Case report

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JEJUNO-JEJUNAL INTUSSUSCEPTION CAUSED BY SKIN MELANOMA METASTASES: A CASE REPORT

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Skin melanoma is a relatively rare malignant tumor with a raising incidence in last decades. Biological course is characterized by lymphatic and hematogenous spread, but metastases in intestine and mesenteric lymph nodes are frequent. These metastases can lead to acute intestinal occlusion as a sign of acute abdomen requiring surgical emergency.

Patient 68 year old, admitted with clinical and radiology signs of acute intestinal occlusion, underwent emergency surgery. The jejuno-jejunal intussusception was found and the lead point of intussusception was intramural melanoma metastasis 4cm in diameter. The small bowel resection length was 40cm. Postoperative course was without complications. Five years before, patient underwent surgery for melanoma of the skin of left scapular region.

Acute intestinal occlusion in patients operated from skin melanoma could seldom be caused by hematogenous intra-abdominal metastases of skin melanoma.

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Key words: *intestinal occlusion, invagination, melanoma*

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Introduction

Only in 15% of intussusceptions, lead points in the small bowel are malignant lesions. They are frequently metastatic in nature and commonly caused by a melanoma (1).

According to recent reviews, the small intestine is a frequent site of melanoma metastases and this is the main cause of secondary intestinal tumors. Superficial spreading melanoma is the most common type of melanoma (70% to 80%) and therefore responsible for most gastrointestinal metastases, which can develop even more than 10 years after resection of the primary cutaneous lesion. Around 60% of the patients who suffer from melanoma have small bowel metastases at the moment of death, but in only 1% to 4% of the cases they are detected as complications occur (2).

The intussusception caused by metastatic melanoma is more often as primary melanoma (3).

Clinical findings are indolent with intermittent crampy abdominal pain leading to acute obstruction with abdominal distension, pain and vomiting. Less than 20% have blood in stool. The diagnosis in plain radiography is highly demanding showing the absence of lower liver edge sign (absence of the subhepatic angle) in upper right abdomen. Other signs are: target sign, crescent sign and a bowel obstruction. The target sign is a mass in the right upper quadrant. It sometimes does not have a target appearance and just resembles a solid mass. It is sometimes called a pseudokidney sign because it may have the shape of an oval mass in the right upper quadrant. The crescent sign is caused by the intussuscepting lead point (intussusceptum) protruding into a gas filled pocket, which often results in a crescent shaped gas pocket. But if the pocket is large, it may not be crescent shaped.

On ultrasound, it could be seen as concentric alternating echogenic and anechogenic bands and defined as target sign, looking as doughnut or bull`s eye (signs are synonyms) The echogenic bands are formed by mucosa and muscularis whereas the submucosa is responsible for the hypoechoic bands. Other sign could be defined as pseudokidney sign, where other part mimics cortical part of kidney but inner part mimics medulla.

Case report

Male Caucasian, 68-year old, underwent excision of nodular skin melanoma (Breslow IV -6 mm in depth) in left scapular region 5 years ago. Patient refers to surgeon complaining of diffuse abdominal

pain and acute abdominal distension from last night. The pain is continuous, sharp and diffuse in the abdomen. He vomits nonbilious and the body temperature was normal. The symptoms had progressively worsened over the last month with colicky intermittent pain mostly in periumbilical region and no changes in bowel habits. Blood samples confirmed raised lactate-dehydrogenase 545U/L and hypochromic microcytic anemia (Hb 9.4g/dl; hematocrit 29.0%).

On physical examination, his vital signs were normal. The hallmark physical findings were a sausage-shaped mass palpable in periumbilical region and moderate abdominal distension. There were no clinical signs of peritonitis. Murphy's sign was negative.

Diagnosis of intestinal occlusion was proved by erected plain abdominal radiography with huge dilated bowel loop with air-fluid levels, distention of the whole small intestine and a solid pseudo-mass in the right upper quadrant (arrow head in Figure 1).

The ultrasound examination reveals pathognomonic bull-eye sign (arrow head in Figure 2).

Therefore, an emergency laparotomy was performed through median incision and jejuno-jejunal intussusception was found (Figure 3).

