

CASE REPORT

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A pancreatic mucinous cystadenoma in a man with mesenchymal stroma, expressing oestrogen and progesterone receptors

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Abstract A 43-year-old man presented with abdominal discomfort caused by relapsing pancreatitis. Radiological examination revealed a multilocular cystic mass in the tail of the pancreas, which was resected. Gross examination showed a multilocular cystic lesion measuring 2.5 cm in diameter and containing clear fluid. Microscopically, a mucinous cystadenoma with mesenchymal stroma was diagnosed. The lesion showed two different components: a cyst lined by a columnar, mucin-secreting epithelium and a moderate cellular stroma composed of spindle cells. The stromal element appeared similar to primitive mesenchyme. Immunohistochemical staining confirmed this origin through vimentin expression and showed moderate to strong nuclear staining with oestrogen and progesterone receptor antibodies. Cystadenomas are rare tumours of the pancreas, but mesenchymal stroma is uncommon in such tumours; it is more frequently described in the liver and the bile ducts, and primarily in women.

Key words Pancreas · Mucinous cystadenoma · Mesenchymal stroma · Oestrogen receptor antibody expression · Progesterone receptor antibody expression

Introduction

Cystic lesions of the pancreas are not common [3, 8, 10, 12], inflammatory pseudocyst being the most common.

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True cysts can be divided into retention cysts, congenital cysts and proliferative cysts, the last comprising cystadenomas and cystadenocarcinomas [16]. Cystadenomas account for 10% of all benign cystic lesions of the pancreas and are divided into serous and mucinous types [4, 6]. Mucinous cystadenomas of the pancreas occur mostly in middle-aged women. They are usually asymptomatic or give rise to non-specific symptoms [3, 7, 9, 12]. Few cases of pancreatic cystadenomas with hypercellular, mesenchymal underlying stroma have been reported in the literature [3, 7, 12, 16–19], and they are more commonly encountered in the liver and the bile ducts [20]. These lesions occur almost exclusively in women [1, 5, 11, 12, 15, 17]. Oestrogen receptor expression has been found in some cases [11, 13, 17, 19].

The presence of oestrogen and progesterone receptors in cystadenomas with mesenchymal stroma has been studied only to a limited extent [17, 19]. In the following case report we present a pancreatic mucinous cystadenoma with mesenchymal stroma, expressing oestrogen and progesterone receptors, in a male patient.

Case report

Clinical history

A 43-year old man was admitted in February 1996 with recurrent episodes of upper abdominal pain, vomiting, weight loss and steatorrhoea.

Alcohol intake had been moderate to high for over 10 years. Transient episodes of epigastric pain had been occurring since 1990, and a diagnosis of acute pancreatitis with pseudocyst formation was made for the first time in June 1995. On admission, clinical examination revealed no major abnormalities except for weight loss.

Laboratory investigations showed an increase of both amylase, up to 288 U/l (normal < 220 U/l), and lipase, up to 990 U/l (normal < 190 U/l), while tumour markers (CEA, CA19.9) were negative. On ultrasonography and computed tomography, the cystic lesion in the tail of the pancreas had increased from 3.2 cm to 4.0 cm in diameter and contained multiple septations and solid areas. Magnetic resonance imaging confirmed the presence of a septated cystic mass giving rise to some irregular dilatation of the proximal part of the main pancreatic duct. These findings were interpreted as being consistent with a mucinous cystadenoma or cystadenocarcinoma. The tail of the pancreas was therefore resected with preservation of the spleen.

