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Case Report

Acute duodenal obstruction after hemorrhaging of a duodenal duplication cyst

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Duodenal duplication cysts are relatively rare congenital anomalies that occur most often in infants and children. We report a case of a 13-year-old boy who presented with duodenal obstruction caused by bleeding of a duodenal duplication cyst. Operative findings and hispathological examination confirmed the diagnosis. We treated him with subtotal excision of the duodenal duplication cyst and mucosal stripping and ablation of the remaining mucosa.

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Introduction

Alimentary tract duplications are relatively rare congenital anomalies that occur most often in infants and children. Most of the duplications are found at the jejuno-ileal site, and duodenal duplications occur in less than 1 of 100.000 births[1]. Duodenal duplication has been described as causing pancreatitis, bleeding and duodenal obstruction[1]. We present the case of a 13-year-old boy with acute duodenal obstruction caused by duodenal duplication cyst bleeding.

Case report

A 13-year-old boy started to have abdominal pain and vomiting two days before visiting the family doctor. Ultrasonography indicated gastric outlet obstruction caused by a hemorrhaging retroperitoneal cyst and he was referred to our hospital. He had no significant family or past history, including trauma. Physical examination revealed right hypochondrial tenderness with a firm mass. There was no rebound or guarding. Laboratory findings revealed elevation of serum amylase levels (730U/L). Abdominal ultrasonography showed gastric distension and mild dilation of the intrahepatic bile duct. A nasogastric tube was inserted, and 310 mL of gastric juice with bile was aspirated. On abdominal computed tomography (CT), a giant cyst of 8cm x 5cm x 8cm in size was observed obstructing the duodenum, extending to the ascending colon and the upper rectum. The cyst adhered to the second part of the duodenum which had no communication to the duodenal lumen (Figure 1 and 2). Contrast-enhanced CT demonstrated extravasation of contrast medium into the cyst (Figure 3).

An operation was performed on presumed diagnosis of obstruction of the second part of the duodenum caused by retroperitoneal cystic hemorrhage. The firm cyst located below the duodenum was unmovable (Figure 4). The stomach and the first portion of the duodenum proximal to the cyst

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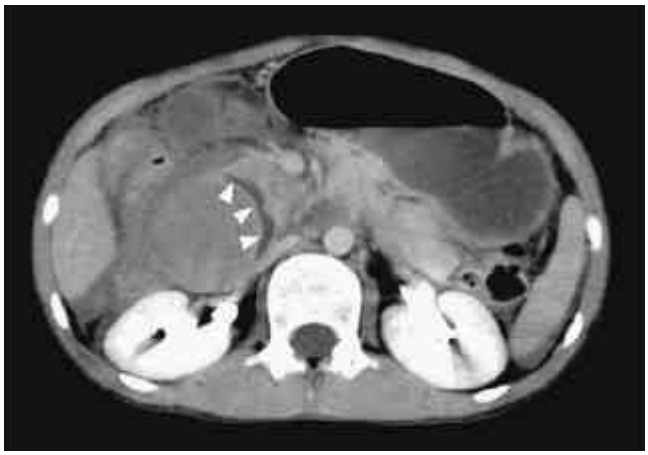

Figure 1. Axial contrast-enhanced CT
CT scan shows duodenal compression by the cyst (arrowheads).

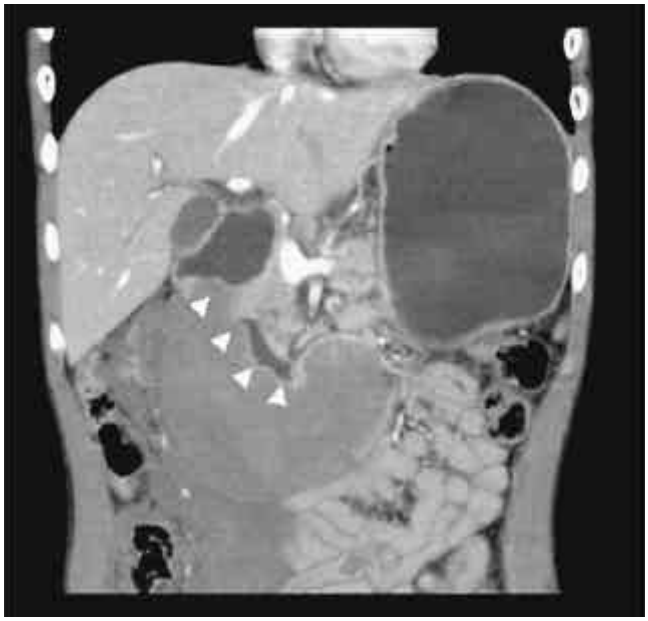

Figure 2. Coronal contrast-enhanced CT
The cyst compresses the second portion of the duodenum (arrowheads) and the stomach and duodenal bulb are dilated.