The manual reposition of intussusception was not possible and a segmental jejunal resection (40 cm in length) was performed. The lead point for intussusception was intramural metastasis of melanoma. There were a lot of black colored, round shaped nodular metastases in the mesentery and two melanotic lesions were present in the jejunal wall, approximately 75cm distal to the duodeno-jejunal junction (Treiz ligament) (Figure 4). The bowel continuity was obtained by end-to-end anastomosis and a drain was placed in abdominal cavity before closing.

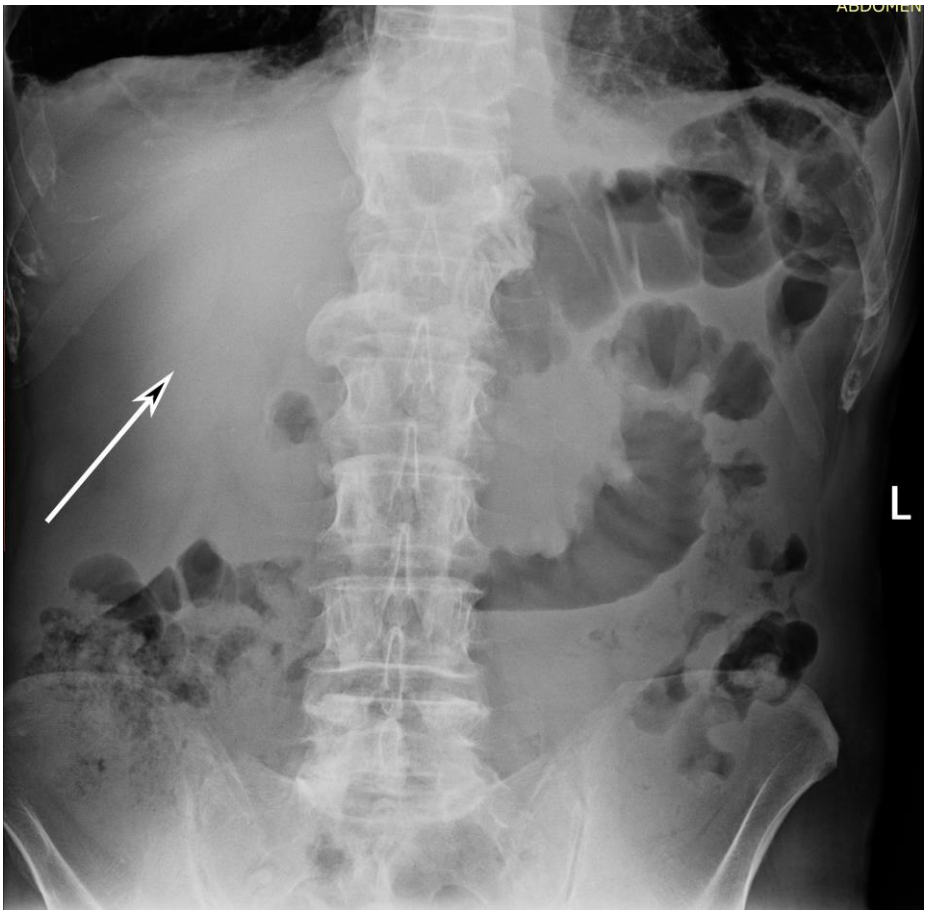

Figure 1. Abdominal radiograph illustrating a dilated small bowel loop and solid mass in right upper quadrant (arrow head) with scanty bowel gas elsewhere

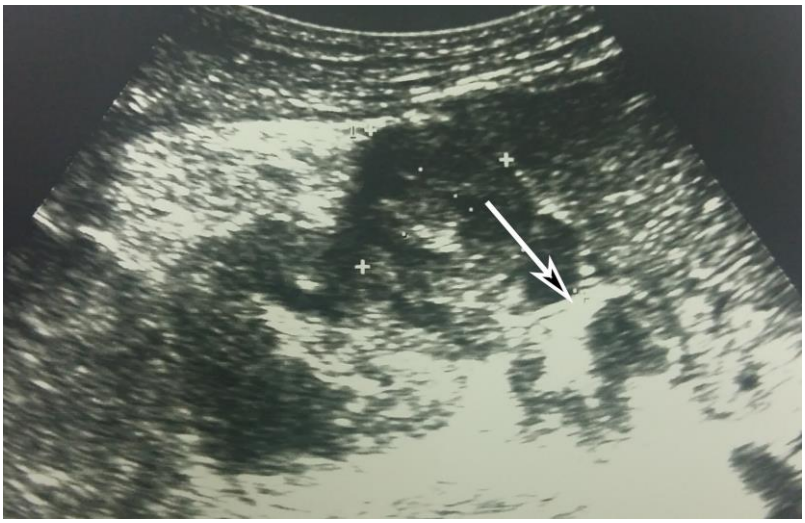

Figure 2. Ultrasound depicts target sign (arrow head)

