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CASE STUDY

GASTRIC AND PANCREATIC HETEROTOPIA PRESENTING AS PATHOLOGICAL INTUSSUSCEPTION IN AN ADULT FEMALE

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Abstract: Gastric and pancreatic heterotopias are rare entities. They have been identified throughout the entire length of the gastrointestinal tract and in other structures, including Meckel's diverticulum and the Ampulla of Vater. However, very few cases of combined gastric and pancreatic heterotopias in ileum have been reported till date. Also, intussusceptions in adults are uncommon. Here we report a novel case of combined gastric and pancreatic heterotopia in ileum in a 32-year-old female. The patient presented with complaints of pain in abdomen and bleeding per rectum. On computed tomography, ileo-ileal intussusception was seen in the distal ileum. Exploratory laparotomy revealed an ileal polyp. The ileal segment with the polyp was resected and sent for histpathological examination. It was diagnosed as heterotopic gastric and pancreatic tissue in ileum. It highlights the importance of histopathological examination for definitive diagnosis of this rare clinical entity.

KEYWORDS: Gastric heterotopias, intussusception, pancreatic heterotopias, polyp

INTRODUCTION:

Heterotopias are defined as the presence of tissues outside their typical location without vascular or anatomical continuity with the organ proper. [1,2] The incidence of heterotopic tissue in the gastrointestinal tract varies from <1% to around 13%. [1,3] They have been identified throughout the entire length of the gastrointestinal tract and in other structures, including Meckel's diverticulum and the Ampulla of Vater.[2] However, combined gastric and pancreatic heterotopia is rare and very few cases have been reported in the ileum.[2]

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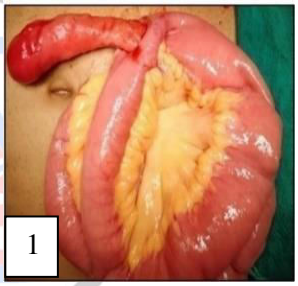
Clinically, patients may present with pain, anaemia, melena or weight loss in cases of pancreatic heterotopias when its size reaches 1 cm in the stomach or duodenum.[4] Gastric heterotopias often become clinically significant due to sequelae of acid secretion and/or polyp formation. The majority of patients with combined pancreatic and gastric heterotopias form polypoid or mass-like lesions in the gastrointestinal tract, causing clinical symptoms due to their physical location or mass effects.[3] These entities have been described as a leading point for intussusception in the pediatric literature.[1] Intussusceptions due to heterotopic tissues in polyp in adults is uncommon.[5,6] Here, we report a novel case of combined gastric and pancreatic heterotopias identified in ileal polyp which was complicated by ileo-ileal intussusceptions in an adult female.

CASE REPORT:

A 32-year-old female presented with the complaints of pain in abdomen and bleeding per rectum for four days. On physical examination, mild abdominal distension was noted. There were no haemorrhoids or anal fissure. The rectal examination was normal. A contrast-enhanced computed tomography scan of the abdomen revealed ileo-ileal intussusception in the distal ileum. On exploratory laparotomy, a polyp 2x2x1 cm was noted proximal to ileo-caecal junction as the lead point for intussusceptions [Figure 1]. Resection of the intussuscepted segment with the polyp was done. The resected ileal segment with the polyp was sent for histopathological examination.

