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# Case Report

## Heterotopic Pancreas- an Unusual Cause of Ileoileal Intussusception: a Case Report

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## ABSTRACT

**Heterotopic pancreas is an uncommon developmental anomaly of upper gastrointestinal tract. Heterotopic pancreas tissue is very rarely found in ileum. Intussusception in children is usually idiopathic, but definitive aetiology can be established in 90% of adult cases. We are reporting a case of pancreatic heterotopia presenting as a lead point of ileo-ileal intussusception in a 1year 3month year old boy.**

***Keywords:*** **Heterotopic Tissue, Pancreas, Ileum, Intussusception**

## Introduction

Pancreatic heterotopia is defined as presence of pancreatic tissue at ectopic location which lacks anatomical and vascular continuity with main pancreas (1). Presence of heterotopic pancreas has been documented in 0.2% of all upper abdominal surgeries (2). Intussusception is an invagination of a part of intestine into the lumen of adjacent gut. "It is one of the major causes of intestinal obstruction during infancy and childhood" (3). Intussusception caused by heterotopic pancreatic tissue is exceedingly rare (2, 3). We are presenting a case of ileo-ileal intussusception caused by heterotopic pancreatic tissue in a young child.

## Case Report

A 15 month old boy was admitted in emergency surgery ward in our hospital with complaints of severe colicky abdominal pain and repeated bilious vomiting for 24 hours. He also passed blood mixed mucous during trying of defecation for twice. He was partially treated by paediatrician at local hospital but pain was not relieved by usual dose of anti-spasmodic and analgesic medications. On clinical examination, his vitals were stable and abdomen was flat, tender with evidence of hyper peristalsis. A firm, tender and sausage shaped mass was palpable at right

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paraumbilical region. Among the routine blood examinations, he had neutrophilic leukocytosis (total leukocyte count 14,700/mm$^3$ with neutrophil count 79%). Routine blood biochemistry and serum electrolytes were within normal limit. Clinical history and examination findings were suggestive of intussusception.

On transabdominal sonography, an abnormal loop of bowel was found at right paraumbilical region and extension into infra-umbilical area with marked probe tenderness. On the basis of sonographic evidence of acute bowel intussusception, emergency exploratory laparotomy was performed with lower midline incision. An area of ileo-ileal intussusception was noted one meter proximal to ileo-coecal junction. Wall of the gut showed patchy areas of brownish black discolouration. A 10cm segment of small gut including ileo-ileal intussusception was resected to preventpossibility of wet gangrene. End to end primary ileo-ilealanastomosis was performed. The child responded well to post-operative supportive management. Specimen of resected gut was sent for histopathological examination.

In the histopathology laboratory, we received two small bowel segments measuring 5×2×2cm and 4×2×2cm respectively. Both the specimens were brownish black in colour. A solid, whitish polypoid mass measuring 10x6x5mm was found at lead point of intussusceptions (Fig.1, 2). On microscopy, mucosal surface of the gut was necrosed and ulcerated (Fig.3). There was sub-mucosal vascular congestion and dense neutrophilic infiltration in all layers of gut (Fig.3). Sections from the polypoid area showed lobulated structures comprised of acinar glands, ductules and islet cells in the muscular layer of intestine (Fig. 3, 4, 5) similar to the histology of normal pancreatic tissue. These structures were interpreted as heterotopic pancreatic tissue. Final histopathological diagnosis of heterotopic pancreatic tissue leading to intussusception was reported.

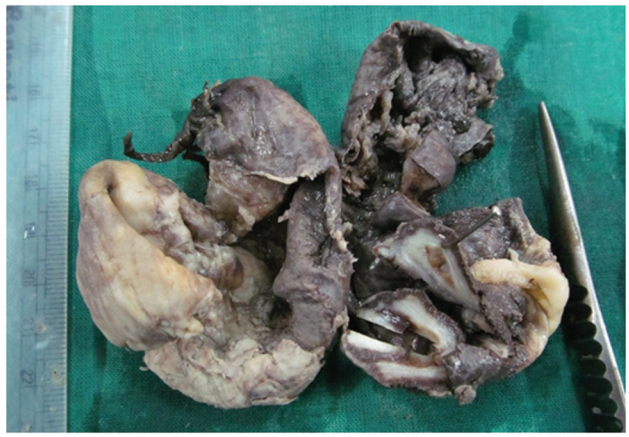

**Fig. 1:** Gross picture of the gut portions show brown black discolouration of necrosed areas and cut open grey-white nodular mass with a central duct **(**Original**).**

