Case Report

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Duodenal Metastasis From a Rectal Malignant Melanoma Primary: A Case Report

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ABSTRACT

Duodenal melanoma constitutes a very small number of gastrointestinal malignant melanomas. The small intestine is a common site for the metastatic spread of cutaneous melanomas; however, its metastatic involvement from a primary rectal melanoma origin has seldom been reported. We present the histopathologic features of an unusual case of primary rectal melanoma metastasizing to the duodenum.

Introduction

The malignant melanomas of the Gastrointestinal (GI) tract are rare. The anorectum is a recognized site for primary GI melanomas [1] diagnostic, and therapeutic aspects of malignant melanoma metastases to the GI. Furthermore, the most common site of melanoma metastases to the GI tract is the small intestine with the ileum and jejunum, as the most affected sites [2]. Regional lymph node and distant metastasis are prevalent in anorectal melanomas; however, there has seldom been reported as a case of metastatic spread to the small bowel. We report a case of metastatic duodenal melanoma with a rectal origin.

Case Presentation

A 59-year-old man was admitted to our gastroenterology clinic in October 2019, with complaints of rectal

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bleeding, weight loss, and anemia. His medical history presented a diagnosis of primary rectal malignant melanoma made 8 months before his referral to us. At that time, the tumor had originally presented as a rectal polyp, measuring 4 cm in the greatest dimension, for which excisional biopsy was performed. The patient neglected treatment adherence during this time and received no surgical, chemoradiation, or hormone therapy interventions. The colonoscopy performed at the time the patient was referred to our center demonstrated a semi-circumferential polypoid ulcerative mass located within 5 cm of the anal verge, which rendered the scope to pass. The obtained biopsy failed to represent the tumor on pathological examination.

A conducted upper GI endoscopy revealed multiple black discolored lesions at the second portion of the duodenum measuring 3-6 mm in diameter as well as one at the duodenal bulb equal to 8 mm. On the histopathologic examination of the duodenal biopsy, the mucosa was infiltrated by atypical epithelioid cells, containing brown granular cytoplasmic pigments (Figure 1A); thus, such observations resembled those of the primary rectal polyp previously diagnosed as malignant melanoma (Figure 2A & 2B). As per the immunohistochemistry study, the tumor cells illustrated strong positivity for HMB45, Melan A, and SOX 10 (Figure 1B-D). The S100 marker was also positive; however, CK AE1/AE3 indicated negative reactivity. The patient expired two months later without receiving therapeutic interventions.

Discussion

The malignant melanomas of the GI tract are rare and mainly secondary to a cutaneous or ocular origin [3]. The primary melanomas of the GI tract derive from the neural crest cells in the anus, esophagus, small intestine, and stomach [1, 3] diagnostic, and therapeutic aspects of malignant melanoma metastases to the GI. Melanoma can also be found in the rectum and sigmoid colon due to the local migration of primal skin melanocytes [4].

Anorectal melanomas are the most frequent primary melanomas of the GI occurring more often between the sixth and eighth decades with a female predilection [5, 6]. Most cases present as a pigmented polypoid lesions with symptoms, such as rectal bleeding, pain, and change in bowel habits [7]. Rapid growth with early metastasis to regional lymph nodes, as well as distant metastasis to the lung, liver, brain, and bones, are detected in these tumors. Such alternations are due to the high number of blood and lymphatic vessels in this area [7, 8]. The small intestine also harbors a high density of blood vessels and lymphatic channels making it susceptible to hematogenous spread of tumor cells from other sites. Numerous small bowel malignancies are metastatic [9]. Among these secondary tumors, extra-intestinal malignant melanoma is the most prevalent type, metastasizing to the small intestine [9, 10]. Metastatic small bowel melanoma ranges from 35% to 70% and equally involves the ileum and jejunum [2].

