E-ISSN: 2616-3470
P-ISSN: 2616-3462

www.surgeryscience.com
2022; 6(4): 18-21
Received: 21-07-2022
Accepted: 25-08-2022

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Heterotopia of pancreas leading to ileo-ileal intussusception in an adult: A case report

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DOI: https://doi.org/10.33545/surgery.2022.v6.i4a.945

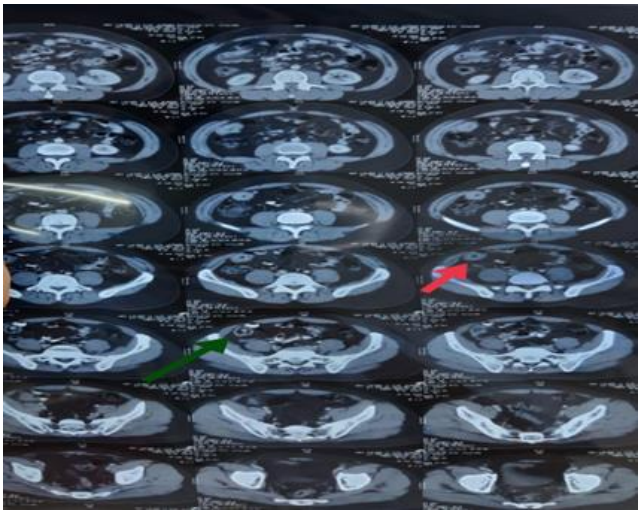
Abstract
A heterotopic pancreas as the lead point of ileo-ileal intussusception is extremely rare. A 26year old male patient previously healthy male, presented to the emergency room with the complaint of pain in the lower abdomen since 2 weeks, colicky type, progressive in nature, associated with melena for 5 days and non-bilious vomiting for 3 days. A preoperative diagnosis of ileo-ileal intussusception was made on ultrasound and cect abdomen and an emergency exploratory laparotomy was done. At laparotomy an ileo-ileal intussusception was found and a polyp noted in distal ileum as a lead point. On histopathology this polyp was found to be heterotopic pancreas.

Keywords: Heterotopic pancreas, lead point, intussusception

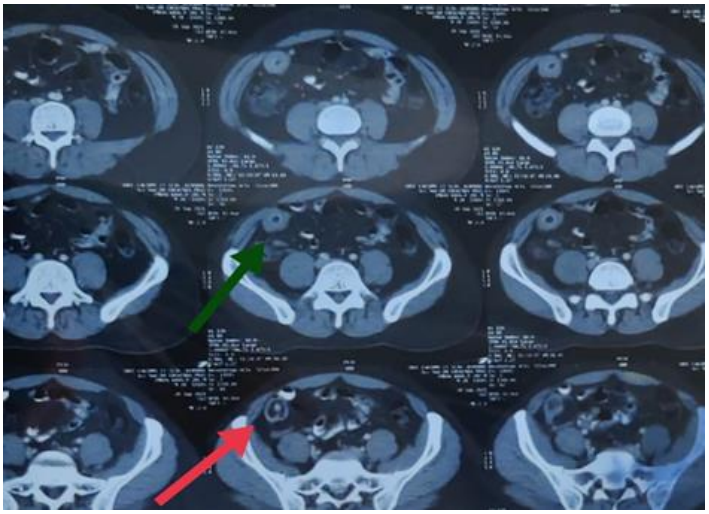
Introduction
Heterotopic Pancreas, is a developmental aberration described as pancreatic tissue present in ectopic sites that is not connected to the main Pancreas [1]. It is found in in 0.6-13% of autopsies [2] and as an incidental finding in about 1 of every 500 of upper abdominal operations [1]. A heterotopic pancreas is usually is rare entity in Ileum, if present it is seen in Meckel's Diverticulum. Intussusception is the frequent cause of intestinal obstruction in infancy. Accounts for only 2% of bowel obstruction in adult population. This is a case report of Intussusception in a 26year old caused by heterotopic pancreas in a Ileal polyp.

Case report
A 26year old male patient was admitted to hospital with history of pain in the lower abdomen since 2 weeks, colicky type, progressive in nature. Pain was relieved on taking antispasmodics and antacids. Pain is associated with melena for 5 days and non-bilious vomiting for 3 days. No Mass was palpable on abdominal examination, per rectal examination shown malena. Ultrasound abdomen showed bowel in bowel appearance in right lumbar region. CECT scan of abdomen and pelvis with oral contrast showed bowel in bowel appearance in Right Iliac Fossa and diagnosed with Ileo-Ileal Intussusception.
Patient underwent Diagnostic laparoscopy bowel in bowel appearance was noted in right iliac fossa. Laparoscopic reduction was attempted but not successful. So proceeded with exploratory laparotomy on mid-midline incision. The Intussusception was manually reduced, on examination a polyp was noted in distal ileum along with significant narrowing of bowel. So, resection of the segment of bowel along with polyp and end to end anastomosis was done. intraoperatively whole bowel examined no polyps noted, postoperatively ct shows no evidence of polyps.