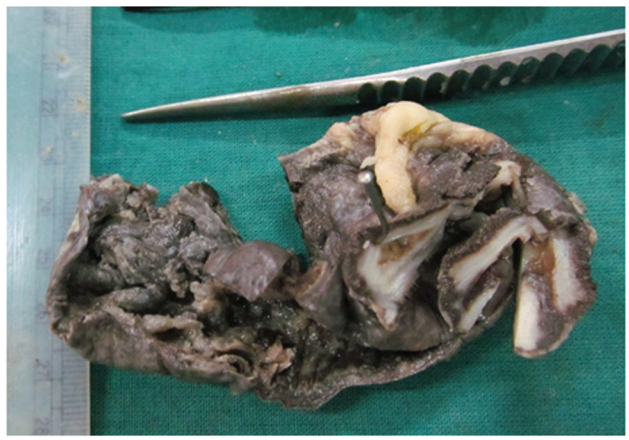

**Fig. 2:** Gross picture of the gut loop with cut open grey-white nodule as a lead point of intussusception (Original).

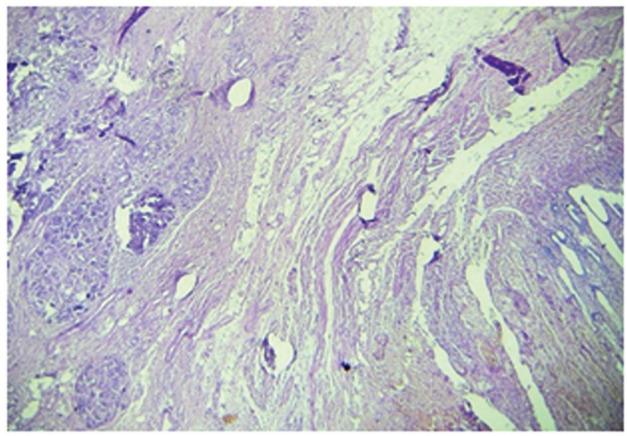

**Fig. 3:** Microscopy shows necrosis and congestion of ileal epithelium and sub-epithelial tissue with lobular mass like structures at muscularis propia(H & E stain, ×40).

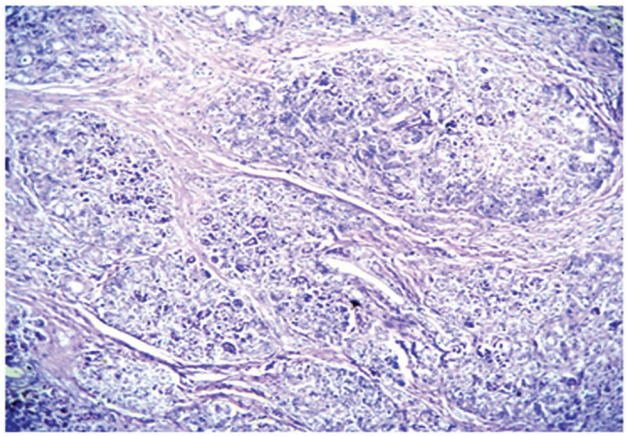

**Fig. 4:**Microscopy shows acini, islet cells and ductules arranged in lobular architecture similar to normal pancreatic tissue (H & E stain, ×100).

