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Atypical Presentation of Jejunojejunal Intussusception Due to Ectopic Pancreas in a Young Male

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Authors' contributions

This work was carried out in collaboration among all authors. Author BH participated in data collection and wrote the article. All authors read and approved the final manuscript.

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Case Study

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ABSTRACT

Background: Intussusception may be caused by a variety of conditions and more commonly involves the ileum. Jejunojejunal intussusception is a rare condition and may originate from intramural tumors. Ectopic pancreatic tissue may be found throughout the entire gastrointestinal track causing no symptoms.
Case Report: A 29 year old African American male presented with cramping abdominal pain and nausea to the emergency room. As a child he had multiple trips to the ER for abdominal pain but no diagnosis had been made. Five years earlier a small bowel intussusception was found on CT-scan but exploratory laparotomy was negative. The patient had remained asymptomatic until 3 months prior to the new episode. CT-scan showed recurrent intussusception of the proximal small bowel. An extensive work-up did not reveal diagnosis but the patient was readmitted multiple times for abdominal pain and again signs of intussusceptions were found. He underwent exploratory laparoscopy and no intussusception but a small yellowish lesion at 10cm past the ligament of Treitz was detected. A segmental jejunal resection containing the 1.5cm lesion was performed. Pathology revealed a transmural lesion consistent with ectopic pancreatic tissue. The immediate postoperative course was uneventful and the patent improved; however, he continued to have episodes of

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abdominal cramping but further workup was negative and we believe he suffered from a motility disorder due to the chronic dilatation of the duodenum and proximal jejunum associated with the recurrent intussusceptions.

Discussion: We present a rare case of ectopic pancreas causing recurrent jejunojejunal intussusceptions in an adult; these small lesions may be difficult to find and should be surgically removed as the recurrent obstructions may cause long term damage to the proximal intestinal segments. Laparoscopy should be the preferred approach for such cases.

Keywords: Intussusception; ectopic pancreas; laparoscopy.

1. INTRODUCTION

Intussusception may be caused by a variety of conditions including infection, inflammation and tumors and more commonly involves the ileum [1,2]. Jejunojejunal intussusception is a rare condition and may originate from intramural primary or secondary tumors [3]. Ectopic pancreatic tissue may be found throughout the entire gastrointestinal track and is often asymptomatic [4,5]. However, it may become symptomatic and cause pain, hemorrhage or obstruction, which occurs usually during childhood [6,7]. Several cases of small bowel intussusception associated with ectopic pancreatic tissue have been published [8-14]. Surgery is indicated and recently the laparoscopic approach has been accepted by many surgeons [9]. We here in report on an unusual presentation of a jejunojejunal intussusception caused by ectopic pancreatic tissue.

2. CASE REPORT

A 29 year old African American male presented to the emergency room (ER) with a history of recurrent small bowel intussusception for which he had undergone exploratory laparotomy five years ago but during surgery no pathology causing the intussusception was found. The patient was asymptomatic for several years until three months prior to the ER visit when he started to have abdominal pain, cramping, and nausea. CT scan showed a proximal small bowel intussusception just distal to the duodenojejunal junction. Within few hours of bowel rest he improved and the pain completely subsided and he tolerated a clear liquid diet. An extensive work-up was initiated which included CT-enterography, push endoscopy and capsule endoscopy which all were negative. On further questioning, the patient reported that as a child his mother had to bring him to the ER on multiple occasions when he developed abdominal cramps. He never was referred to a pediatric gastroenterologist and no attempts were made to work up the intermittent severe abdominal symptoms as a child. As an adolescent he started to abuse recreational drugs, however, he grew appropriately and finished school and worked as a chef. During the work up for the recent episode he lost his job due to the multiple occasions of absence and he lost a significant amount of weight. As no cause for the recurrent intussusceptions was found on the extensive work up, recreational drug abuse was entertained as a trigger. He had another and more prolonged episode of abdominal pain and CT-scan demonstrated recurrent intussusception (Fig. 1).

