



Case report

Intussusception of the third portion of the duodenum secondary to a primary duodenal malignancy: A case report

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ARTICLE INFO

Keywords:

Adult
Duodenum
Intussusception
Duodenal cancer
Case report

ABSTRACT

Introduction and importance: Intussusception is invagination of the bowel and is rare in adults, representing only 5% of all cases of intussusception. Duodenal intussusception is also very rare. To the best of our knowledge, there is only one previous report of a malignant tumor in the fourth portion of the duodenum as the lead point of intussusception (Vaibhav et al., 2021 [1]). This reports a duodenal-duodenal intussusception caused by a malignant tumor in the third portion of the duodenum.

Case presentation: A 36-year-old woman with abdominal pain was diagnosed with duodenal intussusception by abdominal ultrasonography and computed tomography scan. Double balloon endoscopy showed that the intussusception had spontaneously reduced, and a tumor was found in the third portion of the duodenum. Open resection was performed because of the tumor location. Pathologic examination revealed adenocarcinoma with an adenoma of the duodenum. The patient had no evidence of recurrence for 4 years after resection.

Clinical discussion: This patient had a primary duodenal malignancy and presented with intussusception. Duodenal intussusception is a rare condition requiring prompt intervention, usually requiring surgery. In this patient, since endoscopic resection was difficult, partial resection of duodenum was performed by open surgery assisted by double balloon enteroscopy. The pathological diagnosis of the resected specimen was primary duodenal malignancy.

Conclusion: This is a report of adult duodenal intussusception caused by a primary duodenal malignancy. This rare condition posed an intriguing challenge for the optimal operative approach.

1. Introduction

Intussusception involves the full thickness of the bowel wall invaginating into distal bowel. Intussusception is common in children and rare in adults, accounting for only 5% of all cases of intussusception [2]. In addition, due to the fixed retroperitoneal location of the duodenum, duodenal intussusception is particularly rare [3].

The lead point of an intussusception is usually a Meckel's diverticulum, a Peutz-Jeghers polyp or a benign or malignant tumor [4]. The present report describes an extremely rare duodenal-duodenal intussusception in which the lead point is a primary malignancy in the third portion of the duodenum. This report is in line with the SCARE criteria [5].

2. Presentation of case

A 36-year-old woman with an intellectual disability and 16 years status post cholecystectomy presented to an outside facility with abdominal pain. Physical examination showed tenderness in the epigastric region but no palpable masses and normal bowel sounds. Abdominal ultrasonography and computed tomography scan examinations showed that the third portion of the duodenum had a concentric circular area with a target sign and a 3.5 × 3.5 cm mass distally, consistent with intussusception (Fig. 1). She was referred for further evaluation and treatment. Laboratory tests were within normal limits. A double balloon endoscopy examination showed that the intussusception had spontaneously reduced, and a polyp was seen in the third portion of the duodenum. A biopsy of the polyp showed a tubule-villous adenoma

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<https://doi.org/10.1016/j.ijscr.2022.107085>

Received 10 February 2022; Received in revised form 11 April 2022; Accepted 11 April 2022

Available online 13 April 2022

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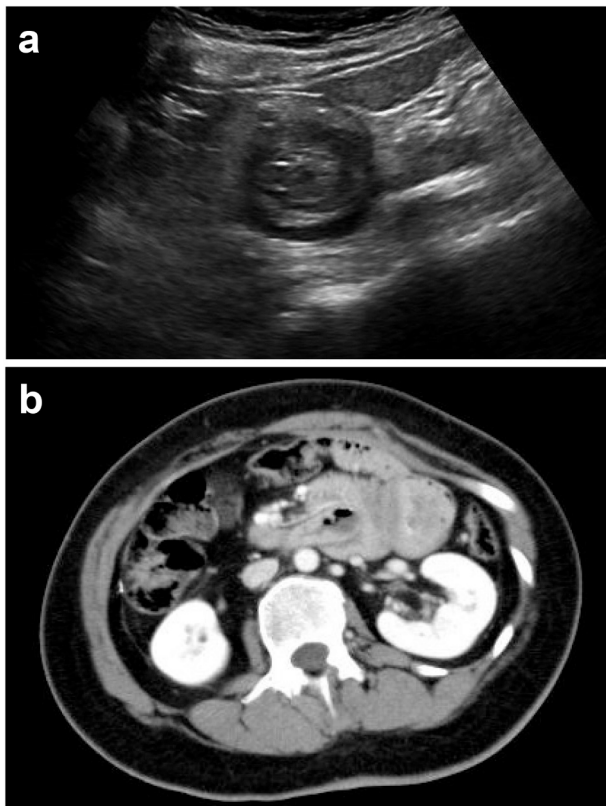


Fig. 1. Ultrasonography and computed tomography imaging.
 (a) Target sign.
 (b) Intussusception and tumor.

