

Case Report

Duodenoduodenal intussusception: two case reports with different lead points

Rutu A. Brahmbhatt*, Drupad G. Patel, Pranav B. Chaudhary

Department of General Surgery, Smt Municipal Medical College, Ahmedabad, Gujarat, India

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*Correspondence:

Dr. Rutu A. Brahmbhatt,

E-mail: rutu.brahmbhatt19@gmail.com

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ABSTRACT

Intussusception occurs when one portion of the bowel invaginates into an immediately adjacent segment, almost invariably it is the proximal into the distal. Duodenoduodenal intussusception is a rare condition that is in general caused by a tumour either benign or malignant. However duodenal intussusception is an extremely rare entity as duodenum is a fixed retroperitoneal structure. The lead point of intussusception is usually a Meckel's diverticulum, a Peutz-Jeghers polyp or intestinal adenomas. We describe two case reports of duodenoduodenal intussusception secondary to a tubulovillous adenoma and juvenile hamartomatous polyp ((Peutz Jegher's polyposis) that caused intussusception in a 17-year-old female and 11-year-old male respectively. Both cases had underlying abnormalities acting as lead points with different etiologies. We resected the tumour mass from both patients successfully. This report describes the rare case of isolated adenoma of the 3rd part of the duodenum and Peutz Jegher polyp in the jejunum presented as duodenoduodenal and duodenojejunal type of enteroenteric intussusception fully treated by segmental resection of the duodenum and jejunum and doing well on follow up.

Keywords: Duodenoduodenal intussusception, Tubulovillous adenoma, Juvenile hamartomatous polyp (Peutz-Jeghers polyposis), Intussusception lead points, Target sign, Contrast-enhanced computed tomography

INTRODUCTION

Intussusception is the invagination of the bowel and is common in children and rare in adults, accounting for only 5% of all cases of intussusception.¹ Intussusception usually involves the small bowel and rarely large bowel. Symptoms include abdominal pain, which may wax and wane, vomiting, bloating, and bloody stool. It may result in small bowel obstruction. Other complications may include peritonitis/bowel perforation.²⁻⁵

Duodenal intussusception is an extremely rare entity as duodenum assumes a fixed, retroperitoneal position during embryological intestinal rotation and duodenal tumours leading to intussusception are rare.⁶ The lead point of intussusception is usually a Meckel's diverticulum, a Peutz-Jeghers polyp or a benign or

malignant tumour.⁷ Here, we present two cases of true duodenal and duodenojejunal intussusception secondary to duodenal adenoma and peutz jegher polyp respectively without malrotation abnormality. Diagnosis of duodenoduodenal intussusception is usually delayed because of its longstanding, intermittent, and nonspecific symptoms and most cases are diagnosed at emergency laparotomy.⁸ We hereby present two cases of duodenoduodenal intussusception in paediatric age group.

CASE REPORTS

Case 1

A 17-year-old female patient presented with complaints of vomiting and upper abdominal pain for the last 2-3 years which had aggravated over the last 3 months.

Vomiting occurred within 1-2 hours of food intake and was non-bilious and projectile. Upper abdominal pain increased after food intake and was relieved after vomiting. There was a significant weight loss of 14 kg in 3 months and a loss of appetite. There was no history of intestinal obstruction or surgical intervention. On examination, the patient was anaemic and had icterus with a palpable mass in the epigastric region. Abdominal ultrasonography (USG) showed target appearance in the left hypochondrium with bowel within bowel appearance. On upper GI scopy (Figure 1 A and B) a mass was seen causing luminal obstruction at 3rd part of the duodenum along with duodenoduodenal intussusception. Contrast-enhanced computed tomography (CECT) with oral contrast showed evidence of long segment intussusception noted at level of distal 2nd part of duodenum with intussusceptum being second part of duodenum, ampulla of vater, head and uncinate process of pancreas while intussusception is 3rd and 4th part of duodenum along with 4×3×6 cm sized homogenously enhancing polypoidal lesion within the intussusceptum.

