

Case Report

Duodenal intussusception due to periampullary duodenal adenoma: A case report

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ABSTRACT

Duodenal intussusceptions are uncommon due to their fixed location in retroperitoneum except when associated with bowel malrotation abnormalities. We present a case of recurrent duodenojejunal intussusception in a 34-year-old female diagnosed on imaging. The lead point, in our case, was periampullary duodenal tubulovillous adenoma which was confirmed on endoscopy and subsequent punch biopsy. The patient underwent segmental duodenal resection anastomosis preserving the ampulla with reduction of the intussusceptum and fixation of the jejunal loops and its mesentery.

Keywords: Duodenal intussusception, Periampullary adenoma, Computed tomography, Magnetic resonance imaging

CASE REPORT

A 34-year-old woman presented to our hospital with complaints of recurrent episodes of epigastric fullness and vomiting after meals. These symptoms gradually improved spontaneously after few hours of meal intake. There was no significant history of illness in past or similar complaints in family members. Physical examination revealed pallor but no significant tenderness or palpable lump in the abdomen. Blood analysis showed moderate microcytic anemia (hemoglobin level 7 g/dL) and mildly raised total bilirubin level ~1.2 mg/dL (direct bilirubin was 1.5 mg/dL). Rest of the liver function tests was normal (serum alanine transaminase levels – 32 IU/L and aspartate aminotransferase levels – 36 IU/L). Serum alkaline phosphatase level was raised slightly, 400 IU/L (normal: 100–300 IU/L) and amylase was not elevated. Further, investigations revealed the cause of anemia to be iron deficiency anemia, and hence, ultrasound abdomen was done to look for cause of cholestasis. It revealed prominent common bile duct (CBD) (Diameter 11 mm); however, no cause of obstruction was seen. Magnetic resonance cholangiopancreatography (MRCP) was performed to look for cause of obstruction which showed long segment duodenojejunal intussusception. There was leftward shift of the head of pancreas, CBD, and the pancreatic duct causing proximal dilatation of CBD. No obvious lead point was identified [Figure 1]. Contrast-enhanced computed tomography (CECT) confirmed the findings of the MRI; however, still the lead point could not be identified. There was no evidence of malrotation or bowel ischemia [Figure 1]. Upper gastrointestinal endoscopy showed a well-defined polypoidal mass with thin stalk arising from the medial wall of the duodenum approximately 2 cm from the ampulla in the caudal direction. No intussusception was seen at the time of endoscopy [Figure 2]. Punch biopsy was taken from the lesion which suggested tubulovillous adenoma on pathological examination

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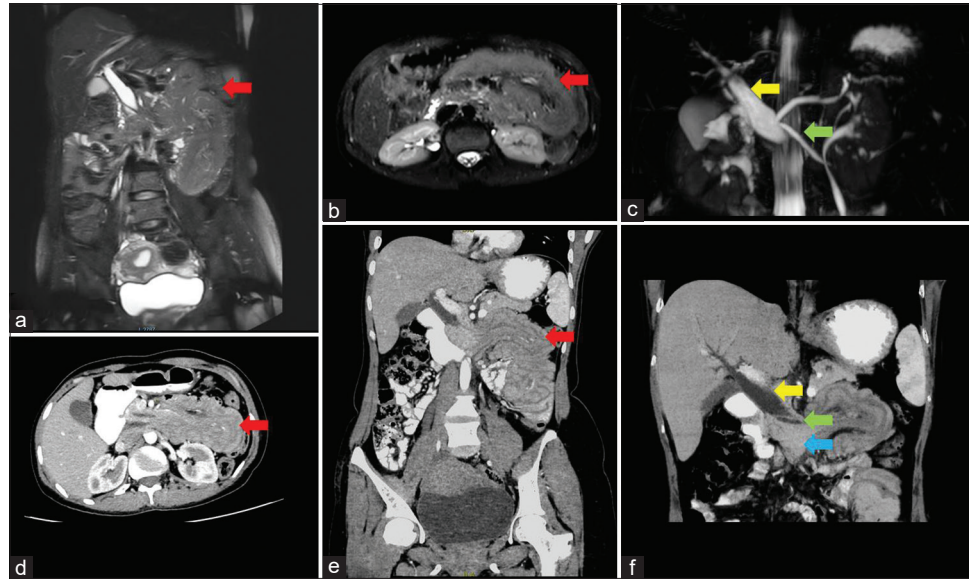


Figure 1: Coronal and axial, MRI, and CT images show telescoping of the duodenum into jejunum (red arrows in a, b and d, e). MRCP and MinIP CT images show dilated common bile duct (yellow arrow in c and f), prominent pancreatic duct directed toward left (green arrows in c and f), and head of the pancreas pulled leftward (blue arrow in f). MRI: Magnetic resonance imaging, CT: Computed tomography, MRCP: Magnetic resonance cholangiopancreatography, and Min IP: Minimum intensity projection.

