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Epidermoid Cyst in an Intrapancreatic Accessory Spleen: Three Case Reports and Review of the Literatures

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Abstract The development of an epidermoid cyst in an intrapancreatic accessory spleen is an extremely rare lesion, with only 17 cases being reported in the English literature. All such cases were located in the pancreatic tail, some of which showed carbohydrate antigen 19-9 (CA19-9) immunoreactivity in the lining of the epithelium. A few of them indicated an elevation of the serum CA19-9 level. Here we report three cases of an epidermoid cyst in an intrapancreatic accessory spleen. Cases 1 and 2 were 57-year-old and 70-year-old women, while case 3 was a 37-year-old man. All three cases were asymptomatic. Serum CA19-9 levels showed within normal limits (case 1), slightly elevated (case 2), and clearly elevated (case 3). They underwent a distal pancreatectomy with splenectomy (cases 1 and 2) and without splenectomy (case 3). Grossly, the surgical specimen was a well-demarcated, multiple (case 1) or solitary (cases 2 and 3) cystic mass in the pancreatic tail. A high level of fluid CA 19-9 was detected in case 1. Microscopically, the cystic walls were lined with squamous and cuboidal epithelium, which were surrounded by normal splenic tissue and hyalinized fibrous tissue. The lining squamous epithelium was revealed as nonkeratinizing (Cases 1and 2) or keratinizing (Case 3). Immunohistochemically, CA19-9 was positive in the monolayer and surface layer of the cuboidal epithelium, but negative for the keratinizing squamous epithelium. As for the histogenesis, it is suggested that the cystic lining of the epithelium may derive from the pancreatic duct which protrudes into the accessory spleen.

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Introduction

Although the occurrence of an accessory spleen is not uncommon, the development of an epidermoid cyst in an intrapancreatic accessory spleen is extremely rare. Since Davidson et al. [1] reported the first case of an epidermoid cyst originating from an intrapancreatic accessory spleen in 1980, there have been 16 additional cases reported in the English literature [2–17]. All of them have been the subject of only sporadic case reports. Therefore, this is the first report discussing more than one confirmed case of an epidermoid cyst in an intrapancreatic accessory spleen.

Such cysts in the present study were located in the pancreatic tail [1–17]. Each cyst was lined with epithelium surrounded by normal splenic tissue in the pancreas. Some cases showed carbohydrate antigen 19-9 (CA19-9) immunoreactivity in the epithelial lining. Furthermore, a few cases displayed an elevation of the serum CA19-9 level [6, 11–13]. Because of the difficulty in differentiating this lesion from a cystic neoplasm of the pancreas by an imaging study, none of these cases was correctly diagnosed before surgery.

In the present paper, we report three cases of an epidermoid cyst in the intrapancreatic accessory spleen. The clinicopathological and immunohistochemical characteristics are discussed and compared with cases previously reported in the literature, and this is the first report to describe the D2-40 positivity in the epidermoid cyst arising from an intrapancreatic accessory spleen. Furthermore, we investigate their histogenesis base on their histological and immunohistochemical appearance.

Clinical History

Case 1

A 57-year-old Japanese woman with no symptoms was admitted to the hospital for a follow-up of hypertension, and a cystic lesion of the pancreatic tail was incidentally detected during abdominal ultrasonography. No history of trauma or pancreatitis was recorded. Physical examination and blood chemistry findings upon admission showed no abnormalities. Tumour markers such as carcinoembryonic antigen (CEA) and CA19-9 were within normal limits. Enhanced computed tomography (CT) confirmed a cystic mass lesion measuring 6 cm in diameter in the tail of the pancreas, and the cystic wall showed a partial enhancement. Following a clinical diagnosis of a pancreatic cystic tumour, a distal pancreatectomy with splenectomy was carried out.

