

Case Report

A case of retrograde jejuno-duodenal intussusception caused by jejunal lipoma: a case report and review of literature

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ABSTRACT

Adult intussusception is a rare entity, comprising 5% of all cases of intussusception. The presentation of adult intussusception commonly varies, ranging from chronic abdominal pain to acute presentation mimicking symptoms of bowel obstruction. This report describes a case of retrograde jejuno-duodenal intussusception in a 48-year old healthy male with a history of several episodes of diffuse abdominal pain and an acute episode of melena of 3 days duration. Examination revealed a soft, nondistended abdomen with palpable mass at upper abdomen associated with tenderness on palpation. Computed tomography scan findings revealed a 10cm length of jejuno-duodenal intussusception likely secondary to a jejunal lipoma. Exploratory laparotomy revealed resolved intussusception secondary to a lipoma which was treated with segmental intestinal resection. In this case report, we describe a rare case of retrograde jejuno-duodenal intussusception caused by a jejunal lipoma and performed a literature review on retrograde intussusception.

Keywords: Duodenojejunal, Retrograde, Intussusception, Jejunal lipoma

INTRODUCTION

Intussusception is defined as the telescoping of a segment of the gastrointestinal tract (the intussusceptum) within the lumen of the adjacent segment (the intussusciptens).¹ Adult intussusception is a rare entity, representing 5% of all cases of intussusception and accounts for 1%-5% of intestinal obstruction in adults.² There are two types of intussusception- anterograde intussusception and retrograde intussusception.¹ Anterograde intussusception is more common than retrograde intussusception, and it is classified according to location (enteroenteric, ileocolic, ileocecal, or colocolic) and cause (benign, malignant or idiopathic).² In adult intussusception, it can be further classified based on the presence of a lead point.² In adults, most of the intussusception is caused by a lead point and require surgery, whereas in the pediatric population most of it is primary and benign, and it is treated by either pneumatic or hydrostatic reduction.

In adult intussusception with a lead point, benign tumor constitutes majority of the cases observed in the small intestine, whereas in colon it is commonly associated with malignancy.³

Retrograde intussusceptions are extremely rare and represent 1% of all types of intussusception.¹ It is an antiperistaltic invagination of a distal bowel lead point through a more proximal segment of bowel, and it is most commonly reported after gastric resection and gastrojejunostomy.⁴ It is believed to be caused by altered peristalsis in focal areas, leading to dysrhythmic contractions and causing retrograde intussusception.⁵ However, the exact mechanism triggering retrograde intussusception is not known. The presentation of retrograde intussusception varies with location, presenting with acute esophageal obstruction in gastroesophageal intussusception, to symptoms of acute pancreatitis seen in jejuno-duodenal intussusception.^{6,7} A

few cases of retrograde jejuno duodenal intussusception have been described in the literature, with its etiology most likely due to post-operative complications or placement of gastrostomy tubes, complication from duodenojejunoscopy, polyp or jejunal duplication cyst.⁷⁻¹¹ In this case report, we report a rare case of retrograde jejuno duodenal intussusception caused by a jejunal lipoma.

CASE REPORT

In this case report, we are reporting a 48 years old male presenting with abdominal pain for 4 days with melena for 3 days. He described his pain as diffuse in location and intermittent in nature, exacerbated by eating. He does not have any loss of weight, loss of appetite, weakness, fatigue, nausea or vomiting. Medical history and surgical history are otherwise not significant. Family history is negative for malignancies. Physical examination revealed a well appearing, well-nourished male with no acute distress. Vital signs include a blood pressure of 132/64 mmHg, heart rate of 86 beats per minute, respiratory rate of 15 breaths per minute and temperature of 37°C. Abdominal examination is significant for tenderness in epigastrium and upper abdomen, with a possible sausage-like mass in upper abdomen. There was no guarding or rebound tenderness, and bowel sounds were normoactive.

Complete blood count, complete metabolic panel, coagulation profile and tumor markers such as CEA and CA 19-9 were checked, and was significant for hypokalaemia. Computer aided tomography of the abdomen and pelvis revealed intussusception of the small bowel within the left upper quadrant, involving the distal duodenum and proximal jejunum, measuring approximately 10 cm in length. A 2.8 x 2.1 cm fat density mass was noted in the distal aspect of the intussuscepted bowel that might represent a lipoma. EGD/Enteroscopy revealed a large, polypoid, friable ulcerated mass in the proximal jejunum just beyond the ligament of Treitz (Figure 1, 2, 3). The lesion was marked with blue dye for surgical resection.

