Turk J Gastroenterol 2010; 21 (4): 439-442

Intestinal malignant melanoma presenting with small bowel invagination: A case report

İnvajinasyon ile prezente olan intestinal malign melanoma: Olgu sunumu

Mutlu DOĞAN[1], Süleyman ÖZDEMİR[2], Ethem GEÇİM[3], Esra ERDEN[4], Fikri İÇLİ[1]

Departments of [1]Medical Oncology, [3]General Surgery, and [4]Pathology, Ankara University School of Medicine, Ankara
Department of [2]General Surgery, [2]Ufuk University School of Medicine, Ankara

Gastrointestinal tract metastasis of any malignancy is rare. Cutaneous or ocular malignant melanomas are the most common tumors that metastasize to the gastrointestinal tract. Major symptoms of these metastatic lesions are bleeding and obstruction of the gastrointestinal tract. However, malignant melanoma arising in intestinal mucosa causing intestinal obstruction is a rare clinical entity. Herein, we present a case of primary gastrointestinal tract malignant melanoma who presented initially with iron deficiency anemia, which consequently triggered an invagination of jejunal and ileal segments causing obstruction symptoms, three months later.

Key words: Intestinal malignant melanoma, invagination, small bowel obstruction

Malignitelerin gastrointestinal traktus metastazları nadir görülür. Kutanöz veya oküler malign melanoma gastrointestinal traktusa en sık metastaz yapan tümörlerdir. Gastrointestinal traktusa metastaz yapan tümörlerde en sık görülen semptomlar; kanama ve obstrüksiyondur. Ancak, intestinal mukozadan gelişen ve intestinal obstrüksiyona neden olan malign melanoma ender görülen bir klinik durumdur. Burada, demir eksikliği anemisi ile prezente olan, ardından 3 ay sonra jejunal ve ileal segmentlerde invajinasyonu tetikleyerek obstrüksiyon semptomlarına neden olan primer gastrointestinal traktus malign melanomalı olgu sunulmuştur.

Anahtar kelimeler: İntestinal malign melanoma, invajinasyon, ince barsak obstrüksiyonu

INTRODUCTION

Small bowel metastasis and obstruction due to a tumor originating outside the peritoneal cavity are rare (1). Ocular or subcutaneous malignant melanomas (MM) are the most common neoplasms that metastasize to the small intestine (2). Primary intestinal MM presenting with iron deficiency anemia followed by invagination is a rare case in the literature.

CASE REPORT

A 68-year-old male patient was admitted to the hospital with iron deficiency anemia, and he was diagnosed as mild acute erosive gastritis and bulbitis on upper gastrointestinal tract (GIT) endoscopy. Colonoscopy revealed no pathology. After three months of medical treatment, he was admitted again with acute abdominal pain and vomiting. Abdominal distention was prominent on physical examination. Intestinal dilatation prominent on the small bowel with hyperperistalsis and a 6 cm solid mass were seen on abdominal sonography. Axial computed tomography (CT) demonstrated a polypoid mass (7x5x3.5 cm) in the jejunum, which was hindering transition to the distal bowel segment (Figure 1).

A diagnostic laparotomy was performed, and during surgical exploration, intussusceptions of the jejunum and ileum due to two fungating tumors were observed (Figure 2). A 35-cm and 15-cm of small bowel segments with tumoral invasion were resected with mesenteric attachments and an end-to-end anastomosis was performed. Histopathology revealed MM of the small intestine (Figure 3a,b). Tumor cells had diffuse specific cytoplasmic immunostaining with HMB-45 and S-100 (Figure 4a,b,c,d). The tumor was 9 cm in diameter and was invading the muscular layer. There were metastatic lymph nodes in the mesentery.

Address for correspondence: Mutlu DOĞAN
Ankara University School of Medicine
Cebeci Hospital
Department of Medical Oncology
06590 Dikimevi, Ankara, Turkey
Phone: + 90 312 595 71 12 • Fax: + 90 312 319 22 83
E-mail: mutludogan1@yahoo.com

