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Small bowel intussusception secondary to metastatic melanoma 15 years after complete excision of the primary tumor



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ABSTRACT

INTRODUCTION: Primary intestinal melanoma is a rare entity, however the gastrointestinal tract, and particularly the small bowel, is a common site of recurrence from cutaneous melanoma.

PRESENTATION OF CASE We report the case of a 48 year old woman with small bowel intussusception secondary to metastatic cutaneous melanoma, 15 years after excision of the primary tumor. The patient underwent an emergency small bowel resection with negative margins on final pathology.

DISCUSSION: Surgical resection is a palliative, yet necessary, procedure in the setting of small bowel obstruction due to intussusception secondary to intestinal metastatic melanoma. In case of bowel metastasis, presenting symptoms are nonspecific and do not provide significant clues to the differential diagnosis of the underlying disease. In some patients, complete surgical resection of early diagnosed bowel metastases is associated with prolonged survival. Systemic chemotherapy in these patients does not provide survival benefit.

CONCLUSION: The occurrence of bowel relapse after very long disease free interval, while highly unlikely in most tumors, should always be considered in the differential diagnosis of patients with previous history of cutaneous malignant melanoma presenting with gastrointestinal symptoms.

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1. Background

Intussusception is an infrequent cause of mechanical intestinal obstruction in adult patients. We describe the case of a jejunal intussusception secondary to small bowel metastasis from cutaneous melanoma 15 years after complete excision of the primary tumor.

2. Case report

A 48-year old woman came to our Emergency Department complaining of diffuse abdominal pain. The symptoms had progressively worsened over the last month, with colicky abdominal pain mostly in the upper quadrants, exacerbated by food, and associated with nausea, occasional vomiting and no changes in bowel habits. Over the last year the patient was being investigated for hypochromic microcytic anemia (she underwent an upper endoscopy 3 months earlier that was normal and a colonoscopy had been scheduled). Past medical history was remarkable for a left

shoulder Breslow IV (7 mm in depth) nodular malignant melanoma completely excised, with a negative 15 years follow up. Patient also underwent a left laterocervical lymph node biopsy 10 years earlier and pathology report was consistent with reactive sinus histiocytosis. Her family history was positive for breast cancer.

Evaluation at the Emergency Department included complete blood count which confirmed hypochromic microcytic anemia (Hb 9.7 g/dl; hematocrit 30.5%). An abdominal X-ray did not reveal any specific finding. Abdominal ultrasound showed a concentric image (consistent with the so called “doughnut sign”) suggestive of small bowel intussusception. A CT scan confirmed a proximal jejuno-jejunal invagination, with dilated stomach, duodenum and first jejunal loop (Fig. 1). Therefore, an emergency laparotomy was performed and the intussuscepted bowel segment was resected (Fig. 2). Postoperative course was unremarkable and the patient was discharged on postoperative day 10. Pathology report was consistent with ulcerated melanoma metastasis with epithelioid morphology, with the tumor approaching within less than 1 mm of the serous margin and both longitudinal resection margins free from tumor (Fig. 3). Mean mitotic activity was 4–5 mitosis/10 HPF, and no vascular or lymphatic invasion was noted. None out of eight lymph nodes resected was involved with tumor. Immunohistochemistry was positive for S-100, HMB-45, Melan-A, and Vimentine, while was negative for Cytokeratin AE1/AE3. A

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Fig. 1. CT scan showing small bowel intussusception with evidence of change in diameter of the bowel.



Fig. 2. Surgical specimen showing the invaginated bowel segment.



Fig. 3. Open surgical specimen revealing a 5 cm x 5 cm neoplasm with superficial fibrinous exudate.

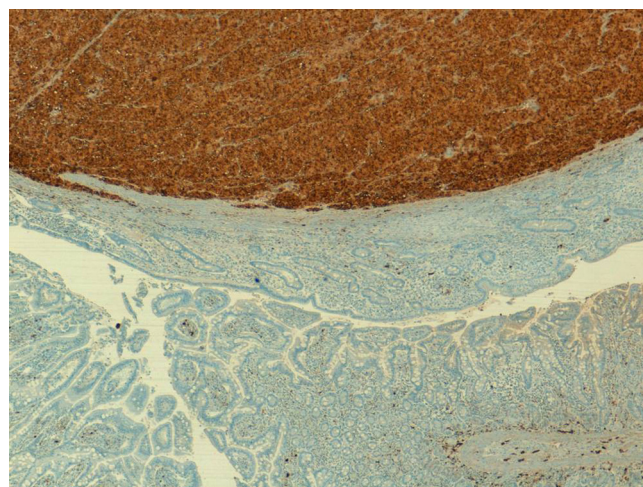


Fig. 4. Immunohistochemistry for S100 showing intense and diffuse nuclear and cytoplasmic expression, consistent with melanoma (40x).