Case 2

Upon an image examination, a 70-year-old Japanese woman with no symptoms but with a history of lung and breast cancer was found to have a cystic lesion of the pancreatic tail. Among her tumour markers, CA19-9 (48 U/ml) was slightly increased, whereas CEA was within normal limits. CT confirmed a cystic mass lesion measuring 2.6 cm in the tail of the pancreas. A distal pancreatectomy with splenectomy was carried out after a clinical diagnosis of a mucinous cystic tumour in the pancreas.

Case 3

A 37-year-old Japanese man with no symptoms was incidentally detected to have a cystic lesion of the pancreatic tail during a medical check-up. Among his tumour markers, CA19-9 (647 U/ml) showed an elevation, whereas CEA was within normal limits. Enhanced CT confirmed a cystic mass lesion measuring 10 cm in diameter in the tail of pancreas, and the cystic wall showed a partial enhancement (Fig. 1). A distal pancreatectomy was carried out upon a clinical diagnosis of a pancreatic cystic lesion, such as a serous cystic tumour or lymphoepithelial cyst.

Materials and Methods

The specimens were fixed in 20% buffered formalin, embedded in paraffin, and stained with hematoxylin and eosin (H&E) as well as periodic acid Schiff (PAS), periodic acid Schiff with diastase pretreatment (d-PAS), and alcian blue stain according to the standard procedure.

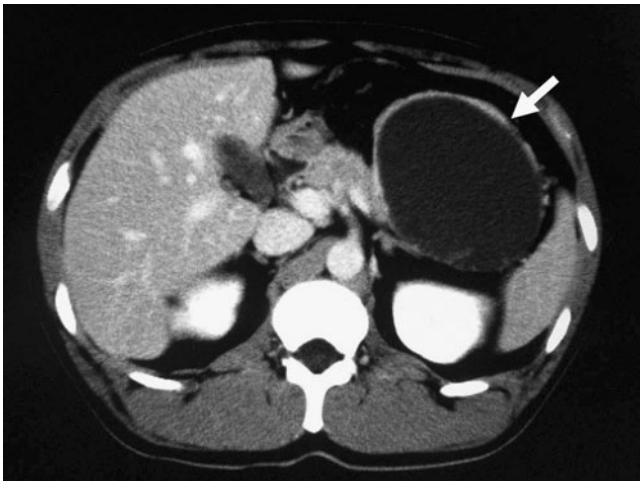

Fig. 1 Enhanced CT in case 3 shows a cystic mass lesion (arrow) measuring 10 cm in the tail of the pancreas together with a partial enhancement of the cystic wall

Immunohistochemical studies were performed on tissue sections using the labeled streptavidin biotinylated antibody (LSAB) method with an autostaining system (Ventana Benchmark System, Tucson, AZ, USA) according to the manufacturer's protocol. The following antibodies were employed: cytokeratin (CK) 7 (Novocastra, clone OV-TL 12/30, 1:50), CK 13 (Novocastra, clone KS-1A3, 1:100), CK18 (Novocastra, DC-10, 1:40), CK20 (Novocastra, clone Ks20.8, 1:50), epithelial membrane antigen (EMA) (Dako, clone E29, 1:50), CA19-9 (Dako, clone 116-NS-19-9, 1:200), CEA (Dako, clone ll-7, 1:40), calretinin (Novocastra, clone 5A5, 1:100), and D2-40 (Nichirei, prediluted). Table 1 summarized the immunohistochemical results in three present cases of epidermoid cyst in an intrapancreatic accessory spleen.

Results

Case 1

Grossly, the surgical specimen was a well-demarcated, multicystic mass measuring 6.0×5.0×4.0 cm in the pancreatic tail (Fig. 2a). The cystic wall was grayish-white with a brown component that resembled normal spleen, and there was an accessory spleen in the splenic hilum. The cysts contained colorless serous and partially mucinous fluid. A high level of fluid CA 19-9 (1,880 U/ml) was detected.

