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CASE REPORT

Intraluminal duodenal diverticulum in a child concomitant with an entrapped coin and a duodenal polyp

Hui-Ling Peng [a], Cheng-Tau Su [a,*], Chia-Yu Chang [b], Beng-Huat Lau [c]

[a] Department of Diagnostic Radiology, Shin Kong Wu Ho-Su Memorial Hospital, Taipei, Taiwan
[b] Department of Pediatric Surgery, Shin Kong Wu Ho Su Memorial Hospital, Taipei, Taiwan
[c] Department of Pediatrics, Shin Kong Wu Ho Su Memorial Hospital, Taipei, Taiwan

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Summary Intraluminal duodenal diverticulum (IDD) is a rare congenital anomaly. We present the report of an 8-year-old girl who had an entrapped coin in an IDD for 3 years that was associated with recurrent pancreatitis. Besides, a duodenoduodenal intussusception was found during the course of investigation and it seemed that a concomitant duodenal polyp contributed to the development of the intussusception. In view of the rarity of each of the aforementioned situations and the improbability of these conditions occurring together, this unusual and possibly unique case is therefore reported here.

1. Introduction

An intraluminal duodenal diverticulum (IDD) is a rare developmental anomaly that is usually asymptomatic. Complications such as intestinal obstruction, hemorrhage, cholangitis, or pancreatitis have been reported in patients with IDD, but these mostly occur only in adult patients.[1] Retention of a coin in the IDD has only been sporadically described.[2] Moreover, duodenoduodenal intussusception is a rare entity because the duodenum is fixed in the retroperitoneum. When encountered, it is usually secondary to a neoplasm that acts as a lead point.[3] The aim of this report is to describe an IDD with a coin entrapped in it for 3 years that caused recurrent pancreatitis in a young girl in whom duodenoduodenal intussusception was possibly caused by a duodenal polyp.

Conflicts of interest: The authors declare that there are no conflicts of interest related to the subject matter or materials discussed in this article.

* Corresponding author. Department of Diagnostic Radiology, Shin Kong Wu Ho-Su Memorial Hospital, Number 95, Wen Chang Road, Shih Lin District, Taipei 111, Taiwan.
E-mail address: hungko@ms21.hinet.net (C.-T. Su).

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2. Case report

An 8-year-old girl had a 1-day history of epigastric pain, nausea, and vomiting. Laboratory test results suggested an acute pancreatitis with elevated white blood count of 18,500/mm^3 (normal value 4000–10,000/mm^3), elevated amylase level of 797 U/L (normal value 28–109 U/L), and elevated lipase level of 1005 U/L (normal value 13–60 U/L). An abdominal radiograph showed a round, metallic foreign body in the upper abdomen. A detailed historical and radiographic review revealed that the metallic object was a coin that was ingested 3 years earlier. The abdominal pain and vomiting subsided promptly after conservative management, and laboratory results returned to normal within 4 days. No attempt was made to deal with the coin retention in this admission and the patient was discharged home with appointment for follow-up. However, 4 weeks later, the patient was re-admitted because of a further bout of pancreatitis (with amylase and lipase levels of 1521 U/L and 3620 U/L, respectively). Abdominal sonography revealed intestinal intussusception that was not reduced by administration of normal saline enema. Initial computed tomography (CT) showed a duodenoduodenal intussusception at the second portion of the duodenum and a fluid-filled lesion situated distally to it (Fig. 1A and B). Because of the presence of an artifact caused by the coin, we could not comment on the exact anatomical and pathological structure of this lesion. At laparoscopy, no extraluminal foreign body or abscess was found and the intussusception resolved spontaneously after general anesthesia administration. Gastroduodenoscopy revealed a pouch-like lesion arising from the wall of the second portion of the duodenum with a coin entrapped inside (Fig. 2A). A 2-cm polypoid lesion was present at the orifice (1 cm in diameter) of the pouch (Fig. 2B). A 5-dollar Taiwan coin (2 cm in diameter and 0.1 cm in thickness) was successfully removed by endoscopy. An upper gastrointestinal series showed a contrast-filled "wind sock" arising in the second portion of the duodenum and extending to the third portion. A peculiar filling defect near the opening of the wind sock also showed a polypoid mass (Fig. 3). A subsequent oral-contrast CT revealed a contrast-filled pouch that was surrounded by a hypodense halo and lying in the duodenal lumen with a polypoid filling defect located at its proximal portion (Fig. 4). Based on these endoscopic and radiological findings, the diagnosis of IDD with a duodenal polyp was made. The diverticulum and the polyp at its base were then surgically removed by performing a longitudinal duodenotomy with careful visualization of the papilla of Vater (Fig. 5). The mucosal defect of the duodenum was repaired with interrupted absorbable stitches and the duodenotomy was closed transversely. Histologic analysis of the specimens confirmed a 2 cm × 1 cm × 1 cm inflammatory polyp and a 7 cm × 4 cm × 3-cm IDD. The postoperative course was uneventful and the patient was healthy at follow-up.

