Ileo-ileal Intussusception Caused by Pancreas Heterotopia

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ABSTRACT

Adult intussusception occurs rarely and is quite different from pediatric occurrence in both etiology and treatment. It usually has a leading point which could be either a neoplasm or not. This report presents the diagnosis and management of one patient, a 51-year-old man with symptomatic ileo-ileal intussusception caused by pancreas heterotopia. Pancreas heterotopia is unusual and can be a leading point of small-bowel intussusceptions.

Keywords: Pancreas heterotopias, heterotopic pancreas, ectopic pancreas, ileo-ileal intussusception

Siriraj Med J 2012;64:101-104
E-journal: http://www.sirirajmedj.com

INTRODUCTION

Intussusception is defined as invagination of one segment of the gastrointestinal tract into a subjacent one. Prevalence in adults is rare, estimated at only 5% of all intussusceptions.[1] About 90% of intussusceptions in adults are caused by a definite underlying disorder.[2] Most small-bowel intussusceptions are secondary to benign lesions, with malignant lesions in about 15% of cases.[3] Benign causes include neoplasms such as gastrointestinal stromal tumors (GISTs), nonneoplastic polyps, congenital lesions such as Meckel's diverticulum and intestinal duplication, inflammatory lesions, and submucosal hematoma from trauma.[4] Pancreas heterotopias or heterotopic pancreas (HP) or ectopic pancreas is unusual, but not rare, which occurs predominantly in the stomach, duodenum and proximal jejunum. Lesions sited more distally in the distal jejunum and ileum are more rare and generally asymptomatic. Intussusception caused by HP is exceptionally rare. From the authors' best knowledge, there were only a few case reports about isolated HP causing ileo-ileal intussusception in adults.

We report an adult case of intermittent colicky pain caused by ileo-ileal intussusception secondary to isolated HP.

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Received 23 November 2011
Revised 13 February 2012
Accepted 13 February 2012

CASE REPORT

A 51-year-old man went to a general hospital due to colicky epigastric pain for eight weeks. He underwent ultrasonography, which revealed right renal calculus. The colicky pain did not disappear after he took medication for treatment of renal calculus. He also had of hematochezia four times over two days, but he never had fainting or syncope. He never had nausea, vomiting or bowel habit change. He was admitted in the general hospital. Esophagogastro-duodenoscopy (EGD) and colonoscopy were done and were unremarkable. He further underwent CT abdomen and GI follow through (GIFT). The retrospective review of these studies, showed there was an intraluminal filling defect at the distal small bowel, sized about 3 cm (Fig 1, 2). The diagnosis was a large polypoid tumor. The day of admission at Siriraj Hospital (about 1 week after doing CT of the whole abdomen), clinical examination showed an intermittent, movable, mildly painful, firm consistency mass sized about 5 cm at the right side of the abdomen.

Initial blood tests including complete blood count (CBC), biochemistry and coagulation showed unremarkable results. Emergency abdominal ultrasonography was performed to exclude intussusceptions. The ultrasound examination revealed invagination of bowel loops, which contained concentric rings of high and low echogenicity, with doughnut sign and hay-fork sign appeared at the right abdomen, compatible with intussusceptions (Fig 1, 2 and 3a-3c). When compared with the prior CT abdomen, this suspected ileo-ileal type. A round-shaped, homogeneous hyperechoic mass was also seen at the tip of the intussusceptum, sized 1.2x1.2x1.3 cm, which represented the leading point. The diagnosis at that time was intussusceptions with leading