Manuscript received: 28.05.2009 **Accepted:** 01.10.2009

doi: 10.4318/tjg.2010.0133

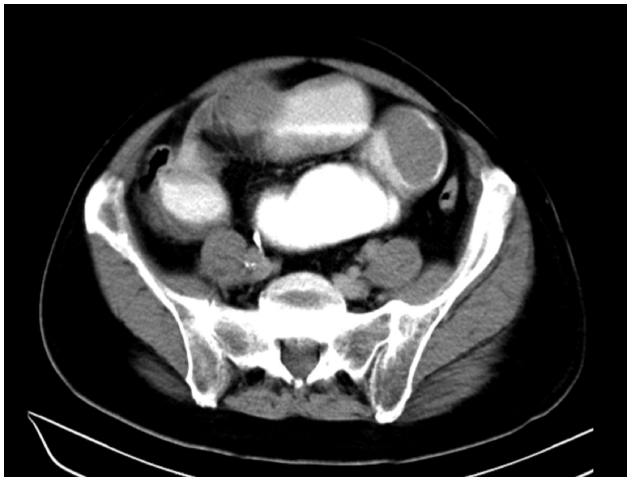

Figure 1. CT images of intestinal invagination: mass lesion in dilated jejunal loop segment.

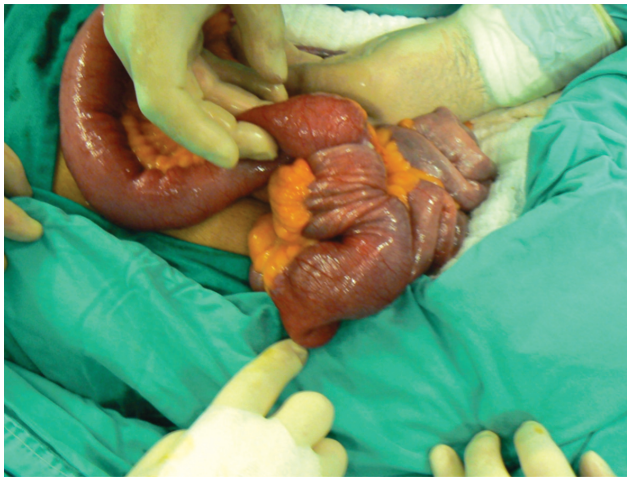

Figure 2. Tumor causing invagination of the jejunum.

He had no other lesions that might have been primary on detailed examination of other sites, such as skin, oropharynx, eye, or rectum. The fluorine-18 fluorodeoxyglucose positron emission tomography/CT (F-18 FDG-PET/CT) showed normal F-18 FDG uptake in the postoperative period.

He was planned to receive adjuvant high-dose interferon (IFN)-α 2b (20 MU/m2/day, 5 days of the week for 4 weeks followed by 10 MU/m2/day, 3 days of the week for 48 weeks). IFN therapy was continued for four months. When liver and lung metastases were discovered on abdominal CT, IFN therapy was stopped and temozolomide (150 mg/m2 x 5 days for 4 weeks, repeated every 6 weeks) was started. Though the lesions remained stable for three months, multiple subcutaneous and brain metastases appeared. Brain metastasis failed to respond to cranial radiotherapy, and he died with disease progression 11 months after the diagnosis.

DISCUSSION

Primary intestinal MM is a rare disease. In a large series of 84836 cases of MM, only 1.3% originated from GI mucosa (4). Sachs et al. (4) defined the diagnostic criteria for primary intestinal MM as a solitary lesion in the small intestine without metastasis except for regional lymph nodes. They re-