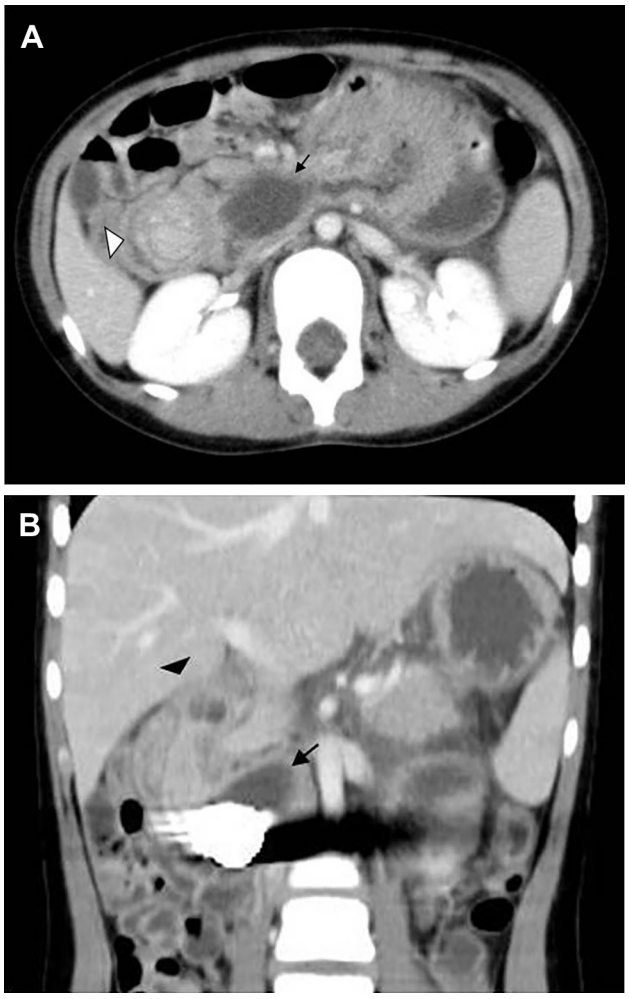

Figure 1 (A) Initial contrast computed tomography (computed tomography dose index 3.86 mGy) showing the "target" appearance in the region of the duodenum, which confirmed the diagnosis of duodenoduodenal intussusception (white arrowhead). The image shows a fluid-filled lesion that appeared to arise from the duodenum (black arrow). (B) Coronal multiplanar reformatted image showing the duodenoduodenal intussusception (arrowhead). The image also shows a fluid-filled lesion with a metallic artifact lying distally to it (black arrow).

3. Discussion

Coins are the most frequently ingested foreign bodies in children.[4] In general, coins smaller than 2.5 cm in diameter will usually pass spontaneously in children in the absence of stenotic lesions or abnormalities in the gastrointestinal tract.[4] The coin in our case was 2 cm in diameter, and therefore could be excreted from the gastrointestinal tract without causing damage. However, it had been entrapped for 3 years at the duodenum because of an IDD, a rare type of duodenal diverticulum. Duodenal diverticula are classified into extraluminal duodenal diverticulum (EDD) and IDD. EDD is acquired and more common. In EDD, a sac of mucosal or submucosal layer herniates through a muscular defect in the duodenal wall. However, the precise mechanism of EDD development is not known yet.[5] In contrast to EDD, IDD is a rare congenital abnormality that has the sac protruding into the duodenal lumen, and thus far only about 150 cases have been described in the literature.[6] During early fetal