Microscopically, the cysts were surrounded by normal splenic tissue and hyalinized fibrous tissue (Fig. 3a) consisting of red and white pulps (Fig. 3b), and the cystic walls were lined with nonkeratinizing stratified squamous epithelium and focally cuboidal epithelium. The cuboidal epithelium presented as monolayer (Fig. 3c) or several

Table 1 List of the immunohistochemical results in three present cases of epidermoid cyst in an intrapancreatic accessory spleen

Antigen	Antibody clone	Source	Dilution	Results					
				Case 1		Case 2		Case 3	
				SE	CE	SE	CE	SE	CE
CK 7	OV-TL 12/30	Novocastra	1:50	−	+	−	+	−	+
CK 13	KS-1A3	Novocastra	1:100	+	+	+	+	+	+
CK 18	DC-10	Novocastra	1:40	−	+	−	+	−	+
CK 20	Ks20.8	Novocastra	1:50	−	−	−	−	−	−
EMA	E29	Dako	1:50	−	+	−	+	−	+
CA19-9	116-NS-19-9	Dako	1:200	−	+	−	+	−	+
CEA	ll-7	Dako	1:40	−	−	−	−	−	−
Calretinin	5A5	Novocastra	1:100	−	−	−	−	−	−
D2-40	D2-40	Nichirei	Prediluted	+	+	+	−	+	−

CK cytokeratin; *EMA* epithelial membrane antigen; *CA19-9* carbohydrate antigen 19-9; *CEA* carcinoembryonic antigen; *SE* squamous epithelium; *CE* cuboidal epithelium; −, negative; +, positive

layers, and showed transitional findings to the squamous epithelium. The ratio of cuboidal epithelium was about 30% of the total lining epithelium. Epithelial component demonstrated no atypia. The splenic capsule was well-defined and attached to the pancreatic parenchyma through a thin fibrous tissue including the pancreatic duct. PAS, d-PAS and alcian blue stain revealed cytoplasmic positivity in a small number of cubical cells.

Immunohistochemically, the lining squamous and cuboidal epithelia were positive for CK 13, while the lining cuboidal epithelium was positive for CK 7, CK18, and EMA. CA19-9 was positive for both the monolayer (Fig. 4a) and surface layer of the cuboidal epithelium. CK20, CEA and calretinin were negative, while D2-40 was partially positive only in the basal layer of the lining epithelium (Fig. 4d). Regarding the immunohistochemical pattern, the lining cubical epithelium was similar to that of the pancreatic duct.

Case 2

Grossly, the surgical specimen showed a well-demarcated, solitary cystic mass measuring 1.7×1.0×0.8 cm in the pancreatic tail. The cysts contained colorless serous fluid and the cystic wall was grayish-white. The brown component that resembled normal spleen was not clear in the cystic wall, whereas there was an accessory spleen in the splenic hilum.

Microscopically, the cyst was partially lined with non-keratinizing stratified squamous epithelium (Fig. 3e), and focally several layers of the cubical epithelium (Fig. 3d), whereas most of the lining epithelium was denuded. Both epithelial components indicated a transitional appearance to one another. The ratio of cuboidal epithelium was about 40% of the total lining epithelium. There were no malignant findings in the lining epithelium. Most of these lining epithelia were surrounded by hyalinized fibrous tissue, whereas normal splenic tissue was seen in a very few areas that were attached to the pancreatic parenchyma. PAS, d-PAS and alcian blue stain showed no cytoplasmic positivity in the lining epithelium.

Immunohistochemically, the linings of both epithelia were positive for CK 13, the lining cuboidal epithelium was positive for CK 7, CK18, and EMA. CA19-9 was positive for the surface layer of the cuboidal epithelium (Fig. 4b),

