

Pancreatic Heterotopia in an Ileal Lipoma - A Pathological Rarity Causing Intestinal Obstruction

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Abstract

Adult intussusception is a rare entity. In adults, 90% have a cause for intussusception which is usually a polyp, adenoma, adhesion, hamartoma, or tumor as a lead point. Rarely described is heterotopic pancreatic tissue with lipoma as a lead point. Here, we report a case of a 35-year-old male patient who presented with diffuse abdominal pain for 1 day. Computed tomography scan of the abdomen and pelvis showed intussusception involving distal small bowel loops at two levels with obstruction of ileal loops between the intussusception. Histopathology of the specimen showed heterotopic pancreatic tissue with submucosal lipoma. A comprehensive review on this rare entity is attempted.

Key words: Gastrointestinal, Heterotopia, Lipoma

INTRODUCTION

Pancreatic heterotopia (PH) is defined as pancreatic tissue outside its typical location without vascular or anatomical continuity with the pancreas proper. These are rare clinical entities which have been identified in virtually the entire length of the gastrointestinal system.¹ However, PH is most common in the duodenum and jejunum.² Involvement of other organs such as the esophagus, terminal ileum, Meckel's diverticulum, colon, ampulla of Vater, gallbladder, spleen, mesentery, liver, bile duct, omentum, and urinary bladder is rare but has been previously reported.³ The clinical significance of PH depends on size, location, and associated complications.

We report a novel case of PH with associated submucosal lipoma, identified in a 3.5 cm ileum mass, which was complicated by infarction. We believe that this is the first report of all three histological entities coexisting in an ileal lesion.

CASE REPORT

A previously healthy 35-year-old male presented to our institution for the evaluation of diffuse abdominal pain for 1 day. There was one episode of non-bilious/non-blood-stained vomiting.

The patient gave a history of two similar episodes which were relieved with medicines. X-ray abdomen erect showed fecal loading and multiple air-fluid levels. Computed tomography (CT) scan of the abdomen and pelvis showed intussusception involving distal small bowel loops (ileal loops) at two levels with obstruction of ileal loops between the intussusception. Laparotomy with resection and anastomosis were done under general anesthesia (Figures 1 and 2).

Gross examination of the specimen revealed a pedunculated polyp in a segment of the small intestine measuring 3.5 cm. The polyp measured 7.5 cm × 3 cm × 2 cm, with the stalk measuring 2.7 cm in length. The polyp and stalk were markedly congested and grossly appeared infarcted. The adjacent ileal mucosa is grossly edematous.

Microscopically, the polypoidal lesion was seen to be covered by infarcted ileal mucosa beneath which was seen, in the submucosa, groups of pancreatic acini, islets of Langerhans and an occasional pancreatic duct. These

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Figure 1: Intraoperative image showing the partly infarcted lipoma removed from the intestinal lumen

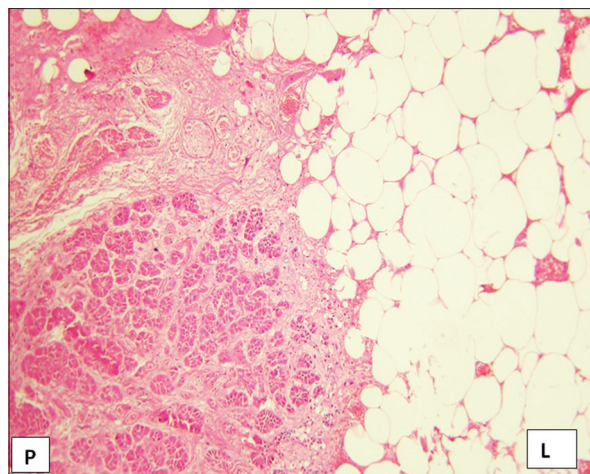


Figure 3: Presence of pancreatic tissue (P) in the lipoma (L) (H and E, x10)

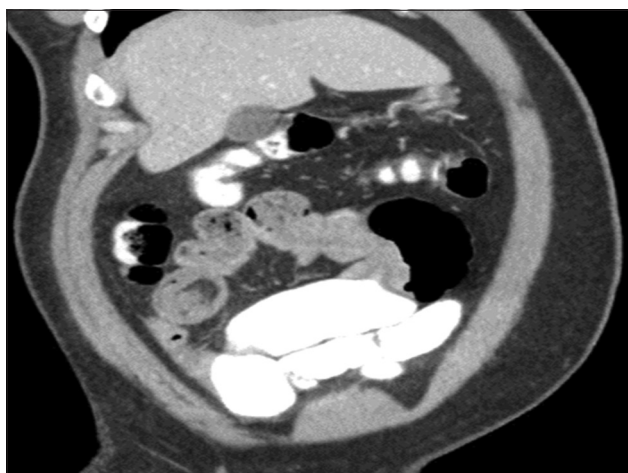


Figure 2: Contrast-enhanced computed tomography abdomen showing ileo-ileal intussusception