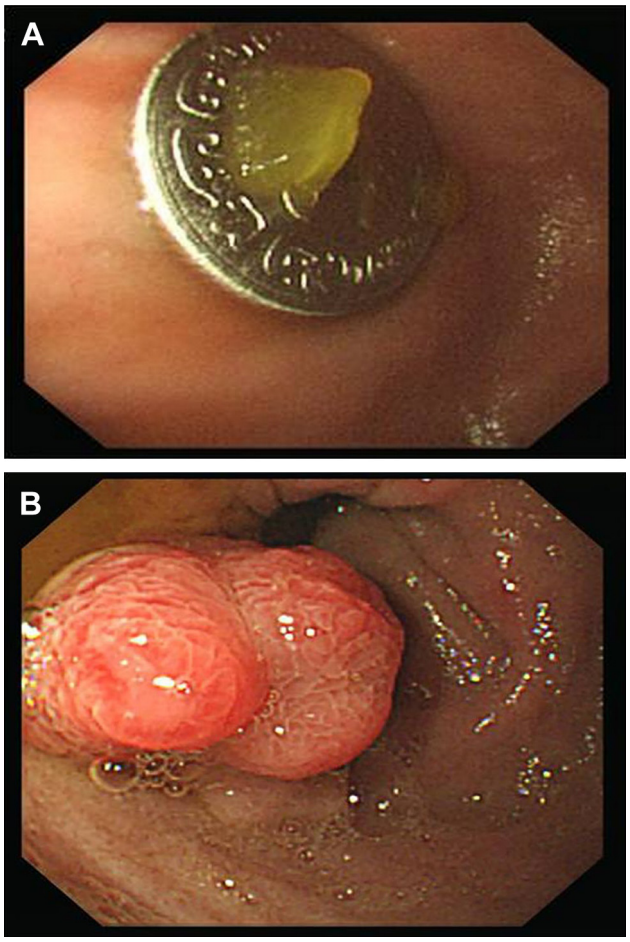

Figure 2 (A) A 5-dollar Taiwan coin (2 cm in diameter and 0.1 cm in thickness) inside the pouch that was removed by gastroduodenoscopy. A thick mucus plug on the surface of the coin is seen. (B) A 2-cm polypoid mass at the orifice of the pouch.

life, duodenal lumen is initially occluded by proliferating epithelial cells and will be recanalized later. Failure of the lumen to fully recanalize may lead to a duodenal diaphragm or duodenal web and over time peristaltic stretching may transform it into an IDD. It usually occurs at the second portion of the duodenum and arises near the ampulla of Vater and is usually asymptomatic in childhood.[5,6] As the IDD enlarges with patient's age or distends with food and repetitive peristalsis, it acts as a large foreign body in the lumen, which contributes to partial or intermittent duodenal obstruction. Postprandial fullness, pain, and vomiting are the frequently reported symptoms in such cases. Besides foreign body entrapment, other reported complications in IDD are duodenal obstruction, upper intestinal bleeding, cholangitis, and pancreatitis.[1,2]

The reported incidence of pancreatitis associated with IDD is <20% and it mainly affects young adults.[7] The pathogenesis of pancreatitis in patients with IDD is not clear. The most widely accepted mechanism is the reflux of the duodenal content through the papilla of Vater. This can be due to pull of the diverticulum neck at the papillary level during duodenal peristalsis and partial obstruction of the

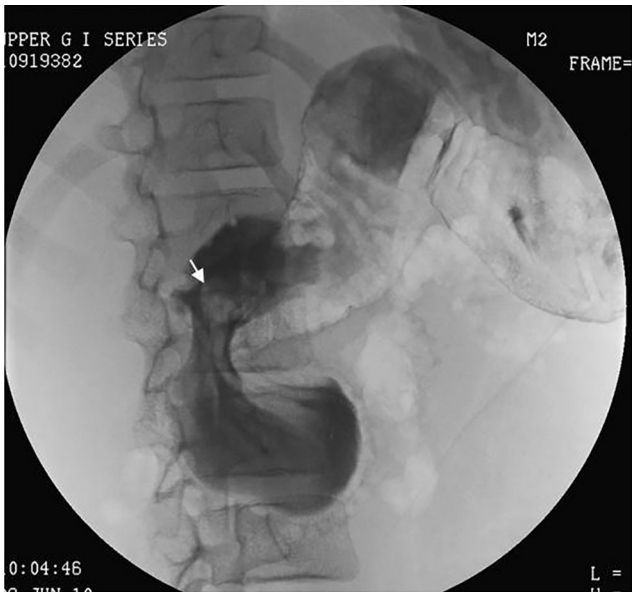

Figure 3 Upper gastrointestinal series showing the typical wind-sock sign. The intraluminal duodenal diverticulum is filled with contrast and the surrounding radiolucent layer is its wall. The outermost layer of contrast is the true duodenal lumen. The peculiar filling defect located at the opening of the pouch is the duodenal polyp (white arrow).

duodenum by the distended diverticulum.[7] Our patient would have probably escaped clinical recognition until later in adulthood had she not ingested the coin. It is felt that the presence of the coin could have further contributed to the distention and enlargement of the IDD and subsequently initiated the clinical manifestations of pancreatitis.