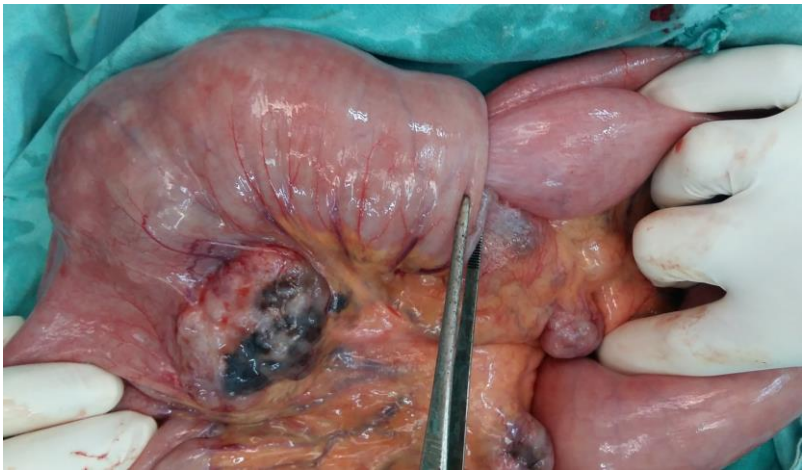

Figure 3. Intraoperative photograph indicating jejuno-jejunal intussusception

Figure 4. Open surgical specimen showing invaginated bowel segment.
The metastatic melanoma as an ulcerated polypoid mass is shown as the lead point

Postoperative course was uneventful. Postoperatively, the patient was treated with intravenous antibiotics and monitored closely for postoperative ileus. Supportive management in the form of analgesia, nasogastric tube and intravenous fluids was required for 72 h until normal dietary intake was reestablished. The patient was discharged on the 10th postoperative day.

The histopathologic examination revealed intestinal malignant melanoma lesions with a maximum diameter of 3.5cm.

Discussion

Incidence of clinically evident small bowel metastatic deposits after skin melanoma surgery is 2% to 5% of patients and is most commonly expressed as an acute intestinal obstruction (4).

Some authors refer the propensity of melanoma to metastasize to the GI tract, and the small bowel is a common site of involvement (35% to 70% of GI metastasis from skin melanoma). In the gastrointestinal tract, the small bowel is the most frequent site of metastasis of melanoma, mainly because of its rich blood supply (5).

In 58% of the patients with malignant melanoma, intestinal metastases were found at autopsy.

That means less than 5% of patients with metastases to the gastrointestinal tract are diagnosed antemortem. Though malignant melanoma is the most common cause of the extra-abdominal source of intestinal metastasis, it is rare to find them presenting as jejuno-jejunal intussusception (6). The magnitude of silent metastases coupled with the fact that these metastases can present with almost any GI symptom highlights the need for high clinical suspicion in patients with a previous malignant melanoma presenting with GI symptoms. The rarity of jejuno-jejunal intussusception is the prime reason for reporting our case.

The clinically evident bowel metastasis in our case appeared 5 years after primary excision what is in accordance with literature review that the average time from excision of the primary cutaneous melanoma to the occurrence of an intestinal metastasis is 3-6 years. Due to the difficulty in exploring the whole length of the small bowel using common diagnostic procedures, a preoperative diagnosis is often challenging to establish. Routine examinations are plain radiography and ultrasound. When the patient presented with abdominal pain, vomiting and distension, the diagnosis of intestinal obstruction in the emergency setting must be confirmed by erect abdominal X-ray. Ultrasonography showed small intestine intussusception. In cases with small bowel intussusception, plain radiography shows signs of bowel obstruction such as dilated loops of bowel or air-fluid level in the bowel lumen, and rarely a mass lesion or intraluminal air trapped between the walls of the intussusceptum and intussuscipiens (air crescent sign). These findings nevertheless lack the specificity and sensitivity to diagnose intussusception (7). Plain abdominal radiographs are of limited value in the diagnosis of intussusception due to their reduced sensitivity and specificity. However, they are often performed as part of the initial investigations for patients presenting with an acute abdomen. One of the radiographic features consistent with intussusceptions is signs of intestinal obstruction proximal to the lead point. In our case, we referred the air fluid level and solid mass in upper right abdomen.