During laparotomy, (Figure 2 A-C) the stomach along with the first and second part of the duodenum was dilated. Duodenal intussusception was found with the distal duodenum and head of the pancreas acting as intussusceptum and the proximal duodenum as the intussusceptions. On opening 3rd part of the duodenum a pedunculated cauliflower-like polypoidal mass of 8×6 cm was found obstructing the 3rd part of the duodenum on antimesenteric side. Trans duodenal polypectomy was done along with Roux en Y duodenojejunostomy and jejunojejunal anastomosis was performed. Specimens were sent for histopathological examination.

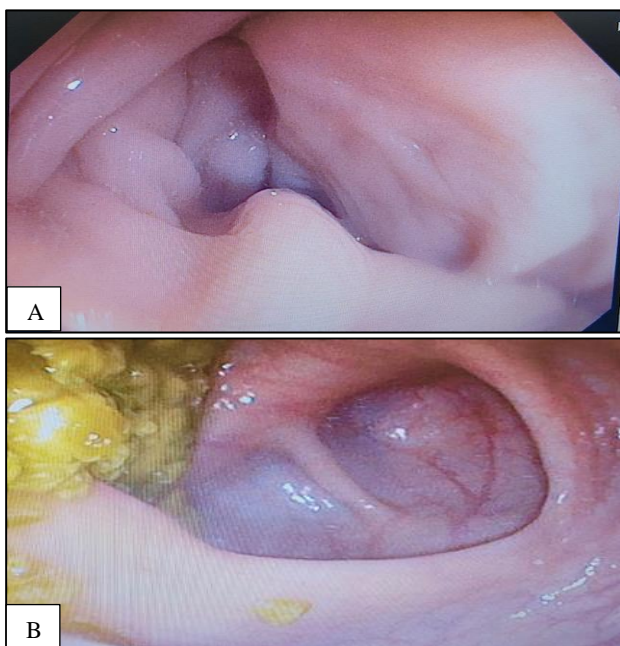


Figure 1 (A and B): UGI scopy suggestive of luminal obstruction of the duodenum. UGI scopy suggestive of duodenoduodenal intussusception.

A gross examination of the specimen confirmed intussusception along with the presence of a polypoidal mass lesion in the third part of the duodenum. The rest of the bowel was unremarkable (Figure 3 A and B).

Histopathological examination (Figure 4 A and B) of the mass revealed features of tubulovillous adenoma with no unequivocal high-grade dysplasia or metaplasia. There is no evidence of stalk infiltration.

