

Challenges in managing duodenal intussusception: A rare cause of gastric outlet obstruction in adults

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ABSTRACT

Intussusception is a rare occurrence in adults and only 5% being adults. Ileocolic intussusception is the most common type in adults whereas duodenal intussusception (DI) is the rarest subtype due to natural retroperitoneal fixation of the duodenum. There are only a few case-reports available in the literature. Here, we aimed to present our experience of five cases with DI in adults, there operative challenges and strategies to deal with the same. Age of presentation ranged from 18-45 years, and four out of five were females. Most common presentation was partial of recurrent gastric outlet obstruction (GOO) along with weight loss. They were all diagnosed on contract-enhanced-CT scan of abdomen, and all had a lead point in form of a benign polypoidal mass arising from duodenum. Two patients were managed with local excision of polyp and retroperitoneal fixation of redundant duodenum, whereas pancreaticoduodenectomy and segmental duodenal resection was required for the other two patients. One patient was lost to follow-up without definitive treatment as was minimally symptomatic. All four operated patients had uneventful recovery post-operatively and are doing well in follow-up. DI is an infrequent occurrence and a rare etiology of GOO in adults. Most of them having benign lead point, require surgical excision of lead point with fixation of duodenum to retroperitoneum. Anatomical proximity of important structures like CBD and pancreatic duct especially at perivaterian location makes surgical resection challenging. With thorough anatomical knowledge and appropriate management, this condition can be managed well.

Keywords: Duodenal intussusception, duodenal polyps, intussusception, gastric outlet obstruction

INTRODUCTION

Intussusception, i.e., invagination of a portion of the intestine into another segment in a telescope-like fashion. It is a rare occurrence in adults, with an incidence of 5% and represents 1-5% of all causes of small bowel obstruction in adults (1,2). Duodenal intussusception (DI) is an exceedingly rare entity due to its retroperitoneal fixation and only a few case reports are available in the literature, mostly describing a single case (3,4). It is a challenging situation due to rarity, nonspecific symptoms, and difficult anatomic considerations. Here, we aimed to present our experience with DI in adults, operative challenges, and strategies to deal with the same (Table 1).

Case 1

Thirty-year-old female patient presented with recurrent episodes of postprandial colicky left upper abdominal pain, distension and bloating sensation for two months with significant unintentional weight loss, but no anorexia. She had an episode of obstructive jaundice, which resolved spontaneously with resolution of pain. Physical examination did not reveal any significant findings. Her serum alkaline phosphatase level was elevated (700 IU/L). Magnetic resonance cholangiopancreatography (MRCP) revealed medial stretching of the duodenum, common bile duct (CBD), and pancreas, as well as elongated duodeno-jejunal (DJ) flexure. Bilobar intrahepatic biliary radicals and CBD were dilated. Further imaging with contrast enhanced computer tomography (CECT) confirmed DI, but lead point was not identified. Ultrasonography aided in visualization of the polypoidal mass as lead point. On exploration, duodenum and proximal jejunum were dilated with an intussuscepted bowel segment into the proximal jejunum in left upper abdomen. After reduction, duodenotomy was made on lateral wall of the 2nd part

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Table 1. Patient profile							
Age (Years)	Sex	Presentation	Lead Point Location	Surgery	Histopathology	Post-op	Outcome (Follow-up Period)
30	Female	Recurrent GOO, Jaundice, Weight loss	D2*	Local resection of polyp and duodenal fixation to retroperitoneum	Adenomatous polyp	Uneventful	Doing well (six months)
45	Female	Recurrent GOO	D2*	Pancreaticoduodenectomy	Hamartomatous polyp	Uneventful	Doing well (two years)
32	Male	Recurrent GOO, Weight loss	D4*	Segmental resection of intussuscepted bowel with polyp	Adenomatous polyp	Uneventful	Doing well (three years)
18	Female	Recurrent GOO, Weight loss	D2*	Local resection of polyp and duodenal fixation to retroperitoneum	Hyperplastic polyp	Post-op gastroparesis, resolved with conservative management	Doing well (five years)
27	Female	Mild dyspeptic symptoms	D1*	Denied surgery	Adenomatous polyp	Denied surgery	Lost to follow up
*D: Duodenum, Number: Part of duodenum.							

of duodenum (D2). Lead point was a 6 x 5 cm perivaterian polyp. Since the opening of the papilla was not clearly defined, cholecystectomy was done, and papilla was identified by flushing saline via cystic duct, which was 1.5 cm away from polypoidal mass. Excised polypoidal mass, including 1 cm margin all around was sent for frozen section and reported as tubulo-villous adenomatous polyp low-grade dysplasia with clear margins. Duodenotomy was closed, and duodenum was fixed to the retroperitoneum. Post-operative course was uneventful. Oral contrast study on 5th-post-operative-day (POD) revealed no leakage of contrast and free flow from duodenum to the jejunum. The patient was discharged on the 7th-POD. She is doing well at six months of follow-up (Figure 1A-H).

