

# A Rare Case of Epidermoid Cyst in the Pancreatic Tail Invaginated from the Splenic Hilum: The Long-term Changes in the Imaging Findings

Yoshiaki Sugiyama<sup>1,2</sup>, Toru Kawamoto<sup>1,3</sup>, Junpei Sasajima<sup>1</sup>, Kazuya Koizumi<sup>1,4</sup>,  
Hidenori Karasaki<sup>5</sup> and Yusuke Mizukami<sup>5</sup>

---

## Abstract

---

Epidermoid cysts arising from both the pancreas and spleen are rare. We herein report a case of a surgically resected epidermoid cyst in the pancreatic tail invaginated from the spleen. A multi-locular cyst, 2 cm in diameter, without a solid component was discovered incidentally in the pancreatic tail. During the 11-year follow-up, the emergence of satellite cystic lesions with distinct appearances was seen, and surgical resection was selected despite the lack of any associated symptoms or evidence of cytological abnormalities. Histologically, these cysts were lined with benign multi-layered flattened epithelium surrounded by a thin layer consisting of cells positive for CD8 and CD68 and connecting to the spleen.

**Key words:** pancreatic cyst, splenic epidermoid cyst, long-term follow-up, magnetic resonance imaging

(Intern Med 55: 3591-3594, 2016)

(DOI: 10.2169/internalmedicine.55.7466)

---

## Introduction

---

A non-parasitic splenic cyst is a rare condition with an incidence of 0.07% as reported in a review of large autopsy cases (1). Splenic epidermoid cysts are rare benign primary cysts that account for 10% of non-parasitic splenic cysts (2). Epidermoid cysts occasionally arise from accessory spleens, and those lesions in the pancreas are usually associated with intrapancreatic accessory spleens (3). We herein describe a rare case of an epidermoid cyst in the pancreatic tail that invaginated from the spleen. Surgical resection was performed due to drastic changes in imaging findings after an 11-year follow-up.

---

## Case Report

---

In 1999, a 46-year-old Japanese woman with a history of acute nephritis was referred to our hospital for investigation

of her renal dysfunction. She was diagnosed with chronic glomerulonephritis, and multi-locular cysts in the pancreatic tail were also discovered incidentally. The patient had no history of abdominal pain or a fever, and the findings on a physical examination were unremarkable. The blood tests showed elevated levels of blood urea nitrogen and creatinine, but normal levels of cancer antigen 19-9, carcinoembryonic antigen, and Dupan-2. Magnetic resonance imaging (MRI; Fig. 1) and endoscopic ultrasonography (EUS; Fig. 2) showed a 2-cm cystic lesion in the pancreatic tail. Cyst fluid obtained by EUS-guided fine needle aspiration (EUS-FNA) revealed no malignant cells, and follow-up was recommended for the patient. In 2000, a new cystic lesion located between the initial cysts and the spleen was discovered (Fig. 1, arrow), and the lesion grew slightly over the next three years. In 2010, additional 2-cm cystic lesions were discovered (Fig. 1, arrowhead) with lower intensity on T2-weighted MRI than the other cysts (Fig. 3, arrowhead).

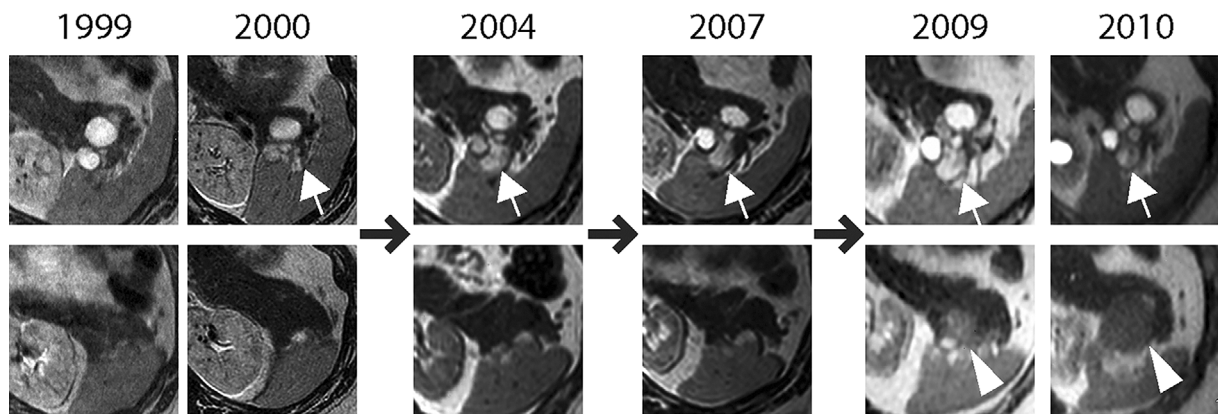
The patient remained asymptomatic, and blood tests