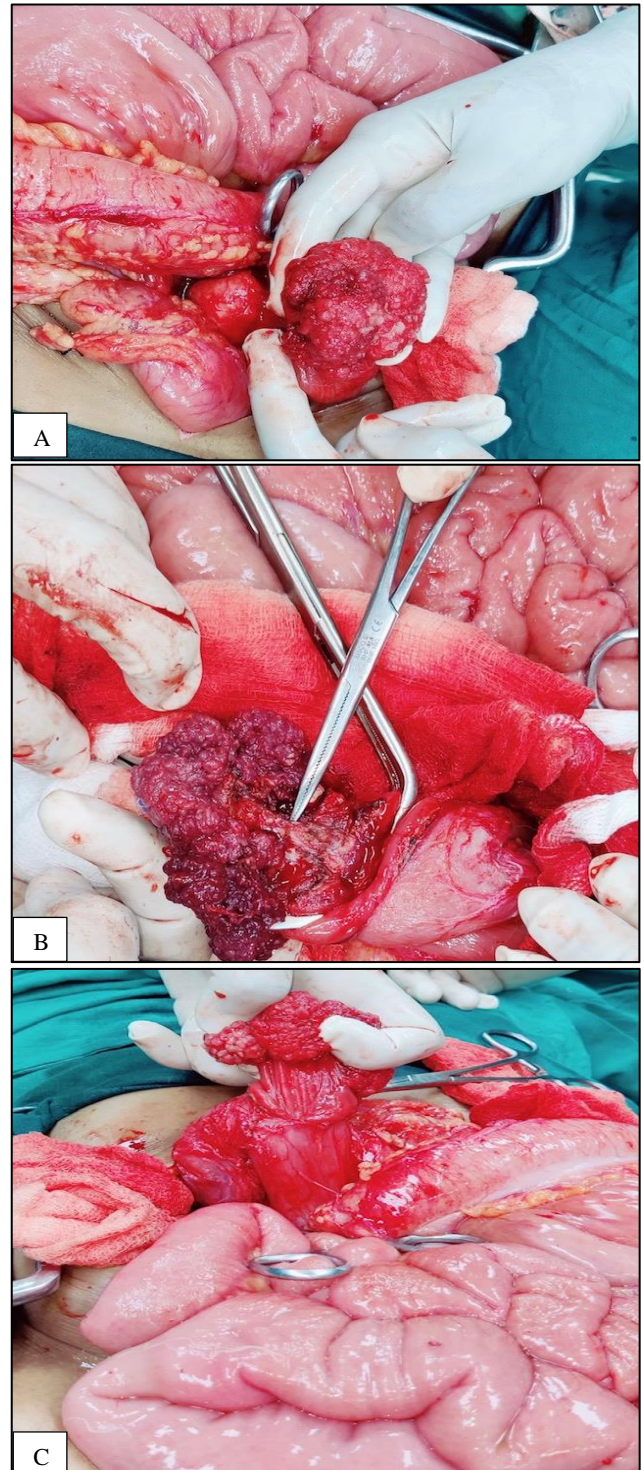


Figure 2 (A-C): Intraoperative images.



Figure 3 (A and B): Gross specimen.

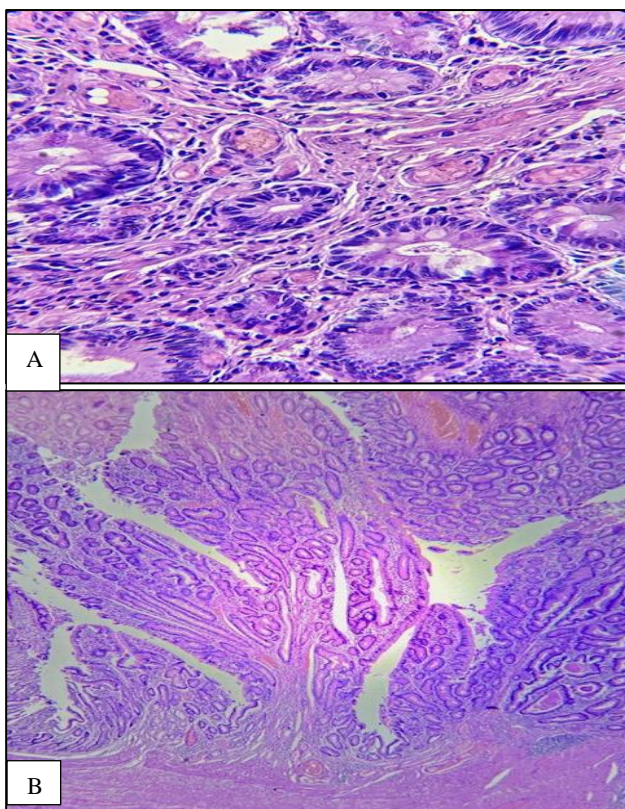


Figure 4 (A and B): Histopathological slide. showing proliferation of Glands with lining of pseudostratified columnar epithelium with elongated and hyperchromatic nuclei. Suggestive of tubulovillous adenomatous polyp of duodenum.

The post-operative period was uneventful and the patient was discharged on the 16th postoperative day after she could comfortably take solids. At the 4-month follow-up, she is doing well.