Case 2

Forty five-year-old female patient presented with recurrent episodes of postprandial abdominal bloating and fullness for 10 years. Palpation revealed a 5 x 4 cm non-tender lump in right hypochondrium. Ultrasonography suggested possibility of intussusception with a polypoidal lead point. Further evaluation with CECT revealed intussusception of D2 along with pancreatic head, dilated CBD and pancreatic duct (PD), into the $3^{\rm rd}$ and $4^{\rm th}$ parts of the duodenum (D3, D4), reaching up to DJ flexure. A 5 x 4 cm polyp was noted as lead point. On exploration, stomach and D2 were distended, with DI noted in right upper abdomen. After reduction of intussusception, duodenotomy was made and a large polypoidal mass noted arising from medial wall of papilla acting as lead point. CBD was dilated. As the mass was arising from ampulla, pancreaticoduodenectomy

was performed. Histopathological examination (HPE) revealed a hamartamatous polyp. Post-operative course was uneventful, and the patient was discharged on 10th-POD. The patient is doing well after two years of follow-up (Figure 2A-C).

Thirty two-year-old male patient had recurrent episodes of small amounts of bilious vomiting 2-3 hours after food intake for one year, along with significant unintentional weight loss, but no anorexia. Physical examination was normal. CECT revealed distended stomach and D2, along with DJ intussusception. On exploration, DJ-flexure was found intussuscepting retrogradely into D3 and D4. Segmental resection of the intussuscepted bowel loop was done, and continuity restored with side-to-side duodenojejunal anastomosis. On cutting the specimen, a solitary sessile polyp measuring 4.5 x 4 cm was noted in the D3, 3 cm away from the resected ends. HPE revealed adenomatous polyp with lowgrade dysplasia. Post-operative course was uneventful, and the patient was discharged on 6th-POD. The patient is doing well after three years of follow-up.

Case 4

Eighteen-year-old female patient had recurrent episodes of postprandial epigastric pain, fullness, large volume gastric vomiting, and melena every 1-2 months for 2-3 days for two years and suffered significant unintentional weight loss without anorexia. Physical examination was normal. Oesophagogastroscopy revealed large lobulated growth arising close to the papilla; biopsy was inconclusive. Further evaluation with CECT

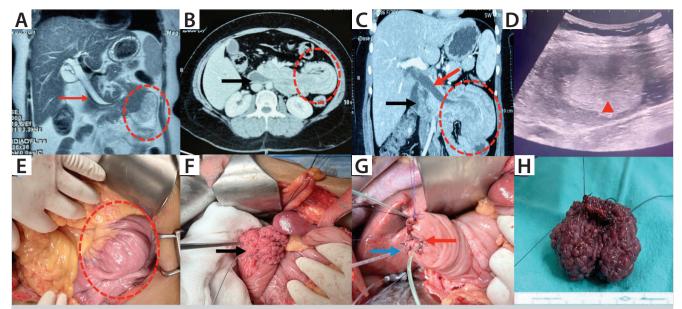


Figure 1. Case 1: **A.** Magnetic resonance cholangiopancreatography (MRCP) Coronal section depicts an elongated, dilated CBD (red arrow) tapering towards left of midline with central intrahepatic biliary radical dilatation and a suspicious clumped up bowel loops (marked with dotted circle). **B.** Computed Tomography (CT) scan of the abdomen, axial section of suggests dilated 2nd part of duodenum (black arrow), collapsed 3rd part of duodenum with duodenojejunal intussusception (dotted circle). **C.** Coronal section of the CT scan suggests dilated common bile duct (red arrow) pulled across midline to left side, dilated 2nd part of duodenum (black arrow), collapsed 3rd part of duodenum with duodenojejunal intussusception (marked by dotted circle). **D.** Ultrasonography of the abdomen showing the polypoidal mass as lead point (red arrow head). On exploration **E.** Intussusception of duodenum was noted into jejunum (red dotted circle), intussusception was reduced. **F.** A polypoidal mass in perivaterian region (black arrow) was identified as lead point on lateral duodenotomy. **G.** Choledocoduodenostomy done. **H.** Gross specimen of the excised polyp.