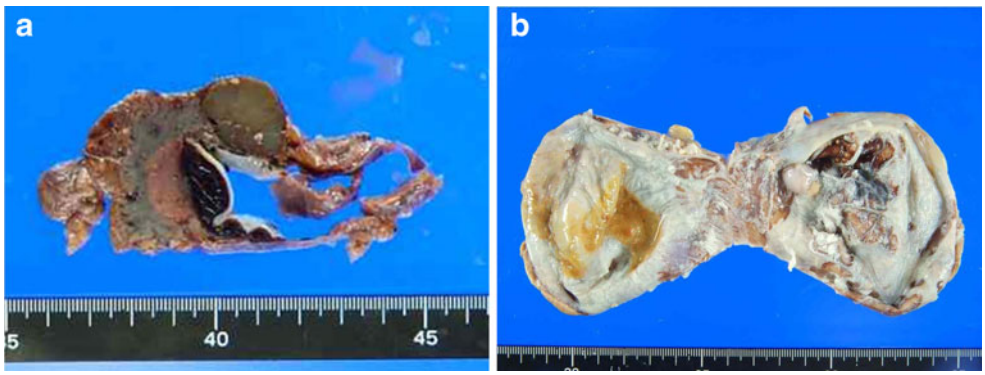

Fig. 2 Gross appearance. **a** Case 1 shows a multicystic mass measuring 6.0×5.0×4.0 cm in the pancreatic tail and a cystic wall grayish-white in color having a brown component that resembles a normal spleen. **b** Case 3 shows a solitary cystic mass measuring 10.0×7.0×7.0 cm in the pancreatic tail and cysts containing gelatinous tan material admixed with a whitish keratinous substance