(Fig. 2).

Since endoscopic resection was complicated due to the location of the lesion, open resection was performed. Surgery began with intraoperative double balloon enteroscopy. Resection of the tumor was attempted using double balloon endoscopy under general anesthesia, but it was not considered safe to continue and open resection undertaken. An incision was made around the ligament of Treitz, and after mobilization, a 5 cm enterotomy created in the proximal jejunum to identify the tumor while performing double balloon endoscopy. The tumor was pedunculated and removed with a margin (Fig. 3). The enterotomy was closed in two layers, and the integrity of the closure confirmed by endoscopy. The patient made an uneventful recovery and was discharged on postoperative day 10. Postoperative pathology revealed adenocarcinoma in an adenoma of the duodenum (type1, 46 × 25 × 25 mm, tub1, pT2, ly0, v0, pHM0, pVMO) (Fig. 4). The patient is currently seen in follow-up at an outpatient clinic, with no evidence of recurrence four years after resection.

3. Discussion

Intussusception is rare in adults and duodenal intussusception is particularly rare. Duodenal intussusception rarely occurs because the duodenum is relatively fixed in the retroperitoneum [3]. For that reason, intussusception is often associated with malrotation [6] and the lead point is usually a Meckel's diverticulum, a benign tumor such as a hamartoma, an adenoma or stromal tumor [4]. There are reports of intussusception due to benign adenomas [7,8], but the present patient had a primary duodenal malignancy. There is one report of a malignant duodenal tumor in the fourth portion as the lead point of an intussusception and segmental duodenal resection was performed [1]. The tumor identified in the present patient was in the third portion and we resected the tumor with a margin. Intussusception of the horizontal part of the duodenum is particularly rare [1,8].

Proximal small bowel intussusception is generally transient and does not result in obstruction [9]. Duodenal intussusception is unusual because of limited mobility of the duodenal wall [3]. The typical clinical

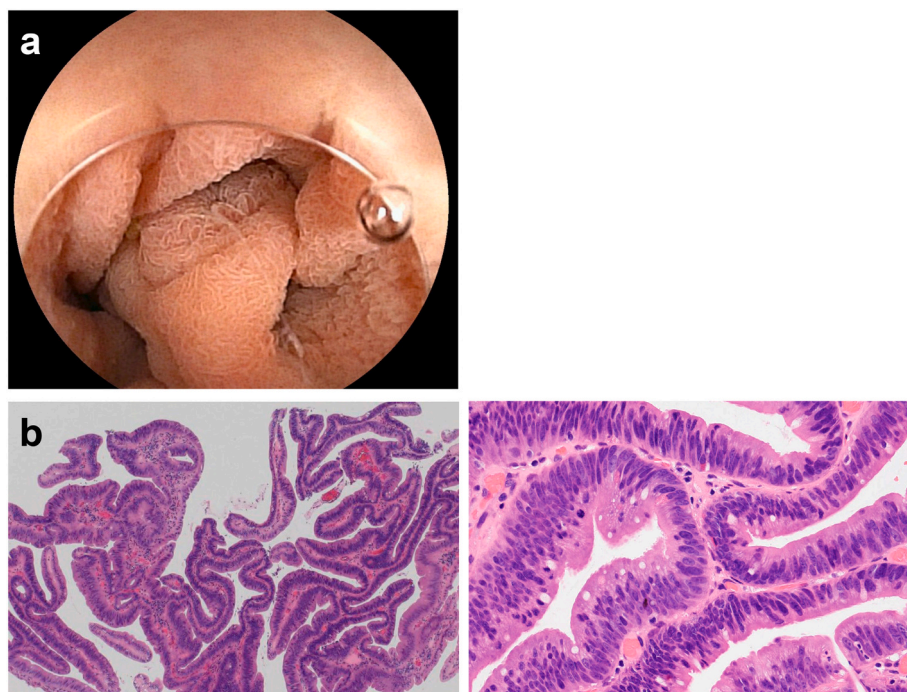


Fig. 2. Double balloon endoscopy examination.
 (a) Intussusception, third portion of the duodenum.
 (b) Pathology, tubulovillous adenoma.

