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ABSTRACT

Introduction: Pancreatic heterotopia has been described most frequently in stomach and duodenum. Isolated occurrence of heterotopic pancreas in ileum is exceptional and is usually discovered incidentally during surgery. In rare cases, heterotopic pancreas has been reported as a lead point of ileoileal intussusception.

Case Report: A one-year-old boy was presented with recent onset of constipation and abdominal distention. Radiologic investigation suggested a diagnosis of midgut volvulus with ileoileal intussusception. Resection anastomosis of the intussusception was performed. Histologic examination of the resected specimen showed heterotopic pancreas in the wall of intussusceptum at its tip. No other significant pathology was found.

Conclusion: Ileal heterotopic pancreas is a rare cause of intussusception at any age and is usually not evident clinically. Hence, careful examination of the lead point of intussusceptum in resection specimens is mandatory to delineate the underlying etiology of these cases.

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INTRODUCTION

Intussusception is defined as the telescoping of a segment of the gastrointestinal tract into subjacent one. It is the most common cause of intestinal obstruction in children and is often idiopathic [1]. Intussusception with intraluminal heterotopic pancreatic tissue as the lead point is a rare condition [2]. It usually presents in stomach, duodenum and proximal jejunum. Occurrence more distally in the distal jejunum and ileum is exceptionally rare and usually asymptomatic [1, 3]. Intussusception due to heterotopic pancreas has been described in a few cases, including pediatric as well as adult patients [1, 2]. On an extensive review of literature, we found less than 25 cases of isolated heterotopic pancreas of ileum as leading point of intussusception in children. We describe the clinicopathologic features of an infant with ileoileal intussusception due to heterotopic pancreas in ileum. The existing literature is briefly reviewed as well.

CASE REPORT

A one-year-old boy was admitted to our hospital with complaints of fever, constipation, vomiting and distension of abdomen since two days. Physical examination showed marked abdominal distension with venous prominence.

A provisional diagnosis of intestinal obstruction was made. Radiologic investigations showed multiple air fluid levels with features of midgut volvulus and ileoileal intussusception. The child underwent an exploratory laparotomy. Intra-operatively, midgut volvulus with torsion of mesentery and ileoileal intussusception 30 cm proximal to ileocaecal junction was found. The intussusceptum appeared gangrenous. There was no free fluid in the abdomen. Surgical correction of the volvulus with Ladd's procedure and resection of ileal intussusception was done with end-to-end anastomosis. The resected specimen was submitted for histopathology.

We received a segment of ileum measuring 8.5 cm in length. The segment was gangrenous and showed an intussusception 6 cm in length. Intussusceptum was 5 cm long with a blind end and focal mucosal ulceration. Intussuscipiens was 7.5 cm in length and showed normal mucosal folds. A congested nodule measuring 0.6x0.6 cm was identified in the lumen of intussusceptum near the tip of the blind end. Multiple sections from the nodule as well as intestinal wall were processed.

Microscopically, the nodule showed a well-circumscribed polypoid tissue with ulcerated mucosa. In the submucosa, pancreatic acini were identified embedded within the gangrenous wall of the nodule (Figure 1). No islet cells were identified. The histological impression was that of heterotopic pancreatic tissue, type 2 in the submucosa of ileum. There was no other significant pathological change such as inflammation or neoplastic transformation in the pancreatic tissue.

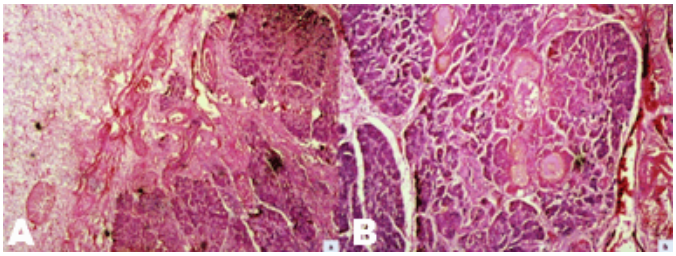

Figure 1: Photomicrograph (A) Showing a nodule composed of pancreatic acini without islet structures (H&E stain, x100), (B) Higher magnification showing the gangrenous acini (H&E stain, x200).