---

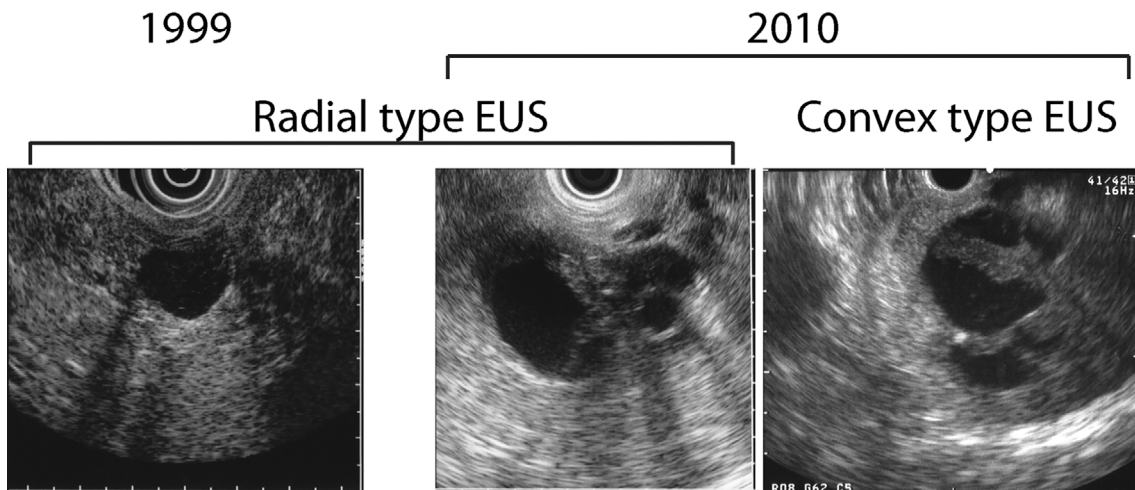
<sup>1</sup>Division of Gastroenterology and Hematology/Oncology, Department of Medicine, Asahikawa Medical University, Japan, <sup>2</sup>Department of Gastroenterology, Nayoro City General Hospital, Japan, <sup>3</sup>Department of Gastroenterology, Hokkaido Prefectural Welfare Federation of Agricultural Cooperative Asahikawa Kosei General Hospital, Japan, <sup>4</sup>Gastroenterology Medicine Center, Shonan Kamakura General Hospital, Japan and <sup>5</sup>Center for Clinical and Biomedical Research, Sapporo Higashi Tokushukai Hospital, Japan

Received for publication March 23, 2016; Accepted for publication April 27, 2016

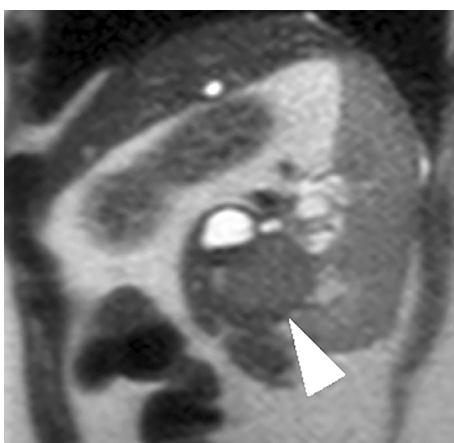
Correspondence to Dr. Junpei Sasajima, junsasaji@gmail.com



**Figure 1.** The serial changes in the MRI findings. T2-weighted MRI of the multi-locular cysts in the pancreatic tail during 11-year follow-up. Several new cystic lesions (arrow) were detected between the index lesions and the spleen in 2000. In 2010, an additional lesion, 2 cm in size, was observed in the splenic hilum (arrowhead); the images in 2009 also showed the presence of the new lesion retrospectively.



**Figure 2.** The endoscopic ultrasound images. Imaging revealed a cystic lesion in the pancreatic tail with multi-locular lesions in 1999. Eleven years later, the lesion had enlarged with thick septa.