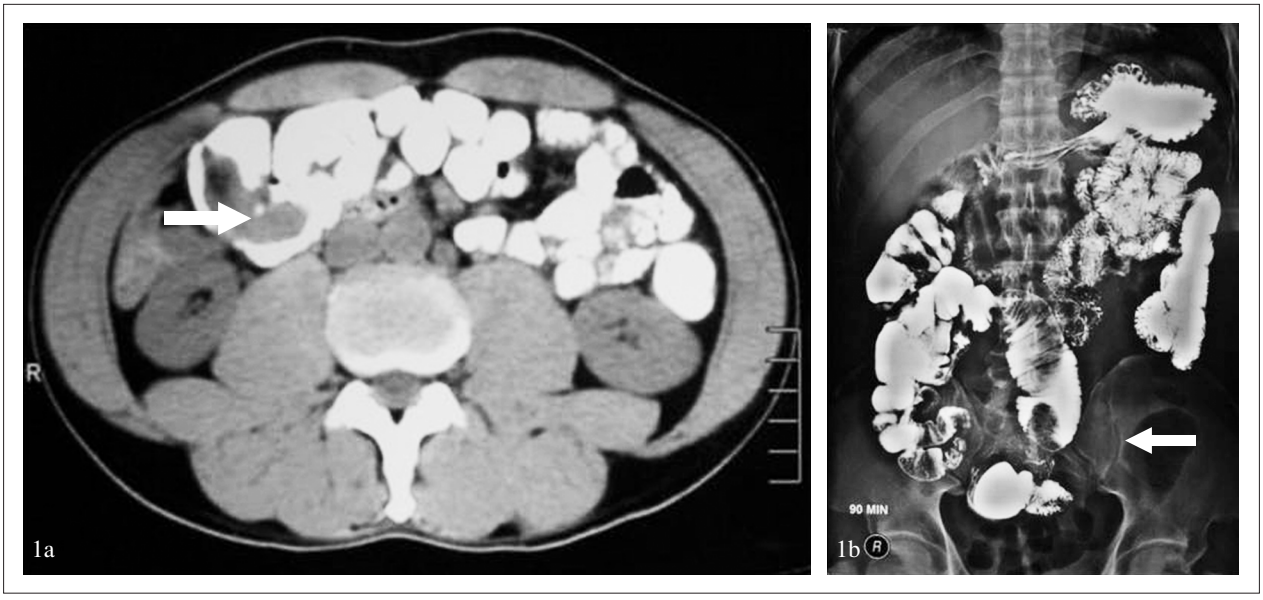

Fig 1a and 1b.

Fig 1a. Non-enhanced CT scan (from outside hospital) showed an intraluminal filling defect at the small bowel, sized about 3 cm. (arrow)

Fig 1b. GI follow through (GIFT) at 90 min delayed image (from outside hospital) revealed a suspected radiolucent filling defect with segmental dilatation at distal ileum. (arrow)

point. On the basis of this, the patient was referred for a laparotomic operation.

At laparotomy, a segment of the ileo-ileal intussusceptions was noted about 10 cm long. Then, the segmental ileum was resected. The intussusceptum was excised and the gross specimen showed an intramural mass, measuring 2x1.2x1 cm, with edematous hyperemic covering mucosa. A section through the polypoid lesion showed an intramural, yellowish, soft nodule (Fig 3b).

Histological examination revealed shallow ulcer and mild inflammation. Ectopic pancreatic tissue was observed in the muscular wall (Fig 3c and 3d).

DISCUSSION

Adult intussusception is quite unusual compared with pediatric occurrence, with a prevalence of only about 5% of all intussusceptions.[1] Diagnostic methods are focused on diagnosis and treatment planning. Intussusceptions that have leading points need definite treatment or surgery.

Intussusception is also an emergency condition in which diagnosis can be confirmed by ultrasonography. This modality is a non invasive technique and can be a portable method. Doughnut sign, and hay-fork sign or sandwich sign are ultrasonographic findings which are highly suggestive of intussusceptions. Ultrasound also demonstrates the leading point, but has a limitation to characterize the nature of the leading point.

