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Case report Metastatic melanoma to the small bowel causing intussusception: A case report

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| ARTICLE INFO  | A B S T R A C T   |
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| <i>Keywords:</i><br>Metastatic melanoma<br>Intussusception<br>Bowel obstruction<br>Immunotherapy<br>Case report | Introduction and importance: Melanoma is a malignant skin neoplasm with a high metastatic potential. Several reports have shown that metastatic melanoma has a predilection to metastasize to the GI tract; however, diagnosing metastatic melanoma as a cause of intussusception has been reported in only few cases with variable presentations.<br><i>Case presentation:</i> We present the case of a 48-year-old woman with a long history of metastatic melanoma who presented with recurrent enteric intussusception due to a melanoma lesion acting as a pathologic lead point despite immunotherapy treatment. We contribute the management plan, diagnostic modalities, and surgical approach of this rare form of adult intussusception in guidance of future management plans.<br><i>Clinical discussion:</i> The variability in presentation of adult intussusception makes diagnosis difficult and the lack of consensus on management and surgical strategies poses challenging hurdles. A diagnostic laparoscopy followed by reduction and resection of the intussuscepted lesion in a small surgical field is an effective and beneficial palliative procedure with favorable outcomes. Our patient developed intussusception despite receiving a trial of dual immunotherapy after chemotherapy.<br><i>Conclusion:</i> It may be insufficient to control disease even with dual immunotherapy after chemotherapy. Further studies are needed to determine the optimal surgical and oncological management in treating gastrointestinal metastasis of malignant melanoma. |

# 1. Introduction

Malignant Melanoma is a skin neoplasm with predilection to metastasize to the gastrointestinal tract, particularly the small bowel [1]. Commonly, patients with intestinal metastasis present with nonspecific symptoms or are even asymptomatic [2]. The tendency of metastatic melanoma to form a gastrointestinal mass that acts as lead point causing intussusception has been reported in few cases and is considered an unusual presentation [2–4]. Only some studies demonstrated metastatic melanoma as the culprit lead point in adult intussusception [5]. The estimated time between diagnosis of the primary lesion and metastasis to the GI tract is up to 4.5 years [6]. This long latency period along with the variability in presentation makes diagnosis difficult. Although surgery is considered the standard of care in adult intussuception [5,7], the literature lacks consensus on the optimal

management and definitive surgical approach. It is thus essential to recognize the possible presentations and add management strategies to the literature. In this report, we demonstrate the rare case of a 48-year-old patient with metastatic melanoma causing intussusception despite receiving dual immunotherapy after chemotherapy.

This case report has been reported in line with the SCARE criteria 2020 [8].

# 2. Case presentation

This is a case of a 48-year-old woman with a medical history of hypothyroidism and peptic ulcer disease. Patient also had a history of a malignant melanoma of the left heel which was first diagnosed in early 2019 after which the patient underwent excision, sentinel lymph node biopsy and skin grafting. Pathology then demonstrated a stage IIB acral

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type invasive malignant melanoma lesion. In late 2019, patient progressed to stage IV melanoma after developing inguinal lymphadenopathy. She was started on dual immunotherapy with Nivolumab and Ipilimumab.

In 2020, she experienced diarrhea and abdominal discomfort that warranted a CT scan and a colonoscopy. At the time, colitis was diagnosed and attributed to Ipilimumab. At that point, the patient had already received 3 cycles of immunotherapy. CT showed stable disease; thus, the patient was observed closely and received a course of prednisone. When disease progression was demonstrated by subsequent PET CT, patient received a rechallenge with Nivolumab for 5 additional cycles with 1 subsequent cycle of Temozolomide. Due to the lack of other treatment options, the decision to rechallenge with dual immunotherapy with a lower dose of Ipilimumab at 1 mg/kg instead of the previous dose of 3 mg/kg was pursued.

In January 2021, the patient presented to the emergency department for colicky abdominal pain and vomiting and had last taken immunotherapy 4 days prior. On abdominal CT scan (Fig. 1), intussusception of the small bowels was shown, caused by a lead point in the mid-to-distal ileal loops. The patient was stable with no signs of acute abdomen on physical exam, and no evidence of bowel compromise or perforation on imaging. Accordingly, patient was managed conservatively and discharged upon resolution of symptoms. The patient experienced a second episode four days later, for which she was managed similarly.

The patient remained asymptomatic until May 2021, when she presented to the emergency department for colicky abdominal pain of 2 days duration associated with one day of obstipation and several episodes of bilious vomiting. CT scan revealed an ileo-ileal intussusception with no signs of perforation or bowel compromise. Due to her recurrent episodes of intussusception, management options were discussed with the patient and the decision was to pursue surgical exploration at our tertiary care center by a highly specialized surgeon in aim of a definitive treatment.