The patient underwent an exploratory laparotomy 2 days later with intraoperative finding of resolved intussusception and a mass located 30 cm distal from ligament of Treitz measuring approximately 5cm in size. Segmental resection of the tumor was done with 5cm margins at the proximal and distal end, and the small bowel was connected with side-to-side staple anastomosis. Histopathological finding of the specimen revealed polypoid submucosal lipoma of 4.5cm x 2.3cm with reactive changes consistent with intussusception. The patient was discharged from hospital 4 days postoperatively with no further complications.

DISCUSSION

The first report of intussusception was made in 1674 by Barbet of Amsterdam.¹² It is defined as the telescoping

of a proximal segment of the gastrointestinal tract into an adjacent distal segment.¹³ Adult intussusception is a rare entity in adults, comprising around 5% of all cases of intussusception and for 1-5% of intestinal obstructions in adult. The overall incidence of intussusception in adults is around 2-3 cases/1000,000 population/year.¹⁴ The male-to-female ratio is approximately 3:1 in children, but the prevalence is equal in adults.¹⁴

Majority of the small bowel intussusception in adults are caused by benign lesions (60%), followed by malignancy (30%) and idiopathic causes (10%).¹⁵ Of lesions that cause intussusception, site of intussusception and presence of anemia (hemoglobin <12) are independent predictors of malignancy.¹⁶



Figure 1: Computed tomography revealing a lesion intussusception of the small bowel with the characteristic “target sign” appearance involving the duodenum.



Figure 2: Computed tomography showing intussusception of the small bowel in the left upper quadrant.

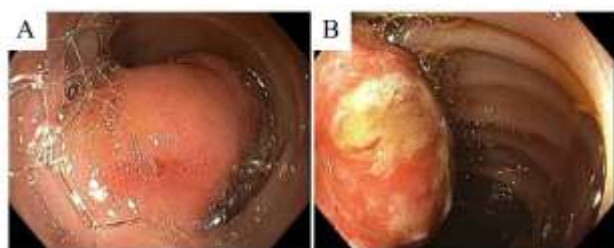


Figure 3: A) Polypoid mass measuring approximately 5cm in size noted in proximal jejunum just beyond ligament of Treitz. B) Polypoid mass appears friable with surface ulceration.

Table 1: Types of retrograde intussusception and its associated risk factors/etiology.

Types of retrograde intussusception	Associated risk factors/ Etiology
Gastro-esophageal	Hiatal hernia ²⁶ Achalasia ²⁷ Congenital obstructing lesions: malrotation with Ladd’s bands, superior mesenteric artery syndrome, anular pancreas ²⁶ Eating disorders complicated by moderate to severe intractable obesity ²⁶ Physical exertion ²⁶ Idiopathic
Duodeno-gastric	Complication from placement of gastrostomy tube ²⁰
Duodeno-deuodenal	Complication of duodenal ulcer ²⁸
Jejuno-duodenal	Complication from duodeno-jejunoscopy ⁷ Complication from placement of gastrostomy tube ⁹ Polyp ¹⁰ Jejunal duplication cyst ¹¹
Enteric	Idiopathic ²⁹ Foreign bodies : roundworm ²⁹ Neoplasms: polyp, secondary melanoma ²⁹ Meckel’s diverticulum ²⁹ Complication from Roux-en-Y gastric bypass ³⁰
Ceco-ileal	Neoplasms: lymphosarcoma, adenocarcinoma ²⁹
Colonic	Neoplasms : benign polyp, malignant papilloma, pedunculated lipoma ²⁹
Jejuno-gastric	Complication from gastrojejunoscopy and Bilroth II reconstruction ³¹ Complication from gastrostomy tube ⁴

Retrograde intussusception

Retrograde intussusception is defined as telescoping of the distal segment of the gastrointestinal tract into the adjacent proximal segment.¹⁷ They are extremely rare and have been reported to occur in less than one percent of intussusceptions of all types.¹ The types of retrograde intussusceptions are listed in Table 1. The exact pathophysiology of retrograde intussusception is not exactly known, but it is believed to be caused by reverse peristalsis secondary to distal obstruction in the presence of bowel fixation by extrinsic inflammatory or neoplastic processes.¹ In our patient, it is likely that retrograde intussusception is caused by the lipoma, which triggers anti-peristaltic activity, and this phenomenon has been described in previous literature.¹⁸

With the duodenum acting as a relative fixed point, it results in invagination of the proximal portion of the bowel by the distal bowel. Causes and associated risk factors for the different types of retrograde intussusception described in the literature are listed (Table 1). Currently, 4 cases of jejunoduodenal intussusception have been described in the literature, caused by a hamartomatous polyp from duodenum, jejunal duplication cyst complication from placement of gastrostomy tube, and complication from duodenojejunoscopy.⁷⁻¹¹ Retrograde jejuno-duodenal intussusception due to a jejunal lipoma has not been reported in the literature.