The next examination should be abdominal ultrasonography as a useful technique in the diagnosis of intussusception in adults and children. The features described include a target and doughnut signs on the transverse view and a pseudokidney sign on a longitudinal view. Ultrasonography carries no radiation risks. The limitations include obesity and bowel gas which may obscure the typical findings (7). We defined target sign in ultrasound in our case.

Although different imaging techniques such as barium examinations and CT, they both may be able to depict larger intestinal lesions. In developed countries, CT of the abdomen seems to be the radiological investigation of choice, with a sensitivity of 71.4% to 87.5% and a specificity of 100% in the prospect of diagnosis of intussusception in adults (7). In adult patients with long-term nonspecific abdominal pain, barium study is contraindicated because it brings the risk of intestinal perforation. Despite of various imaging modalities have been used to help in establishing the diagnosis, it is frequently confirmed only during surgical intervention. Surgery is currently the treatment of choice without a precise surgical strategy. There is not a clear consensus about the optimal surgical approach and there is still controversy about reduction before resection (8). The current controversy remains on the extent of surgical resection vs. reduction of the intussusception. The initial favor to resect enbloc the intussuscepted segment of bowel was based on the theoretical risks of venous embolization of the tumor cells on bowel manipulation and also the risks of perforating the ischemic bowel with contamination of the peritoneal cavity (9-11). Resection without reduction was the standard of care for intussusception caused by tumor and advocated by most surgeons (12, 13). In our case, we also performed resection without attempts to reduction. Some authors advise that simple reduction is acceptable in post-traumatic or idiopathic intussusceptions, where no pathological cause could be identified, obviously after the exclusion of bowel ischemia or perforation (14). Some authors suggest that reduction prior to resection can be safely performed in selected patients with suspected benign disease, especially when small bowel intussusception is presented without ischemia or there is a risk of short gut syndrome after wide en block resections (2). Diagnostic laparoscopy and resection has been used successfully in selected patients. In patients with chronic and subacute presentation with partial small bowel obstruction, laparoscopy offers the benefit of a conservative approach with possible reduction of the bowel but laparoscopy in acutely obstructed patients with bowel distension where visualization may be poor, and bowel manipulation may further risk perforation and increase the morbidity of an operation (15). However, surgery is not curative and it should be considered as a good means

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 (16).

Conclusion

Metastatic melanoma of the gastrointestinal tract, especially bowel intussusception caused by metastatic melanoma should be suspected in patients with history of melanoma of the skin and acute gastrointestinal symptoms. Emergency surgery is the mainstay of treatment and bowel resection is appropriate treatment. A high index of clinical suspicion combined with the appropriate imaging might help in establishing an early diagnosis, emergency surgery and avoiding serious complications like perforation and peritonitis. In the presence of a lead point lesion but no preoperative tissue diagnosis, surgical intervention in the form of bowel resection without reduction is advisable.