Primary small intestinal melanomas are so rare. Practically, all small bowel melanomas are considered secondary to a regressed or undiagnosed origin from a cutaneous, ocular, or anorectal primary [9, 11]. The diagnostic criteria of primary intestinal melanoma include no history of melanoma or atypical melanocytic lesion of the skin, the absence of extra-intestinal metastatic spread of melanoma, and the presence of an in situ component in the intestinal mucosa [2]. On endoscopic evaluation, the metastatic tumor may manifest as multiple small

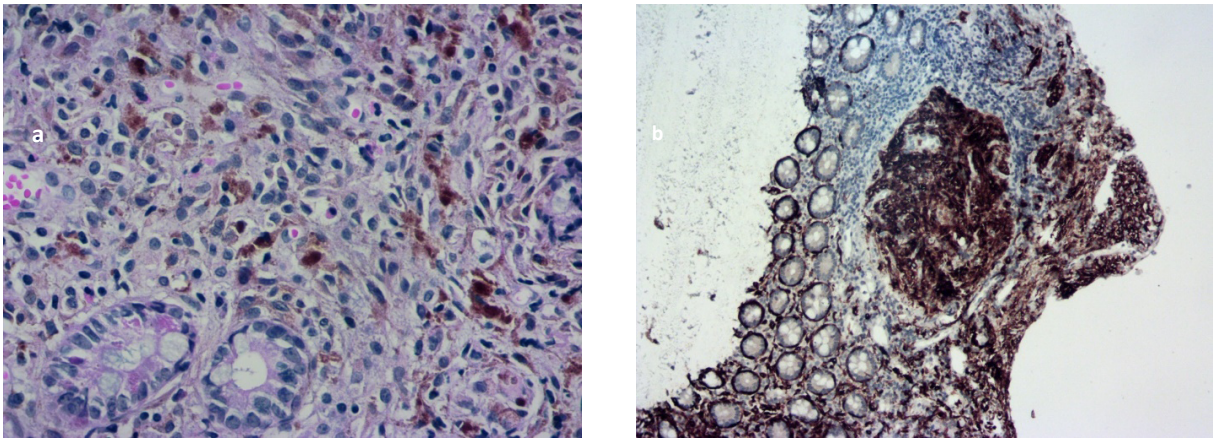

Figure 1. Duodenal melanoma

A. Malignant cells infiltrating the lamina propria (H&E x 100); B. The sheets of tumor cells with irregular nuclei and melanin pigments (H&E x 400).