Most small-bowel intussusceptions have leading points and most are benign conditions.[3] Heterotopic pancreas (HP) is a congenital disorder in which benign pancreatic tissue presents outside the pancreatic frame. It has been suggested that HP results from the separation of pancreatic tissue during the embryonic rotation of the dorsal and ventral buds.[5,6] It usually remains asymptomatic. Although, it is unusual and asymptomatic, there are many reports about

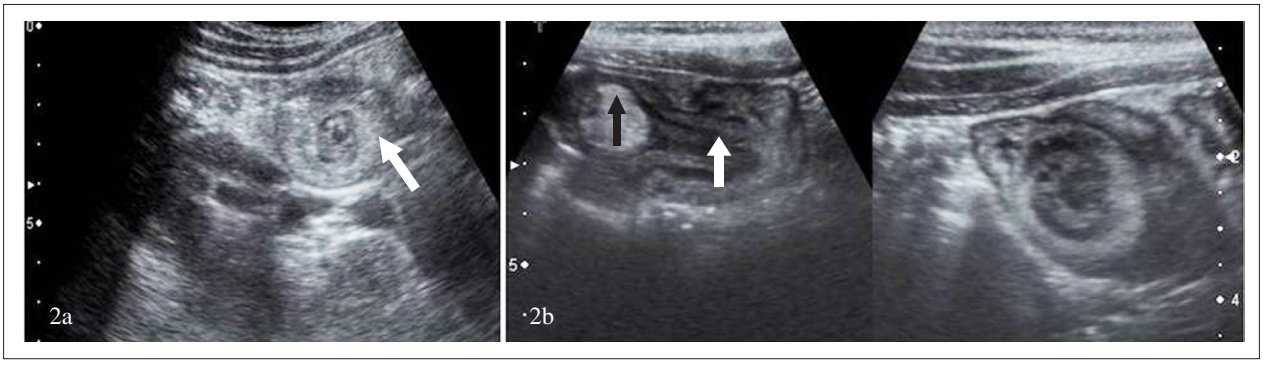

Fig 2a and 2b.

Fig 2a and 2b. Gray-scale ultrasound showed invagination of bowel loops, which contained concentric rings of high and low echogenicity, with doughnut sign (2-a) and hayfork sign (2-b) (white arrow) at the right-sided abdomen, compatible with intussusceptions. A round-shaped, homogeneous hyperechoic mass of the leading point was seen at the tip of the intussusceptum (black arrow).