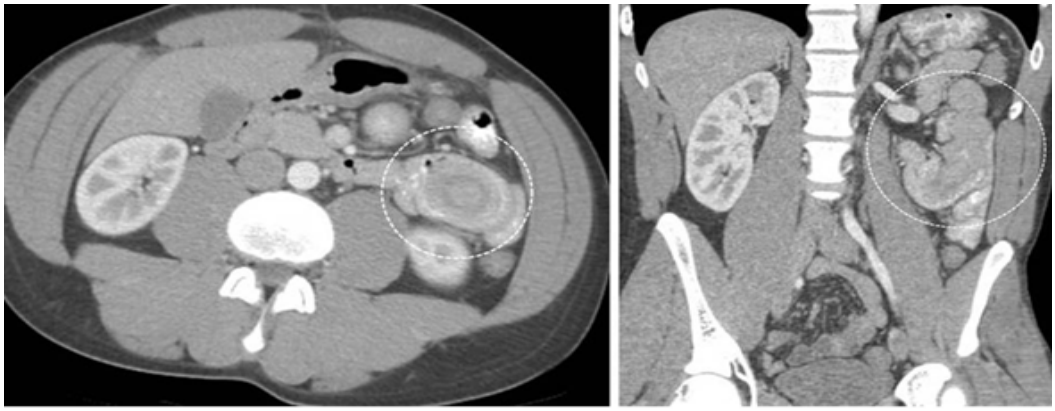

Fig. 1. CT-scan showing proximal jejunojejunal intussusception

Indication for an exploratory laparoscopy was made five days into the recent admission. Five mm trocars were placed in the left and right mid abdomen and the umbilicus. No significant adhesions and no dilated small bowel and no intussusception were seen on exploration. The small bowel was run backwards from the terminal ileum to the ligament of Treitz and no pathology was found. On antegrade exploration, at about 10cm past the ligament of Trietz, a small yellowish lesion could be seen shining through the small bowel serosa and on further examination this was identified as the area of intussusception as the small bowel was slightly dilated proximal to the lesion. Resection of the segment of the jejunum that contained the 1.5cm lesion was carried out (Fig. 2).

An extracorporeal side to side anastomosis using a 4cm epigastric incision was created. Pathology revealed a transmural lesion compatible with Type 1 ectopic pancreatic tissue.

The initial postoperative course was uneventful and after discharge the patient regained weight and had no more pain. However, within few weeks after surgery he came back to the ER with recurrent abdominal cramps. Repeat CT-scan was negative and he improved within 24 hours and was discharged. During the next six months he had multiple such episodes and went to several different emergency rooms and had hospitalizations. Again extensive work up was done but CT-scans showed no more intussusceptions, upper gastrointestinal follow through with barium, endoscopy and a gastric emptying study were all negative. On an abdominal ultrasound, sludge in the gall bladder was found and ultimately it was decided to perform another exploratory laparoscopy with cholecystectomy. Some adhesions from the previous surgery were divided. Mesenteric lymphadenopathy was found (Fig. 3a); pathology of the lymph node biopsy showed reactive lymphoid tissue.

The anastomosis was found patent without any abnormalities and no dilated small bowel (Fig. 3b,c)

Cholecystectomy was done without complications and the patient was discharged the same day.

After a short improvement in symptoms, the patient continued to complain of abdominal cramps. Work up at a referral center was negative and ultimately he was lost to follow up but he kept filling prescriptions for narcotics.