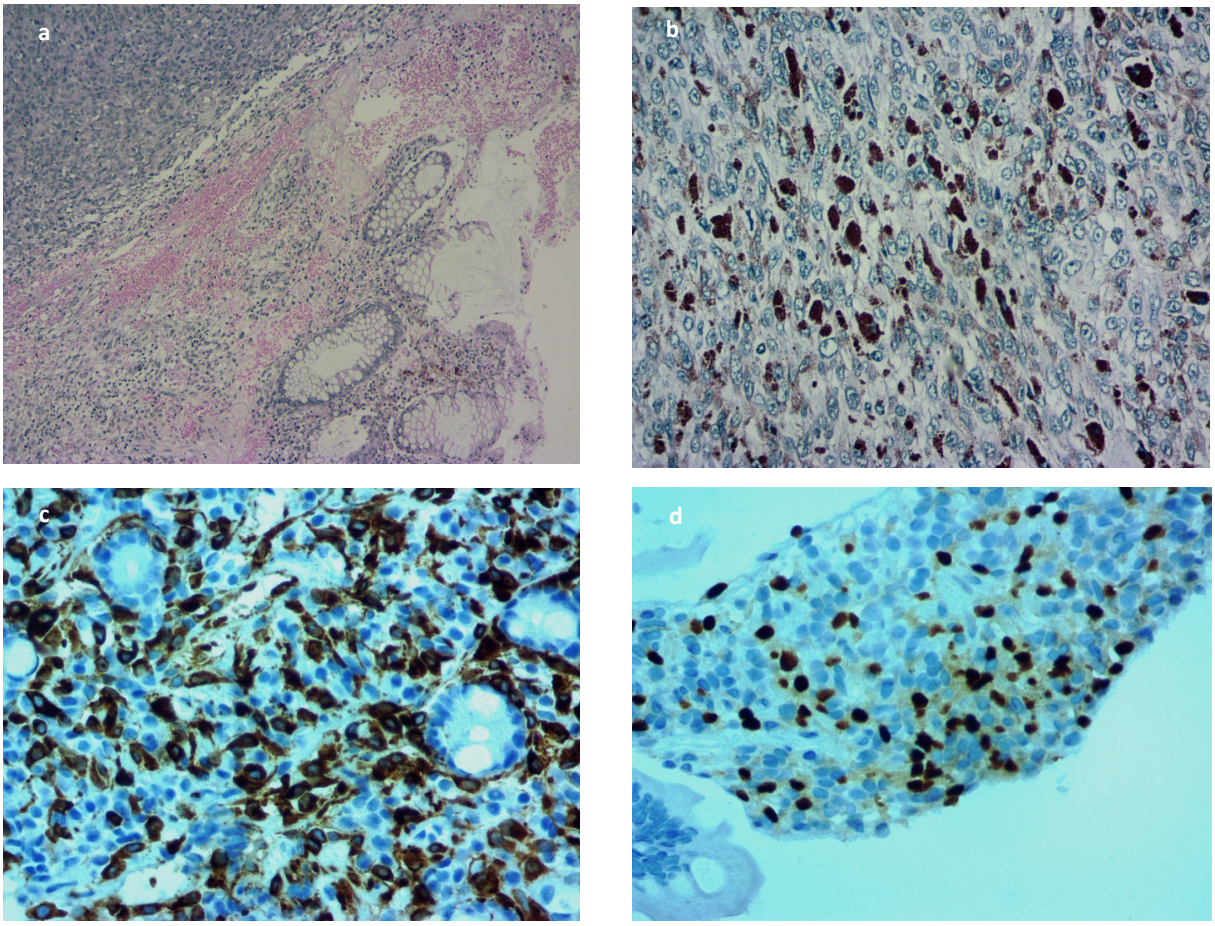

Figure 2. Primary rectal melanoma

A. Duodenal mucosa infiltrated by tumor cells with cytoplasmic melanin pigments (H&E x 400); B. Neoplastic cells are positive for HMB 45 (x100); C. Melan A stains strongly positive (x400); D. SOX 10 staining tumor nuclei (x400).

submucosal nodules, several ulcerated polypoid lesions, or an extrinsic tumor mass; these presentations could be melanotic or amelanotic [12].

Histologically, the tumor cells are discohesive, spindled, or epithelioid-shaped with large pleomorphic hyperchromatic nuclei, eosinophilic nucleoli, and frequent mitosis. Abundant melanin pigments may be observed in tumor cells; however, they are sometimes sparse or absent. The brown or black melanin accumulation in the lamina propria is similar in morphology to iron, lipofuscin, or sulphate deposits [13]. Melanin pigments stain positive with the Fontana-Masson method, but not with the Prussian blue iron stain approach. However, the Fontana-Masson stain method is not specific for melanin and may highlight other non-melanin substances detected in pseudomelanosis duodeni [13]. When a visible pigment is accompanied by atypical cells, malignant melanoma is a more probable option. Other tumors can be confused with melanoma, especially when no obvious pigment is observed. Intestinal neoplasms, such as poorly-differentiated carcinoma, lymphoma, leiomyosarcoma, neuroendocrine tumor, and GI stromal tumor are among potential melanoma mimickers in this respect [14]. Melanocytic markers help in distinguishing and confirming malignant melanoma. Useful immunohistochemical stains for melanoma include HMB45, Melan A, SOX10, tyrosinase, and S100 protein [15]. Ki67 immunostain is also beneficial; it demonstrates a high proliferative index in melanoma in contrast to pseudomelanosis duodeni [13]. Treatment intervention includes surgical resection, immunotherapy, chemotherapy, or only palliative therapy. Patients with intestinal metastatic melanoma present a poor prognosis, having an overall median survival of 6-9 months and a 5-year survival rate of <10% [2].

Conclusion

Small intestinal melanomas are uncommon tumors, i.e. mostly metastatic from a cutaneous primary. Ocular,

anal, and rectal colon origins must also be considered as a source of metastases, while excluding the extremely rare primary melanoma. Histopathologic recognition with immunohistochemistry confirmation and a thorough clinical investigation is required for a definite diagnosis.

Ethical Considerations

Compliance with ethical guidelines

All ethical principles are considered in this article. The participants were informed about the purpose of the research and its implementation stages.

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Conflict of interest

The authors declared no conflict of interest.

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