As a first step, a diagnostic laparoscopy was performed. Intraoperatively, the small bowels were extensively inspected, and multiple dark lesions were found seeding into the mesentery and the small bowels. Inspection and running of the ileal loops revealed intussuscepted small bowels with a large culprit lesion acting as the lead point (Media 1). The intussuscepted bowel segments were delivered through a right lower quadrant incision and divided, and a bowel anastomosis was performed (Fig. 2A). Another large lesion was visualized along the ileal loops and was also resected to avoid bowel complications or obstruction (Fig. 2B). The patient had an uncomplicated postoperative course. She received antibiotics for the first two days following surgery and was discharged 9 days later in a stable condition. Patient was followed up in specialty clinics 2 weeks after her surgery with no reported postoperative complications. Given the wide metastatic spread of this patient's primary tumor, prognosis remained dismal and surgical intervention was done as a palliative mean to alleviate symptoms and



Fig. 1. Abdominal CT scan on presentation demonstrating mid-to-distal ileal loop intussusception.



**Media 1.** Intraoperative running of the small bowel to relieve intussusception revealing the culprit metastatic melanoma lesion.

improve survival.

# 3. Discussion

Intussusception involves the telescoping of a proximal bowel segment into the lumen of an adjoining segment causing intestinal obstruction. The incidence of intussusception varies between age groups, where adult intussusception accounts for almost 5% of all cases of intussusception [2]. In contrast to childhood intussusception, which is mostly idiopathic, most cases in adults are due to a pathologic lead point such as a polyp or neoplasm [9]. In a systematic review and meta-analysis by Hong et al., a metastatic carcinoma acted as a culprit in the majority of intussusception cases [10].

The variable presentation of adult intussusception along with a low index of suspicion for the condition, given its rarity, makes diagnosis challenging [11]. CT scan of the abdomen remains the imaging modality of choice in diagnosis [12]. Our patient presented with symptoms of abdominal pain and bilious vomiting suggestive of bowel obstruction. The differential diagnosis for such a presentation may include but are not limited to gastrointestinal etiologies such as adhesions, hernias, malignancy, volvulus, or inflammatory bowel disease [13]. However, given our patient's medical history and predilection of malignant melanoma to metastasize to the small bowel, bowel intussusception was high on the differential and was evident on CT.

Operative management is the standard of care in adult intussusception because the causative agent in the majority of cases is a malignant lead point [14]. However, recent studies showed that not all patients with adult intussusception require surgery, and instead some are managed conservatively depending on CT findings and clinical presentation [15]. Although a pathologic lead point was detected during our patient's first presentation, we proceeded with conservative management due to the patient's critical status, high tumor burden and patient's preference. Symptom recurrence necessitated surgery as a palliative approach. In fact, several studies have demonstrated that complete surgical resection of gastrointestinal metastatic melanoma lesion can be performed safely and is associated with a survival benefit [16].

The surgical approach is controversial with some studies recommending resection without reduction while others recommend reduction followed by resection [10]. For cases of enteric intussusception, several studies recommend reduction followed by resection. However, resection en-bloc without reduction is recommended for colonic intussusception [10]. Our surgical approach was unique in that diagnostic laparoscopy was performed as a first step instead of an exploratory laparotomy. Once the pathologic lead point was detected, a small incision was done to access the intussuscepted segment and perform the operation in a small surgical field. Since our patient had an ileo-ileal intussusception, we proceeded with reduction followed by resection to avoid long segment removal.

Nivolumab and ipilimumab are immune checkpoint inhibitors that



Fig. 2. (A) Intraoperative view of the enteric metastatic melanoma lesion causing ileo-ileal intussusception. (B) Specimen view of the excised lesion causing ileo-ileal intussusception.

have demonstrated efficacy with metastatic melanoma [17]. Reports show that patients with malignant melanoma receiving nivolumab alone developed GI metastasis despite treatment response at other metastatic sites [18]. Even so, to our knowledge, one case in the literature reports immunotherapy given after chemotherapy, but this was attempted postoperatively [2]. Although the possibility that intestinal metastasis was present before treatment initiation cannot be excluded, our patient developed intussusception while receiving Nivolumab therapy, even after a trial of dual immunotherapy after chemotherapy. Therefore, further studies are needed to define an optimal surgical and oncological treatment plan to limit disease spread and improve survival in such patients.

### 4. Conclusion

Malignant melanoma may result in metastatic lesions that have the tendency to form pathologic lead points leading to intussusception which presents clinically as a gastrointestinal obstruction. A diagnostic laparoscopy followed by reduction and resection of the intussuscepted lesion in a small surgical field is an effective and beneficial procedure for patients at a high surgical risk. In addition, it may be insufficient to control disease even with dual immunotherapy after chemotherapy. Further studies are needed to determine the optimal management plan for gastrointestinal metastasis caused by malignant melanoma.

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# **Ethical approval**

Exemption from ethical approval was provided.

### Consent

Informed consent was obtained from the patient for the publication of this case report. There are no personal identifiers of the patient.

### Author contribution

MJK performed the surgery. HK, BO, and RK collected the data. HK, BO, RTK and RK did the literature review and wrote the manuscript. MJK edited the manuscript.

# **Research registration**

Name of the registry: N/A.

Unique identifying number or registration: N/A. Hyperlink to your specific registration (must be publicly accessible and will be checked): N/A.

### Guarantor

Mohamad Jawad Khalife, MD, FACS, FRCP, FRCS.

#### Declaration of competing interest

None.

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## Provenance and peer review

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# Appendix A. Supplementary data

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