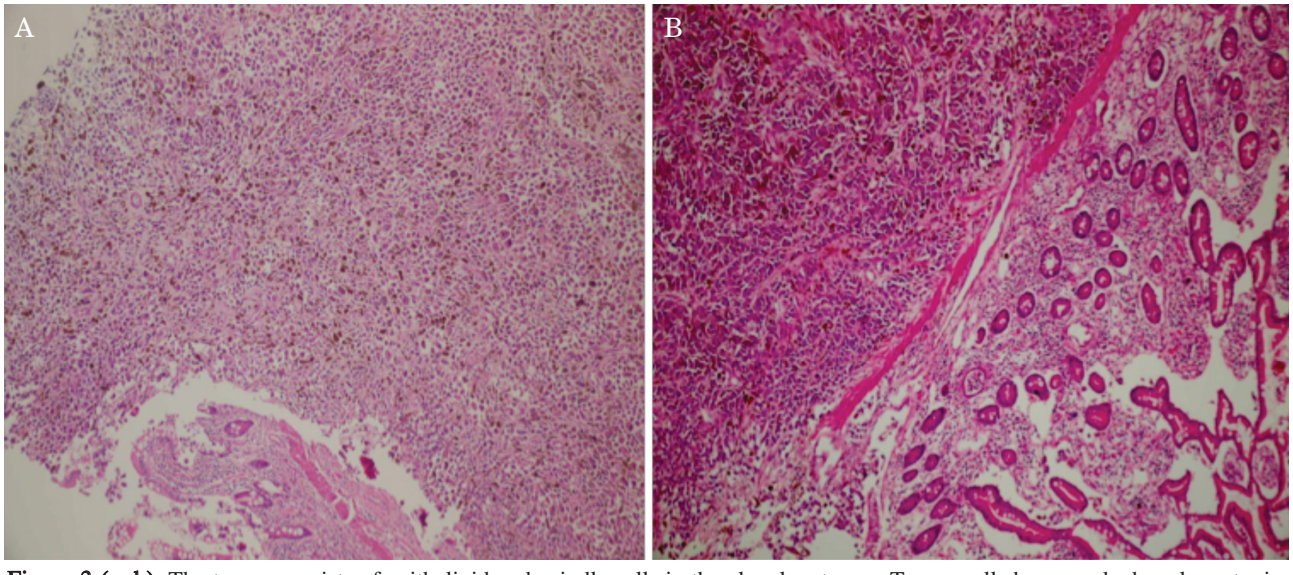

Figure 3 (a, b). The tumor consists of epithelioid and spindle cells in the alveolar stroma. Tumor cells have marked nuclear atypia, hyperchromasia, pleomorphism, multinucleation, large eosinophilic nucleoli, and mitotic figures (HE x10, HE x100, respectively).