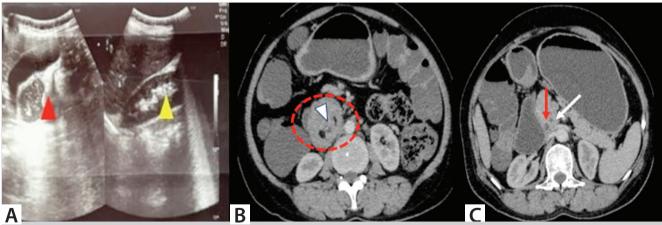


Figure 2. Case 2: **A.** Ultrasonography of the abdomen showing the polypoidal mass as lead point (red arrow head) and target sign (yellow arrow head) suggesting intussusception of bowel. **B.** Axial section of the CT scan suggests duodenoduodenal intussusception (dotted circle) and the lead point (white arrow head) **C.** Further sections on CT scan suggest dilated CBD (red arrow) and pancreatic duct (white arrow).

revealed DI in D2 and prominent CBD. On exploration, 1^{st} part of the duodenum (D1) was dilated, and DI was noted from the D2 up to the proximal jejunum, which was reduced, and duodenotomy made on lateral wall of D2. Papilla and a perivaterian polypoid growth of size 3 x 3 cm were identified. Polyp was excised circumferentially, with a 5 mm margin safeguarding the papilla. Duodenotomy closed, and the redundant duodenum was fixed to the retroperitoneum and parities. HPE revealed

hyperplastic polyp. Postoperatively, the patient had gastroparesis, requiring medical management. The patient tolerated solid food by 10th POD and was discharged on 13th-POD. At five years of follow-up, the patient is doing well (Figure 3A,B).

Case 5

Twenty seven-year-old female patient was evaluated for primary infertility and was found to have possibility of small

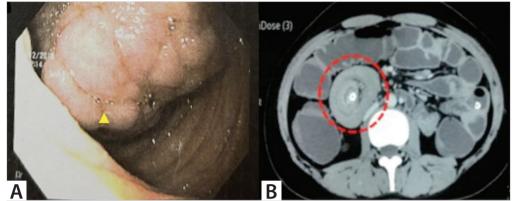


Figure 3. Case 4: A. Oesophagogastroscopy revealed a large lobulated growth (arrow head) arising close to the papilla. **B.** Axial section of the CT scan shows target sign suggesting duodenoduodenal intussusception (dotted circle) with NJ in situ.

bowel intussusception. Other than mild dyspeptic symptoms, she was asymptomatic. CECT revealed distended stomach and herniation of D1 into the D2 and D3, with a pedunculated polyp arising from the D1 as lead point. Oesophagogastroscopy confirmed the presence of 4 x 6 cm pedunculated polyp. HPE revealed an adenomatous polyp with low-grade dysplasia. The atient was offered surgical management, but she denied as she had minimal symptoms and was lost to follow-up (Figure 4).

DISCUSSION

Intussusception, disease of the childhood, occurs with a frequency of only 5% in adults (5). Ileocolic intussusception is the most common, whereas DI is the rarest subtype in adults (6). It is probably due to retroperitoneum fixity of the duodenum and rarity of mass in this region (7,8). In adults, DI is

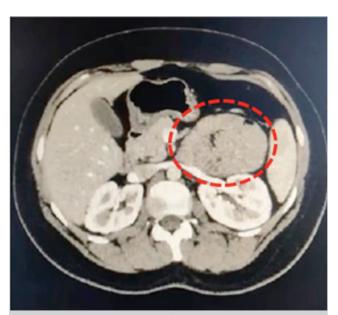


Figure 4. Case 5: Axial section of the CT scan shows target sign suggesting duodenoduodenal intussusception (dotted circle).

frequently seen in young and middle-aged women (9). In our series, the age ranged from 18-45 years, with a median of 31 years, and the female-to-male ratio was 4:1. Usual presentation is with features of recurrent or partial gastric outlet obstruction, including colicky abdominal pain, bloating, nausea, and vomiting, which may vary from being subtle to severe in intensity, as seen in our series as well (10). Biliary and pancreatic systems are generally not involved unless the mass is located in perivaterian region, dragging the CBD and PD along with intussusceptum, leading to their dilatation, which may or may not manifest clinically (11). In our series, four patients had features of recurrent GOO, while one had minimal dyspeptic symptoms. Additionally, three patients had dilated CBD; however, only one had obstructive jaundice due to perivaterian tubulovillous adenomatous polyps dragging the CBD into intussusception. Another patient had palpable lump on physical examination. Patients may also present with history of weight loss due to long-term partial or recurrent GOO, as in our series, three out of five patients had significant weight loss. Diagnosing DI is important as there is high risk of life-threatening complications associated with it, such as ischemia, obstruction, or intraluminal bleeding, requiring emergency treatment. Due to non-specific and often chronic or recurrent symptoms, diagnosis is usually missed or delayed. Hence, clinicians should be suspicious of intussusception if presentation is of partial or recurrent GOO, pancreatitis, or obstructive jaundice along with weight loss, in young females. CECT is the most reliable investigation for preoperative diagnosis (sensitivity: 71.4-87.5%; specificity: 100%) for DI showing 'target' or 'doughnut' signs, as well as a sausage-shaped mass or pitchfork image (12). In our series, all patients were diagnosed preoperatively with CT scan. Additionally, CT also helps in identification of lead-point lesions. A well-defined lead point is seen in >90% of adult intussusception (Al), most of which are benign (>70%) (13). Lead point is present in almost all cases of DI in the form of