Gross examination of the specimen revealed intussusception in a **9 cm** intestinal segment. On cutting open, a polyp measuring 2x2x1.5 cm was identified. Rest of the mucosa appeared unremarkable. Multiple sections were taken and microscopic examination revealed submucosal edema and congestion in the intestinal wall. There was presence of subserosal polyp [Figure 2] composed of adipose tissue, muscle and ectopic pancreatic ducts [Figure 3] and gastric tissue comprising of gastric fundic type epithelium [Figure 4 and 5]. There **were** no architectural or cytological atypia or dysplastic features. These findings of abnormally located, ectopic benign pancreatic and gastric tissue were morphologically consistent with co-existing heterotopic pancreatic and gastric tissue **within the ileum**. No dysplasia or malignancy was identified.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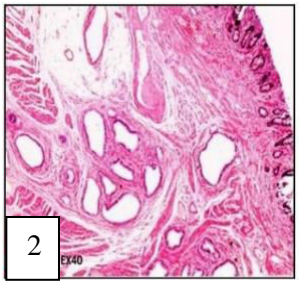
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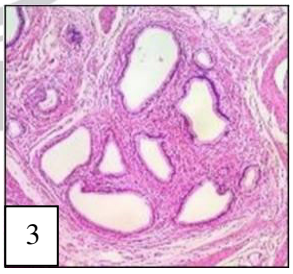
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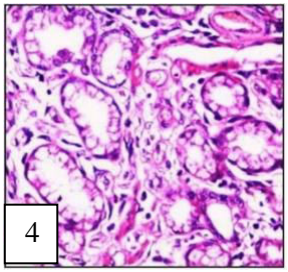
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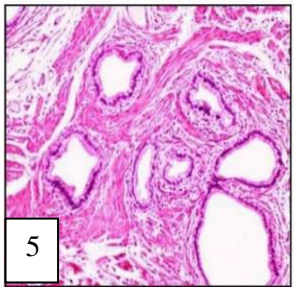
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Figure 1: Intussusception with Polyp, Figure 2: Subserosal polyp (HE X40), Figure 3: Ectopic Pancreatic ducts (Hematoxylin and eosin stain; X100), Figure 4: Ectopic gastric tissue with muscular disorganization (Hematoxylin and eosin stain; X100), Figure 5: Ectopic gastric tissue (Hematoxylin and eosin stain; X400).

DISCUSSION:

The term "heterotopia" has been described as the presence of the tissue outside its usual location. The first case of heterotopic gastric tissue was reported in esophagus in 1805 by Schmidt.[7] Hunt et al in 1934 published the case report on aberrant pancreatic tissue in Meckel's diverticulum.[8] Combined gastric and pancreatic heterotopias had been first described n the transverse colon by Burne in 1958.[9]

Heterotopic pancreatic and gastric tissues are considered as congenital anomaly. Abnormalities during embryological development may result in pancreatic heterotopias. Three theories have been proposed namely (1) the separation of heterotopic pancreatic tissue from primitive pancreas during embryonic rotation; (2) pancreatic tissue is abnormally transported by the longitudinal growth of the intestine from lateral budding of the rudimentary pancreatic tissue while penetrating the intestinal **wall,** [10] and (3) abnormalities in the Notch signaling system that leads to changes in differentiation in the developing foregut **endoderm.** [11]

Developmental anomaly and heterotopic differentiation had been considered to result in heterotopic gastric tissue. Metaplastic differentiation is also one of the described mechanisms for the pathogenesis of this entity.[12]

The combination of pancreatic and gastric heterotopic tissue has been described in esophagus [13] and duodenum but its presence in ileum as component of a polypoid mass in adult makes this case unique. Very few cases of combined heterotopic tissue in ileum being a lead point for intussusceptions in adults have been described.[6] Jiang K et al reported this entity in ileum in 38-year-old female with ileo-ileal intussusception. Similar case had been described in a 61-year-old male in the small intestine by Alfrejat M et al [6] and in jejunum in 55-year-old female by Rather JM **et al.** [14] There are case reports in the pediatric population for combined heterotopias as a cause for **intussusception.** [1] Shemer et al reported in gastric and pancreatic heterotopic tissue in Meckel's diverticulum in a **11-year-old male**. [15]

Radiological imaging and endoscopy are not sufficient to determine and definitively diagnose heterotopias. Histopathological examination plays a very important role for identifying this entity. In our case, the lesion was identified when the patient was operated for the ileal polyp and thorough histopathological examination was done by taking multiple sections. Proper microscopic examination led us to make a definitive diagnosis.

CONCLUSION:

Heterotopic gastric and pancreatic tissue in the ileum is a rare entity and remains difficult to diagnose pre-operatively. Hence, proper histopathological examination is essential in making the correct diagnosis.

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