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Case Report
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Where You Less Expect It: A Rare Case of Intestinal Intussusception

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Abstract

Background: Gastrointestinal metastases from malignant melanoma are recognized in 60% of autoptic cases, but become symptomatic only in 0.8% to 9% of patients.

Case Report: We report the case of a 51 year-old women, complaining of vague abdominal pain arisen from few weeks, who was admitted to the emergency room for vomiting and worsening abdominal pain. The investigations revealed microcytic anemia and a radiological picture of suspected multiple intussusceptions resulting in intestinal obstruction. She underwent emergency surgery, which revealed three polypoid formations of the small bowel and grayish mesenteric lymphadenomegalies. Double intestinal resection and lymph node biopsy were performed. The histological examination showed the presence of intestinal and lymph node localizations of malignant melanoma, more likely metastatic in nature.

Discussion: The small bowel is the most frequent site of gastro-intestinal metastases from malignant melanoma (71% to 91%). These only rarely cause symptoms including abdominal pain, vomiting and anemia. The diagnosis is based on radiological and endoscopic findings and requires histological confirmation. In 2% of cases the primitive is not detectable. Immunotherapy and molecular target therapies are the preferred systemic options, but surgical resection guarantees the best prognostic results.

Conclusion: Intestinal intussusception in adults is a rare pathology, which can hide malignant neoplasms. Among these, even in the absence of a history of skin pathology, gastrointestinal metastases from melanoma should be considered, especially in patients with abdominal symptoms and anemia that cannot otherwise be explained. Surgical resection not only resolves the symptoms, but can reveal unexpected diagnoses.

Keywords: Melanoma; Gastro-intestinal metastases; Bowel obstruction; Intussusception; Anemia

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Introduction

Intestinal intussusception in adults is a rare disease, generally secondary to an organic cause, often represented by a neoplastic lesion [1]. Gastrointestinal metastases from malignant melanoma are relatively frequent, but they very rarely give symptoms and even more rarely cause intestinal intussusception. Primary melanoma of the small intestine, on the other hand, is an almost exceptional and even controversial diagnosis [2], as it could more likely represent a metastasis from an undetectable primitive [3].

Case Presentation

A 51-year-old Caucasian woman went to the emergency room with worsening abdominal pain and vomiting. The patient had already been complaining of vague pains, in mesogastrium and epigastric region, exacerbated by meals, since a few weeks. She reported irregular bowel movements, but with stools seemingly normal in color and consistency; she denied hyperpyrexia and significant weight loss. She was not assuming any medication. The anamnestic collection reported only an appendectomy in childhood, an endoscopic sigma polypectomy and a family history of gastric carcinoma. Checking our computerized records we found out that she had also removed a skin lesion in her left arm 8 years earlier, histologically classified as a compound nevus. Upon clinical examination the patient appeared in good general conditions, normal weight and with normal vital parameters. Her abdomen was barely bloated, slightly painful in the upper quadrants and in the left iliac fossa without signs of peritonism, with a swelling seemingly due to a left colic cord and

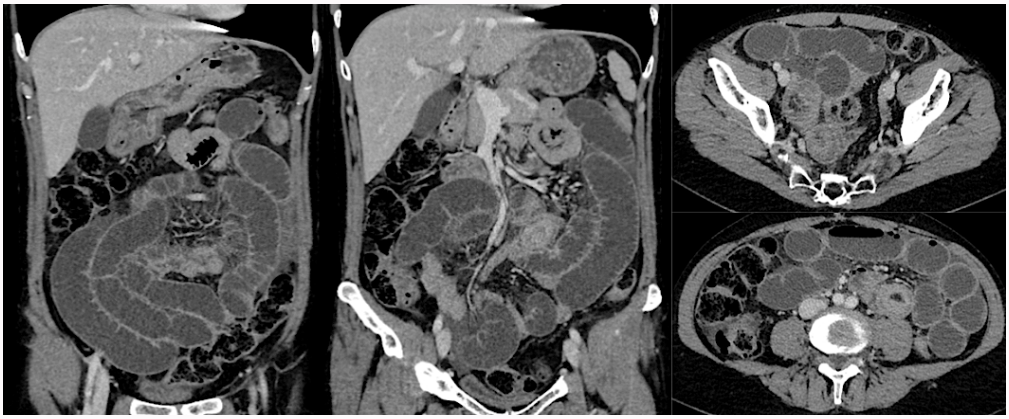

Figure 1: Abdomen CT scan.