Case 2

An 11-year-old male presented with a long history of abdominal discomfort, loss of appetite, and early satiety with complaints of periumbilical pain for 4 years which was mild to moderate in intensity, intermittent, dull aching in nature aggravated by intake of food along postprandial vomiting since 1 year. On physical examination patient was pale and emaciated with hyperpigmentation of lips and oral cavity (Figure 5 A and B). The patient underwent upper GI scopy (Figure 6 A and B) suggestive of multiple sessile polyps more than 100 were seen in the entire stomach with a single large pedunculated 5×5 cm 1 cm thick stalk polyp arising from jejunum. On ultrasonography (USG) there was evidence of bowel within bowel appearance in epigastric and right hypochondriac region. Antrum and pylorus being intussusceptum and duodenum being intussusciens. Contrast enhanced computerised tomography (CECT) confirmed duodenoduodenal and duodenojejunal type of enteroenteric intussusception. Intussusceptum being pylorus of stomach, 1st part of duodenum, and branches of superior mesenteric vessels while intussusciens is 2nd part of the duodenum. Few heterogeneously enhancing lesions with internal non-enhancing necrotic areas are noted in 2nd part of duodenum (intussusciens), suggestive of a mass lesion that acts as lead point. Another long segmental intussusception (for a segment of approximately 24 cm) was noted at the level of DJ flexure and proximal part of jejunum with resultant dilated proximal third and fourth part of duodenum, maximum diameter measures approx. 61 mm suggestive of duodenojejunal type of intussusception. Intussusceptum being 3rd and 4th part of duodenum, branches of superior mesenteric vessels, and multiple heterogeneously enhancing lymph nodes while intussusciens being jejunal loops. Patient was taken up for surgery. On surgical exploration (Figure 7 A and B), pyloro duodenal intussusception seen with a distended 1st part of duodenum, pyloroduodenal intussusception was manually reduced and 5×6 cm sized polyp resected from 1st part of duodenum. Gastroduodenal end-to-end anastomosis was done, 3 polyps identified 5 cm, 150 cm, 170 cm distal to duodeno jejunal flexure. During intraoperative enteroscopy, jejunum was found hypertrophied. Jejunal polyp 5×4.5×3 cm single polypoidal mass identified at 150 cm from DJ flexure polyp resected at antimesenteric border. In addition, enlarged mesenteric lymph nodes are seen in peri jejunal area. Specimens were sent for HPE. On gross examination (Figure 8 A and B) of revealed intussusception with a polypoidal mass lesion. Histopathologic sections (Figure 9 A and B) from duodenal and jejunal polypoidal mass revealed distinctive papillary tree-like arborization of compact smooth

muscles with pseudo invasion seen in mucosa and muscularis propria. No evidence of dysplasia/ malignancy suggestive of juvenile hamartomatous polyposis (Peutz Jegher's polyposis).

Post-post-op period was uneventful and patient was discharged on 12th postop day after patient comfortably tolerated solids. On 3 months follow up he is doing well.

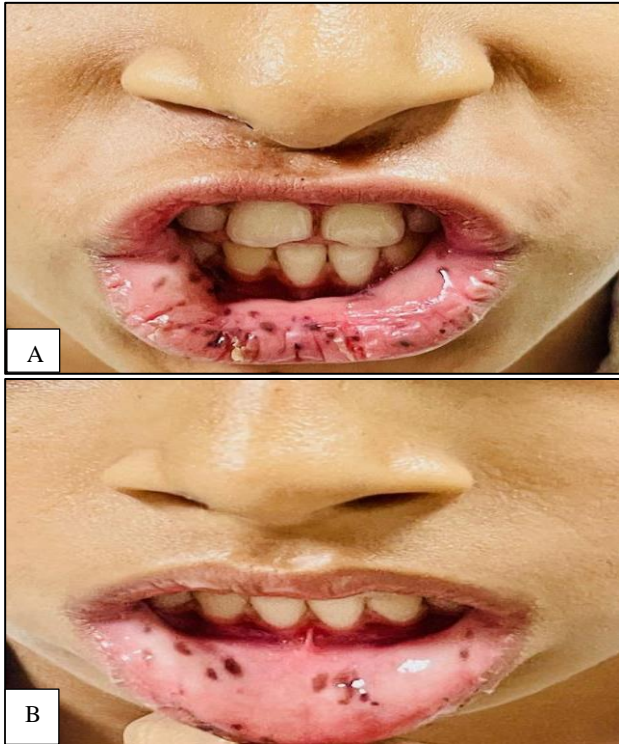


Figure 5 (A and B): Clinical picture of the patient with pigmented lips and mucous membrane.