BRAF V600 mutation was detected (Fig. 4). The multidisciplinary oncology team discarded an adjuvant treatment because of lack of evidence of efficacy. A 3-months PET-CT scan follow-up showed no evidence of relapse.

3. Discussion

Intussusception is an uncommon cause of bowel obstruction and potentially bowel infarction in adults, which occurs when a segment of intestine along with its mesentery invaginates into the lumen of the bowel tract distal to it.^{1,2}

The etiology is idiopathic in about 10% of the case, while 70–90% of the times it is secondary to a tumor which serves as a lead point, either malignant, more frequently in the colon, or benign, usually in the small intestine (carcinoid, lipoma, hamartoma, leiomyoma, inflammatory adenoma).^{1,3} Less common causes are Meckel's diverticulum, adhesions and bowel wall hematoma.²

Primary small bowel malignancies are uncommon, accounting for less than 2% of gastrointestinal (GI) tumors.⁴ The GI tract is a common site of metastasis, with the small bowel representing the second most frequently involved organ (58% of the cases) after the liver.⁵

Cutaneous melanoma has a propensity to metastasize to the GI tract, and the small bowel is a common site of involvement (35–70% of GI metastasis from skin melanoma).^{6–9} Although considered to be rare, primary GI melanomas have been described in four – not always distinct – types (cavitary, infiltrating, exoenteric, and polypoid), often difficult to distinguish from a metastatic form especially when the cutaneous primary is unknown or regressed.^{10,11}

The average time from excision of the primary cutaneous melanoma to the occurrence of an intestinal metastasis is 3–6 years, even though cases of synchronous metastasis or within 6 months of diagnosis have been described, with a mean symptom-free interval reported between 6 and 90 months.^{11–14}

Abdominal ultrasonography is a valuable tool in diagnosing an intussusception secondary to malignancy, showing a multiple concentric pattern, also known as target, doughnut, or bull's eye sign. Small-bowel follow-through and conventional enteroclysis have the capability of identifying intraluminal abnormalities, however they are not accurate in detecting extraintestinal findings, thus requiring further imaging studies.⁹

A CT-enteroclysis can be used for confirming the diagnosis in those cases presenting with intermittent small-bowel obstruction.⁹ Capsule endoscopy, can be a valid adjunction in assessing

small-bowel lesions, with the possibility of investigating segments of intestine not explored by conventional endoscopy, however caution must be exercised when a partial obstruction is suspected.¹⁵

Surgical exploration with bowel resection remains the standard of care, especially for patients with limited spread of the disease.^{12,13} However, surgery is not curative and it should be considered as a good mean of palliation with a chance of improving prognosis (5-year survival up to 40% and a disease-free interval up to 10 years) when free surgical margins can be achieved.^{10,16}

Postoperative chemotherapy is sometimes administered as further palliative treatment although evidence of its effectiveness is currently lacking.^{8,17}

4. Conclusion

Metastases from malignant cutaneous melanoma to the GI tract can occur after very long disease free interval, hence metastatic disease must be considered in patients who present with anemia and/or melena or with obstructive symptoms. Surgical resection is the mainstay of treatment for acute complications of cutaneous melanoma metastatic to the bowel, with reported prognostic benefits in selected cases with limited extent of disease.

Conflict of interest

All authors have no conflicts of interest.

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None.

Ethical approval

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Author contributions

Vincenzo Vigorita, Fabio Ausania and Marco Bertucci Zoccali made the study design and contributed to the preparation of the

manuscript writing. Vincenzo Vigorita, Cristina Facal Alvarez and Blanca De Urrutia Nadal performed the operation. Vincenzo Vigorita, Fabio Ausania and Jose Enrique Casal Nuñez revised and approved the final report

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