Presentation of retrograde intussusception

The presentation of retrograde intussusception varies depending on the location of intussusception, with gastroesophageal intussusception presenting with symptoms of acute esophageal obstruction, and duodenogastric intussusception presenting with signs and symptoms of gastric outlet obstruction.^{19,20} For jejunoduodenal intussusception, the presentation includes abdominal pain, vomiting, acute pancreatitis, and symptoms of biliary obstruction.

Diagnosis of retrograde intussusception

For diagnosis of intussusception, CT scan is the radiological investigation of choice, with a sensitivity of 71.4%-87.5% and a specificity of 100% in the prospect of diagnosis of intussusception.¹⁴ CT features of intussusception include: 1) Target like mass in which the inner central area represents the invaginated intussusceptum that is surrounded by the thick-wall intussusciens. 2) Oral contrast material trapped between the opposing walls of the intussusceptum and intussusciens. 3) A soft tissue mass secondary to intussusception, possibly with a lead point telescoping into the intussusciens. 4) Bowel wall thickening or intramural air may be seen in case of blood supply restriction.²¹ Other modalities have also been used to diagnose intussusception, such as barium studies,

abdominal ultrasonography, and endoscopic approaches such as EGD, enteroscopy, and colonoscopy.¹⁴

Currently, the findings of retrograde enteric intussusception could not be fully diagnosed on computed tomography and sonography. However, they can be diagnosed via barium study showing “reverse claw sign”, which represents part of the sheathing portion of the intussusciens filling but not the lumen of the intussusceptum.¹ Based on the limitation of the imaging modalities, it is not expected that the confirmed diagnosis of retrograde jejuno duodenal intussusception be made using CT scan, as the appearance of “target sign” cannot reliably distinguish between antegrade and retrograde intussusception. The diagnosis of retrograde intussusception should be made with the combination of image study and the clinical finding.

Management of retrograde intussusception

In the management of small bowel intussusception, it is important to differentiate between intussusception that is self-limiting and cases that necessitate surgery, as increased use of CT scans led to detection of asymptomatic intussusception that is self-limiting. Intussusception of length that is less than 3.5cm is predictive of spontaneous resolution; hence it is likely to be self-limiting and not requiring surgery.²² Lvoff et al has concluded that due to increased detection of self-limiting small bowel intussusception with CT scans, it is important to note that intussusception with length of less than 3.5cm is more likely to be self-limiting and not requiring surgery.²² In the presence of a coexisting surgical pathology, surgical resection is usually indicated, with frozen section of the specimen obtained to exclude malignancy.^{23,24} In retrograde intussusception, spontaneous resolution is more likely due to antegrade peristalsis, which accounts for the resolution of intussusception in our patient during laparotomy.²⁵ As retrograde intussusception has higher likelihood of spontaneous resolution, we tattooed the site of lesion which was visible during endoscopy, allowing easier identification of the site of lesion intraoperatively.

CONCLUSION

Retrograde intussusception is a rare condition in adult intussusception which is difficult to diagnose preoperatively due to the limitations of current imaging modalities. The management of retrograde intussusception is similar to antegrade intussusception, and site-marking of the lesion preoperatively will facilitate easier identification of the lesion intraoperatively.

