E-ISSN: 3068-5052
Journal of Regenerative Medicine & Biology Research

ATHENAEUM SCIENTIFIC PUBLISHERS

Clinical Note

A Case of an Intrapancreatic Accessory Spleen in a Child

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Citation: Ohno M, et al. A Case of an Intrapancreatic Accessory Spleen in a Child. J Reg Med Biol Res. 2025;6(3):1-3.

https://doi.org/10.46889/JRMBR.2025.6307

Received Date: 08-12-2025

Accepted Date: 22-12-2025

Published Date: 29-12-2025

Clinical Note

A 13-year-old boy with a suspected intraperitoneal tumor presented with left flank pain and a fever of 38.5°C. Blood test results were as follows: white blood cell count, 17.2 10^3/μL; C-reactive protein, 14.53 mg/dL; alpha fetoprotein, 1 ng/mL; human chorionic gonadotropin, 1.4 mIU/mL; neuron specific enolase, 9.9 ng/mL; carcinoembryonic antigen, 0.6 ng/mL; carbohydrate antigen 19-9, 8.1 U/mL and duodenal pancreatic cancer antigen-2, 25 U/mL. Ultrasound revealed two large hypoechoic cystic masses with internal septa (Fig. 1). Computed Tomography (CT) showed a potential tumor, which was a well-defined, multilocular cystic lesion with a septum-like structure and walls with uneven thickness, with a splenic artery supply. Located in the retroperitoneum, the tumor was in contact with the spleen, colonic splenic flexure and kidneys. Slight pancreatic tail infiltration suggested a pancreatic tumor possibility (Fig. 1). Magnetic Resonance Imaging (MRI) showed no signs of infiltration into the pancreatic duct. The tumor also had a contrast-enhanced mural nodule, suggesting the possibility of a teratoma. After hospitalization, antibiotic treatment was provided, relieving inflammation. However, cyst size remained unchanged, prompting tumor resection with a differential diagnosis of teratoma, pancreatic tumor and lymphangioma.

The tumor was found at the splenic flexure through transillumination of the gastric omentum (Fig. 1). Thus, the omentum was incised to expose the tumor. The tumor was carefully dissected as it adhered to the pancreatic tail, splenic hilum and greater omentum. Rapid pathology testing of the cyst wall confirmed the absence of malignancy. Slight damage to the splenic hilum and pancreatic tail was repaired, while the pancreatic duct was undamaged. A drain was inserted and the surgery was completed.

Pathological findings (Fig. 1) showed that the cyst contained thick fibrous tissue. An accessory spleen was present within the cystic wall. No evidence of pancreatic tumor or teratoma was observed. The postoperative course was uneventful and the patient was discharged 6 days after surgery, with no recurrence 6 months post-surgery.

Accessory spleens are often observed as small round nodules near the splenic hilum or pancreatic tail. However, cases where large tumors are located in the retroperitoneum or pancreatic parenchyma are extremely rare. Accessory spleens within the pancreatic parenchyma in the pancreatic tail are called Intrapancreatic Accessory Spleens (IPASs). Halpert, et al., reported that out of 2,700 autopsy cases, 291 (10.8%) had accessory spleens and 78 (2.9%) had IPASs [1]. IPASs are broadly divided into two: those primarily composed of solid components, including normal splenic tissue and those primarily composed of cystic components combined with lesions, such as epidermoid and epithelial cysts, in solid components. Davidson, et al., reported a case of an epidermoid-cyst IPASs; since then, approximately 40 cases have been reported [2-4]. The mean patient age was 46.1 years (range 12-70 years). Most patients were female (58.3%) and the majority were Asian (77.7%), suggesting a potential racial factor [3]. According to our investigation, only one reported case involved a child-a 12-year-old girl-who was treated with distal pancreatectomy [4].

IPASs show the same contrast enhancement pattern as the spleen on CT and MRI. Furthermore, in Superparamagnetic Iron Oxide (SPIO)-enhanced MRI examinations, low signals are observed for IPASs in T2-weighted imaging, just like those for the spleen. SPIO-enhanced MRI is a diagnostic imaging method with excellent spatial and tissue resolution and no radiation exposure. Meanwhile, nuclear medicine examinations include 99 mTc hepatosplenic tin colloid scintigraphy and 99 mTc damaged red blood cell scintigraphy. Radiation exposure and the small size of the accessory spleen can make visualization challenging. In our case, an accessory spleen was present in the mural nodule of the cyst, which was well-enhanced by CT and MRI (Fig. 1). As an IPASs was not considered, SPIO-enhanced MRI and scintigraphy were not performed previously.

Surgery is recommended for dermoid cysts of splenic origin if the cyst is large and poses a risk of rupture [5]. Despite the difficulty of making a definitive preoperative diagnosis in our case, we successfully performed surgery by tumor removal. No reports of recurrence have been documented. Herein, we presented a rare case of an IPAS in a child. When a mass is located in the pancreatic tail or retroperitoneum, the possibility of an IPAS should be included in the differential diagnosis.