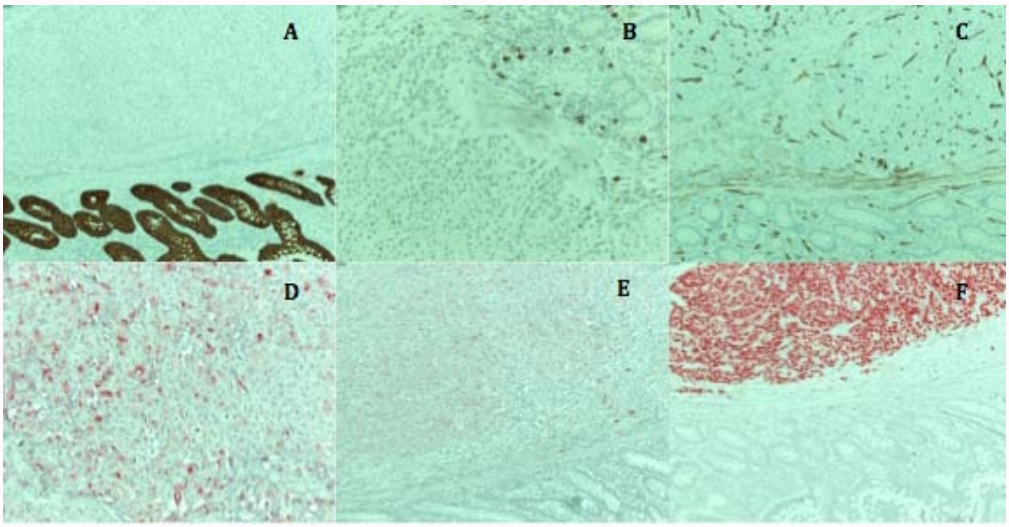

Figure 2: Immunophenotype: A. CK8-18; B. CD 117; C. CD34; D. HMB 45; E. S-100; F. SOX10.

very torpid peristalsis. The blood tests showed just marked microcytic anemia (Hb 7.3 g/dl, MCV 72.8fL) while phlogosis indexes, hydro-electrolytic and hemogenic profiles were all within normal ranges. A single unit of RBC was then transfused. She performed an abdominal X-ray, which showed some short air-fluid levels in the central quadrants, likely of the ileum, and faecal material in the large bowel. She then performed a gastroscopy, which revealed no signs of active or previous bleeding. We then prescribed an abdominal contrast enhanced CT scan that highlighted multiple, dilated small bowel loops with some air-fluid levels and three jejuno-ileal tracts with thickened walls and convoluted course resembling possible invaginations, two of which located in the left mesogastrium and one in the small pelvis (Figure 1).

It was therefore decided for emergency surgery. At the laparotomy there was a frank bowel obstruction with modest free citrine effusion. On palpation, a voluminous intraluminal nodular lesion, of about 3 cm, was determining the supposed ileal invagination; upstream of it, about 30 and 45 cm from the Treiz ligament, 2 further similar neoformations were found. A diffuse mesenteric lymphadenopathy was also appreciated; a voluminous dark-colored lymph node of about 3 cm, located at the root of the mesentery, near the first jejunal loop, and another smaller one, of similar pathological appearance, were excised for biopsy. A double ileal resection of 10 and 20 cm in length was performed with subsequent double mechanical side-to-side is peristaltic anastomosis. The post- operative period was uneventful and the patient was discharged on the 7th postoperative day.