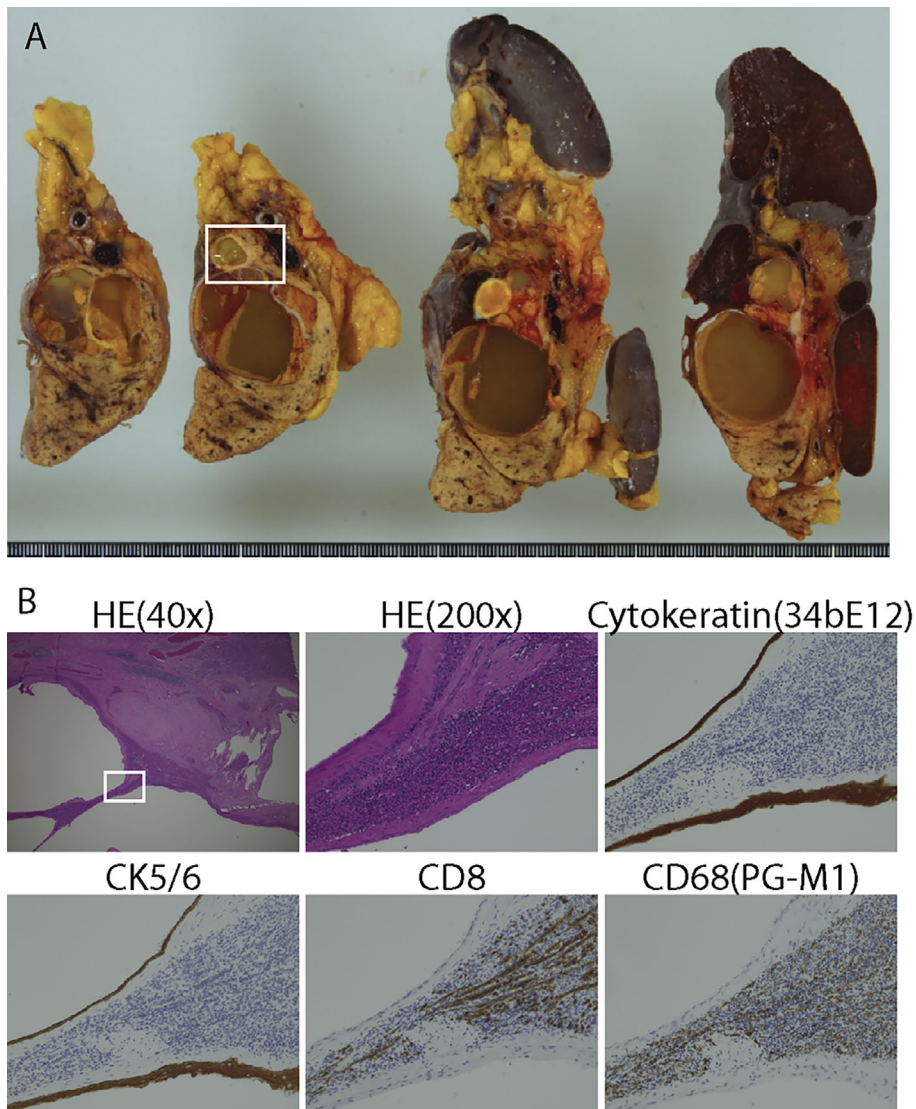
**Figure 3.** A multi-planar reconstruction image of the MRI findings before distal pancreatectomy. Multiple cysts were observed between the pancreatic tail and splenic hilum. The intensity of the additional cystic lesions (arrowhead) was lower than that of the other cysts.

showed that only the levels of elastase were elevated. EUS also revealed multi-locular cysts with thickened septa surrounded by a capsule (Fig. 2). Re-sampling of the cyst fluid by EUS-FNA once again was negative for malignancy. Since bleeding into the pancreatic cysts was suspected, distal pancreatectomy was performed.

A pathological examination revealed a lesion measuring 5.5×5.0×3.5 cm in size in the pancreatic tail containing multi-locular cysts. These cysts were lined with benign multi-layered flattened epithelium that was positive for high-molecular-weight keratin (34bE12) and cytokeratin 5/6. Most of the cysts were surrounded by a thin layer of red-colored tissue consisting of cells positive for CD8 and CD68. This layer was connected to the spleen; therefore, this multi-locular pancreatic cyst was diagnosed to be an invaginated splenic epidermoid cyst (Fig. 4).