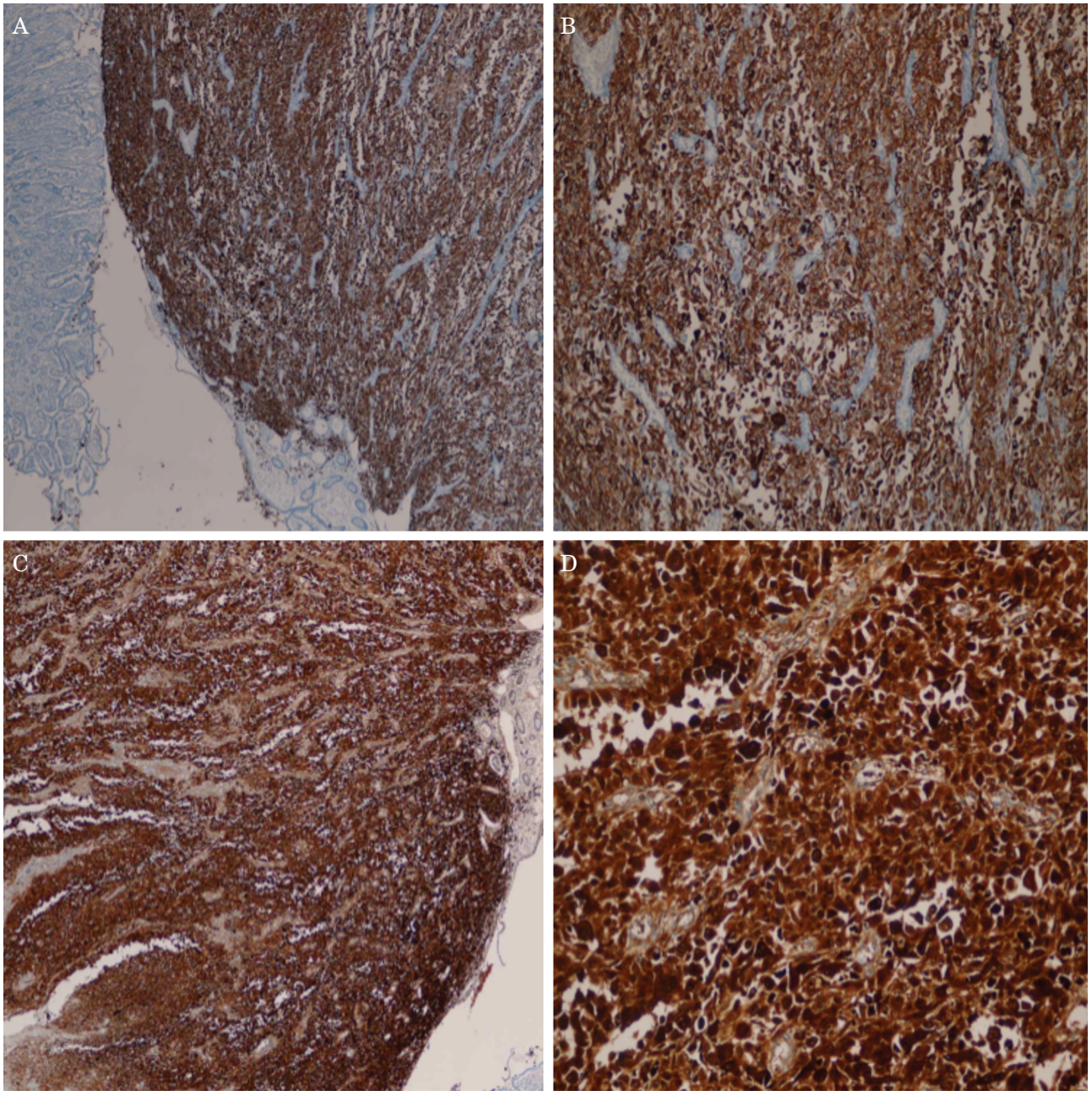

Figure 4 (a, b, c, d). Specific immunohistochemical staining with HMB-45 and S-100 (HMB-45 x40, HMB-45 x100, S-100 x40, S-100 x100, respectively).

ported disease-free survival as 12 months after diagnosis.

F-18 FDG-PET/CT has a high sensitivity (97%) for detection of visceral metastasis from MM. Presentation with iron deficiency anemia and small bowel invagination later, with normal F-18 FDG uptake on PET/CT after resection of intestinal disease, suggest that our case was a primary intestinal MM.

Intestinal MM rarely presents with GI bleeding or obstruction related to invagination. It is difficult to evaluate the distal part of the small intestine after the duodenum, and therefore the diagnosis may be delayed, as in our case. If a patient has iron deficiency anemia without obvious pathology on the upper and lower GI endoscopy, he should be evaluated more carefully considering primary or metastatic intestinal tumors.

It is reported that MMs arising from mucosal surfaces are more aggressive than their cutaneous form (5). This may be related to the rich lymphatic and vascular supply of the intestines. Intestinal MM in our case also presented an aggressive behavior without any response to standard treatment. Delayed diagnosis might have also affected the prognosis.

Resection of the involved segment of GIT in MM, even in the metastatic patients, is generally recommended (3,6). Sanki et al. (6) reported that resection of GI metastasis of MM may contribute to survival, and it should be applied to the patients who have limited disease. Likewise, there is data supporting the benefit of adjuvant IFN in these patients (7). It seems to be more beneficial especially in the cases with lymph node involvement. However, our patient had significant disease progression at the fourth month of IFN treatment.

In conclusion, there is limited experience in the optimal treatment of primary intestinal MM. The poor prognosis of our patient suggests that standard treatments for cutaneous MM are not effective in the MM with intestinal localization.

REFERENCES

1. Hao XS, Li Q, Chen H. Small bowel metastases of malignant melanoma: palliative effect of surgical resection. Jpn J Clin Oncol 1999; 29: 442-4.
2. Kadakia SC, Parker A, Canales L. Metastatic tumors to the upper gastrointestinal tract: endoscopic experience. Am J Gastroenterol 1992; 87: 1418-23.
3. Chang AE, Karnell LH, Menck HR. The national cancer data base report on cutaneous and noncutaneous melanoma: a summary of 84,836 cases from the past decade. The American College of Surgeons Commission on Cancer and the American Cancer Society. Cancer 1998; 83: 1664-78.
4. Sachs DL, Lowe L, Chang AE, et al. Do primary small intestinal melanomas exist? Report of a case. J Am Acad Dermatol 1999; 41: 1042-44.
5. Lagoudianakis EE, Genetzakis M, Tsekouras DK, et al. Primary gastric melanoma: a case report. World J Gastroenterol 2006; 12: 4425-7.
6. Sanki A, Scolyer RA, Thompson JF. Surgery for melanoma metastases of the gastrointestinal tract: indications and results. Eur J Surg Oncol 2009; 35: 313-9.
7. Ollila DW, Essner R, Wanek LA, Morton DL. Surgical resection for melanoma metastatic to the gastrointestinal tract. Arch Surg 1996; 131: 975-9.
8. Schuster LM, Green R, Fraker D. Primary and metastatic diseases in malignant melanoma of the gastrointestinal tract. Curr Opin Oncol 2000; 12: 181-5.