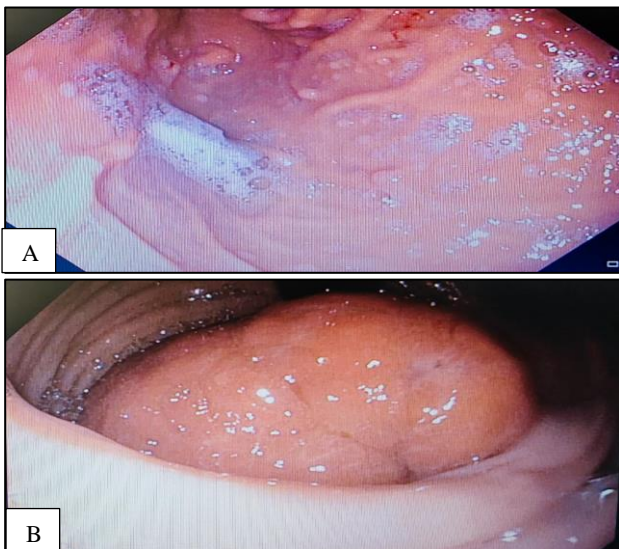


Figure 6 (A and B): Upper GI scopy suggestive of multiple duodenal polyps and polypoidal mass in jejunum, showing multiple small polyps on upper GI scopy. A polypoidal mass in jejunum.

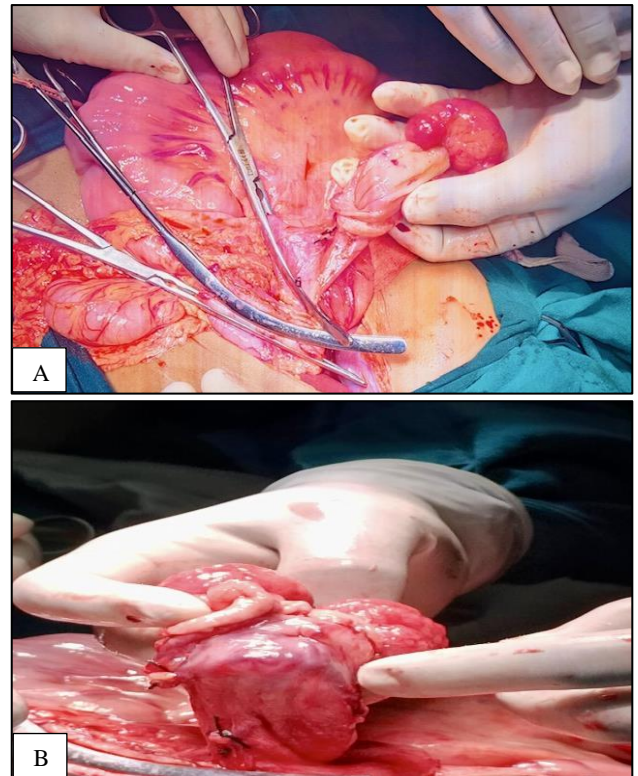


Figure 7 (A and B): Intraop image of jejunal polyp. Pedunculated jejunal polyp causing intussusception.

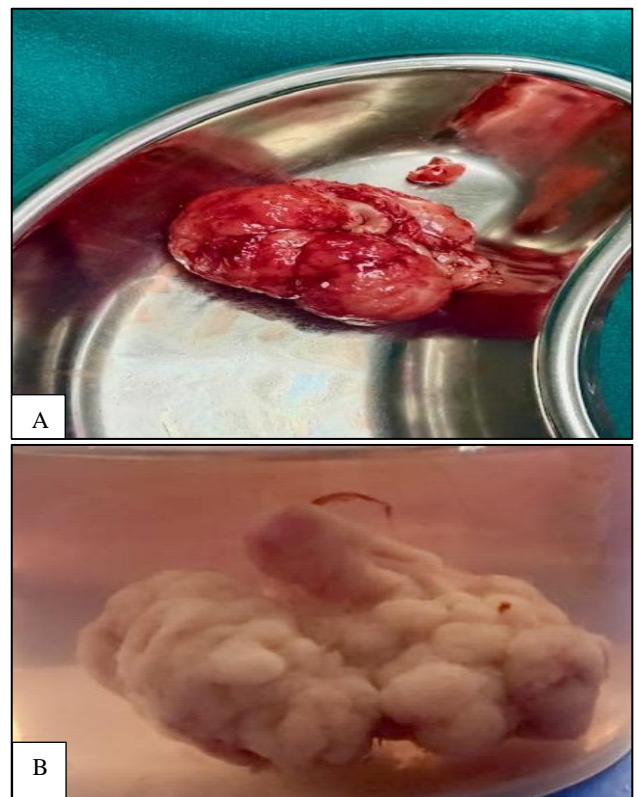


Figure 8 (A and B): Gross specimen showing cauliflower-like pedunculated mass. Formalin mounted specimen cauliflower-like pedunculated mass.