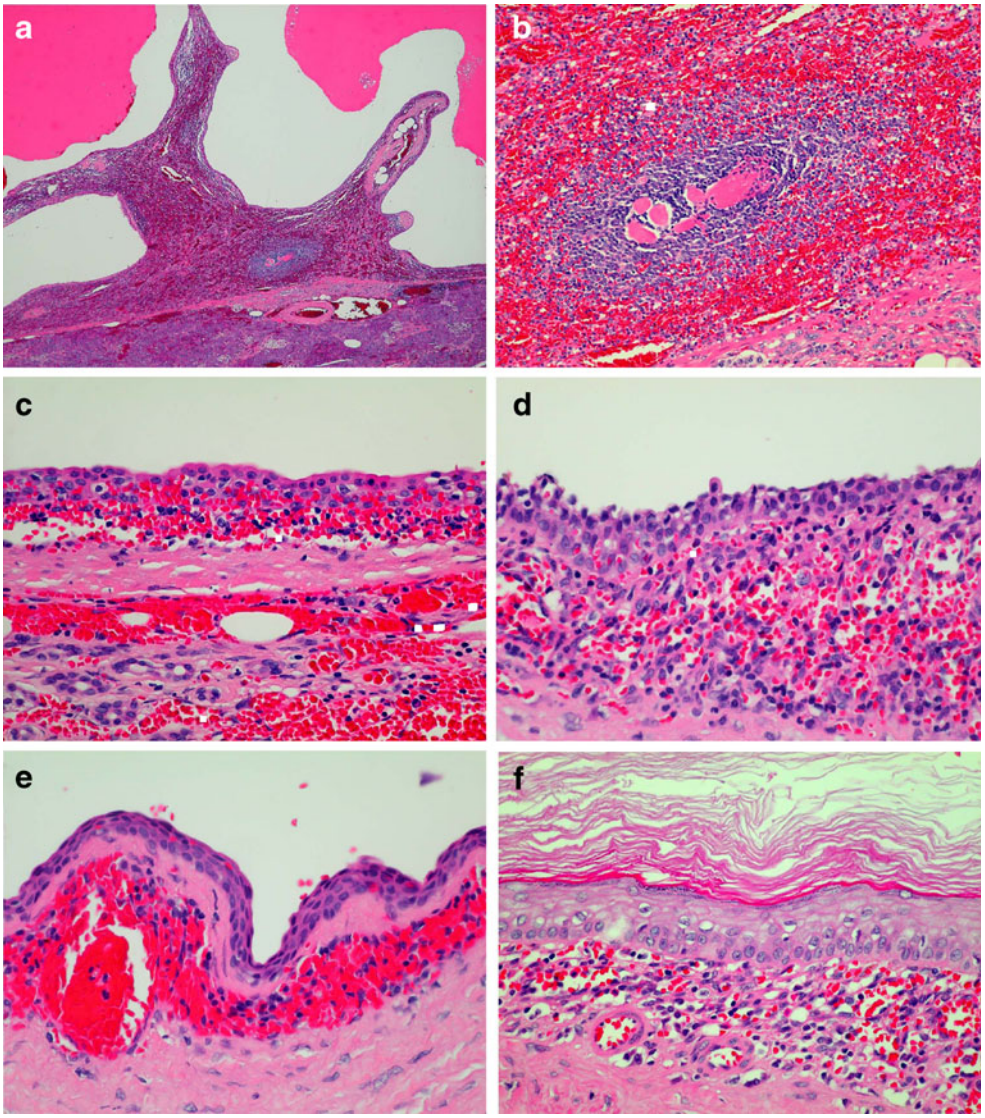

Fig. 3 Microscopic appearance. **a** The cysts are surrounded by normal splenic tissue and hyalinized fibrous tissue (×40). **b** The accessory spleen in the pancreas consists of both red and white pulp (×200). **c** The lining epithelium shows a monolayer of cubical epithelium (×400). **d** The lining epithelium shows several layers of cubical epithelium (×400). **e** The lining epithelium shows a nonkeratinizing stratified squamous epithelium (×400). **f** The lining epithelium shows a keratinizing stratified squamous epithelium (×400)

CK20, CEA and calretinin were negative, and D2-40 was positive only in the basal layer of the stratified squamous epithelium.

Case 3

Grossly, the surgical specimen was seen as a solitary cystic mass measuring 10.0×7.0×7.0 cm in the pancreatic tail (Fig. 2b). The cystic wall showed grayish-white in color partially mixed with a brown component. The cysts contained a gelatinous tan material admixed with a whitish keratinous substance.

Microscopically, the cysts were lined with the keratinizing stratified squamous epithelium (Fig. 3f) and focally several layers of the cubical epithelium. The cuboidal epithelium revealed transitional findings to the stratified squamous epithelium. The ratio of the cuboidal epithelium was about 5%. The lining epithelium was denuded over a small area, and a foreign body granuloma was seen. No malignancy was found in the lining epithelia. These were surrounded by hyalinized fibrous tissue and focally normal splenic tissue. The splenic capsule was well-defined, and attached to the pancreatic parenchyma through the thin fibrous tissue, including the pancreatic duct. Under PAS, d-PAS and alcian blue stain, no cytoplasmic mucin was seen in the lining epithelium.

Immunohistochemically, the lining squamous and cuboidal epithelia were positive for CK 13, and the lining cuboidal epithelium was positive for CK 7, CK18, and EMA. CA19-9 was positive for the surface layer of the cuboidal epithelium. In contrast, CA19-9 was negative for the keratinizing squamous epithelium (Fig. 4c). CK20, CEA and calretinin were negative, and D2-40 was positive only in the basal layer of the lining squamous epithelium.