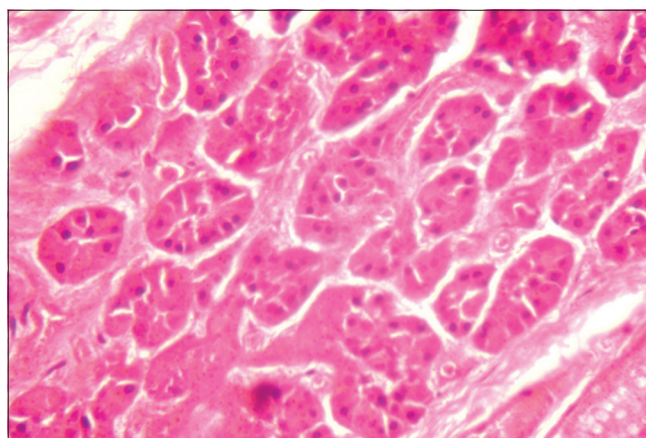


Figure 4: Pancreatic tissue, focally infarcted (H and E, x40)

pancreatic elements were haphazardly arranged and are surrounded by a vaguely circumscribed lesion composed of lobules of mature fibro-adipose tissue interspersed with delicate fibrovascular septa. The pedicle of the polyp showed the presence of smooth muscle bundles and thick-walled blood vessels. The polyp showed focal evidence of infarctoid necrosis (Figures 3 and 4). These findings of abnormally located, ectopic, benign pancreatic tissue were morphologically consistent with coexisting PH residing within a submucosal lipoma. No dysplasia or malignancy was identified. Post-laparotomy, CT scan of the abdomen and pelvis showed reduction of intussusception with no obvious recurrent mass.

DISCUSSION

Heterotopia indicates findings of the presence of normal tissue at a site where it usually does not reside and it lacks anatomic and vascular connections with the parent organ.

The mechanism of formation of heterotopia is obscure; hypothesis includes errors in embryologic development.³ The most commonly reported heterotopias in the gut are gastric and PHs. The frequency of PH in autopsy series shows large variability (0.55-25%), which indicates that the lesion often goes undetected and is largely harmless.⁴ The incidence of heterotopic pancreas is 0.2% of upper abdominal operations and 2/3rd are detected incidentally at the time of surgery in cases of isolated PH.⁵ Approximately half of cases of small bowel PH are asymptomatic.³ Symptomatic PH has a peak incidence in the fourth to sixth decades of life with a male to female ratio of 3:1. PH can cause pain, bleeding, obstruction, and intussusception, and it can be complicated by the development of virtually any disease of the orthotic pancreas.^{6,7} PH is most common in the duodenum, upper jejunum, and stomach. It can be found throughout the alimentary tract. PH in the ileum is rare.⁸ In the gut, PH appears as round or lobulated intramural nodules usually measuring <3 cm. It is usually found in the submucosa but has also been found in the muscularis propria or serosa.⁴ Barbosa *et al.* reported the

first case of ileal heterotopic pancreas with intussusception in 1946 and 16 cases of ileal heterotopic pancreas and gastrointestinal bleeding have been reported till date in the literature. Diagnosis is usually made after histological examination of the suspected tissue in symptomatic patients which shows collections of normal acini, ducts, and often islets of Langerhans.^{8,9}

Lipomas of the gastrointestinal tract (GIT) are benign tumors and were first reported by Bauer in 1757. The rarity of these tumors and lack of specific symptoms and signs often poses a diagnostic challenge.¹⁰ Lipomas are found anywhere along the GIT, however, most commonly occur in the ileum.¹¹ Smaller lesions do not cause many symptoms. However, larger lesions invariably lead to intussusception. The terminal ileum is the most common site for lipomas to occur. The submucosal type is the most common followed by intermuscular and serosal types.¹²

Depending on location, resection with restoration of continuity by means of an anastomosis has been shown to be adequate for the treatment for ileal pancreas.¹³

CONCLUSION

Previous authors have described cases of heterotopic pancreas as a lead point for intussusception in adults.^{14,15} However, the mass in our case contained pancreatic heterotopic tissue with an associated benign mesenchymal neoplasm lipoma complicated by infarction. This combination makes this case unique.

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