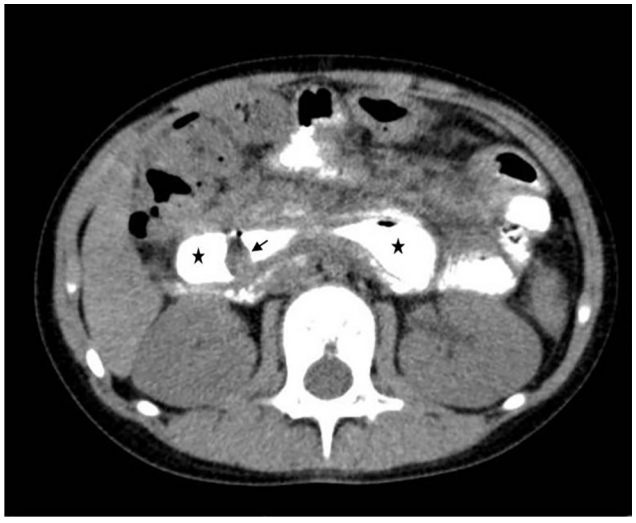

Figure 4 Oral-contrast computed tomography (computed tomography dose index 3.86 mGy) showing a characteristic halo image. The pouch-like intraluminal duodenal diverticulum is filled with contrast and seen lying in the duodenum (stars). The hypodense halo surrounding the pouch represents the diverticulum wall. A polypoid mass located at the proximal portion of the pouch is also seen (black arrow).

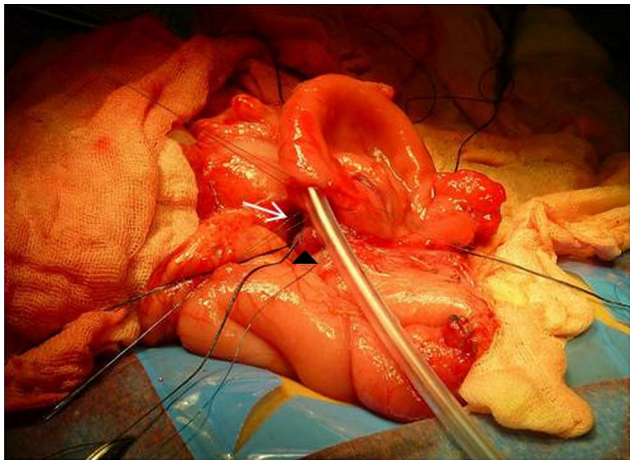

Figure 5 After duodenotomy, an intraluminal duodenal diverticulum was found at the wall of the second portion of the duodenum (white arrow) with a polypoid mass located in front of its orifice (black arrowhead).The true lumen of the duodenum had been catheterized.

Although IDD is a rare cause of pancreatitis in children, it should be included in the differential diagnosis. Thus, an upper gastrointestinal study is mandatory for evaluation of anatomic anomalies when other diagnostic tools fail to identify the cause of the pancreatitis in children.[8] As in our case, the pathognomonic radiologic finding of IDD is a wind sock or teardrop appearance of the contrast collecting within the duodenal lumen, which was lined by a narrow, radiolucent band.[8] Endoscopy, endoscopic retrograde cholangiopancreatography, magnetic resonance imaging, and CT scanning can also be used to make the diagnosis.[5,7,8] Surgical resection, either by open or endoscopic means, is the preferred option for managing symptomatic IDD.

In addition to IDD, our patient also had duodenoduodenal intussusception, which is a very rare complication because the duodenum is fixed in the retroperitoneum. It usually occurs secondary to a neoplasm that acts as a lead point.[3] Griffin et al[2] described an IDD that had migrated beyond the ligament of Treitz causing intussusceptions at the distal portion of the duodenum. They found that distention of the diverticulum led to subsequent intussusception that caused duodenal obstructions in their case.[2] Intussusception in our patient occurred at the second portion of the duodenum, which is exceptionally rare. It is debatable whether intussusception is secondary to IDD per se or the duodenal polyp. Because the polyp was situated proximally to the orifice of IDD, the peristaltic waves would have reached the polyp earlier. As a result, it is more likely that the polyp had acted as a lead point for mucosal enveloping.

In conclusion, IDD is a rare disorder in which a foreign body can often be entrapped. If an ingested foreign body is not excreted in the stool, the possibility of retention in the intestinal tract should always be considered and a thorough investigation is mandatory. Moreover, IDD as the cause or a contributing factor should be kept in mind in the differential diagnosis of pancreatitis in children. Finally, duodenoduodenal intussusceptions, although rare, does occur, and the underlying cause of this extremely uncommon situation should be identified.

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