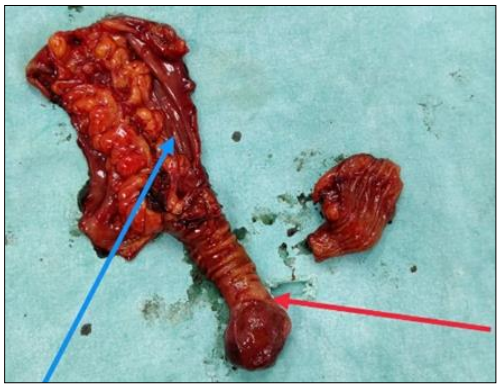
Specimen was sent to histopathology examination
Gross specimen shows pedunculated polyp in a segment of ileum of about 8 cm with a pedunculated poly of 4 cm in diameter. The polyp and stalk were congested and surrounding mucosa was edematous.
Post operatively patient did well and was discharged after seventh post operative day.
Histology of the resected specimen showed Pancreatic tissue in the Polyp.
Microscopically pancreatic tissue was seen in the polypoidal mass. Pancreatic acini along with pancreatic ducts were noted in the polyp.



Green arrow showing polyp in the ileum

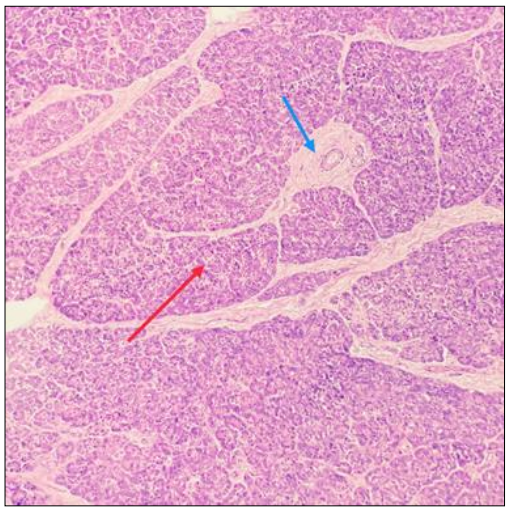


Red arrow showing target sign

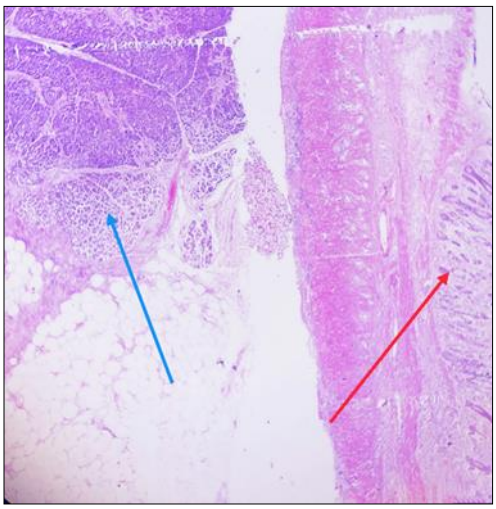


Blue arrow: Ileal segment. **Red arrow:** Ileal polyp

A) Gross specimen of resected Ileal segment along with ilea polyp.



b) Shows Blue arrow shows pancreatic duct and red arrow shows pancreatic acini.



c) Blue arrow shows pancreatic tissue and red arrow shows intestinal epithelium.

Discussion

Intussusception is the most common cause of intestinal obstruction in infancy and children under age of 5 years but it accounts for about 2% of bowel obstruction in adults. The cause of intussusception is idiopathic in children between age of 6 months to 24 months, one hypothesis suggests hypertrophy of Peyer's patches beyond the age group of 2 years we should consider the possibility of pathological Lead point (PLP). The incidence of pathological lead point increases with age. Pancreas heterotopica is an uncommon PLP. According to certain theories, the separation of pancreatic tissue during the embryonic rotation of the dorsal and ventral buds causes heterotopic pancreas. The oesophagus, lungs, gallbladder, spleen, umbilicus, fallopian tubes, lymphnodes, mediastinum, tongue, and submandibular salivary gland are among the less frequent HP locations.