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REFERENCES

1. Nunez D, Bjarnason T. Barium findings in retrograde intussusception. *Am J Roentgenol.* 1975;125:715-22.
2. Marinis A, Yiallourou A, Samanides L. Intussusception of the bowel in adults: A review. *World J Gastroenterol.* 2009;15:407-11.
3. Barussaud M. Clinical spectrum and surgical approach of adult intussusceptions: a multicentric study. *Int J Color Dis.* 2006;21.
4. Gasparri M, Pipinos I, Kralovich K. Retrograde jejuno gastric intussusception. *South Med J.* 2000;93:499-500.
5. Pavalkis SP. In *Current Concepts in Colonic Disorders* (ed. Lule DG.) (InTech, 2012). doi: Saulius Paskauskas and Dainius Pavalkis (2012). *Adult Intussusception, Current Concepts in Colonic Disorders*, Dr. Godfrey Lule (Ed.), In Tech, DOI: 10.5772/25416. Available from: <http://www.intechopen.com/books/current-concepts-in-colonic-disorders/adult-intussusception>.
6. Cohen HJ, Weiss G. Gastroesophageal intussusception. *J Pediatr.* 2016;34:367-70.
7. Chen LE, Bhalla S, Warner BW. Retrograde jejuno duodenal intussusception: a rare cause of acute pancreatitis after surgery for duodenal atresia. *J Pediatr Surg.* 2008;43:E31-3.
8. Rangunath K, Robers A, Senapati S. Retrograde jejuno duodenal intussusception caused by a migrated percutaneous endoscopic gastrostomy tube. *Dig Dis Sci.* 2004;49:1815-7.
9. Ciaccia D, Quigley R, Shami P. A case of retrograde jejuno duodenal intussusception caused by a feeding gastrostomy tube. *Nutr Clin Pract.* 1994;9:18-21.
10. Ozer A, Sarkut P, Ozturk E. Jejuno duodenal intussusception caused by a solitary polyp in a woman with Peutz-Jeghers syndrome: a case report. *J Med Case Rep.* 2014;8:13.
11. Norton KI, Luhmann KC. Retrograde jejuno duodenal intussusception associated with a jejunal duplication cyst in a newborn. *Pediatr Radiol.* 1993;23:360-1.
12. Yalamarthi S, Smith RC. Adult intussusception: case reports and review of literature. *Postgrad Med J.* 2005;81:174-7.
13. Agha F. Intussusception in Adults. *Am. J. Roentgenol.* 1986;146:527-31.
14. Potts J, Samarae A. Al. Small bowel intussusception in adults. *Ann R Coll Surg Engl.* 2014;96:11-4.
15. Renzulli P, Candinas D. Idiopathic Small Bowel Intussusception in an Adult. *Can Med Assoc J.* 2010;182:148.
16. Cera SM. Intestinal Intussusception. 2008;1:106-13.

17. Gupta RK, Agrawal CS, Yadav R, Bajracharya A., Sah PL. Intussusception in adults: Institutional review. *Int J Surg.* 2011;9:91-5.
18. Groper MJ. Retrograde enteric intussusception. *Ann Surg.* 1940;112:344-51.
19. Lukish JR, Eichelberger MR, Henry L, Mohan P, Markle, B. Gastroesophageal intussusception: a new cause of acute esophageal obstruction in children. *J Pediatr Surgery.* 2004;39:1125-7.
20. Osuntokun B, Falcone R, Alonso M, Cohen MB. Duodenogastric Intussusception: A rare cause of gastric outlet obstruction. *J Pediatr Gastroenterol Nutr.* 2004;39:299-301.
21. Charalambous G, Katergiannakis V, Manouras A. Jejunojejunal Lipoma Causing Intussusception. *Case Rep Gastroenterol.* 2012;6:684-8.
22. Lvoff N, Breiman RS, Coakley FV. Distinguishing Features of Self-limiting Adult Small-Bowel Intussusception Identified at CT. *Radiology.* 2003;227.
23. Catalano O. Transient Small Bowel Intussusception. *Br J Radiol.* 1997;70:805-8.
24. Charalambous G, Katergiannakis V. Jejunojejunal Lipoma Causing Intussusception. *Case Rep. Gastroenterol.* 2012:684-688.
25. Kavia S, Charig M, Desai A. Adult retrograde intussusception. *Clin Radiol Extra.* 2004;59:59-60.
26. Gowen GF, Stoldt HS, Rosato FE. Five risk factors identify patients with gastroesophageal intussusception. *Arch Surg.* 1999;134(12):1394-7.
27. Wong MD, Davidson SB, Ledgerwood AM, Lucas CE. Retrograde gastroesophageal intussusception complicating chronic achalasia. *Arch Surg.* 1995;130(9):1009-10.
28. Kefeli A, Basyigit S, Yeniova AO, Uzman M, Aktas B. Retrograde duodenoduodenal intussusception: An uncommon complication of peptic ulcer. *Chin Med J. (Engl).* 2015;128:2981.
29. Akehurst AC. Retrograde intussusception. A report of a case and a review of 103 cases in the literature. *Br J Surg.* 1955;43:207-13.
30. Simper SC, Erzinger JM, McKinlay RD, Smith SC. Retrograde (reverse) jejunal intussusception might not be such a rare problem: a single group's experience of 23 cases. *Surg Obes Relat Dis.* 2008;4:77-83.
31. Singh S, Singh A, Bhagat S, Singh B. Retrograde jejuno-gastric intussusception. *Niger J Surg.* 2015;21:70-2.

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