References

1. Chang CC, Chen YY, Chen YF, Lin CN, Yen HH, Lou HY. Adult intussusception in Asians: clinical presentations, diagnosis, and treatment. J Gastroenterol Hepatol 2007; 22(11):1767-71. [CrossRef][PubMed]
2. Alvarez AF, Nicolás M, Goransky J, Vaccaro CA, Beskow A, Cavadas D. Ileocolic intussusception due to intestinal metastatic melanoma. Case report and review of the literature. Int J Surg Case Rep 2011; 2(6): 118-21.[CrossRef][PubMed]
3. Kouladouros K, Gärtner D, Münch S, Paul M, Schön MR. Recurrent intussusception as initial manifestation of primary intestinal melanoma: Case report and literature review. World J Gastroenterol 2015; 21(10): 3114-20.[CrossRef][PubMed]
4. Vigorita V, Ausania F, Bertucci Zoccali M, Alvarez CF, Nadal Bde U, Nuñez JE. Small bowel intussusception secondary to metastatic melanoma 15 years after complete excision of the primary tumor. Int J Surg Case Rep 2015; 6:26-8.[CrossRef]
5. Gill SS, Heuman DM, Mihas AA. Small intestinal neoplasms. J Clin Gastroenterol 2001;33(4):267-82. [CrossRef][PubMed]
6. Malik P, Mandia R, Maheshwari R, Sharma DS. Jejunojejunal intussusception: An unusual presentation of malignant melanoma. Australas Med J 2014; 7(10): 416-8.[CrossRef][PubMed]
7. Potts J, Al Samaraee A, El-Hakeem A. Small bowel intussusception in adults. Ann R Coll Surg Engl 2014; 96(1):11-4.[CrossRef][PubMed]
8. Wang N, Cui XY, Liu Y, Long J, Xu YH, Guo RX, et al. Adult intussusception: a retrospective review of 41 cases. World J Gastroenterol 2009; 15(26):3303-8. [CrossRef][PubMed]
9. El-Sergany A, Darwish A, Mehta P, Mahmoud A. Community teaching hospital surgical experience with adult intussusception: Study of nine cases and literature review. Int J Surg Case Rep 2015;12:26-30. [CrossRef][PubMed]
10. Hanan B, Diniz TR, da Luz MM, da Conceição SA, da Silva RG, Lacerda-Filho A. Intussusception in adults: a retrospective study. Colorectal Dis 2010; 12(6):574-8. [CrossRef][PubMed]
11. Kim JS, Lim JH, Jeong JH, Kim WS. Conservative management of adult small bowel intussusception detected at abdominal computed tomography. Korean J Gastroenterol 2015; 65(5):291-6. [CrossRef][PubMed]
12. Honjo H, Mike M, Kusanagi H, Kano N. Adult intussusception: a retrospective review. World J Surg 2015; 39(1):134-8.[CrossRef][PubMed]
13. Ongom PA, Opio CK, Kijjambu SC. Presentation, aetiology and treatment of adult intussusception in a tertiary Sub-Saharan hospital: a 10-year retrospective study. BMC Gastroenterol 2014;14:86. [CrossRef][PubMed]
14. Patel S, Eagles N, Thomas P. Jejunal intussusception: a rare cause of an acute abdomen in adults. Ann R Coll Surg Engl 2014; 96(1):11-4.[PubMed]
15. Siow SL, Mahendran HA. A case series of adult intussusception managed laparoscopically. Surg Laparosc Endosc Percutan Tech 2014; 24(4):327-31. [CrossRef][PubMed]
16. Gore RM, Silvers RI, Thakrar KH, Wenzke DR, Mehta UK, Newmark GM, et al. Bowel Obstruction. Radiol Clin North Am 2015; 53(6):1225-40. [CrossRef][PubMed]

Prikaz bolesnika

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JEJUNO-JEJUNALNA INVAGINACIJA UZROKOVANA METASTAZAMA MELANOMA KOŽE: PRIKAZ SLUČAJA

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Melanom kože je relativno redak tumor, ali je incidencija ovog maligniteta u stalnom porastu. U biološkom toku opisane su limfogene i hematogene metastaze, a često mogu nastati metastaze u creva i limfne čvorove mezenterijuma. Ove metastaze mogu dovesti do crevne okluzije, koje mogu uzrokovati akutni abdomen kao urgentno hirurško oboljenje.

Bolesnik star 68 godina primljen je zbog kliničkih i radioloških znakova i akutne intestinalne okluzije i hitno je operisan. Nađena je invaginacija jejunuma, a vodeći deo invaginata činila je intramuralna metastaza melanoma prečnika 4cm. Učinjena je resekcija tankog creva dužine 40cm i post-operativni tok je protekao uredno. Pet godina pre prijema, bolesnik je operisan zbog melanoma na koži leđa.

Akutne crevne okluzije kod bolesnika koji su operisani od melanoma kože mogu retko biti uzrokovane hematogenim intraabdominalnim metastazama melanoma.

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Ključne reči: *crevna okluzija, invaginacija, melanom*