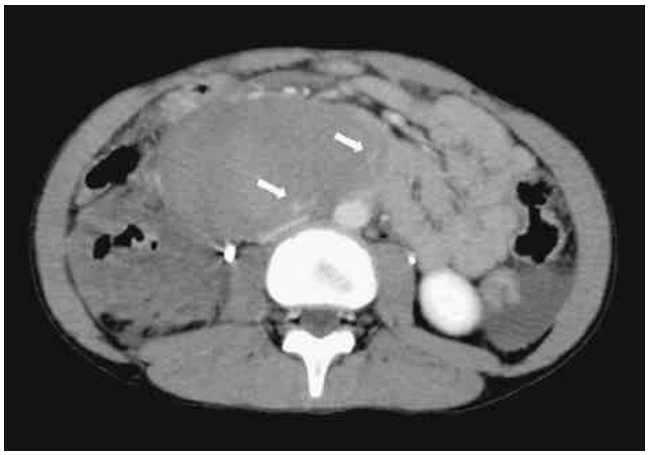

Figure 3. Axial contrast-enhanced CT
Extravasation of contrast media is observed in the cyst (arrows).

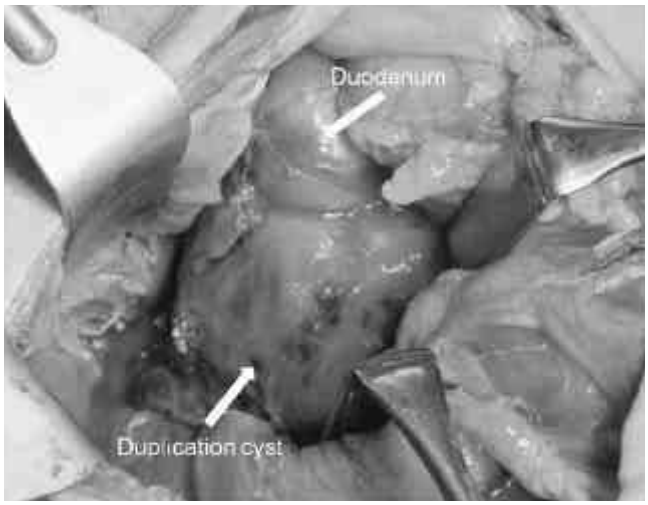

Figure 4. Operative finding
The firm cyst adjoining the second portion of the duodenum compresses the duodenum.

were significantly dilated. The cyst was opened wide, and multiple thrombi were removed from the cavity. No communication was identified between the cyst lumen and the duodenum lumen. The cyst wall adhered to a wide area of the second and third portion of the duodenum. To avoid duodenal continuity, we performed partial resection of the cyst wall and mucosal stripping and ablation of the remaining mucosal structure on the adjoining duodenum. Histopathology confirmed the diagnosis of the duodenal duplication cyst by identifying the two muscular layers and the intramuscular ganglion cells of the extirpated cyst. There was no gastric mucosa or peptic ulceration in the resected specimen.

The patient had an uneventful recovery. Postoperative radioisotope examination showed no accumulation of remnant gastric mucosa.

Discussion

Alimentary tract duplications have been described by Fitz in 1884[2] and Ladd in 1937[3]. They are most commonly found in the jejunum or ileum and least often in the duodenum, and may arise during the fetal period[1, 4]. Several theories have been proposed to explain the etiology of alimentary tract duplications, including errors in the recanalization of the primitive intestine, failure in the regression of embryonic diverticula and errors in the splitting of the notochord[3-6]. As with other duplications, the most common types of duodenal duplication are cystic and non-communicating. The symptoms and signs of duodenal duplication depend on their type and location. Asymptomatic masses are rare, but can occasionally

be found on physical or radiographic examination. The presence of gastric or pancreatic mucosa in the cyst may lead to peptic ulcers, perforations and bleeding[3]. Bleeding in communicating duplication manifests as hematemesis or melena; bleeding in non-communicating duplication manifests as compression of adjacent structures.

Severe compression can cause gastrointestinal obstruction. Duodenal duplication may cause acute pancreatitis due to compression of the ampulla of Vateri[7].

Physical examination of the abdomen and laboratory studies showed no characteristic features. Preoperative diagnosis of duodenal duplication can be difficult. Ultrasonography may show "double wall" or "muscular rim" signs and suggest a diagnosis of cystic typed duplication. "Double wall" or "muscular rim" refers to the appearance of a cyst mimicking the gastrointestinal tract with an echogenic inner margin corresponding to mucosa surrounded by a hypoechoic rim of tissue representing the smooth-muscle layer.

Identification of this sign on sonography of an abdominal mass has been regarded as characteristic of an enteric duplication cyst[8,9]. CT and MRI also can provide an accurate definition of the cyst, vascular attachment and contents of the cyst[10]. Radioisotopes have been used to locate the ctopic gastric mucosa in the cysts.

Treatment for duodenal duplication is complete primary excision, thus preventing any further risk that may arise from residual ectopic gastric or pancreatic mucosa[11, 12]. Malignant changes can occur in the mucosa of an enteric duplication[13]. Complete excision of a duodenal duplication is sometimes difficult because the duodenum is an organ that is fixed behind the retroperitoneum and also has delicate anatomical communication to the pancreatobiliary duct. Management of duodenal duplication should be decided by the volume, type, location and proximity to the duodenal wall or pancreaticobiliary duct. In the case of incomplete excision of duodenal duplication, complete mucosal stripping or ablation of the remaining structure must be carried out. In the present case, we detected no residual gastric mucosa after postoperative radioisotope examination.

Conclusion

Duodenal duplication should be considered as the differential diagnosis for a patient with a cystic lesion adjoining the duodenum. Desirable surgical treatment is complete excision of the duplication but, if not possible, subtotal excision and internal derivation is recommended.

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