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## Multiple synchronous adenocarcinomas of the small bowel in a young patient: A case report

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## ABSTRACT

**INTRODUCTION:** Adenocarcinoma of the small bowel is a rare neoplasm presented usually in elder patients as a single tumor. Its presentation as multiple tumors and in young patients is exceptional and there aren't any guidelines to orient its therapy.

**PRESENTATION OF CASE:** We present the rare case of a sixteen-year-old woman that presents to the emergency department with an intussusception due to a small bowel tumor. The resected specimen showed multiple adenocarcinomas. A complete endoscopic and PET-CT study showed other 5 lesions from the duodenum to the ileum that were resected. Genetic counseling showed no pathogenic changes. The final staging was T2N0M0 and only surveillance was indicated. The patient is now 3 years without any recurrence.

**DISCUSSION:** Multiple adenocarcinomas of the small bowel are a very infrequent presentation of the disease. Most common risk factors include Crohn disease and adenomas. Its presentation is usually vague with a delay in its diagnosis. The treatment remains mainly surgical with limited use of adjuvant therapy. The most important prognostic factor is lymph node involvement with 5-year survival that can range from 3%–60% depending on the stage.

**CONCLUSION:** This case represents an exceptional presentation of a very rare pathology with few cases described in the literature. There isn't one single best study to stage the patient and surgery is still the standard of treatment while adjuvant therapies studies are being conducted. The young age and lack of predisposing factors or mutations leaves an open field for investigation.

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## 1. Introduction

This case report is in line with the SCARE guidelines for surgical case reports [1]. Small bowel malignancy is an uncommon condition comprising no more than 2% of all tumors of the gastrointestinal tract. Its annual incidence is approximately 13.9 cases per million persons. Clinical presentation is often vague and non-specific delaying diagnosis [2].

The treatment of small bowel adenocarcinomas (SBA) is primarily surgical with limited role in adjuvant therapy. Most cases commonly present as single tumors in elder patients; however, we

present a rare case of multiple adenocarcinomas of the small bowel in a young patient treated in our academic private institution.

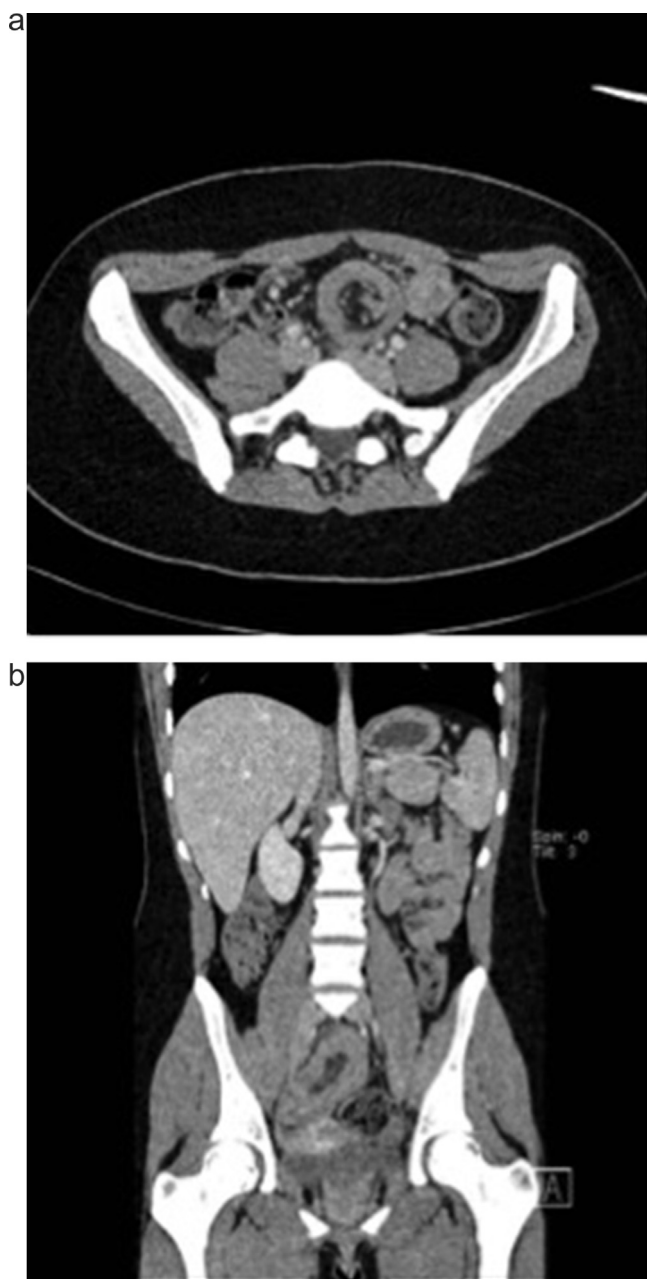
## 2. Presentation of the case

A 16-year-old female without personal or family history of digestive diseases, cancer or genetic disorders presented to the emergency department. She complained of increasing diffuse abdominal pain over 12 h without nausea, vomiting, diarrhea or weight loss. She was afebrile, hemodynamically stable, with moderate diffuse abdominal tenderness and distention. A computerized tomography showed an intussusception of the small bowel without any other findings (Fig. 1a and b).

