Metastatic melanoma causing recurrent intussusception and perforation of small bowel: case reports and literature review

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Practice points

- Intestinal intussusception and bowel perforation due to metastatic cutaneous melanoma are rare.
- Reintussusception of resected jejunal segment in very short time is very rare.
- Surgeons should be aware of both intussusception and perforation in patients presenting with symptoms of bowel obstruction and abdominal pain in malignant cutaneous melanoma.
- In the management of intussusception, symptomatic patients with no evidence of bowel obstruction require surgery with bowel resection.
- For patients with bowel perforation from metastatic melanoma, treatment options should be preferred according to hemodynamic status of patients and presence of contamination. Surgical closure in small perforated areas could be a treatment of choice for palliation.
- For patients with sign of bowel obstruction due to intussusception, resection of intussuscepted segment is treatment of choice.

Aim: To present cases of recurred jejuno-jejunal intussusception and jejunal perforation due to melanoma.

Materials & methods: Case 1: A 43-year-old male under treatment for malignant melanoma was presented with abdominal pain and distention. Ten centimeter intussuscepted jejunum was resected. Second exploration was done due to failure to pass gas and stool. The reintussusception was detected and resection of reintussuscepted jejunum was performed. Case 2: A 63-year-old male was presented with abdominal pain. Abdomen computed tomography showed free air in the abdomen suggesting intestinal perforations. Perforated area at 80 cm in the jejunum sutured.

Conclusion: We present the seemingly first report of reintussusception of resected segment in a very short time. Surgeons should be aware of both intussusception and perforation in metastatic melanoma.

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Keywords: intussusception • malignant cutaneous melanoma • metastasis • obstruction • perforation • reintussusception • small bowel

Gastrointestinal tract metastases are found in approximately 20% of stage IV metastatic cutaneous melanoma patients, but previous autopsy studies have shown a prevalence as high as 58% in deceased patients. The most common sites of metastases are in the small bowel, followed by the large bowel and the stomach [1]. Small bowel metastatic deposits attributed to malignant melanoma are found in 2–5% of patients with malignant melanoma of the skin. Intussusception caused by cutaneous malignant melanoma is a very rare condition [2,3]. The prognosis of metastatic melanoma is poor [2]. Metastasis from melanoma causing bowel perforation is rare and presents as an acute abdomen. Bowel perforation from melanoma metastases showed an intraoperative finding of multiple widespread brown lesions [4].
Materials & methods
This study included a case presentation of jejuno-jejunal intussusception and reintussusception operated twice and a case of jejunal perforation due to metastatic melanoma. Both patients signed an informed consent for this procedure. To our knowledge, this is first published case of intussusception and reintussusception of metastatic melanoma in short time.

Case 1
A 43-year-old male patient diagnosed with cutaneous malignant melanoma in 2014 (T4N0M0, positron emission tomography). Patient had interferon, dexamethasone, granisetron and ipilimumab treatment and radiotherapy in 2016. Two malignant melanoma metastatic lesions were noticed in cranial MRI and patient had radiotherapy to whole brain in 2017. Abdomen computed tomography (CT) in 2018 showed small intestine intussusception (115 × 42 × 41.5 mm) in a segment adjacent to the left colon (Figure 1). Tube thoracostomy on left side was done due to massive pleural effusion before laparoscopic abdominal surgery. The pleural effusion was a malignant effusion from melanoma. In exploration, a 10-cm intussuscepted intestinal segment was found in the jejunum. With pulling the intussuscepted bowel, completely narrowed intestine with an inflammation and fibrosis area of 2 cm was noticed (Figures 2 & 3). Intussuscepted segment was pulled out of abdomen with 4-cm skin incision. Twenty-five centimeter of small intestine was cut with linear staples and anastomosed end to end with 3-0 vicril and
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Figure 2. Intraoperative appearance of small bowel intussusception.

Figure 3. Specimen view of intussuscepted bowel.

3-0 prolene. Pathology of specimen showed a lesion with a polypoid appearance 3.2 cm in diameter with diagnosis of malignant melanoma. Patient was discharged at postoperative sixth day with good bowel sound and normal defecation. At 22 days of first operation, patient had failure to pass gas and stool and had abdominal pain and nausea. Abdominal plain x-ray showed air fluid levels (Figure 4). In exploration, the reintussusception was detected at 5 cm in the area where resection and anastomosis was performed at 40 cm distal Treitz (Figure 5). The focus of the recurrent intussusception was at the anastomosis and there was no new metastatic lesion. There was no residual disease in the second resected specimen. Resection of reintussuscepted jejunum with side-to-side anastomosis was performed using Covidien linear staples (Figure 6). Patient was discharged at postoperative fifth day and transferred to oncology unit for oncological treatment.
Figure 4. Air fluid level in plain x-Ray before second operation.