[Figure 2]. The patient underwent segmental duodenal resection-anastomosis preserving the ampulla with reduction of the intussusceptum and side-to-side fixation of the jejunal loops and its mesentery. The patient was discharged weeks after the operation with no complaints.

Histological examination of the specimen demonstrated a $5 \times 4.2 \times 3$ cm polypoidal mass arising from D2 segment which showed tubulopapillary growth resting on fibrocollagenous core and lined by tall columnar mucin secreting epithelium showing nuclear stratification. No invasion into stroma or fibrous core was seen suggestive of tubulopapillary adenomatous polyp. The tumor resection margins were all clear and there was no lymph node involvement.

DISCUSSION

Intussusception is the phenomenon of invagination of the proximal bowel segment into the immediate distal bowel associated with long mesentery, causing free mobility of the intestine.^[1,2] Duodenal intussusception is unusual to occur because the second and third parts of duodenum are retroperitoneal and fixed in location. It can, however, occur in cases with malrotation.^[3] Previously described case reports of duodenal intussusception, not associated with malrotation,^[4,5] mention common misinterpretation of distal migration of duodenal tumors with mucosal prolapse as intussusception.^[3] It is a process of elongation of mucosa

and formation of stalk at the base of tumor which migrates distally along with tumor with forward peristalsis, leading to deformation of the walls of the distal bowel segment.

Classically, imaging diagnosis of intussusception has been described on barium studies as “coiled spring sign” or a “target sign” on CT/MRI or ultrasonography. Mucosal prolapse can mimic all these signs even without actual occurrence of intussusception.^[1,6] For a definitive diagnosis of intussusception CT is a better modality as it demonstrates invagination of proximal bowel into distal bowel along with its mesentery (containing fat, mesenteric vessels and nodes).^[1] There have been various cases reporting duodenal intussusception secondary to duodenal tumors such as adenoma, gastrointestinal stromal tumors,^[7] duodenal duplication cysts and Brunner’s gland hamartomas, carcinoid, and few secondary to ulcers.^[1,8,9] Our case describes the classical imaging findings of duodenal intussusception in a patient being worked up for cholestasis as described on MRI and CT findings. In addition to invagination of mesenteric fat with intussusceptum, there was invagination of ampulla and pancreatic parenchyma in the region of head causing dilatation of the CBD and pancreatic duct without malrotational abnormality.

Endoscopic and intraoperative findings in our case suggested true intussusception, along with mucosal prolapse. The tumor was identified in the D2 segment of duodenum in medial wall 2 cm caudal to the ampulla. When the tumor migrated distally with peristalsis, mucosa, duodenal wall, and the structures