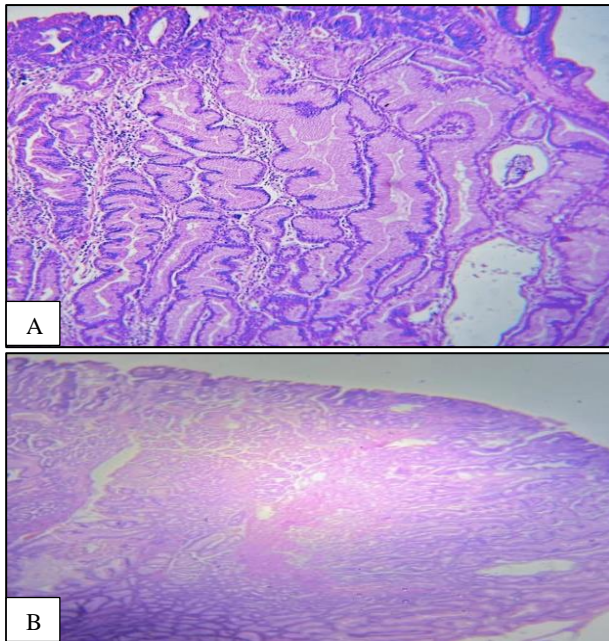


Figure 9 (A and B): Histopathological slide suggestive of hamartomatous polyp (Peutz Jegher's polyp). Slide showing edematous, inflamed lamina propria, and cystically dilated glands suggestive of juvenile hamartomatous polyp and arborising tree like pattern.

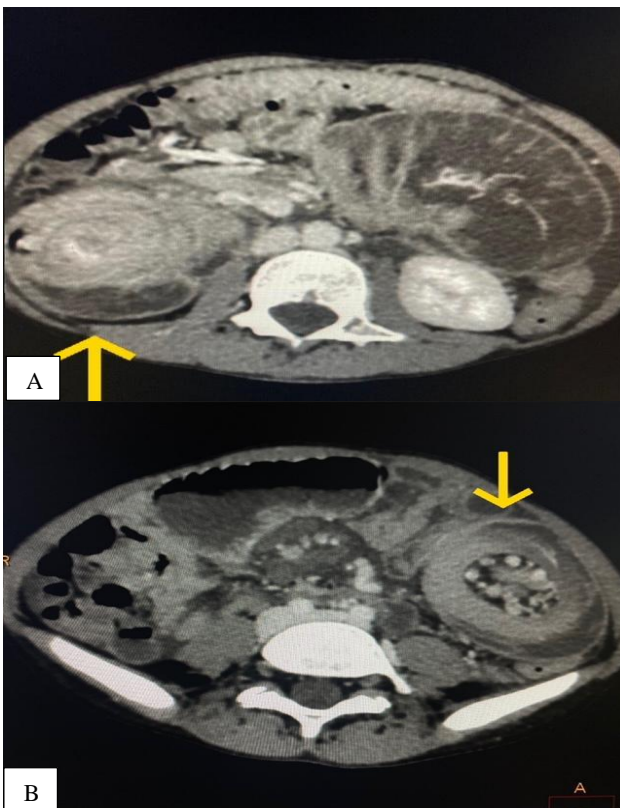


Figure 10 (A and B): Contrast-enhanced computed tomography showing an intussusception caused by a large polyp in a patient with Peutz-Jegher's syndrome: coronal sections.

DISCUSSION

Intussusception is defined as the invagination of one loop of the bowel into the lumen of an adjacent loop, the proximal invaginating segment being called intussusceptum which telescopes into the lumen of the distal segment or intussusceptions. However duodenal intussusception is an extremely rare entity as duodenum is a fixed retroperitoneal structure. The exact mechanism of duodenal intussusception is unknown; however, it is thought that any lesion in the duodenal wall or irritant within the lumen that alters normal peristaltic activity can initiate an invagination. The lead points for the intussusception are attributable to benign, malignant, or idiopathic causes.⁹

Unlike duodenoduodenal intussusception, which occurs within the same segment of the duodenum, duodenojejunal intussusception involves the duodenum telescoping into the jejunum, which is the upper part of the small intestine. Both tubulovillous adenoma and Peutz Jegher's polyp acted as lead points in causing duodenal intussusception.