Various pathological lesions have been identified as leading points in intussusception which include Meckel's diverticulum

(most common cause), intestinal polyps, duplication cysts, lipoma, leiomyoma, lymphoma and lymphosarcoma. Very rarely Heterotopic Pancreas acts as pathological lead point. It is highly uncommon to have an isolated heterotopic pancreas of the ileum without a Meckel's diverticulum causing intussusception, as was documented here.

Heterotopic or Ectopic pancreatic tissue is defined as pancreatic tissue that lacks physical connection to pancreas and has independent blood supply. It is present in 0.6-13% of autopsies and an incidental finding in abdominal operations. Ectopic pancreatic tissue is most commonly found in stomach, duodenum, proximal jejunum, in rare instances other sites like Meckel's diverticulum, ileum, colon, gallbladder, common bile duct, spleen. We searched the literature on Pubmed/google scholar for adult ileal intussusception with heterotopia of pancreas before our case and the results are summerised below.

Table 1: Adult Intestinal intussusceptions with heterotopia of pancreas

S. No.	Ref	Age in year	Gender	Location
1	Gurubulak *et al.* 2007 [7]	22	Female	Ileo-ileal intussusception
2	Tekin *et al.* 2008 male [11]	49	Male	Ileo-ileal intussusception
3	Ganapathi *et al.* 2010 [12]	26	Male	Ileo-ileal intussusception
4	Chuang *et al.* 2010 [9]	26	Female	Ileo-ileal intussusception
5	T Ruangchaijatuporn *et al.* 2012 [5]	51	Male	Ileo-ileal intussusception
6	S Musthafa *et al.* 2015 [6]	32	Male	Ileo-ileal intussusception
7	Flynn A *et al.* 2017 [4]	35	Male	Ileoileal intussusception
8	Xiang *et al.* 2019 (5 patients retrospective study) [3]	-	-	ileoileal intussusception
9	A.Sciannamea *et al.* 2020 [10]	33		
10	Linping Cao 2021 (3 patients retrospective study) [8]			Ileo-ileal intussusception.

Heterotopic Pancreas is classified into three types by Heinrich based on histology. TYPE 1-Heterotopic tissue consisting of all components of pancreatic tissue (acini, ducts and islet cells). TYPE 2-Heterotopic tissue consisting of ducts and acini only but no islet cells. TYPE 3- Heterotopic tissue consisting of ducts only. Later Gasper-Fuentes *et al.* modified the classification into four types. In our case it is TYPE 2 where pancreatic acini, ducts are present but no islets cells.

Majority of patients are asymptomatic, however heterotopic pancreas can cause symptoms based on its location, size and complications. Most common HP symptoms are abdominal pain, nausea, vomiting, melena, blood stool, and weight loss. The presenting complaints in our case were abdominal pain, vomiting, and Melena. Complications in intestine include Intussusception, intestinal obstruction, gastrointestinal bleeding and malignant transformation; malignant transformation is a rare entity.

There is no specific laboratory finding few authors believe that imaging is specific in diagnosing heterotopic pancreas and also to differentiate HP from other space occupying lesions. A small oval-shaped intramural mass with an unclear margin or differential lobe with enhancement characteristics of HP consistent with that of a normal pancreatic tissue. In our case we could only establish on Intussusception, polypoidal lesion was a intra op finding. The magnetic resonance (MR) imaging appearance of heterotopic pancreas mimics that of the orthotopic pancreas. The characteristic high signal intensity of the pancreas at T1-weighted MR imaging is particularly helpful for differentiating heterotopic pancreas from other lesions. Endoscopic ultrasonography and endoscopic biopsy are also helpful in diagnosis of heterotopic pancreas. However, post-operative pathological findings are ultimately used to diagnose heterotopic pancreas.

Surgical excision is the only curative treatment for HP-related intussusception and gastrointestinal bleeding, and it has opportunity for histological diagnosis of potential malignancy.

Conclusion

Intussusception in patients beyond the age of 2 years one should consider a possibility of pathological lead point. Ileo-ileal intussusception due to heterotopic pancreas in a polypoidal mass as pathological lead point is relatively rare entity. In adult cases of intussusception with a history of gastrointestinal bleeding and melena, the possibility of heterotopic pancreas should be considered as a differential diagnosis. CT examination with oral contrast could provide useful preoperative information. Surgical excision is the treatment of choice.

Conflict of Interest

Not available

Financial Support

Not available

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How to Cite This Article
Patel AS, Suneel AT, Singh J, Chitravanshi S. Heterotopia of pancreas leading to ileo-ileal intussusception in an adult: A case report. National Journal of Clinical Orthopaedics. 2022;6(4):18-21.