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Letters to the Editor

Experience in Pancreaticopleural Fistula Caused by Intestinal Duplication Presenting as Pancreatic Pseudocysts

Dear Editor,

Intestinal duplications are rare congenital anomalies and we encountered a large intestinal duplication mimicking pancreatic pseudocyst with chronic bilateral pleural effusion caused by intestinal duplication pleura fistula, which is exceptionally rare.

A 22-month-old boy was hospitalised for several times due to bilateral pleural effusion and pancreatic pseudocyst, however, no fistula was identified. There was no history of trauma. The patient was treated conservatively and his symptoms were rapidly relieving after percutaneous drainage of pancreatic pseudocyst, then he was discharged. A follow-up CT scan with contrast by drainage tube identified a pancreaticopleural fistula was actually existing (Figure 1). Due to the recurrent episodes, pseudocystojejunostomy with a Roux-Y loop was performed (Figure 2). Pathologic examination of the full-thickness biopsy showed that the cyst was lined with intestinal mucosa and muscularis mucosa with muscle layers. The boy was completely asymptomatic on one-year follow-up.

Lewis and Thyng[1] supposed that intestinal duplications are the result of diverticula of the embryonic intestine which fail to regress. During fetal development, if a diverticulum forms on or near the pancreatic bud, which may arise from the anlage of the pancreatic duct, then a duplication with pancreatic communication will appear. However, the precise mechanism remains to be elucidated. Some ectopic pancreatic tissue may exist in the mucosal lining of duplications which makes the secretion of intestinal duplication and the pleura fluid with high amylase. However, no pancreatic tissue was found by pathological examination of the full-thickness biopsy. Maybe that was just partial cystic wall and could not represent the whole cyst.

The underlying mechanism of this case maybe that a posterior disruption of the cyst which flowed through the retroperitoneum and dissected through the esophageal hiatus into the mediastinum to form a pleural fistula, which in turn ruptured into the pleural cavity and formed intestinal duplication pleura fistula.[2] Vague presentation makes it difficult to distinguish intestinal duplication from other mass in the upper left abdomen. Even radiographic studies

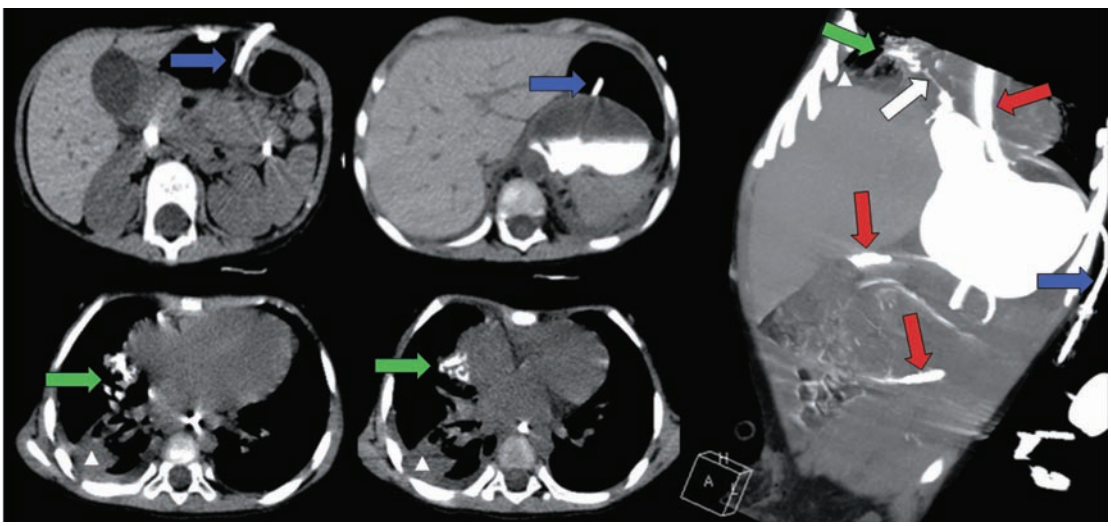

Figure 1 CT with contrast reveals a fistula (white arrow) goes from the cyst up to mediastinum, a jejunal nutrition tube (light grey arrows), pleural effusion (white triangles) and a drainage tube (black arrows), also, the contrast appears in the chest cavity (dark grey arrows).

suggested the possibility of pancreatic pseudocysts and pancreaticopleural fistula rather than a rare intestinal duplication pleura fistula. Magnetic resonance cholangiopancreatography (MRCP) is useful in diagnosing intra-abdominal cysts, especially in depicting pancreatic disease. However, the boy did not take MRCP for some reasons.

The boy responded poorly after 6-month conservative treatment and required surgery eventually. During the operation, the deep position of cyst along with tight adhesion made it too difficult to resect the cyst completely. Forced removal of the cyst might be risky. To protect adjacent tissues, pseudocystojejunostomy with a Roux-Y loop was performed finally. The outcome is good. Surgical management is recommended, also including asymptomatic duplication cysts, for these may cause potential complications such as malignancy, etc..[3] Surgical options for intestinal duplication include simple duplication excision, partial bowel resection with duplicated bowel anomaly and fenestration drainage of the cyst. Rare differential diagnosis of intestinal duplication cyst should be considered in case of recurrent pancreatitis and pancreatic pseudocyst with unclear aetiology.

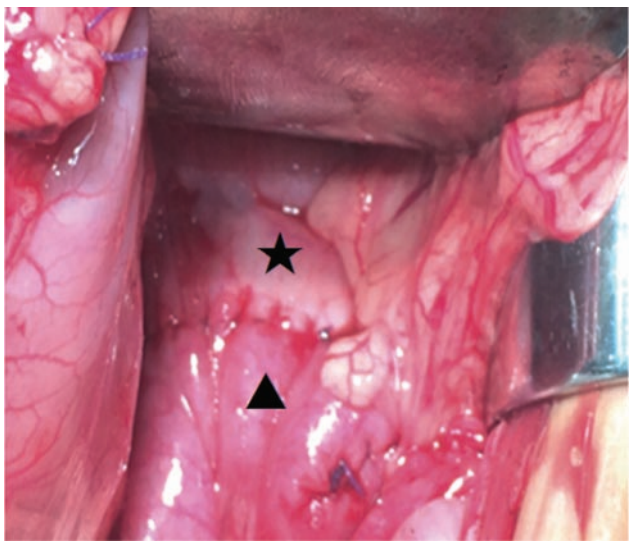

Figure 2 Intraoperative findings show the cyst (star) which looks like a section of digestive tract and jejunum (triangle).

Conflict of Interest

There are no competing interests.

References

1. Lewis FT, Thyng FW. Regular occurrence of intestinal diverticula in embryos of pig, rabbit and man. Am J Anat 1908;7:505-19.
2. Dhebri AR, Ferran N. Nonsurgical management of pancreaticopleuralfistula. JOP 2005;6:152-61.
3. Hsu H, Gueng MK, Tseng YH, Wu CC, Liu PH, Chen CC. Adenocarcinoma arising from colonic duplication cyst with metastasis to omentum: A case report. J Clin Ultrasound 2011; 39:41-3.

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