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Case Report

Neuroendocrine Tumor, A Rare Cause of Adult Intussusception: A Case Report

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Abstract Adult intussusception is a rare condition occurring only in about 5% of adults. When it occurs, it usually involves a pathological lead point, which may be benign or malignant. The malignant conditions are commoner, with adenocarcinoma taking the lead. Preoperative diagnosis is usually challenging due to the absence of the classical signs, among which is the passage of red-currant jelly stool that is present in children. We present a 49-year-old man with ileocolic intussusception as a result of a neuroendocrine tumor.

Keywords: Neuroendocrine tumor, Adult intussusception

Introduction

Relative to the pediatric age group, where intussusception is only second to acute appendicitis as a cause of acute abdomen,[1] its occurrence in adults is rare, and the diagnosis is also challenging.[2,3] Adult intussusception makes up about 5% of all cases of intussusception and about 1% of the causes of bowel obstruction.[4] The etiology of intussusception in adults may involve pathologic lead points, and these are mainly from tumors that may be malignant or benign. They may include carcinomas, Meckel's or colonic diverticuli, and sometimes polyps.[5,6] Intussusception may lead to intestinal obstruction, bowel ischemia, perforation, and sometimes peritonitis. In children, however, the majority of the cases of intussusception have no specific cause.[7] The clinical and radiological diagnoses are also challenging pre-operatively in the adult because the classic triad of abdominal pain, bleeding per rectum, and a palpable abdominal mass are rarely seen. Despite the availability of imaging techniques, there are still missed diagnoses.[8,9] Affordability and accessibility are equally contending issues. Surgery is traditionally considered the primary treatment option, and this can be through open or laparoscopic procedures where the facilities are available.[10,11] Surgical exploration is needed to ascertain the cause and to exclude an underlying tumor. However, there is no consensus about indications for intra-operative reduction and the extent of intestinal resection.[12,13]

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We present a case of adult intussusception (AI) with a pathologic lead point that was histologically confirmed as a neuroendocrine tumor.

Case Report

A 49-year-old male presented with a four-day history of colicky abdominal pain located at the right lower abdomen and radiating toward the back. There is usually an associated mass noticed by the patient at the peak of the abdominal pain, with some relief when the mass reduces spontaneously. He had episodes of vomiting that contained recently ingested meals. There was no abdominal distension and no passage of bloody or mucoid stool. He has had several episodes of similar pain in the past that were relieved with the use of analgesics and intravenous fluids, and sometimes, the pain resolved spontaneously. No co-morbidities and no family history of similar condition.

On examination, he was conscious and alert, in painful distress, not pale, and afebrile (T-36.9°C), but had tachycardia with a pulse rate of 104 per minute. The abdomen was full and moved with respiration. There was tenderness and rebound tenderness with guarding at the right iliac fossa and a vaguely palpable mass found at the right iliac fossa. Digital rectal examination was unremarkable. He had leucocytosis with a white cell count of 13,000/mm^3, and the serum electrolytes were essentially normal. Abdominopelvic ultrasonography was suggestive of ileocolic intussusception with the presence of the target sign. He had an emergency laparotomy. Intraoperatively, an ileocolic intussusception, which was difficult to reduce, was found (Figure 1A). However, further attempts achieved a reduction, which revealed a palpable pale mass about 1 × 1 cm, about 6 cm from the ileocaecal junction (Figure 1B). He had a right hemicolectomy with end-to-end ileocolic anastomosis. Postoperative recovery was uneventful, and he was discharged on postoperative day 7.

