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A case report of anorectal malignant melanoma with mucosal skipped lesion

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ABSTRACT

INTRODUCTION: We report our experience involving a case of relatively rare anorectal malignant melanoma with skipped lesion.
PRESENTATION OF CASE: The patient was a 72-year-old man who had visited a local clinic complaining of a mass in the anal region, whereupon he was referred to our hospital on suspicion of a malignant melanoma. Close examination revealed a 25-mm black type 1 tumor one-third the size of the circumference of the anal canal and located externally to it. We performed transanal resection of the tumor and confirmed a diagnosis of malignant melanoma. Notably, multiple macular black lesions spaced away from the main lesion were observed during surgery in half of the circumference of the anal canal, from the tumor to the pectinate line. A biopsy of the area also revealed malignant melanoma; therefore, we performed abdominoperineal resection. Pathological diagnosis indicated a submucosal depth; the patient was thus diagnosed with T4 N2c M0 stage IIIb malignant melanoma and was followed on an outpatient basis.
DISCUSSION: Patients with anorectal malignant melanoma have very poor prognoses owing to early lymph node metastasis and hematogenous metastasis. Our case illustrates that small anorectal malignant melanoma lesions can spread from the main lesion and invade the mucosa; examinations may sometimes miss such skipped lesions.
CONCLUSION: Skipped lesions can occur in anorectal melanomas; thus, careful scrutiny of such lesions is required. Moreover, lesion resection is critical for anorectal malignant melanomas.

1. Introduction

Anorectal malignant melanoma is a relatively rare disease that is prone to hematogenous and lymphatic metastasis, often resulting in distant metastasis by the time of diagnosis. In this study, we report our experience with a patient who underwent abdominoperineal resection for anorectal malignant melanoma. Cases in which skipped lesions are observed during surgery or pathological examination are considered rare; therefore, we report this case and discuss the relevant literature.

2. Presentation of case

The patient is a 72-year-old man who complained of a sensation of a mass in his anal region. He has a history of endocarditis but no remarkable family history. The patient first sensed a mass in the anal region one year prior and believed it to be a hemorrhoid. On exacerbation, he visited a physician who observed prolapse of a black pedunculated tumor during rectal examination and thus referred the patient to our department. Physical findings included a 20-mm black pedunculated tumor discovered external to the anal verge. Blood tests revealed no blood count or biochemical abnormalities. Carcinoembryonic antigen and carbohydrate

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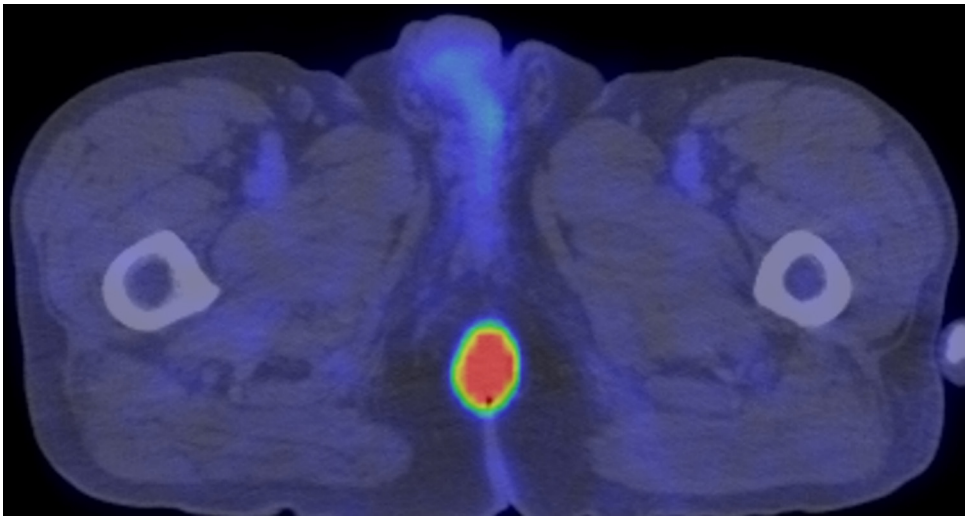

Fig. 1. Positron emission tomography-computed tomography: Abnormally high accumulation of fluorodeoxyglucose (25.1 SUV max) was found in the anal region.