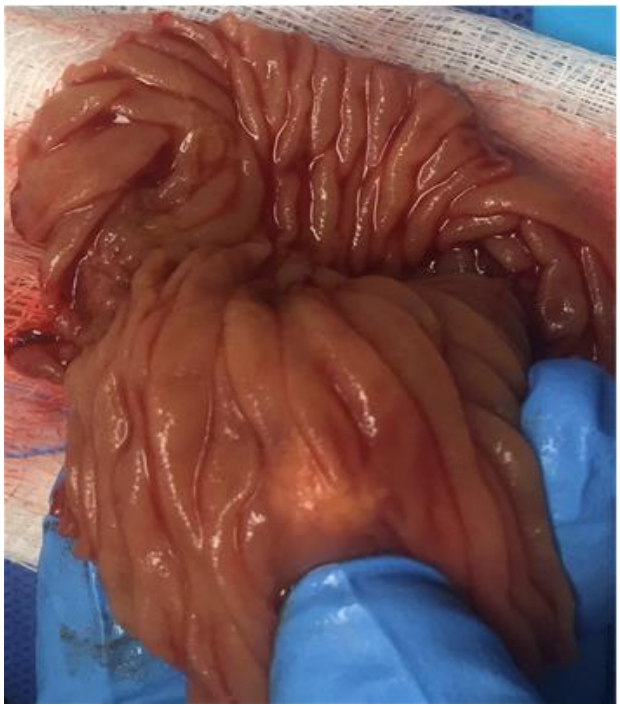

Fig. 2. Resected specimen: small yellowish submucosal lesion

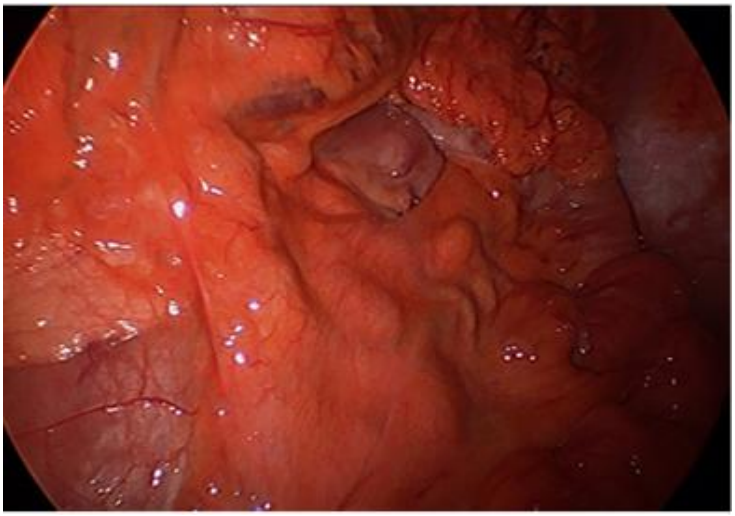

Fig. 3a. Second laparoscopy: mesenteric lymphadenopathy

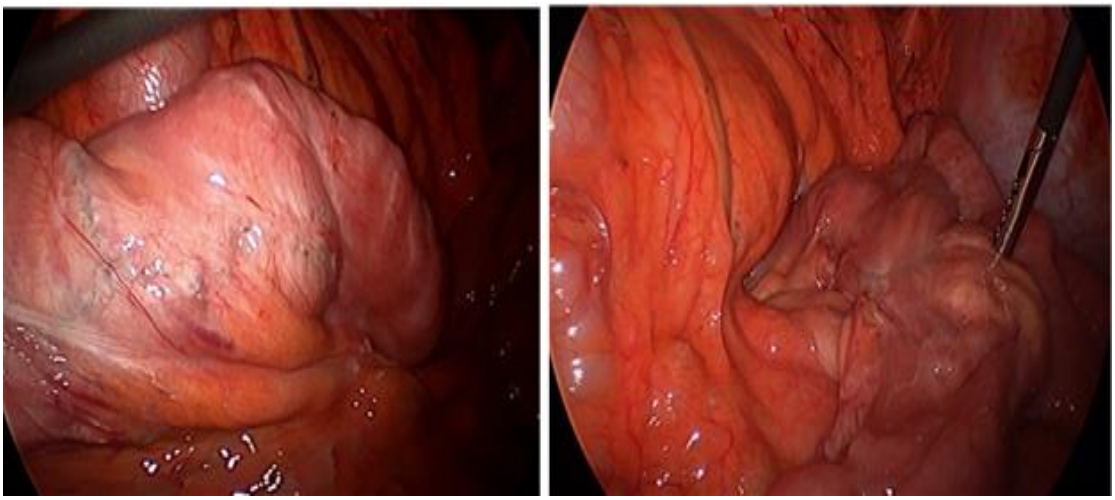

Fig. 3b,c. Second laparoscopy: anastomosis (anterior and posterior view) patent without pathology and no proximal small bowel dilatation

3. DISCUSSION

We present a rare case of ectopic pancreas causing recurrent jejunojejunal intussusceptions in an adult. These small lesions may be difficult to find on multiple imaging studies and may be overlooked even on laparotomy such as in our case. Ultimately, segments of recurrent intussusception should be surgically removed to obtain a diagnosis and due to the fact that the recurrent obstructions may cause long term damage to the proximal intestinal segments [1-3,9].

We do believe that such a functional problem caused the ongoing abdominal pain even after the area of recurrent intussusceptions including the causing lesion had been resected. However, we have taken in consideration that the patient received too many prescriptions for narcotics and ultimately became a “drug seeker”. The case does raise issues with regard to access to appropriate work up as his symptoms started during child hood but he never was referred to a pediatric gastroenterologist. This delay in the diagnosis and the fact that the lesion was missed on an exploratory laparotomy may have resulted in further damage of his proximal gastrointestinal tract. The patient had more than 20 CT-scans during a five year period whenever he went to the ER and this is not only expensive but also is associated with excessive exposure to iodine

contrast as well as radiation. Cases similar to ours have been published [7,8,10,12-14] and ours does emphasize that for rare conditions such as in our patients, a traditional approach may not lead to an accurate diagnosis. The recurrent intussusceptions were well known and a focused work up to foster a diagnosis should have been undertaken [1,2]. Possibly an endoscopic ultrasound may have been of use, however, is technically difficult to do in the proximal jejunum.

The frustration and helplessness of all involved physicians caused by the multiple negative tests climaxed when drug abuse was suggested to cause the patients condition. Ultimately, an exploratory laparoscopy was able to establish the diagnosis and the small bowel resection was tolerated well by the patient [9]. As ectopic pancreas is such a rare condition, the final pathology came as a surprise; however, as mentioned, multiple similar cases have been reported [4,7,10,14]. The best hint that this was an inborn condition came from the patients anamnesis as he had his symptoms since childhood.

4. CONCLUSION

To summarize, when confronted with recurrent intussusceptions, physicians must foster a diagnosis, which may require exploratory laparoscopy or laparotomy. A more focused approach and access to a specialist may have led to diagnosis and treatment of the rare condition at childhood and may have prevented significant pain and suffering for the patient and excessive costs for the health care system.

CONSENT AND ETHICAL APPROVAL

As per international standard or university standard guideline patients consent and ethical approval has been collected and preserved by the authors.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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