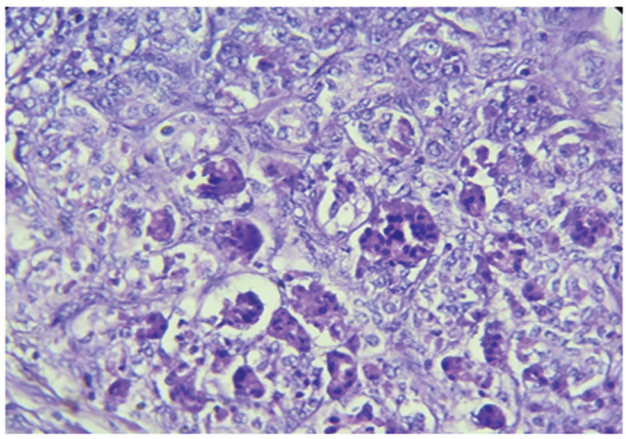

**Fig. 5:**Microscopy shows different components of the tissue-acini and islet cells of heterotopic pancreatic tissue (H & E stain, ×400).

## Discussion

Intussusception is a surgical emergency, commonly occurring in paediatric age group. Majority of the paediatric intussusceptions are idiopathic (2). In contrast, 90% of adult intussusceptions have definitive causes. Heterotopic pancreatic tissue is a rare congenital anomaly. Persistence of primitive endodermal evagination within the gut wall during the rotation of dorsal and ventral pancreatic buds can lead to heterotopic location of pancreatic tissue (4- 6). According to descending order of incidence, heterotopic pancreatic tissues are commonly found in duodenum, stomach and jejunum (4, 7). Ileum is an uncommon site for pancreatic heterotopia and Meckel's diverticulum is the commonest location for ileal pancreatic heterotropia (2, 3, 6). After the first case reported by Jean Schultz in 1729, pancreatic heterotopia was studied by many authors (4, 7). Barbosa *et al.* first used the term 'heterotopic pancreas' and their study of 41 cases included only a single case of intussusception in an infant. Reported incidence is 1 in 500 abdominal surgeries with male preponderance (3, 7). Singh *et al.* said that usually the heterotopic pancreatic tissue presents as a yellow nodule of 1-5mm with mucosal covering and exhibits a central hole which represents rudimentary pancreatic duct (4). In our case we have found a greyish white nodular mass of 10×6×5 mm in dimension and central hole has been seen (Fig.1). Bromberg *et al.*(5) observed that heterotopic pancreatic tissue is more frequently found in sub mucosal and muscularis propia layer but rarely in subserosal and serosal layer. In our case we have found heterotopic pancreatic tissue in muscularis propia layer which is similar to previous researchers. Most of the pancreatic heterotopias are asymptomatic and incidentally diagnosed during unrelated surgical procedures or autopsy (2- 4). Many authors suggest that closer proximity to mucosa and larger lesions (greater than 15mm) are related to symptomatic presentation (6, 7). Chronic vague abdominal pain due to bowl dysmotility, inflammation, haemorrhage and rarely obstruction might be the clinical presentations (7). Only occasional cases of ileal pancreatic heterotopia as a lead point of ileo-ileal intussusceptions have been reported (8, 9). Heinrich classified heterotopic pancreas into three types: type 1 (acini, islets and ducts), type 2 (acini, ducts but no islets) and type 3 (ducts alone)(10). According to Heinrich's classification, our case is type1 pancreatic heterotopia. Diagnosis of pancreatic heterotopia is quite difficult, especially in asymptomatic cas-

es. Ultrasound (USG) and Computerised Tomographic (CT) scans are indicated for evaluation of the nature of intestinal obstruction and formulation of management protocol. But USG, CT scans or even endoscopy have no specific role in detection of heterotopic pancreatic tissue (11). Almost all cases are diagnosed on histopathological examination (12). In our case also, clinical and radiological features failed to reveal the presence of heterotopic pancreatic tissue, similar to previous authors. Surgical resection of involved portion of gut with end to end anastomosis is the definitive management for ileo-ileal intussusceptions to prevent further occurrence, particularly when there is any lead point or gut is devitalised at the time of laparotomy (7, 11).

Though rare, pancreatic heterotopia can be an important cause of ileo-ileal intussusception. Emergency laparotomy and excision of the involved bowel is necessary to prevent recurrence. Histopathological examination of resected specimen of gutis always mandatory for establishment of diagnosis of pancreatic heterotopia as a possible cause and should be encouraged in all cases to pinpoint underlying pathology. We want to emphasize the inclusion of pancreatic heterotopia as a rare but potential cause of paediatric ileo-ileal intussusception.

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