

Heterotopic pancreas presenting as ileo-ileal intussusceptions- A rare case review



Medical Science

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ABSTRACT

BACKGROUND:

A heterotopic pancreas as the lead point of ileo-ileal intussusception is extremely rare. A heterotopic pancreas (HP), a developmental anomaly, is defined as pancreatic tissue found on ectopic sites without contiguity with the main pancreas.

CASE REPORT:

A 17-year-old previously healthy boy, presented to the emergency room with the complaint of severe abdominal pain for the last 6-8 hours. A preoperative diagnosis of ileo-ileal intussusception was made on ultrasound and an emergency exploratory laparotomy was done. At laparotomy an ileo-ileal intussusception was found and a polyp noted as a lead point. On histopathology this polyp was found to be heterotopic pancreas.

CONCLUSION:

HP remains a rare cause of small bowel obstruction. Its management remains no different to that of intussusception from any cause and the possibility of malignant disease should always be remembered when planning surgery.

INTRODUCTION

Intussusception is a condition characterized by the telescoping of a segment of intestine into the other, most commonly the proximal into the distal. It clinically presents as features of small bowel obstruction such as abdominal pain, constipation, vomiting and abdominal distention, in varying proportions. Intussusception is a type of closed loop obstruction, which can lead to gangrene. A heterotopic pancreas (HP), a developmental anomaly, is defined as pancreatic tissue found on ectopic sites without contiguity with the main pancreas. The presence of heterotopic pancreas is unusual with an estimated incidence of 0.2% of upper abdominal operations. HP occurs predominantly in the stomach, duodenum and proximal jejunum. A heterotopic pancreas of the ileum is rare and usually found in a Meckel's diverticulum, which may cause intussusception in childhood. An isolated heterotopic pancreas as the lead point of intussusception is extremely rare especially in adults. Even after extensive literature search we could retrieve very few cases of isolated heterotopic pancreas as the lead point of intussusception. Here, we have described a rare case of small ileal ectopic pancreas that led to ileoileal intussusception and ileus.

Case report

A 17 year old male was admitted with history of abdominal pain, bilious vomiting for 3-4 days. The patient was resuscitated haemodynamically.

Ultra sonography abdomen and erect abdomen x-ray revealed findings suggestive of ileoileal intussusception.

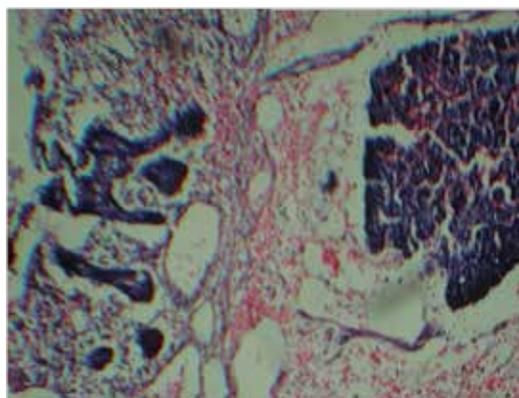
Emergency exploratory laparotomy was performed. Peroperative findings were ileoileal intussusception 20-25 cm proximal to ileocaecal junction. Gangrenous segment of around 5 cm of the ileum was resected and primary anastomosis done. Resected segment was sent for histopathological examination. Patient was discharged on 7th postoperative day and was followed uneventfully.

Intra Op Photograph of intussusception



Histological examination of the polyp revealed thick irregular bundles of displaced smooth muscle tissue within which were entwined ductular mucinous and pancreatic exocrine acinar tissue. The histological profile was typical of pancreatic heterotopia.

Microscopic photograph of Heterotopic Pancreas Tissue



DISCUSSION

Intussusception is primarily a disease of children with only about 5% of cases occurring in adults [1]. An underlying pathological process is usually identifiable in over 90% of cases in adults (compared to an idiopathic cause in the majority of paediatric cases) [2]. Felix *et al.* has published the most comprehensive review on intussusception, with more than 1,200 cases from 11 series [3]. Fifty-five percent of cases occur within the small bowel with 45% in the large bowel. Malignant tumours produced nearly half the cases of colonic intussusception. In contrast, predominantly benign tumours caused small bowel intussusceptions.

The causes of intussusception are summarised in Table.

Table 1. Causes of (small and large bowel) intussusception in adults [7]

Cause	Colonic intussusception	Small bowel intussusception
Malignant tumours (eg. carcinosarcoma, adenocarcinoma, lymphoma)	48%	17%
Benign tumours (eg. leiomyomas, pancreatic heterotopia)	21%	40%
Other causes (eg. idiopathic, post-operative)	31%	43%

Others have reported similar statistics although Agha reported nearly 60% of malignant small bowel tumours as a cause of intussusception in a small cohort of 25 patients [4]. Postoperative intussusception in adults is also usually rare, albeit that there are several anecdotal case reports. Other reported causes of adult intussusception include Meckel's diverticulum, coeliac disease, lymphoma, adenocarcinoma, carcinoid and idiopathic [3, 5, 6].

Klob first described the histological appearance of a heterotopic pancreas in 1859 [7]. The estimated occurrence of heterotopic pancreas is one per 500 upper abdominal operations and up to 5% of autopsy cases [8, 9]. The embryological derivation of heterotopic pancreas is well described [10]. The pancreas is formed from several primitive endodermal evaginations of the primitive duodenal wall. The dorsal diverticulum becomes the body and tail and the ventral portion the head of the pancreas. If one or more of these evaginations remain within the wall of the bowel then these can be carried as longitudinal growth of the intestine continues, leading to ectopic tissue anywhere from the stomach (most common) to the jejunum or ileum (least common). In the

latter, HP is usually associated with a Meckel's diverticulum.

Although there are conflicting reports in the literature, approximately half of cases of small bowel HP are asymptomatic [8]. Intussusception caused by HP is rare but has been described previously [8, 11, 12]. Most adult series that have described this complication have noted HP to be located within the ileum where the concomitant existence of a Meckel's diverticulum is thought to exacerbate the ability of the HP tissue as a lead point for the intussusception.

In contrast to this, our report of a ileoileal intussusception by HP illustrates an exceptionally rare cause of small bowel obstruction in the adult. As in this case, it appears that only lesions greater than 15 mm become symptomatic [8, 13].

In adults, successful management of intussusceptions from any cause will invariably involve resection of the lead-point tissue and at times, segmental resection of the involved intestine (as in this case). Surgical resection is often recommended for large bowel intussusceptions in view of the higher rates of malignancy. This possibility should also be borne in mind in cases of small bowel intussusceptions despite a reported lower incidence.

In summary, HP remains a rare cause of small bowel obstruction. Its management remains no different to that of intussusception from any cause and the possibility of malignant disease should always be remembered when planning surgery.

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