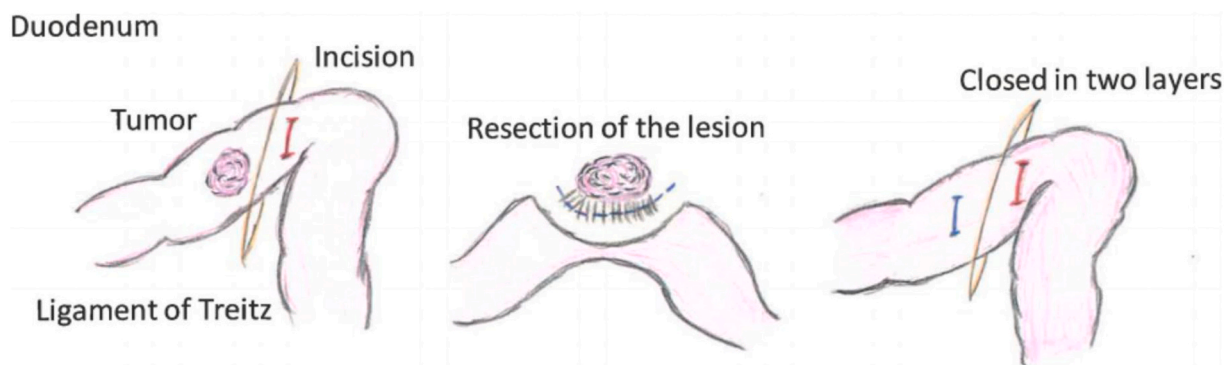


Fig. 3. Surgical procedure.

Resection of the lesion was performed through an enterotomy and closed in two layers.

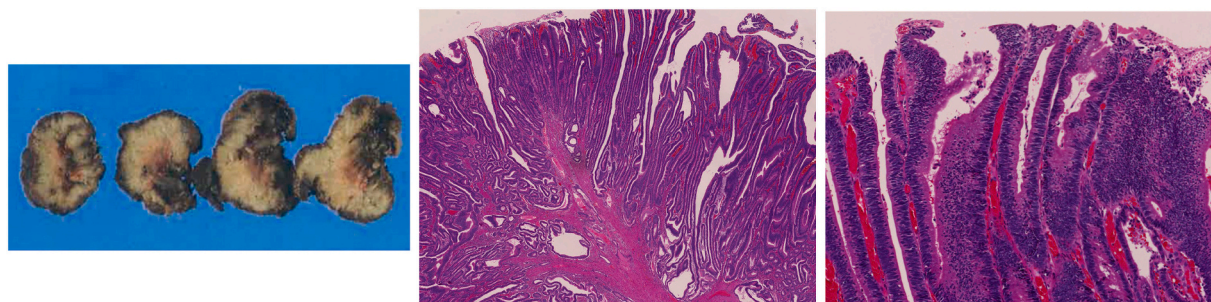


Fig. 4. Adenocarcinoma with adenoma of the duodenum.

Postoperative specimen and pathology. (type1, 46 × 25 × 25 mm, tub1, pT2, ly0, v0, pHM0, pVM0).

manifestation of duodenal intussusception is intestinal obstruction [10]. Some patients may present with anemia, low fever, bloody stool, weight loss or abdominal masses [11]. Abdominal computed tomography scan is the most reliable modality for the diagnosis of intussusception. It shows the presence of typical bowel telescoping signs, described as “target”, “doughnut”, or “sausage-shaped” signs [12]. This patient initially presented with abdominal pain as the main symptom, but it resolved. However, it was useful because we were able to identify the target sign by ultrasonography and computed tomography scan at the time of onset, which led to the diagnosis.

Duodenal intussusception can lead to obstruction so intervention should be prompt [13]. It is generally believed that duodenal intussusception in adults should be treated by surgery without delay [14]. The surgical procedures used for duodenal intussusception include endoscopic resection and laparotomy or laparoscopy. In this patient, since endoscopic resection was difficult, the tumor was resected through an enterotomy assisted by double balloon enteroscopy. We did resection of the lesion because a biopsy before operation showed adenoma. Pathologic examination of the specimen showed a primary malignant tumor of the duodenum. The most common surgical procedure performed for duodenal malignancies with invasion deeper than the mucosa is pancreaticoduodenectomy. However, it has been reported that the 5-year survival rates of pancreaticoduodenectomy and local duodenal resection for duodenal tumors are similar, and the incidence of postoperative complications is relatively high after pancreaticoduodenectomy [15–17]. This patient has been followed up without evidence of recurrence for 4 years after resection.

4. Conclusion

Intussusception in adults is rare, and intussusception in the duodenum is even more rare. In adults, intussusception most often occurs due to a physical lesion, so intussusception with advanced duodenal

cancer is possible. To the best of our knowledge there are no previous case reports of intussusception in the third portion of the duodenum secondary to a primary duodenal malignancy.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Sources of funding

None.

Ethical approval

Ethical approval has been given by our institution.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

All authors have contributed equally for interpretation of data, writing and editing to the submission.

Research registration

Not applicable.

Guarantor

Hirokazu Matsuura, Naohiro Sata.

Declaration of competing interest

None.

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