The patient underwent laparoscopic exploration. There was no free fluid, liver lesions or carcinomatosis. A jejunum-ileum intussusception was seen and a small laparotomy was performed. Multiple small polypoid tumors in the jejunum were the cause of the intussusception and 25 cm of small bowel were resected

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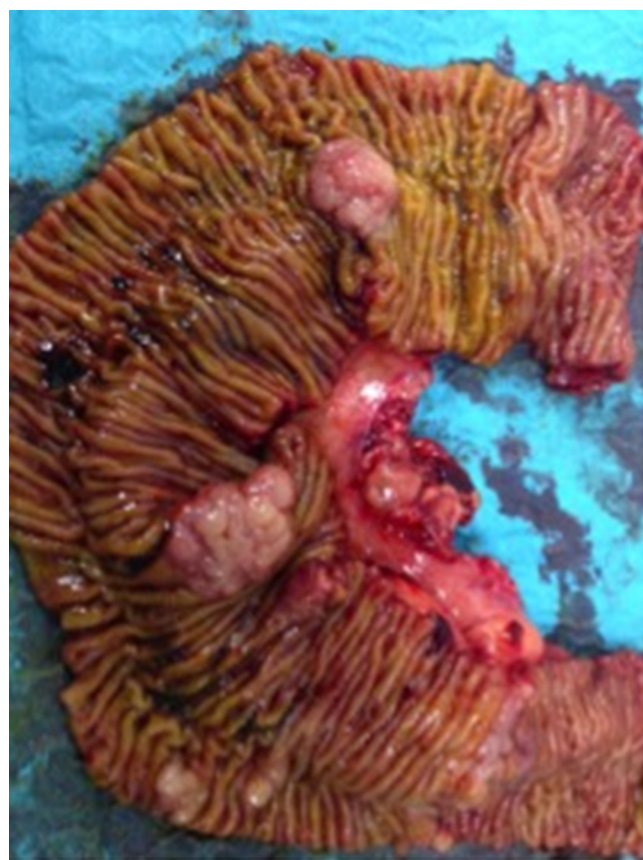


**Fig. 1.** Abdominal CT with i.v. contrast (a) axial and (b) coronal view that shows small bowel thickening associated with an intussusception.

including all the lesions in the specimen without an extended lymphadenectomy and an end-to-end anastomosis was performed (Fig. 2).

Biopsy showed a mucinous adenocarcinoma, an epithelial tumor according the WHO classification of tumors of the small intestine, in 2 tumors of 4,3 cm and 3 cm and well-differentiated tubular adenocarcinoma in 10 tumors of 0,5 cm–1 cm. The tumors infiltrated up to the muscular layer without vascular or perineural penetration. All the tumors of the specimen resected had a wide free margin and one harvested lymph node was free of malignancy.

Afterwards, the patient was controlled with an upper digestive endoscopy that showed a 2,5 cm tumor in the third portion of the duodenum. Colonoscopy was normal. Capsule enteroscopy showed the same tumor at the duodenum, but another 5 in the jejunum and 1 in the ileum. The patient underwent a PET-CT that showed



**Fig. 2.** Resected segment of jejunum with different polypoid tumors that harbor adenocarcinoma.

5 hypercatabolic lesions from the duodenum to the ileum without suspicious lymphadenopathies or metastases (Fig. 3a and b).

Pre-test genetic counseling was offered in order to investigate a genetic inherited etiology and to provide more information regarding medical management and future risks related cancers. Benefits, limitations and implications of the results were discussed and the patient consented to have testing for a panel of genes associated with early onset polyposis. Genetic testing was done at Invitae and the following genes were evaluated for sequence changes and exonic deletions/duplications: APC, BMPR1A, CDH1, EPCAM, MLH1, MSH2, MSH6, MUTYH, PMS2, PTEN, SMAD4, STK11, TP53. Test results were negative with no pathogenic changes detected in the genes analysed thru next generation sequencing. Therefore, the cause of the patient's condition remains unknown. Caution should be taken in interpreting the results since a negative test result reduces but does not eliminate the possibility that the patient's condition has a genetic component.

She finally underwent open surgery with intraoperative endoscopy and resection of all the remaining lesions of the duodenum and the small bowel with segmental resections and extended lymphadenectomy with primary anastomoses. Biopsy was positive for adenocarcinoma in six tumors, all positive lesions had well-differentiated tubular adenocarcinoma. The tumors infiltrated up to the muscular layer without vascular or perineural penetration and without any compromise of lymph nodes. All tumor borders were free of malignancy and 46 harvested lymph nodes were negative. The patient was staged as T2N0M0 and the oncologic committee decided only follow-up. Control PET-CT after three years is negative for recurrence.

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