Figure 5. Intraoperative view of intussuscepted segment during second operation.
Case 2

A 63-year-old male patient diagnosed with malignant melanoma at the end of 2017. Patient had dexamethasone, granisetron and ipilimumab treatment and had palliative radiotherapy to whole brain in 2018. Patient referred to our clinic due to abdominal pain with signs of peritonitis. Abdomen CT showed noteworthy free air densities in the abdomen suggesting intestinal perforations. Emergency surgery was performed. Fecaloid contamination was present in the abdomen during exploration. A perforated area was noticed at 80 cm in the jejunum and was sutured. The operation was purely for palliation and there is no benefit for resection of bowel and lymph node resection. There were mesenteric edema and ground pigmented lesions in the jejunal segment (Figure 7). From the pigmented lesions, a sample for biopsy was taken. Final diagnosis was malignant melanoma. The patient was discharged at fifth day for oncological treatment. Macroscopy of specimen showed a brown fibrous encapsulated cross-section in dark pink color piece of tissue with diagnosis of malignant melanoma. Patient followed treatment with oncology and in May 2018 radiotherapy planned.

Discussion

The small intestine has an abundant blood supply allowing for metastasis from cutaneous melanomas [5]. Primary intestinal melanoma tends to occur in the ileum as a solitary intramural lesion, whereas secondary intestinal melanoma tends to arise in both the jejunum and ileum with multiple polypoid submucosal lesions. The lesions may be ulcerated and crucially they may be amelanotic [6,7].

Preoperative diagnosis of metastatic or small intestine melanoma tends to be difficult. The previous report of bowel perforation from melanoma metastases showed an intraoperative finding of multiple widespread brown lesions [8].

Anastomotic site intussusception is an extremely rare complication following resection of proximal jejunum. The probable predisposing factors are abnormal motility in proximity to duodenojejunal flexure, larger diameter and two-layer Albert–Lembert type anastomosis [9]. High motility and relatively large enteric diameter of the proximal small intestine may influence the occurrence of intussusception [10].
In the management of intussusception, asymptomatic patients with no evidence of disease do not require surgery and intussusception will likely resolve spontaneously. Asymptomatic patients with known metastatic melanoma may be initially observed, but a low threshold for surgery should be maintained. Symptomatic patients with known metastases should undergo surgery [11]. Our case was symptomatic patients with known metastases. Bowel resection was done for intussusception and 3 weeks later intussusception recurred again and second operation resection and anastomosis was undertaken. For patients with bowel perforation from metastatic melanoma, treatment options should be preferred according to the hemodynamic status of patients and presence of contamination. Surgical closure and/or wide intestinal resection with lymph nodes is treatment choice. When surgical closure is only preferred, this needs to be considered and revised as surgery in this setting is purely for palliation and there is no benefit to lymph node resection. For patients with sign of bowel obstruction due to intussusception, resection of intussuscepted segment is the treatment of choice. The median survival in patients with intestinal metastases is inferior to 7 months compared with other sites of metastasis [8]. Our patient survived 6 months after surgery for intussusception and reintussusception. Our second patient with bowel perforation is now 3 years postoperative and under treatment.

Conclusion
Intestinal intussusception and bowel perforation due to metastatic cutaneous melanoma are rare. We believe this is the first report of reintussusception of resected segment in a very short time. Surgeons should be aware of both intussusception and perforation in patients presenting with symptoms of bowel obstruction and abdominal pain in malignant cutaneous melanoma.

Future perspective
The prevalence of malignant cutaneous melanoma seems to be increasing. Surgeons dealing with gastrointestinal area will therefore likely encounter more malignant cutaneous melanoma complications such as intussusception and perforation.

Author contributions
Y Yagmur operated on the patients and planned and wrote the article. MA Açıkgoz helped in planning, writing and collecting material for the literature.

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Ethical conduct of research
The authors state that they have obtained appropriate institutional review board approval or have followed the principles outlined in the Declaration of Helsinki for all human investigations. The authors state that they have obtained verbal and written informed consent from the patient/patients prior to surgery and for the inclusion of their medical and treatment history within this case report.

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