polypoidal mass, which may differ in histopathology. We could identify benign polyps as lead points in all cases. Three were tubulo-villous adenomatous polyps with low-grade dysplasia; one was hamartamatous polyp, and another one was hyperplastic polyp. Next investigation offered is upper GI endoscopy to find out the lead point, if possible, with biopsy to confirm histopathology. Although there is no consensus guideline for managing DI, surgical management is the preferred treatment modality considering the presence of lead point in most cases and chances of associated complications (9,14). Anatomical fixity and location of lead point close to ampulla in DI, makes surgery challenging. Perivaterian mass, located within 2 cm of the papilla, have an increased risk of CBD and PD injury. In general, a widely accepted approach is the reduction of the intussusception, excision of lead point if benign, and fixation of redundant segments of bowel to avoid recurrence (15). In case of suspected or known malignancy, radical excision is required. Most of the lead points in adult enteric intussusception are benign and only <30% are malignant (16). Hence, a more conservative surgical approach should be chosen to prevent mortality and morbidity of major resections after confirming benign pathology on frozen section histopathology. In our series, two patients underwent reduction of intussusception with local excision of polyp, one patient having polyp arising from papilla, abetting local polypectomy, underwent pancreaticoduodenectomy and one required segmental duodenal resection for polyp arising from D3. All patients had smooth post-operative course and are doing fine in follow-up, except one whose further course is not known, as patient lost follow-up without definitive treatment.

CONCLUSION

Duodenal intussusception is an infrequent occurrence and a rare aetiology of gastric outlet obstruction in adults. Most of them have a benign lead point, requiring surgical excision of the lead point with fixation of the duodenum to the retroperitoneum. Anatomical proximity of important structures like CBD and pancreatic duct especially at perivaterian location makes surgical resection challenging. With thorough anatomical knowledge and appropriate management, the condition can be managed well.

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OLGU SERİSİ-ÖZET

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Duodenal intussusepsiyon tedavisinde zorluklar: Yetişkinlerde gastrik çıkış obstrüksiyonunun nadir bir nedeni

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ÖZET

İntussepsiyon/invajinasyon yetişkinlerde nadir görülen bir durumdur ve sadece %5'i yetişkinlerde görülür. İleokolik invajinasyon erişkinlerde en sık görülen tip iken duodenal invajinasyon (Dİ) duodenumun doğal retroperitoneal fiksasyonu nedeniyle en nadir görülen alt tiptir. Literatürde sadece birkaç olgu sunumu mevcuttur. Burada yetişkinlerde Dİ'li beş olguya ilişkin deneyimlerimizi, karşılaşılan ameliyat zorluklarını ve bunlarla başa çıkma stratejilerini sunmayı amaçladık, Başvuru yaşları 18-45 arasında değişmekteydi ve beş olgunun dördü kadındı. En şık başvuru şekli kilo kaybı ile birlikte kısmi tekrarlayan gastrik çıkış obstrüksiyonu (GÇO) idi. Hepsine kontrastlı karın bilgisayarlı tomografisi taramasında tanı konuldu ve hepsinde duodenumdan kaynaklanan beniqn polipoidal kütle seklinde bir öncü nokta vardı. İki hasta polipin lokal eksizyonu ve duodenumun retroperitoneal fiksasyonu ile tedavi edilirken, diğer iki hasta icin pankreatoduodenektomi ve segmental duodenal rezeksiyon gerekti. Bir hasta minimal semptomatik olduğu için kesin tedavi uygulanamadan takipten çıktı. Ameliyat edilen dört hastanın tümü ameliyat sonrası sorunsuz iyileşti ve takiplerinde sorun görülmedi. Dİ nadir görülen bir durumdur ve yetişkinlerde GÇO'nun nadir bir etiyolojisidir. Çoğu benign sürükleyici lezyona sahip olan bu hastalarda, duodenumun retroperitona fiksasyonu ile birlikte sürükleyici lezyonun cerrahi eksizyonu gerekmektedir. Özellikle ampulla cevresi lokalizasyonda koledok ve pankreatik kanal gibi önemli yapıların anatomik yakınlığı cerrahi rezeksiyonu zorlastırmaktadır. Kapsamlı anatomik bilgi ve uygun yönetim ile bu durum iyi bir şekilde yönetilebilir.

Anahtar Kelimeler: Duodenal intussusepsiyon, duodenal polipler, intussusepsiyon, gastrik çıkış obstürksiyonu

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