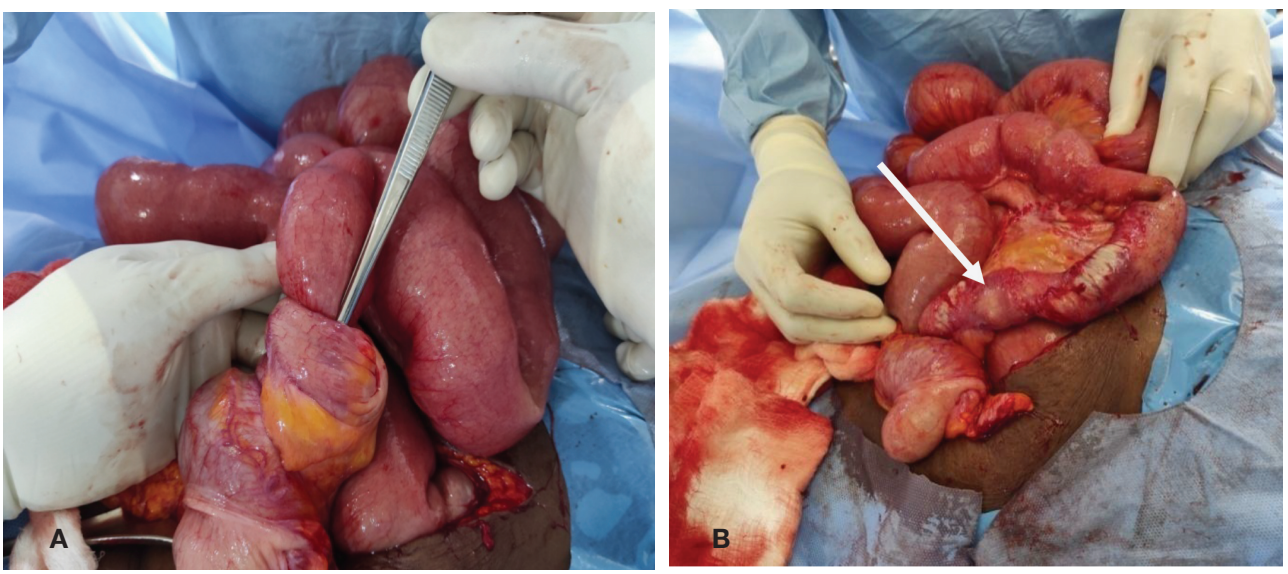

Figure 1 (A) Ileum looping into the caecum and ascending colon.
(B) Shows a mass at the terminal ileum, about 6 cm from the ileocaecal junction.

The histopathologic diagnosis was well-differentiated neuroendocrine carcinoma, as several sections from the ileal mass showed extensive ulceration of the lining of the epithelium by malignant neuroendocrine cells. It was infiltrated by sheets and nests of small round cells with scanty cytoplasm, hyperchromatic nuclei with stippled chromatin patterns, and indistinct nucleoli. Atypical mitotic figures and apoptotic bodies were noted. Tumour cells were seen invading the subserosa, and there are many areas of lymphovascular invasion. The resection margins were free. (Figures 2A and 2B). Immunohistochemistry was positive for chromogranin A (Figure 3).

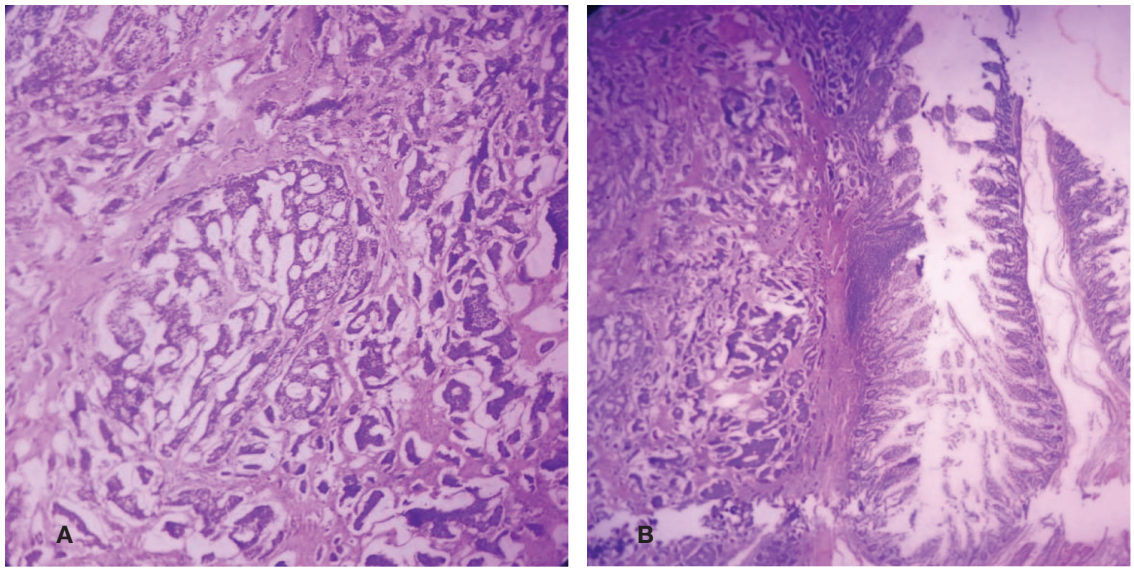

Figure 2 (A) Photomicrograph shows a nest of tumor cells infiltrating through the submucosa to the subserosa. Haematoxylin and Eosin (H&E) stain × 40
(B) Photomicrograph shows the muscular propia and subserosa invasion by tumor cells with stippled chromatin nuclei pattern. Haematoxylin and Eosin (H&E) stain × 400