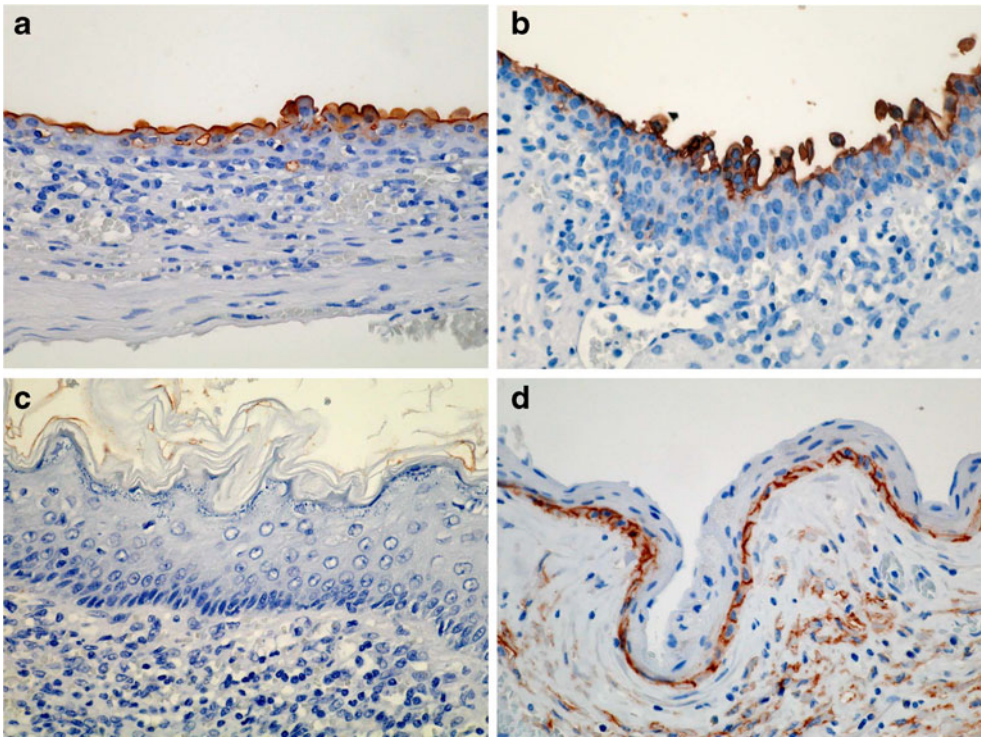

Fig. 4 Immunohistochemical findings (×400). **a** CA19-9 was positive for the monolayer in the cuboidal epithelium. **b** CA19-9 was positive for the surface layer in the cuboidal epithelium. **c** CA19-9 was negative for keratinizing squamous epithelium. **d** D2-40 was positive for the basal layer in the lining epithelium

Discussion

Epidermoid cysts of the spleen are rare entities comprising less than 10% of true nonparasitic splenic cysts [18]. Approximately 20% of accessory spleens occur in or around the tail of the pancreas [19]. An epidermoid cyst in an intrapancreatic accessory spleen is extremely rare, which only 17 cases reported in the English literature. To the best of our knowledge, none of the lesions were found in the extrapancreatic accessory spleen.

Table 2 summarized the 20 cases of epidermoid cyst in an intrapancreatic accessory spleen, including the present cases. They were comprised of 8 men and 12 women, with ages ranging from 32 to 70 years (mean, 47 years). Twelve cases were asymptomatic, whereas the others showed symptoms such as abdominal pain, nausea, or weight loss. All of the lesions were located in the tail of the pancreas, and their mean diameter was 4.8 cm, ranging from 1.8 to 15.0 cm. Both unilocular and multilocular cases have been reported, with cysts containing fluid or keratinaceous material. Similar to an epidermoid cyst of the spleen [20], the lining epithelium of the epidermoid cyst in an intrapancreatic accessory spleen consisted of stratified squamous or cuboidal epithelium, with some cases exhibiting keratinization or mucinous cells.

Elit et al. [21] reported a rare case of squamous cell carcinoma in an epidermoid cyst of the spleen. However, to the best of our knowledge, malignant changes have not been previously reported in an epidermoid cyst arising from an intrapancreatic accessory spleen. Moreover, the present three cases showed no malignant findings such as those in squamous cell carcinoma.

An elevation of the serum CA 19-9 level was observed in 6 cases [6, 11–13]. This clinical finding makes it difficult to preoperatively differentiate between an epidermoid cyst in an intrapancreatic accessory spleen and pancreatic cancer. Higaki et al. [6] reported that the serum CA19-9 levels markedly decreased to normal ranges after surgery in patient diagnosed with an epidermoid cyst in an intrapancreatic accessory spleen, a result suggesting that the serum CA19-9 originated in the epidermoid cyst in an intrapancreatic accessory spleen. Three cases, including the present case 1, revealed a CA 19-9 elevation in the contents of the cyst [7, 16]. Moreover, twelve cases, including the present three, showed CA19-9 immunoreactivity in the cystic lining epithelium. Serum CA 19-9 level elevations may be associated with the total amount of CA 19-9 production in the cystic lining epithelium. However, in the present three cases, the percentage of CA19-9 immunoreactivity in the cystic lining epithelium revealed no obvious association with the serum CA 19-9 level. In the present case 1, CA 19-9 in the contents of the cyst showed an elevation without a serum CA19-9 elevation. This result might suggest that CA19-9 showed exsorption into the cyst, not infusion into the vessel.