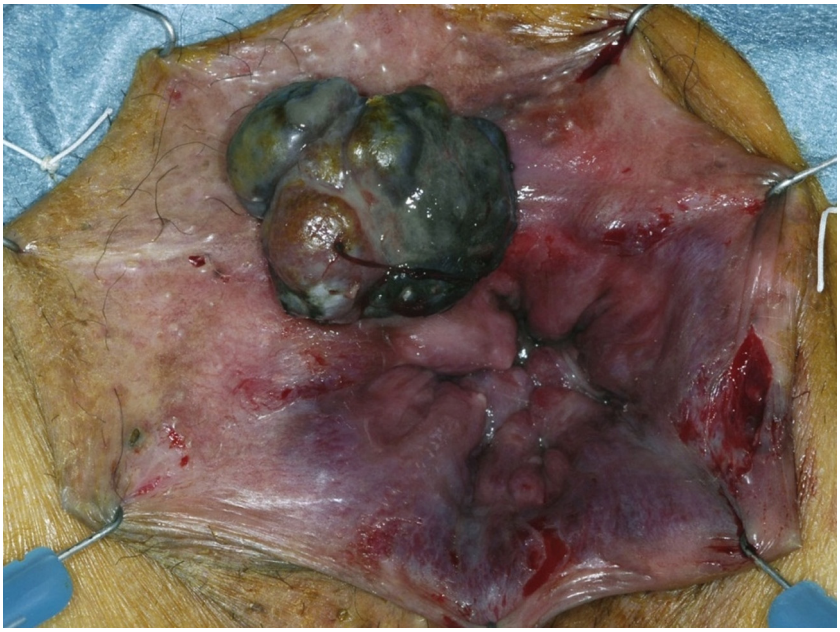

Fig. 2. Physical findings: The base of a black pedunculated raised lesion was found slightly distal to the pectinate line.

antigen 19-9 levels were also within normal ranges. On colonoscopy, a 20-mm pigmented tumor was discovered external to the anal canal. Lesions of the rectal mucosa and the anal canal were not observed. Contrast computed tomography detected no lymphadenopathy or distant metastasis other than the 20-mm tumor in the anal region. Magnetic resonance imaging detected a well-defined mass in the anal region. Positron emission tomography-computed tomography detected an abnormal accumulation (25.1 SUV max) consistent with a tumor (Fig. 1). Based on the above test results, we suspected an anal malignant melanoma and elected to perform an excisional biopsy; this was performed in the lithotomy position under lumbar anesthesia. When the anus was spread, mottled black changes approximately 1–2 mm in size that were non-contiguous with the main lesion were observed in over half of the circumference of the opening adjacent to the dentate line (Fig. 2). We resected the main lesion with a 1-cm horizontal margin, along with the deep portion at a depth that included a small portion of the internal sphincter muscle layer. Additionally, we performed a biopsy of the dentate line tissue containing the mottled black changes that were not contiguous with the main lesion (Fig. 3). Biopsy pathology revealed that the main lesion was a malignant melanoma (25 mm; sm, ly+, v−, HM+, VM−) while the mottled black changes were melanoma in situ. Due to the discovery of the latter, additional resection was deemed necessary. Laparoscopic abdominoperineal resection and D3 dissection were performed on day 35 post-biopsy. Surgical pathology revealed

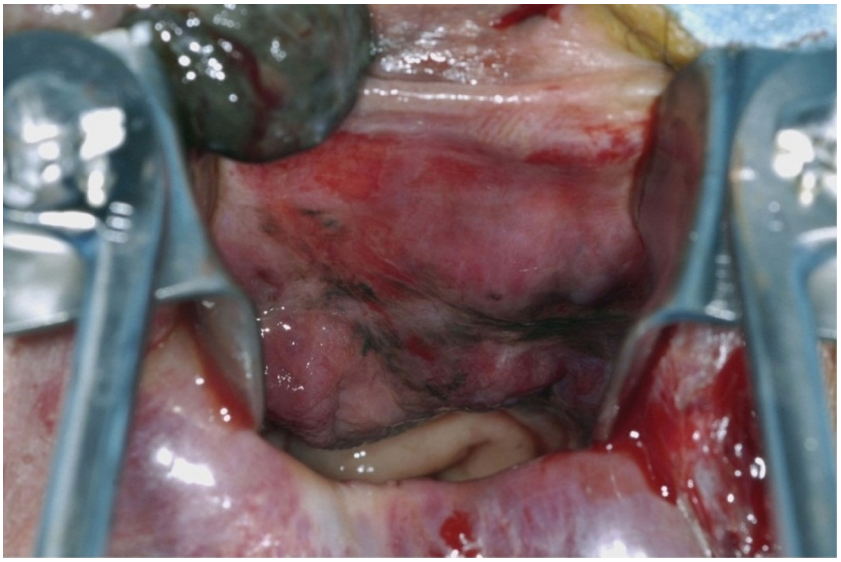

Fig. 3. Biopsy procedure: biopsies were performed on the scattered mottled black changes at the pectinate line level that were separate from the main lesion.