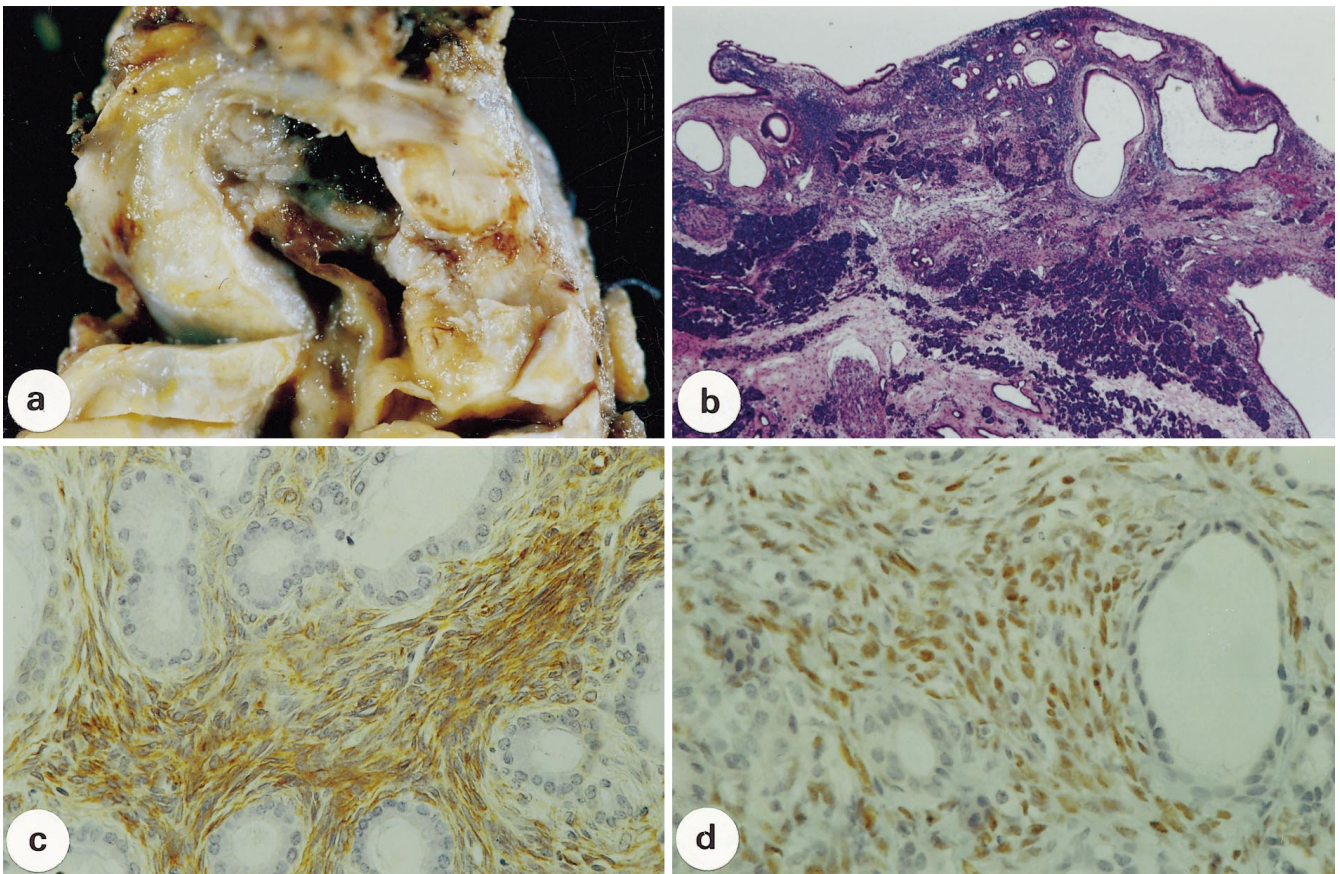
Materials and methods

The surgical specimen was fixed with 10% formaldehyde and embedded in paraffin for haematoxylin and eosin and immunostaining. Immunohistochemical staining was performed on semiserial sections, using an indirect immunoperoxidase technique. The following antibodies were used: monoclonal antibodies directed against oestrogen receptors (monoclonal; dilution 1/30, Dakopatts, Glostrup, Denmark), monoclonal antibodies directed against progesterone receptors (monoclonal; dilution 1/40, Novocastra, Laboratories Newcastle-upon-Tyne, England), vimentin (polyclonal; dilution 1/30, Dakopatts), alpha smooth muscle actin (monoclonal; dilution 1/40, Dakopatts) and desmin (polyclonal; dilution 1/50, Dakopatts).

Pathological findings

Gross pathology showed a multilocular cystic lesion with a diameter of 2.5 cm. The cyst was filled with clear fluid and well surrounded by a fibrous capsule, which varied in thickness but did not exceed 1 cm. Focally solid areas were encountered in the capsule. The lesion was surrounded by macroscopically normal pancreatic tissue (Fig. 1a).

Fig. 1 **a** Macroscopical examination showed that the multicystic lesion was surrounded by a rather thick capsule, **b** The two components (cysts and the stromal component) are closely intermingled. The lower part of this low-power photograph shows adjacent slightly atrophic pancreatic parenchyma. H & E, $\times 4$. **c** Antibodies directed against vimentin show diffuse cytoplasmic positivity of the stromal component. Vimentin, $\times 25$. **d** The positivity with antibodies directed against oestrogen receptor is extensive and almost exclusively nuclear. Oestrogen receptor, $\times 25$



Routine microscopic examination demonstrated a multilocular cyst with a dense, hypercellular underlying stroma. The cyst was lined by a single layer of columnar, mucin-secreting epithelial cells (Fig. 1b). The epithelium showed no cytological atypia and did not infiltrate into the underlying stroma. This stromal component was composed of oval to spindle-shaped cells, with round to elongated nuclei and a small amount of cytoplasm. These cells showed no atypia or mitoses. The hypercellular stroma appeared similar to primitive mesenchyme. The pancreas, adjacent to the cyst, showed a mild degree of atrophy.

Immunohistochemistry showed strong positivity of the stromal component for vimentin, for oestrogen receptors (Fig. 1c, d) and for progesterone receptors. In contrast, stainings with antibodies directed against desmin and alpha smooth muscle actin were negative.

Discussion

In our case there are two essential histological features: a cyst lined by a columnar to cuboidal mucin-secreting epithelium and a moderately to densely cellular stroma, resembling primitive mesenchyme and composed of spindle-shaped cells. The occurrence of this hypercellular stroma in pancreatic cystadenomas is not very common. It has been proposed that this stromal component originates in primitive mesenchymal cells [19]. We have shown immunoreactivity of this mesenchymal stroma for vimentin.

Cystadenomas with mesenchymal stroma are more frequently encountered in the liver and the bile ducts. These lesions mostly affect women [1, 5, 13] and oestrogen receptor expression of the stroma is found in some of them [11, 14]. In our case, immunoreactivity of the mesenchymal stroma for oestrogen and progesterone was found.

The histogenesis of cystadenomas with mesenchymal stroma is uncertain, and there is still no explanation for the female predominance. Since oestrogen receptor expression is found in some of the hepatobiliary and pancreatic tumours, a role for hormonal factors in the pathogenesis has been suggested [14]. The only 'hormonal factor' that could be traced in our otherwise healthy male patient was a somewhat excessive alcohol consumption, which may increase oestrogen production in peripheral fat. The patient did not show any external signs of feminization.

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