In all three present cases, D2-40 was positive in the basal layer of the cystic lining epithelium, a finding not previously reported. Beside the lymphatic endothelium, D2-40 immunoreactivity has appeared in other normal tissues, including mesothelial cells, osteocytes, glandular

Table 2 Summary of case reports on epidermoid cyst in an intrapancreatic accessory spleen

Number	Author	Year	Age (years)	Sex	Site	Size (cm)	Symptoms	Serum CA19-9 (IU/ml)	IHC for CA19-9
1	Davidson et al [1]	1980	40	Male	Tail	7.0×3.0×2.5	Weight loss, nausea	No information	No information
2	Morohoshi et al [2]	1991	32	Female	Tail	6.0×5.0	Left abdominal pain	Within normal limit	Positive
3	Nakae et al [3]	1991	37	Female	Tail	6.5×5.5×4.6	Epigastric pain	No information	No information
4	Tang et al [4]	1994	38	Male	Tail	2.3×2.1	None	Within normal limit	No information
5	Furukawa et al [5]	1998	45	Male	Tail	2.0	None	No information	No information
6	Higaki et al [6]	1998	46	Female	Tail	3.0×3.0	Left back pain	201	Positive
7	Tatayama et al [7]	1998	67	Female	Tail	3.0	Abdominal pain	Within normal limit	Positive
8	Sasou et al [8]	1999	49	Female	Tail	4.3×2.6	None	Within normal limit	Positive
9	Choi et al [9]	2000	54	Female	Tail	15×11	Epigastric comfort, nausea, vomiting, weight loss	No information	No information
10	Tsutsumi et al [10]	2000	51	Male	Tail	2.5×2.5×2.0	None	Within normal limit	No information
11	Horibe et al [11]	2001	48	Male	Tail	2.0×1.0	None	53	Positive
12	Sonomura et al [12]	2002	45	Female	Tail	3.5	Epigastric distress	159	Positive
13	Kanazawa et al [13]	2004	58	Female	Tail	2.5	None	62	Positive
14	Ru et al [14]	2007	41	Male	Tail	2.5	None	No information	Positive
15	Itano et al [15]	2008	40	Male	Tail	4.0×3.2×3.0	None	Within normal limit	No information
16	Servais et al [16]	2008	52	Female	Tail	11.5×10.5×8.5	None	No information	Positive
17	Gleeson et al [17]	2008	32	Female	Tail	1.5×1.2	Right upper abdominal pain	No information	No information
18	Present case 1		57	Female	Tail	6.0×5.0×4.0	None	Within normal limit	Positive
19	Present case 2		70	Female	Tail	1.8×1.0×0.8	None	48	Positive
20	Present case 3		37	Male	Tail	10.0×7.0×7.0	None	647	Positive

IHC immunohistochemistry

myoepithelial cells, and ependymal cells [22]. Furthermore, basal epidermal keratinocytes have been known to be positive for D2-40. Therefore, D2-40 positivity in the basal layer of an epidermoid cyst arising from an intrapancreatic accessory spleen shows one of the characteristics of basal epidermal keratinocytes.