DISCUSSION

Intussusception in pediatric age group is usually idiopathic in origin, with pathological lesion at the lead point being identified in a very small proportion of cases [4, 5]. The incidence of a pathologic process as the leading point of intussusception varied from 2–12% in the previous studies in literature [6]. The various pathologic lesions identified as leading points in pediatric intussusception include Meckel's diverticulum (most common) followed by intestinal polyps, duplication cysts and lymphoma [7, 8]. In a study by Puri and Guiney, 27 (9.2%) of 292 cases of pediatric intussusception revealed pathologic lesions and 48% of these had Meckel's diverticulum as the cause of intussusception [8].

Heterotopic pancreas is defined as pancreatic tissue that has neither anatomic nor vascular relation with the normally located pancreas [9]. Various theories suggest its origin as a result of separation of pancreatic tissue during embryonic rotation of dorsal and ventral buds [10]. Persistence of this separated pancreatic tissue results in heterotopia anywhere from stomach (most common) to jejunum or ileum (least common) [2]. Heterotopic pancreas in the ileum is a rare entity and when identified is usually associated with Meckel's diverticulum. Isolated heterotopic pancreas of the ileum (in absence of any other pathologic finding) is extremely rare and is usually an incidental finding during surgery [4, 8, 9, 11]. Dolan et al. have suggested that symptoms due to heterotopic pancreas are related to its size and mucosal relation [12]. Lesions of heterotopic pancreas measuring >15 mm in diameter and lying close to the mucosa are most likely to be symptomatic. In rare instances, isolated heterotopic pancreatic tissue in the ileum acts as a leading point for intussusception [1, 4, 6, 11]. Submucosal and muscular wall proximity of the pancreatic tissue is hypothesized to aggravate bowel dysmotility and act as local disturbance leading to intussusception [12, 13]. This is confirmed by the previous reports in literature where most of the cases of intussusception due to isolated pancreatic heterotopia in the ileum were either submucosal or serosal [4, 6]. In our patient also, isolated heterotopic pancreas in the submucosa of ileum acted as the lead point of ileoileal intussusception.

Histologically, heterotopic pancreas has been classified into three types by von Heinrich as: Type 1 (all elements of the normal pancreatic tissue are identified), Type 2 (pancreatic tissue with absence of islet cells) and Type 3 (pancreatic ducts only) [14]. Our patient had type 2 heterotopic pancreatic tissue. Although most cases with heterotopic pancreas are asymptomatic, various pathological changes including pancreatitis, pseudo pancreatic cyst, adenoma and carcinoma have been reported in foci of heterotopic pancreas [15]. None of these changes were identified in our case.

On an extensive review of the existing English literature, we found less than 25 cases of isolated heterotopic pancreas of ileum as leading point for intussusception in pediatric age group [2, 4, 6]. The relative frequency of a pathological lesion causing intussusception increases with age. However, the highest frequency of detection of pathologic lesion in cases of intussusception is still the first year of life [6], as in the present case. In a review of 10 children with ileoileal intussusception due to heterotopic pancreas, seven patients were less than two years of age and of these, four were infants [6].

Most cases of intussusception caused by a pathologic lead point are irreducible by barium enema and require

manual reduction [6]. Hence, it is imperative to palpate for lesions like heterotopic pancreas within the wall of ileum after manual reduction [6]. This is especially essential in patients with submucosal location of heterotopic pancreas since the lesion may not be obvious on visual inspection. Simple excision is considered the treatment of choice for heterotopic pancreas leading to intussusception. This procedure avoids recurrence of intussusception as well as the risk of sequelae of pancreatitis etc [6].

CONCLUSION

In conclusion, isolated pancreatic heterotopia in ileum is rare and intussusception due to such occurrence is an even rarer phenomenon. Pediatric intussusception due to heterotopic pancreas has been described in very few cases. Hence the present case reiterates the need for examination of the tip of intussusceptum or the part of intestine that telescopes for diagnosis of submucosal lesions like heterotopic pancreas as the lead point of intussusception in resection specimens.

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Author Contributions

Sarika Verma – Acquisition of data, Drafting the article, Final approval of the version to be published
Pankaj Bansal – Revising it critically for important intellectual content, Final approval of the version to be published
Vivek Manchanda – Acquisition of data, Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published
Ruchika Gupta – Substantial contributions to conception and design, Analysis and interpretation of data, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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