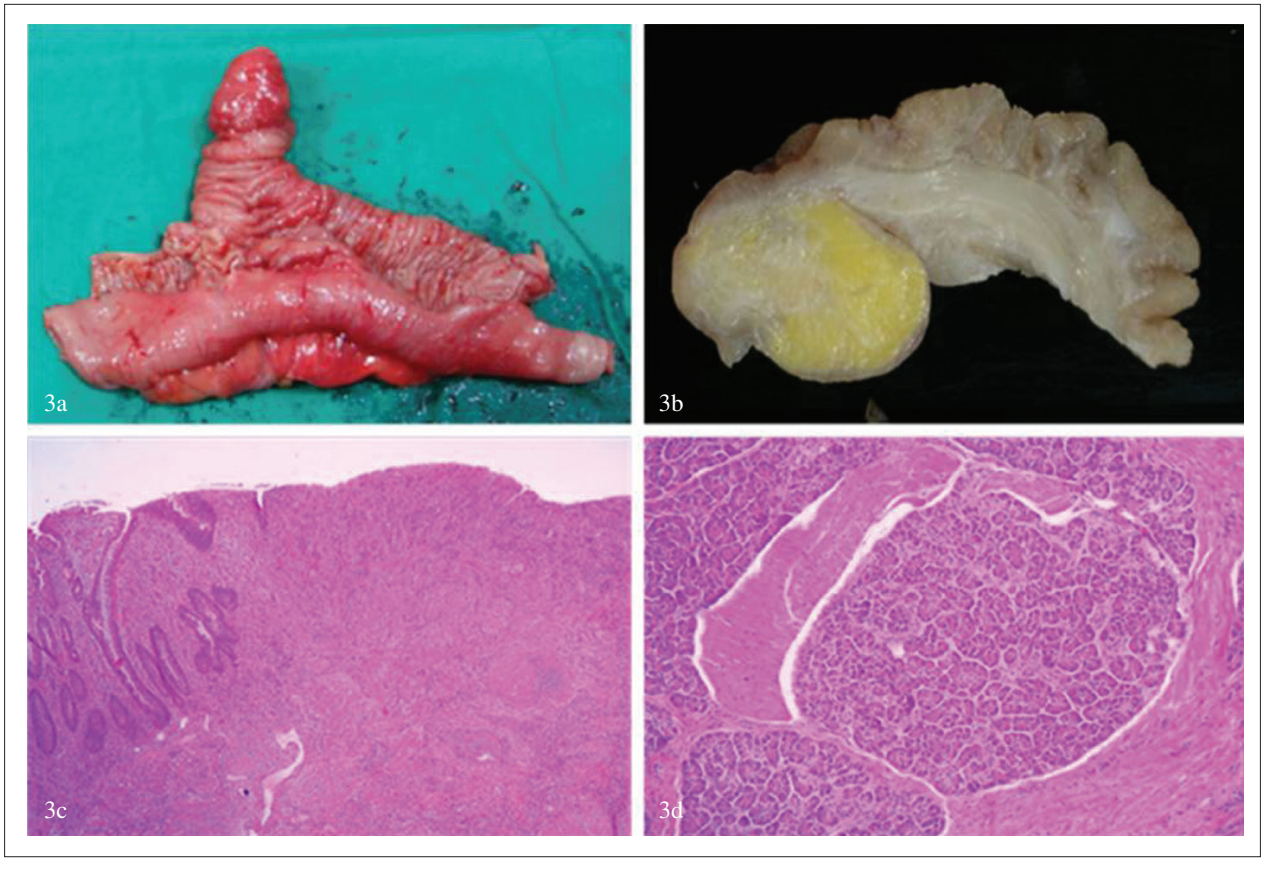

Fig 3a, b, c and d. Left upper, right upper, left lower and right lower images, respectively.

Fig 3a. Gross specimen of the intussusceptum showed an intramural mass, measuring 2x1.2x1 cm, with edematous hyperemic covering mucosa.

Fig 3b. Gross specimen. Cut section at the leading point showed a yellowish intramural nodule (arrow)

Fig 3c and 3d. Histopathology revealed shallow ulcer with mild inflammation and pancreatic tissue in muscular wall, (H&E, magnification 40x).

intussusceptions caused by HP. Most of them were found in pediatric groups, but adult HP is rarely reported.[7-14] Moreover, the lesions located further in the ileum are rarer. From our best knowledge from case reports and literature review, there are only few HP cases with ileo-ileal intussusceptions caused by HP. Ganapathi S et al[7], reported a 26-year-old male case with ileo-ileal intussusception, caused by ectopic pancreas, and hemoperitoneum from a ruptured congenital band attached to the ileal mesentery. Huang WS et al,[10] reported a 25-year-old woman with ileal intussusception and recurrent gastrointestinal bleeding, from an ileal aberrant pancreas. The patient was symptom-free after surgery. Moen J and Mack E[11] reported a case of ileo-ileal intussusception caused by an aberrant pancreatic submucosal mass that remained undetected during several diagnostic tests and two laparotomies. Gurbulak B et al,[13] reported a 22-year-old gravid woman at 31 weeks of gestation, who suffered from ileo-ileal intussusception with heterotopic pancreas as a leading point. After surgery, the patient and her infant made an uneventful recovery. Chuang MT et al,[14] reported a 26-year-old woman with symptoms of bowel obstruction. Her CT scan showed ileoileal intussusception due to a fatty nodule, but the pathology showed the leading point to be ectopic pancreas with abundant fatty infiltration. The successful management of adult intussusceptions will invariably involve the lead-point and segmental intestinal resection. Even though, higher rates of malignancy occur in large bowel intussusceptions, this possibility should also be kept in mind in cases of small bowel intussusceptions despite their lower incidence.

CONCLUSION

Intussusception is an emergency condition and ultrasound is a useful method to give the accurate diagnosis of intussusceptions, which can lead to proper management. This report has also presented the unusual case of ileo-ileal intussuception in an adult in which the leading point is from pancreas heterotopias.

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