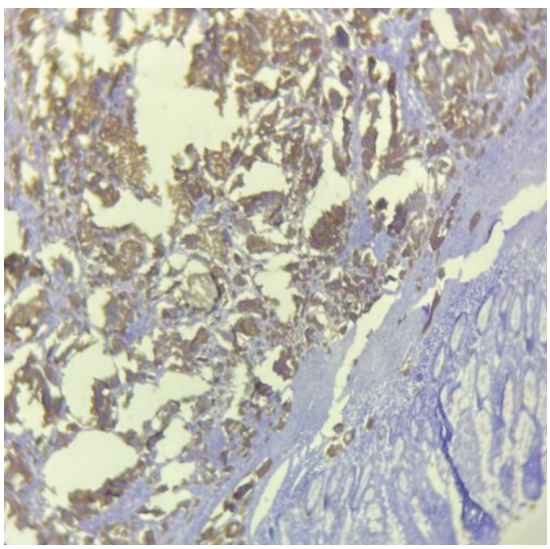

Figure 3 Chromogranin A (CgA) positive tumor cells invading the submucosa × 100

The patient has been on regular follow-up without any complaints.

Discussion

The telescoping of a segment of the bowel into an adjacent one is described as intussusception.[14] A large percentage of adult intussusception occurs in the small and large bowel, while about 10% occur in the stomach or a surgically created stoma.[15] Compared to children, intussusception presents with the classical triad of intermittent abdominal pain, vomiting, and red currant jelly stool, which are not pathognomonic to adult intussusception. Adults may present with non-specific symptoms, which may be acute or chronic, and sometimes with features suggestive of intermittent bowel obstruction.[16] As seen in our patient, the most common symptom is abdominal pain, which may be intermittent vomiting with or without associated nausea.[17] These symptoms may linger from days to years, buttressing the challenge of preoperative diagnosis of adult intussusception.[18]

Radiological investigations play a pivotal role in the preoperative assessment of patients. Abdominal ultrasound may reveal classical images such as target sign or pseudo-kidney sign that may be highly suggestive of intestinal invagination.[18] Computed tomography (CT) scan of the abdomen has also proven helpful in diagnosing intussusception, as it may show a central intussusceptum and an outer intussuscipiens with satisfactory accuracy. However, it has limited value in distinguishing a benign from a malignant or idiopathic lead point.[19] Many studies have reported that more than half of them are malignant. The most common being adenocarcinoma, carcinoid tumors, and leiomyosarcoma.[14,20]

The clinical diagnosis is usually confirmed at surgery, which may be done as open laparotomy or laparoscopy. Laparoscopic surgery for adult intussusception is feasible, safe, and preferred in experienced hands.[21] The choice of either laparoscopic or open technique should, therefore, be based on the clinical status of the patient as well as the surgeon's ability to perform the procedure safely and effectively.

The patient in this report had open laparotomy and reduction of the intussuscipiens followed by resection of the bowel with a rare form of lead point. There is no consensus about what should be done at surgery: en bloc resection against initial reduction followed by a more limited resection.[12,13] Reduction at surgery is thought to help avoid unnecessary bowel resection. Although theoretically, it may increase the risk of potential intra-luminal seeding or venous tumor embolization.[22]

The outcome of surgery in adult intussusception is generally good. However, the long-term prognosis depends on whether the underlying causative pathology is malignant or benign.[23]

Conclusion

Adult intussusception as a result of neuroendocrine tumors is not a common occurrence in day-to-day clinical practice, and the non-specific symptoms it presents with further make the diagnosis difficult. Although computed tomographic scan is the investigation of choice in diagnosing AI, which is not easy to access, abdominal ultrasonography is of benefit. Surgical resection and histological diagnosis are still the preferred and definitive treatment choice.

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