The cytological sample was negative for neoplastic cells, while the histological examination macroscopically confirmed the presence of 3 ileal ulcerated polypoid neoformations. Microscopy showed a subversion of the normal glandular architecture and transmural infiltration of nests of epithelioid cells with angiolymphatic invasion. The intestinal resection margins and visceral serosa were free from neoplastic infiltration, while one of the analyzed nodes was metastatic. The immunophenotype of the neoplastic population was CK8-18, CD117 and CD34 negative and HMB 45, S-100 and SOX10 positive with ki67 85% (Figure 2).

Our pathologist therefore diagnosed intramural locations of poorly pigmented malignant melanoma with epithelioid cells, more likely metastatic. The subsequent molecular analysis showed a BRAF mutation. The patient was then referred to a melanoma oncological center; she underwent several tests including a gynecological, Otorhinolaryngology and ophthalmological evaluation without identifying any suspected primary lesion. Staging head-thorax-abdomen CT revealed no further localizations. The patient was

then diagnosed with gastrointestinal metastases from unknown primary melanoma; she was offered immunotherapy treatment with Nivolumab or with BRAF inhibitors with adjuvant intent to surgery, which was deemed to be radical.

Discussion

Intestinal intussusception, relatively frequent in childhood, represents only 1% of cases of bowel obstruction in adults [4]. Mostly idiopathic in children, in 90% of cases diagnosed in adulthood an underlying cause, often neoplastic, is recognized [1]. Gastrointestinal metastases from malignant melanoma are not infrequent per se, indeed autoptic studies report their presence in 60% of cases [5], predominantly localized in the small bowel [6], probably due to the rich vascularization of this intestinal tract and the high hematogenous spreading capacity of melanoma. Their apparent rarity is determined by their low tendency to give clinical manifestations.

The superficial spreading ocular-cutaneous melanoma is the most commonly subtype responsible for gastrointestinal metastases. Although the primary melanoma can be traced back in most patients, in 2% of cases it cannot be identified [7]. It is hypothesized that in these cases the primary lesion has gone into complete regression or falls below the detection threshold [8]. Instead, primary gastrointestinal melanoma is extremely rare, especially in the small bowel where it is exceptional and considered almost anecdotal by some authors.

The clinical diagnosis of melanoma of the small bowel is very difficult, giving non-specific manifestations such as enterorrhagy, the most frequent symptom, chronic abdominal pain, vomiting, weight loss and microcytic anemia. An acute presentation with bowel obstruction, intussusception and perforation is very rare [3] and abdominal CT scan, although very sensitive in emergency setting, does not allow a preoperative diagnosis of nature.

Surgery is the standard treatment for intussusception, regardless of the benign or malignant nature of the underlying disease. It is more often performed in emergency setting since the diagnosis usually occurs after the occlusive symptoms become obvious. However, there are no guidelines that establish whether it is appropriate to perform a reduction of intussusception before resection [4,9,10]. Although this may be considered adequate when the lead point is likely to be benign, most authors prefer an en-bloc resection without a preceding reduction [9-11]. In our case, also due to the high suspect of malignant disease, a resection of the two intestinal tracts involved was performed with no attempt to reduce the intussusception.

As regards the gastrointestinal melanoma, it would be important to know whether it is a primitive or a metastasis in order to define the best therapeutic path and the different prognostic implications [6]. The median survival of patients with metastatic melanoma is only 6 to 8 months. Therapeutic options include surgical resection, immunotherapy and bio-chemotherapy [12]. The therapeutic goal remains R0 resection, which allows achieving median survivals of up to 64 months [13], significantly higher than those obtainable with palliative procedures or systemic treatments alone [12].

Conclusion

Gastrointestinal metastases from malignant melanoma are not uncommon, but very rarely cause bowel obstruction. Intestinal intussusception, already rare in itself, can hide an even more unpredictable pathology, in particular in patients with no history of skin disease, but that should be remembered especially in case of chronic abdominal symptoms and anemia that cannot otherwise be explained.

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