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Heterotopic Pancreas: A Rare Cause of Ileo-Ileal Intussusception

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Summary

Background:

Heterotopic pancreas is a rare developmental anomaly defined as pancreatic tissue found on ectopic sites without contiguity with the main pancreas. An isolated heterotopic pancreas as a cause of bowel intussusception is extremely rare.

Case Report:

A case of 47-year old male with multiple episodes of melena, constipation and abdominal pain for one year duration is presented. CT eneterography revealed a large circumferential lesion involving the terminal ileum that acted as a leading point to an ileo-ileal intussusception. The resection of the lesion and related bowel segment was carried out. The histopathological examination confirmed the excised lesion as a heterotopic pancreatic tissue.

Conclusions:

Though a rare entity, heterotopic pancreas should be considered in the differential diagnosis of bowel intussusception.

MeSH Keywords:

Choristoma • Ileum • Intussusception • Pancreas

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Background

Heterotopic pancreas (HP) is a developmental anomaly defined as pancreatic tissue found on ectopic sites without contiguity with the main pancreas. An isolated HP as the lead point of bowel intussusception is extremely rare [1]. We reported on a large circumferential multiple HP in the ileum as a leading cause of ileo-ileal intussusception and continuous source of melena, which to the best of our knowledge is quite a rare condition.

Case Report

A 47-year-old male presented with a one-year history of multiple "off-and-on" episodes of melena and constipation. Those were associated with the onset of lower abdominal pain and generalized fatigability. On examination, the patient was pale; the rest of the examination was unremarkable. The investigations showed that the patient had a low hemoglobin level, i.e. 8.5 g/L. CT enterography revealed

a large circumferential lesion measuring approximately 8.0×1.8 cm involving the terminal ileum, which acted as a leading point to an ileo-ileal intussusception (Figure 1A–C). There were also other small multiple satellite lesions. Resection of the segment containing the submucosal lesion was carried out with side-to-side anastomosis.

The resected segment was sent for histopathological examination. The report stated that the segment contained heterotopic pancreatic tissue involving all the layers of the polypoid bowel invagination. There was evidence of extensive mucosal ulceration and granulation tissue formation with attenuation of the muscularis propria at the tip of the polypoid lesion. The patient was discharged and followed up as an out-patient.

Discussion

HP, also known as ectopic, aberrant or accessory pancreas is defined as the presence of pancreatic tissue outside its normal location and without anatomic and vascular continuity

Figure 1. Ileo-ileal intussusception caused by heterotopic pancreas. CT with intravenous contrast: (A) Axial image: Distended bowel loop with intraluminal fat density (like mesentery) with target sign (arrow) suggestive of intussusception. (B) Coronal reformation image: Tubular-like structure at the right iliac fossa shows fat density within. (C) Sagittal reformation image: Ileal loop is seen invaginated into another ileal loop (arrow).

with the main body of the pancreas [2]. Several theories have been proposed to explain the occurrence of HP. The most tenable implicates that during the embryonic rotation of the dorsal and ventral buds, fragments of the pancreas become separated from the main body and are deposited at ectopic sites [3]. HP is usually found incidentally and is generally asymptomatic, but it may become symptomatic when complicated by inflammation, bleeding, obstruction or malignant transformation [4–6]. Although HP can be found throughout the entire gastrointestinal tract, it is most commonly found in the stomach (25–38%), duodenum (17–36%), and jejunum (15–21%). There were rare cases of HP in the esophagus, biliary tract, gallbladder, spleen, and mesentery [7] and even more rarely at other sites including Meckel's diverticulum, ileum, colon, gallbladder, common bile duct, umbilicus, fallopian tubes, liver, spleen and mediastinum [8,9]. HP can be present at any position in the abdominal cavity. It is usually found in the upper gastrointestinal tract, with more than 90% of the cases involving the stomach, duodenum or jejunum [10].