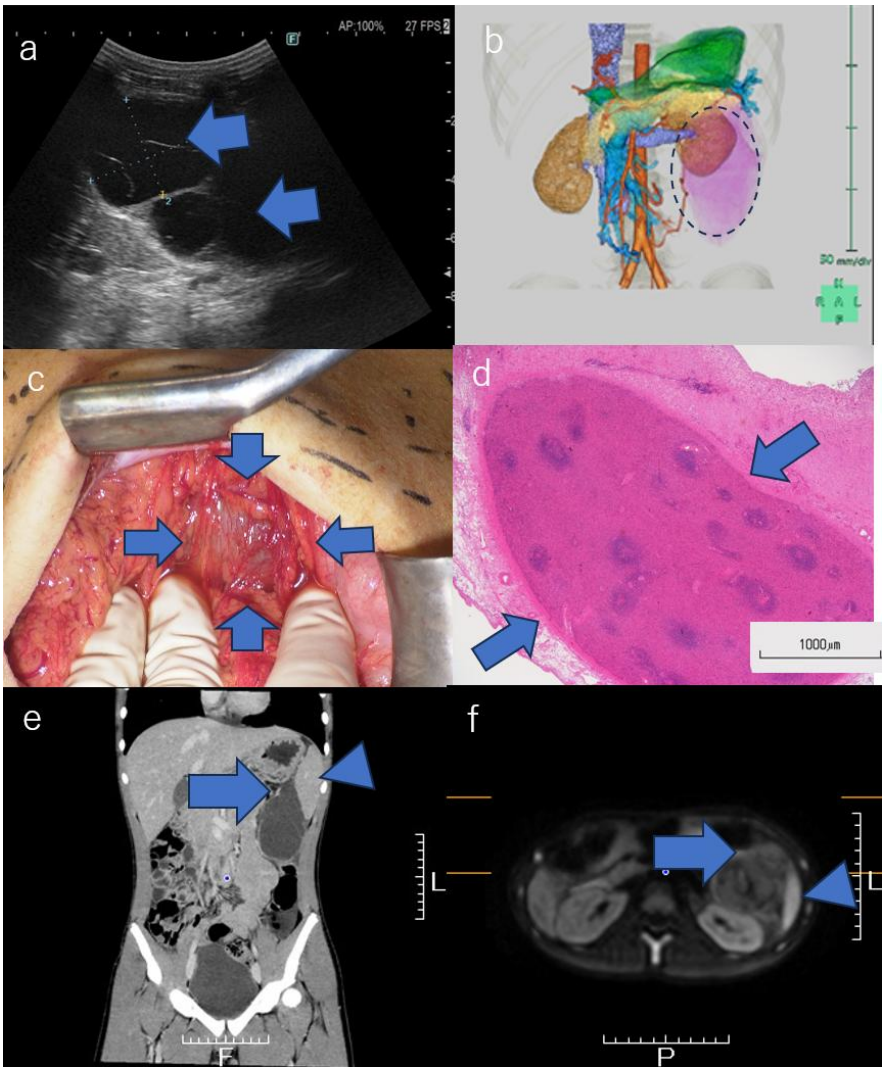

Figure 1: (a) Ultrasound findings show two main large cystic masses (arrow) with internal septa that were observed in the left flank area, measuring approximately 13 cm in maximum overall diameter; (b) Three-dimensional Computed Tomography (CT) shows the cysts (dotted lines) in contact with the tail of the pancreas, the spleen, the splenic flexure of the colon and the kidney; (c) Operative findings: The visualization of the gastric omentum revealed a potential tumor (arrow) at the head of the transverse colon; (d) Pathological findings: The cyst wall had a thick fibrous wall and the epithelium of the lumen was not clear. Splenic tissue (arrow) was observed within the cyst wall. Bar size is 1000 μm; (e) Coronary contrast CT shows small mural nodules (arrow) in the cyst wall, which were of the same density as the spleen (arrowhead); (f) MRI shows high signals in the mural nodules (arrow) on diffusion-weighted imaging, which have the same density as the spleen (arrowhead).

Keywords: Child; Spleen, Accessory; Pancreatic Neoplasms; Retroperitoneal Neoplasms

Conflict of Interest
The authors declare no conflicts of interest.

Funding Statement
None.

Acknowledgments
We appreciate Yuko Nakamura and Yuka Nomura for the data collection.

Informed Consent
Informed consent was obtained from the patient's parents and the case report was approved by the ethics committee of our hospital (Approval No. B0702).

Author Contributions
Y.M. introduced the case to us. M.O., Y.M. and performed the surgery. M.O., Y.M., R.I. and H.W. managed the patient before and after the surgery. All authors have read and approved the final version of the manuscript.

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