, surrounded by ovarian-type subepithelial stroma. The diameter of this multicystic lesion was 2.2 cm. In the surrounding stroma extensive recent and older hemorrhage with fibrosis and histiocytic-resorptive changes with focal dystrophic calcification were demonstrated. These histological features suggest two differential diagnoses: mucinous cystic neoplasia of the pancreas or endometriosis of the pancreas. While regressive changes and ovarian-type stroma are seen in both differential diagnoses, significant stromal hemorrhage and bleeding residues are extremely uncommon in mucinous cystic neoplasia. Smooth muscle cells, which are part of normal endometrial stroma, were also visible. Therefore, the histological diagnosis of a rare case of endometriosis externa involving the pancreas was made.



**Fig. 3.** (a) Photomicrography of the cyst wall ( $\longleftrightarrow$ ) shows typical endometrial tissue with both glandular and stromal components and blood in the lumen of the cyst ( $\star$ ), on the left with normal exocrine pancreatic parenchyma (hematoxylin and eosin stained; original magnification,  $\times 1$ ). (b) Prussian blue reaction with residuals of previous bleeding ( $\longleftarrow$ ;  $\times 2$ ). (c) and (d) positive nuclear immunostaining with estrogen (c) and progesterone receptor (d) specific antibodies in the endometrial stroma and the epithelium ( $\blacktriangleright$ ); (DAB chromogen and hematoxylin counterstain; original magnification,  $\times 4$ ).



## Discussion

Endometriosis is a common condition, manifesting mostly in the pelvis with extragenital manifestations in the pancreas being rare. Firstly described 300 years ago, endometriosis is defined as the presence of endometrial glands and stroma in extrauterine tissue. Symptoms include dysmenorrhea, dyspareunia, and infertility. The histopathogenesis of endometriosis is not fully understood and several theories have been described in the literature, with the two main groups either presuming an endometrial origin of the implants or an origin from other tissues. Endocrine disrupting chemicals as well as coelomic metaplasia, embryonic Mullerian rests, and extrauterine stem/progenitor cells originating from bone marrow have been investigated as possible causes for endometriosis. Furthermore, there are theories of benign metastases or retrograde menstruation. Common factors seem to be a genetic predisposition, estrogen dependence, progesterone resistance, and inflammation (16). Most cases of extragenital endometriosis seem to occur in premenopausal patients, but cases affecting postmenopausal women have been reported (5–15).

Typical imaging features of pelvic endometriosis with hemorrhage are hyperintense foci on T1-weighted (T1W) images with or without fat saturation, or in the absence of bleeding hypointense on T1W and T2-weighted (T2W) images. Since there are only few reports regarding endometriotic cysts in the liver or pancreas, no typical imaging features have been established. A common feature is a cystic lesion, sometimes with hemorrhagic content and in few cases also with a solid component. In our case, the CT scan showed a cystic lesion with a seemingly solid portion and a small calcification. The MRI scan demonstrated a partly serous, partly hemorrhagic lesion in the pancreatic tail with only minimal contrast enhancement of the cyst wall. The hemorrhagic component might have raised the suspicion of endometriosis, but this finding remains unspecific. Since these imaging features are non-specific a definite diagnosis is difficult to establish and a comprehensive workup regarding possible malignant features, tumor markers, fine needle aspiration/biopsy, a previous history of endometriosis, as well as pancreatitis and a possible menstrual cycle dependency of symptoms or changing imaging features over time can be helpful in establishing the diagnosis.

In conclusion, pancreatic endometriosis is a very rare condition, which can affect pre- as well as postmenopausal women. A possible diagnosis of extragenital endometriotic cysts should be considered in women presenting with cystic, hemorrhagic lesions, young

women with fertility issues, changing imaging features, or cyclic symptoms and a previous history of endometriosis. Since imaging features are non-specific the diagnosis remains challenging and in most cases surgery is performed. Thorough diagnostic workup and intraoperative frozen section histology may help avoiding extensive surgery and its associated morbidity and mortality.

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