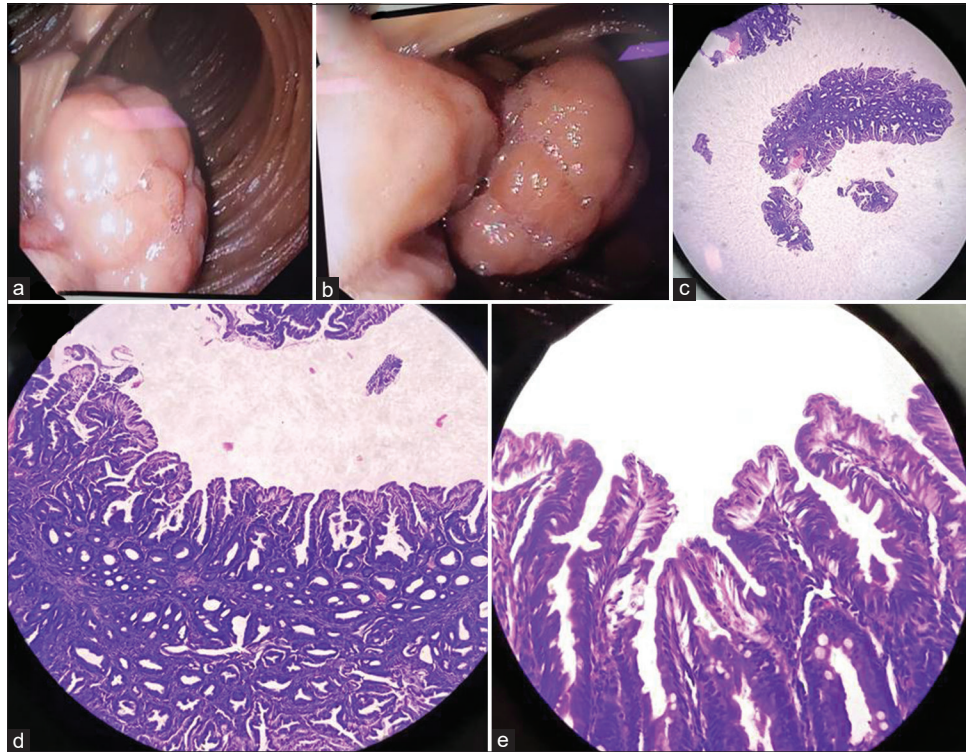


Figure 2: Endoscopic images show lobulated polypoidal lesion arising from medial wall of duodenum in enface (a) and lateral view (b). H&E, ×4 Fragmented punch biopsy from polypoidal lesion in duodenum shows cellular fragment showing tubular and villous architecture (c), H&E, ×10 (d), and ×40 (e) shows higher magnification of cellular fragment comprising of tubulopapillary structure having thin columnar epithelial cells with dysplastic epithelium with cells showing nuclear hyperchromasia. Mild nuclear elongation and focal areas of nuclear stratification and active mitosis. Mucin secretion is preserved in apical portion of epithelial cells.

surrounding the ampulla were all deviated toward the left-lower direction, also suggesting intussusception along with mucosal prolapse. As the enveloping mucosa elongates distally with peristalsis, wall of the duodenum and proximal jejunum also become deformed. The intussusception, however, does not last for a long time and reduces spontaneously, as in this case, causing intermittent symptoms which aggravate after meal. Bowel ischemia was not present in our case. CECT as well as MRI could not identify the polypoidal tumor as lead point in our case.

The exact pathogenesis of the condition is not clear; however, there is associated component of retrograde traction which occurs in addition to the invagination progresses due to lack of mobility of duodenum and pancreas.^[1] As documented in the literature in various case reports as well as in our case, the repeated episodes of intussusception cause intermittent compression of CBD and pancreatic duct causing ductal dilation which lead to cholestasis. If left untreated, it can progress to frank jaundice and can lead to episode of recurrent pancreatitis. It is important to note that the ductal dilation is not caused due to the tumor invasion but due to recurrent mechanical obstruction. In few cases, severe anemia was seen

due to minor chronic bleeding from the tumor;^[1] in our case, however, it was attributable to iron deficiency anemia. Thus, in patients with duodenal intussusception, it is important to carefully assess pancreaticobiliary symptoms in addition to complaints secondary to bowel obstruction.

It is generally believed that adult duodenal intussusception should be treated by surgery without delay. The surgical procedures used for duodenal intussusception include endoscopic resection and laparotomy or laparoscopy. Only in cases where the lesions of the duodenum are limited to the mucosal layer and where the extent of the lesions is <2 cm can endoscopic resection be performed. When the lesion exceeds the mucous layer and the range is more than 2 cm, surgical resection is recommended. In addition, in regard to idiopathic intussusception, simple reduction is recommended after excluding intestinal ischemic necrosis and intestinal perforation.^[10] The choice of operation depends on the situation observed during the operation. The presence of a malignant tumor or the complete resection of the tumor will affect the function of the ampulla, and pancreaticoduodenectomy may be an appropriate choice.^[11] In our case, the polypoidal tumor was excised performing a segmental duodenal

resection anastomoses was done preserving the ampulla along with reduction of the intussusception. A side to side fixation of the jejunal loops was also done to prevent similar episodes in future. A few case reports mention performing pancreaticoduodenectomy as well as surgical ampullectomy in cases, where the adenoma was arising from the ampulla itself to avoid the risk of injury to pancreatic parenchyma;^[10] however, preserving the ampulla was a major surgical challenge in our case and appropriate distance from the ampulla made selective resection anastomoses possible in our case. The case also highlights the importance of surgical approach in cases with duodenal intussusception, where ampulla preservation significantly decreases the post-operative morbidity, and hence, when possible ampulla preserving surgical approach should be adopted in such cases.

CONCLUSION

Duodenal intussusception can occur secondary to pathologies in duodenum such as tumors or ulcers even in the absence of malrotation abnormality. It can also be associated with mucosal prolapse. Pancreaticobiliary symptoms in addition to bowel obstruction should be looked for in patients with duodenal intussusception. Imaging with ultrasound followed by cross-sectional imaging is essential for appropriate patient management.

TEACHING POINTS

1. Duodenal intussusception is usually impossible due to its fixed retroperitoneal location unless associated with bowel malrotation (absent in our case)
2. Imaging is important to look for the lead point incases with intussusception and condition of the intussusceptum for evidence of any signs of obstruction, mesenteric, or bowel ischemia.

MCQs

1. Following can be the presentations of duodenal intussusception due to periampullary lesion?
 - a. Pancreatitis
 - b. Malena
 - c. Jaundice
 - d. All of the above

Answer Key: d

2. Signs of intussusception of imaging include all except?
 - a. Coiled spring sign on Barium study
 - b. Target sign on CT
 - c. Pseudokidney sign on ultrasound
 - d. Thumbprinting sign on Barium study

Answer Key: d

3. Pathogenesis of duodenal intussusception includes all except?
 - a. Bowel malrotation
 - b. Tumor

- c. Aphthous ulcers
- d. Large ulcer

Answer Key: c

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

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