Tubulovillous adenomas may include small, sessile, or polypoid lesions. They are discovered incidentally on endoscopy. Duodenal villous and tubulovillous tumours are often clinically silent.¹⁰ Though they are mostly asymptomatic, sometimes they may present with the signs and symptoms of jaundice (obstructive jaundice), abdominal pain, melena, hematochezia, fever, malaise or weight loss. Physical examination may reveal hard epigastric areas on palpation. Imaging can reveal an ulcerated tumour, dilatation or extensive involvement of the common bile duct and/or pancreatic duct.¹¹ These tumours can also present with some serious complications such as pancreatitis, duodenal obstruction or intussusceptions.^{12,13} Tubulovillous adenomas show a combination of tubular and villous architecture (villous components greater than 25%) and are usually intramural with intraepithelial tumour cells.¹⁴

Peutz Jegher's polyposis is an inherited disorder characterized by multiple distinctive polyps in the gastrointestinal tract, often accompanied by mucocutaneous pigmentation, particularly noticeable around the lips. It is inherited in an autosomal dominant manner, attributed to mutations in the *STK11* (*LKB1*) gene.¹⁵ Clinical symptoms of Peutz jegher's polyposis are characterized by periods of asymptomatic intervals punctuated by complications such as abdominal pain, recurrent intussusception leading to bowel obstruction, rectal polyp prolapse, and often occult bleeding. Small bowel obstruction is a common initial presentation, often necessitating repeat surgeries due to polyp-related complications occurring at relatively short intervals.^{16,17}

The diagnostic modalities used most often are USG, upper gastrointestinal series (UGI), CECT, and endoscopy (ES). Ultrasonography may show a typical

"target sign or doughnut sign". The target sign, as in the presented case, consists of a round soft tissue mass that contains a radiolucent ring of fat within the intussusception. This indicates mesenteric fat around the head of the intussusception.¹⁸ CECT is currently considered the most sensitive radiologic method to make a preoperative diagnosis. It reveals characteristic "bowel within bowel appearance" and "target sign" (Figure 10 A and B) which consist of outer intussusciens, inner intussusceptum, and central fat density formed by the intussuscepted mesenteric fat and mesenteric vessels. In our study, computed tomography was carried out in both patients and it was diagnostic of intussusception. Also in both cases, the original lead points were established on CECT.

The first line of treatment in villous tumours is to excise using an endoscopic approach. Endoscopic snare resection, laser ablation, or endoscopic mucosal resection (EMR) are some of the methods advocated. However, the feasibility of endoscopic removal depends on the size and location of the tumour. Surgery is indicated for large tumours. Traditional open procedures include local and wedge resections or gastrectomies. New surgical techniques include laparoscopic or combined laparoscopic-endoscopic approaches with or without intra-organ surgery. The results of these methods are comparable to that of open surgery. The advantages of these new methods of treatment include reduced pain, shorter duration of recovery, better immune responses, and earlier discharge.¹⁹

Laparotomy is indicated only when endoscopic treatment is impossible or if there is an emergency setting. When surgery is indicated, polypectomy should be performed through enterotomy, while intestinal resection should be reserved for rare cases to avoid short bowel syndrome in the long run. Overall, duodenoduodenal intussusception is a challenging condition to manage, requiring prompt recognition and appropriate intervention to prevent serious complications such as bowel obstruction, ischemia, or perforation. However, with advances in diagnostic imaging and surgical techniques, outcomes have improved significantly in recent years. These case reports aim to add to the small body of research reporting on this topic.

CONCLUSION

The presented cases of duodenoduodenal and duodenojejunal intussusception secondary to tubulovillous adenoma and hamartomatous polyp (Peutz-Jeghers polyposis) underscore the importance of recognizing these rare but potentially serious conditions, especially in pediatric patients. The diagnostic journey involved a combination of clinical suspicion, diagnostic imaging modalities including ultrasonography and contrast-enhanced computed tomography, and definitive histopathological examination following surgical resection. Continued vigilance and collaborative efforts

are crucial for ensuring timely diagnosis and optimal outcomes for patients with these rare entities.

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