The histogenesis of an epidermoid cyst in an intrapancreatic accessory spleen might be identical to that of a splenic epidermoid cyst. There are three hypotheses concerning the histogenesis of such an epidermoid cyst [7]. The first theory suggests the origination from a mesothelial inclusion with subsequent squamous metaplasia [23]. The second indicates a teratomatous derivation or an inclusion of fetal squamous epithelium [24]. The third suggests a derivation from the pancreatic duct which protrudes into the accessory spleen. In the present three cases, the first theory was excluded because the lining epithelium was negative for calretinin as a mesothelial marker. Concerning the second theory, no other teratomatous component such as endodermal tissue was observed in the present cases. As for the fetal squamous epithelium, there were no skin appendages such as hair follicles, sebaceous glands, or sweat glands in the cyst. On the other hand, in the present cases, there were pancreatic ducts in the fibrous tissue surrounding the accessory spleen tissue, and the squamous and cuboidal epithelia indicated a transitional appearance to one another. Furthermore, the immunohistochemical staining pattern of the cystic lining epithelium was the same as that in the pancreatic ducts. In particular, CA 19-9 was characteristically positive for the cystic lining epithelium and pancreatic ducts. These findings suggest the possibility that the epithelium of the epidermoid cyst in an intrapancreatic accessory spleen might derive from the pancreatic duct showing squamous metaplasia. In addition, the present case 1 had a characteristic feature showing a monolayer of cubical epithelia lining the cystic wall that resembled a pancreatic duct, a feature not previously reported. Therefore, it is suggested that a pancreatic duct in the accessory spleen showed dilation, and changed from a monolayer to several layers of cuboidal epithelium. Furthermore, it may reveal squamous metaplasia, and may finally show keratinization. The fact that the epidermoid cyst in an accessory spleen has been reported only in an intrapancreatic accessory spleen might also support this possibility.

The differential diagnosis of such intrapancreatic cysts includes pancreatic pseudocysts, endometrial cysts, serous cystic tumours, mucinous cystic tumours, intraductal papillary mucinous tumours, and lymphoepithelial cysts. Pseudocysts are the most common cystic lesion of the pancreas. In a pseudocyst, the cystic wall consists of granulation tissue without the lining epithelium. Although chronic inflammation including that of lymphocytes and macrophages may be prominent in a pancreatic pseudocyst, it lacks the characteristic histology of splenic tissue. Endometrial cysts are seen only in women with the cystic wall consisting of endometrial glands and endometrial stroma. Serous cystic tumours of the pancreas affect more women than men, and their cystic content is usually a clear fluid. Although histologically such tumours have microcystic or oligocystic architecture with a single layer of cuboidal epithelium, no lymphoid tissue has been identified in the fibrous stroma. Mucinous cystic tumours of the pancreas occur predominantly in females. They contain mucoid fluid, and the cystic lining is composed of mucous columnar cells admixed with goblet cells. Intraductal papillary mucinous tumours present with symptoms of pancreatic duct obstruction, which result in a characteristic clinical manifestation. Additionally, because of their location in the pancreatic head, papillary lesions and a lack of lymphoid tissue, intraductal papillary mucinous tumours are distinctively diagnosed. Lymphoepithelial cysts have been reported predominantly in males, and they are distributed throughout the pancreas [7]. These cysts contain keratinizing material with or without fluid. The cystic wall is lined by mature keratinized squamous epithelium and underlying lymphoid tissue that may contain small lymphocyte aggregations or even follicles with germinal centers. In the latter situation, although they are especially difficult to differentiate from accessory splenic cysts, the lack of red pulp is a useful marker in the diagnosis of a lymphoepithelial cyst. Although other pancreatic tumours may undergo cystic degeneration or a focal cystic change, their typical morphology usually makes a definitive diagnosis relatively easy. In contrast, an imaging diagnosis of an epidermoid cyst in an intrapancreatic accessory spleen before surgery is very difficult. CT images of the cystic wall are similar to those of the spleen, which suggests that a correct diagnosis may be possible with careful examination of the images prior to surgery, as long as relatively large amounts of splenic tissue are present [15]. However, if the amount of splenic tissue is relatively small, an accurate diagnosis before surgery may not be possible.

In conclusion, an epidermoid cyst in an intrapancreatic accessory spleen is extremely rare, making it difficult to diagnose it preoperatively. However, such a cyst should be taken into account in any differential diagnosis of a pancreatic cystic lesion. Moreover, we should be aware of cases revealing serum CA19-9 elevation. Concerning the histogenesis, it is suggested that a cystic lining epithelium may derive from the pancreatic duct which protrudes into the accessory spleen.

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