JOP. J Pancreas (Online) 2007; 8(5):584-587.

CASE REPORT

Heterotopic Pancreas as a Leading Point for Small-Bowel Intussusception in a Pregnant Woman

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ABSTRACT

Context Adult intussusception occurs infrequently and differs from childhood intussusception in its presentation, etiology and treatment. Diagnosis can be delayed because of its longstanding, intermittent, and non-specific symptoms, and most cases are diagnosed at emergency laparotomy.

Case report We present the diagnosis and management of our patient, a pregnant woman, who had adult intussusception due to a heterotopic pancreas .

Conclusion Although relatively rare, intussusception should be included in the differential diagnosis of small bowel obstruction.

INTRODUCTION

Although intussusception is the most common cause of small bowel obstruction in the pediatric population, it is rare in adults [1, 2]. Adult intussusception occurs infrequently and differs from childhood intussusception in its presentation, etiology and treatment. Diagnosis can be delayed because of its longstanding, intermittent and non-specific symptoms, and most cases are diagnosed at emergency laparotomy [2, 3]. In contrast to intussusceptions in children, a demonstrable etiology is found in 70-90% of cases in the adult population. The cause of intussusceptions in adults varies by location. Large bowel lead points are more frequently malignant than small bowel lead points. The small bowel leading points are hamartomas, lipomas, leiomyomas, neurofibromas, adenomas, inflammatory polyps, Peutz-Jeghers syndrome, Meckel diverticulum, parasitic infestation and adhesions [1, 2, 3].
We present the diagnosis and management of a pregnant woman who had adult intussusception due to a heterotopic pancreas.

CASE REPORT

A 22-year-old gravid woman at 31 weeks of gestation, presented with severe abdominal pain and bile-stained vomiting of a 4 day duration. On palpation, there was a distended abdomen, diffuse abdominal tenderness, peritoneal irritation and a decrease of bowel sounds. No masses were palpable. There was no significant medical history. Her body temperature was 37.0°C. Laboratory findings showed a white blood cell count of 17,800 mm^{-3} (reference range: 4,000-10,000 mm^{-3}), hemoglobin 12.3 g/dL (reference range: 11.5-14.5 g/dL), and platelets 200,000 mm^{-3} (reference range: 6,000-206,000 mm^{-3}). All other studies, including electrolytes and urinalysis, were within reference limits. Abdominal radiography showed air fluid levels indicative of a small-bowel obstruction. Computed tomography of the abdomen

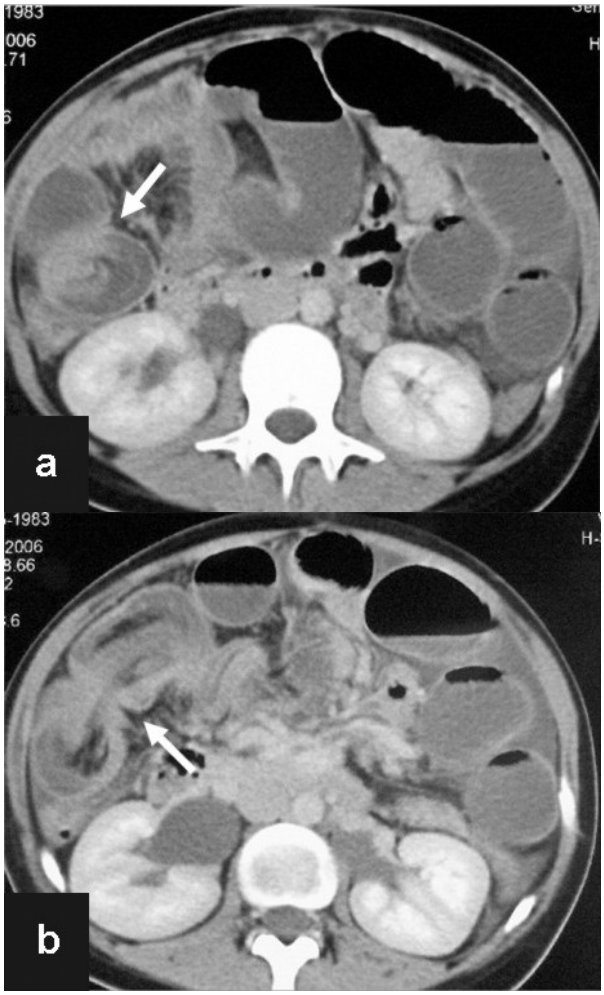

Figure 1. a. Target sign (arrow) in cross-section. **b.** Dilated proximal bowel and target sign (arrow).

revealed 'target lesions' suggestive of small bowel intussusception and free-fluid in the abdominal cavity (Figure 1).

After appropriate resuscitation, a laparotomy was performed using a midline incision, and a cesarean section was carried out immediately. At laparotomy, a ileo-ileal intussusception was noted (Figure 2). The invaginated segment was situated approximately 80 cm from the ileo-ceacal valve and a co-existing Meckel diverticulum was found. However, a Meckel diverticu lum was not found in the necrotic ileal loop. The ileal segment involved was resected and an end-to-end anastomosis was performed. An enterotomy confirmed the presence of a polypoid and non-stalked lesion. The patient and her infant made an uneventful recovery. The microscopic examination of the mass lesion revealed that the mass was composed o f mature pancreatic acini and ducts. The overlying intestinal mucosa was inflamed and atrophic. There were a few ducts draining into the intestinal lumen (Figure 3).