### Discussion

To our knowledge, this is the first report of an epidermoid



**Figure 4.** The pathological findings. (A) Grossly, the cut surface of the pancreas and spleen showed a lesion measuring 5.5×5.0×3.5 cm in the pancreatic tail containing multi-locular cysts. Most of the cystic lesions were surrounded by a thin layer of red-colored tissue connecting to the spleen. (B) A microscopic analysis showed that the cysts were lined with multi-layered flattened non-neoplastic epithelium positive for high-molecular-weight keratin (34bE12) and cytokeratin 5/6. The thin layer surrounding the cysts consisted of cells positive for CD8 and CD68, demonstrating that the layer was normal splenic pulp tissue.

cyst in the pancreatic tail that invaginated from the spleen. Both pancreatic and splenic epidermoid cysts are extremely rare benign primary cysts (1-3). Pancreatic epidermoid cysts usually arise from intrapancreatic accessory spleens and are characterized by non-neoplastic keratinizing epithelium surrounded by splenic parenchyma (3). In this case, the cysts were surrounded by splenic parenchyma and directly connected to the spleen, suggesting an invagination from the splenic hilum rather than arising from an accessory spleen.

Three hypotheses have been proposed regarding the pathogenesis of epidermoid cyst of the spleen or intrapancreatic accessory spleen: the mesothelial invagination theory, the lymph space theory, and the endodermal inclusion theory (2). In our case, the congenital cyst lined and surrounded with splenic parenchyma appeared to be invaginat-

ing into the pancreas from the spleen, conforming to the endodermal inclusion theory. This finding also supports the notion that epithelial splenic cysts do indeed develop metaplasia of heterotopic endodermal inclusion within the spleen (3).

Since the multi-locular cyst in the current case was localized mainly to the pancreas, the preoperative diagnosis was a benign pancreatic cyst. Pancreatic epidermoid cysts are usually identified incidentally and are frequently recognized as neoplastic cysts, such as mucinous cystic neoplasms or intraductal papillary mucinous neoplasms of the pancreas (3). Therefore, immediate surgical resection was performed in the majority of reported cases. Conservative observation was conducted in only four previous reports, but for much shorter periods than in the present case, ranging

from 4 months to 3 years (4-7). In these five patients, including ours, enlargement of the cysts was seen in three cases over time (4, 7), suggesting that epidermoid cysts in the pancreas may sometimes increase in size.

We described a rare case of an epidermoid cyst in the pancreatic tail that invaginated from the spleen, strongly supporting the endodermal inclusion theory. The emergence of satellite lesions and the enlargement of the cyst were observed during the 11-year follow-up, suggesting that epidermoid cysts may increase in size over time.

**The authors state that they have no Conflict of Interest (COI).**

#### Acknowledgement

The authors thank the late Dr. Yoshihiko Tokusashi for providing the pathological diagnosis. Dr. Tokusashi was an outstanding pathologist at Asahikawa Medical University who aided the precise pathological assessment of the resected specimens.

#### References

1. Robbins FG, Yellin AE, Lingua RW, Craig JR, Turrill FL, Mikkelsen WP. Splenic epidermoid cysts. *Ann Surg* **187**: 231-235, 1978.
2. Ingle SB, Hinge Ingle CR, Patrike S. Epithelial cysts of the spleen: a minireview. *World J Gastroenterol* **20**: 13899-13903, 2014.
3. Kim YS, Cho JH. Rare nonneoplastic cysts of pancreas. *Clin Endosc* **48**: 31-38, 2015.
4. Gleeson FC, Kendrick ML, Chari ST, Zhang L, Levy MJ. Epidermoid accessory splenic cyst masquerading as a pancreatic mucinous cystic neoplasm. *Endoscopy* **40** (Suppl 2): E141-E142, 2008.
5. Urakami A, Yoshida K, Hirabayashi Y, et al. Laparoscopy-assisted spleen-preserving pancreatic resection for epidermoid cyst in an intrapancreatic accessory spleen. *Asian J Endosc Surg* **4**: 185-188, 2011.
6. Hamidian Jahromi A, Fallahzadeh MK, Dela Cruz N, Chu Q. Epidermoid cyst arising from an intrapancreatic accessory spleen: a case report and a review of the literature. *J La State Med Soc* **165**: 153-156, 2013.
7. Kumamoto Y, Kaizu T, Tajima H, Kubo H, Nishiyama R, Watanabe M. A rapidly growing epidermoid cyst in an intrapancreatic accessory spleen treated by laparoscopic spleen-preserving distal pancreatectomy: Report of a case. *Int Surg* 2015 (Epub ahead of print).

The Internal Medicine is an Open Access article distributed under the Creative Commons Attribution-NonCommercial-NoDerivatives 4.0 International License. To view the details of this license, please visit (<https://creativecommons.org/licenses/by-nc-nd/4.0/>).