References:

1. Anselme P, Grundfest S, Carey W et al: Pancreatic heterotopias: a rare cause of bowel obstruction. *Surgery*, 1981; 90: 110–13
2. Christodoulidis G, Zacharoulis D, Barbanis S et al: Heterotopic pancreas in the stomach: a case report and literature review. *World J Gastroenterol*, 2007; 13: 6098–100

Despite a relatively frequent occurrence of HP, the vast majority of such cases are asymptomatic [2]. The incidence of a localized pathological leading point [11] for intussusception varies from 2% to 12% in large series. Rarely, jejunal lesions may result in intestinal obstruction or intussusception [12–14].

Isolated HP of the ileum on the other hand is very rare, usually asymptomatic and discovered incidentally during surgery for other conditions, and very rarely as a leading point for intussusception [15–19]. The definitive diagnosis of HP is reached by histopathological examination of the tissue [20].

Conclusions

We are of the opinion that HP, though a rare entity, should be considered in the differential diagnosis of bowel intussusception.

3. Kobayashi S, Okayama Y, Hayashi K et al: Heterotopic pancreas in the stomach which caused obstructive stenosis in the duodenum. *Intern Med*, 2006; 45: 1137–41
4. Agale SV, Agale VG, Zode RR et al: Heterotopic pancreas involving stomach and duodenum. *J Assoc Physicians India*, 2009; 57: 653–54
5. Emerson L, Layfield LJ, Rohr LR et al: Adenocarcinoma arising in association with gastric heterotopic pancreas: a case report and review of the literature. *J Surg Oncol*, 2004; 87: 53–57
6. Hsia CY, Wu CW, Lui WY: Heterotopic pancreas: a difficult diagnosis. *J Clin Gastroenterol*, 1999; 28: 144–47
7. Ormarsson OT, Gudmundsdottir I, Marvik R: Diagnosis and treatment of gastric heterotopic pancreas. *World J Surg*, 2006; 30: 1682–89
8. Tanaka K, Tsunoda T, Eto T et al: Diagnosis and management of heterotopic pancreas. *Int Surg*, 1993; 78: 32–35
9. Kaneda M, Yano T, Yamamoto T et al: Ectopic pancreas in the stomach presenting as an inflammatory abdominal mass. *Am J Gastroenterol*, 1989; 84: 52–54
10. Yuan Z, Chen J, Zheng Q et al: Heterotopic pancreas in the gastrointestinal tract. *World J Gastroenterol*, 2009; 15(29): 3701–3
11. Jacobz A, Nawaz A, Matta H et al: Intussusception secondary to isolated heterotopic pancreas of the ileum: case report and review of the literature. *Ann Saudi Med*, 2002; 22(3–4): 213–15
12. Chandra N, Campbell S, Gibson M et al: Intussusception caused by heterotopic pancreas. *JOP*, 2004; 5: 476–79
13. Gurbulak B, Kabul E, Dural C et al: Heterotopic pancreas as a leading point for small-bowel intussusceptions in a pregnant woman. *JOP*, 2007; 8: 584–87
14. Gupta M, Karlitz J, Raines D et al: Heterotopic pancreas. *J La State Med Soc*, 2010; 162: 310–13
15. Hamada Y, Yonekura Y, Tanao A et al: Isolated heterotopic pancreas causing intussusception. *Eur J Pediatr Surg*, 2000; 10: 197–200
16. Abel R, Keen CE, Bingham JB et al: Heterotopic pancreas as lead point in intussusception: new variant of vitello intestinal tract malformation. *Pediatr Dev Pathol*, 1999; 2: 367–70
17. Carleton CL, Ackerbaum R: Intussusception secondary to aberrant pancreas in a child. *JAMA*, 1976; 236: 1047
18. Erdener A, Avanoğlu A, Ozok G et al: Intussusception in an infant caused by aberrant pancreas. *J Pak Med Assoc*, 1993; 43: 22–23
19. Scholz S, Loff S, Wirth H: Double ileo-ileal intussusception caused by a giant polypoid mass of heterotopic pancreas in a child. *Eur J Pediatr*, 2000; 159: 861–62
20. Trifan A, Tărcoveanu E, Danciu M et al: Gastric heterotopic pancreas: an unusual case and review of the literature. *Gastrointest Liver Dis*, 2012; 21(2): 209–12