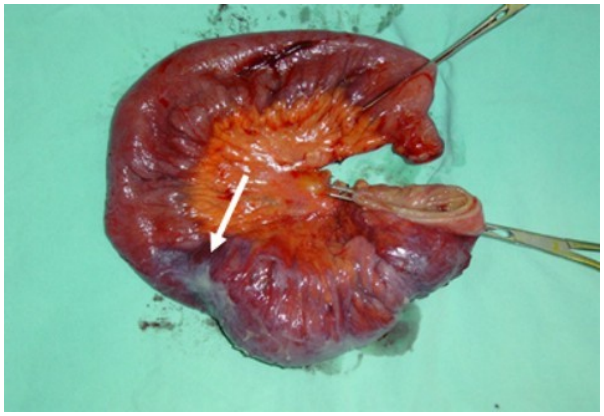

Figure 2. The resected necrotic specimen of the intussusception. The white arrow shows the invagination point.

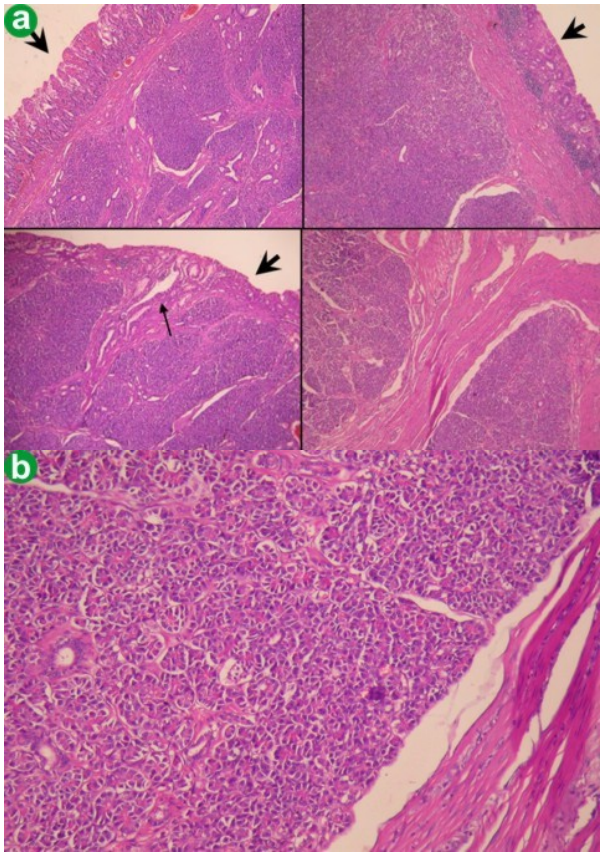

Figure 3. a. Mature exocrine pancreatic tissue within the intestinal wall with atrophic overlying mucosa (arrowheads) and a duct draining into the intestinal lumen (arrow) (H&E, original magnification 40x). **b.** Mature pancreatic acini and ducts (H&E, original magnification 200x).

DISCUSSION

Intussusception is primarily a disease of children with only about 5% of cases

occurring in adults [1, 2]. Diagnosis can be delayed because of its longstanding, intermittent and non-specific symptoms, and most cases are diagnosed at emergency laparotomy. The most useful investigation is abdominal computed tomography (CT). CT revealed a pseudokidney sign and a target sign, with edema of Kerckring's folds in the intussusceptum of the small bowel. The nature of the lead of the invagination varies greatly and includes polyps, hamartomas, lipomas, leiomyomas, neurofibromas, adenomas, inflammatory polyps, tuberculosis, Meckel diverticulum and adhesions [1, 2, 3]. A very rare cause of adult intussusception is heterotopic pancreas [4, 5].

Heterotopic pancreas is a congenital disorder which consists of the presence of normal pancreatic tissue located outside the pancreatic frame. It has been suggested that heterotopic pancreas results from the separation of pancreatic tissue during the embryonic rotation of the dorsal and ventral buds [6, 7]. It usually remains asymptomatic throughout life and is found incidentally during an upper endoscopy or other radiological imaging modalities such as laparotomy. The most common sites of heterotopic pancreata are the stomach, duodenum, jejunum, Meckel diverticulum and ileum. Less common sites include the esophagus, lungs, gallbladder, spleen umbilicus, fallopian tube, lymph nodes, mediastinum, tongue and submandibular salivary gland. Pancreatic abscess, gastric outlet, intussusception and the malignant transformation of heterotopic pancreas have been reported [6, 7, 8].

Intestinal obstruction is uncommon in pregnancy, occurring one in every 2,500-3,500 deliveries [9]. Intestinal obstruction most frequently results from intestinal adhesion and gastrointestinal volvulus. There is a rare report regarding the relationship of the enlargement of a heterotopic pancreas with the hormonal changes in gestation [8]. Ikematsu *et al*. [8] described gastric outlet obstruction caused by a heterotopic pancreas in a pregnant woman. Our patient is the first report of a pregnant woman who suffered from intussusception caused by heterotopic pancreatic tissue in the ileum.

CONCLUSION

CT proves to be the most effective preoperative diagnostic method; however, there is almost always a pathological reason for adult invagination which can often only be clarified intraoperatively, making surgery unavoidable in most cases. Because intussusception is often initially mis-diagnosed in the adult population, it should be kept in mind that small bowel obstruction in adults may be caused by intussusception.

Received June 23rd, 2007 - Accepted July 5th, 2007

Keywords Intussusception; Pancreas; Pregnancy

Conflict of interest The authors have no potential conflicts of interest

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Document URL: http://www.joplink.net/prev/200709/08.html

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