melanoma in situ pPM0 DM0 pRM0 N0. Intraepithelial basolateral proliferation of large atypical melanocytes with nuclear irregularity was observed within an extensive 7.0 × 2.3-cm area around the previously excised main lesion, which constituted a finding of malignant melanoma despite the absence of evident interstitial infiltration (Figs. 4 and 5). The lesion was diagnosed as malignant melanoma (T4, ly+, v-, N2c, M0, f stage IIIb). The patient was discharged on postoperative day 13 following favorable progress. Currently, at 3 months postsurgery, the patient is relapse-free and receives follow-up on an outpatient basis.

3. Discussion

Anorectal malignant melanoma is a relatively rare tumor that accounts for 0.3–1.6% of all malignant melanomas and 0.38–1% of all anorectal malignant tumors [1–3]. It occurs most commonly among individuals in their 60s and is often found in women [4]. The anorectal region, which is the junction of the ectoderm and endoderm, is an embryologically unstable structure. Anorectal malignant melanoma originates from the melanocytes near the dentate line that progresses upward from the submucosa towards the rectum [5]. Non-specific symptoms include melena, sensation of a mass in the anal region, mass prolapse, and abnormal bowel movements. The 5-year survival rate is 14–28.8%, with a median survival period of 12.2–22 months, suggesting a very poor prognosis [4,6]. One cause of this is delayed discovery due to lack of subjective symptoms in the early period and difficulty differentiating the lesion from a hemorrhoid. Another cause is that, because it occurs in an area rich in blood flow, it is prone to early lymphatic and hematogenous metastasis. A biopsy is usually performed for diagnosis; however, only 34% of cases are definitively diagnosed preoperatively [1]. Such a low rate occurs because 10–30% of anorectal malignant melanomas have little or no pigmentation and are thus difficult to differentiate from adenocarcinoma [5]. In such cases, immunohistochemical staining for proteins such as S-100 and HMB45 is useful for diagnosis [7].

The typical treatment is surgical resection; however, standard operative procedures related to the area of resection and lymph dissection have yet to be established. In a Japanese study that included 175 cases, the reported 5-year survival rate for rectal resection (abdominoperineal resection) was 18.7% versus 0% for local excision. Additionally, the 5-year survival rates of patients in the abdominoperineal resection group with maximum tumor diameters of ≤5 cm and >5 cm were 37.6% and 0%, respectively. The 5-year survival rates for patients with depth of invasion within and beyond the muscularis propria were 37.2% and 6.5%, respectively. The presence or absence of lymphatic metastasis has not demonstrated any significant difference in prognosis. The conditions for ensured long-term survival are: 1) maximum tumor diameter <5 cm, 2) wall invasion depth within the muscularis propria, and 3) extensive lymph node dissection regardless of the presence or absence of lymphatic metastasis [8]. In Japan, few studies have assessed the long-term survival of patients who have undergone local excision. Long-term survival has been demonstrated in patients who have undergone abdominoperineal resection with extended lymph node dissection, including patients with lymphatic metastasis; thus, abdominoperineal resection has been widely recommended. In Western countries, there is no significant difference in survival rate between rectal resection with a sufficient safety margin and local excision [9], and most surgical treatment is considered palliative therapy due to the possibility of disease already being advanced at the time of onset and diagnosis [10]. Therefore, some investigators have recommended local excision rather than rectal resection for better quality of life [11,12]. A recent report described how rectal MRI and transrectal endoscopic ultrasonography can better reveal local lymph nodes, thus providing essential information for planning therapeutic strategies [13]. Notably, multiple lesions spaced apart from the main lesion were observed in the anal canal of our patient. Our case emphasizes the necessity of determining the type of surgical procedure as well as the extent of resection by carefully considering the possibility of non-contiguity with the primary lesion.

Melanoma in situ was found pathologically in some of the non-contiguous black spots that were separated from the main lesion in the present case. Such black spots are found in the mucosa of malignant esophageal melanoma and normally indicate tumor progression. Owing to the involvement of increased melanin granules in the mucosal basal layer, it is believed that the tumor originates within that layer [14,15]. The horizontal spreading in the mucosa is consistent with findings of radial growth in cutaneous malignant

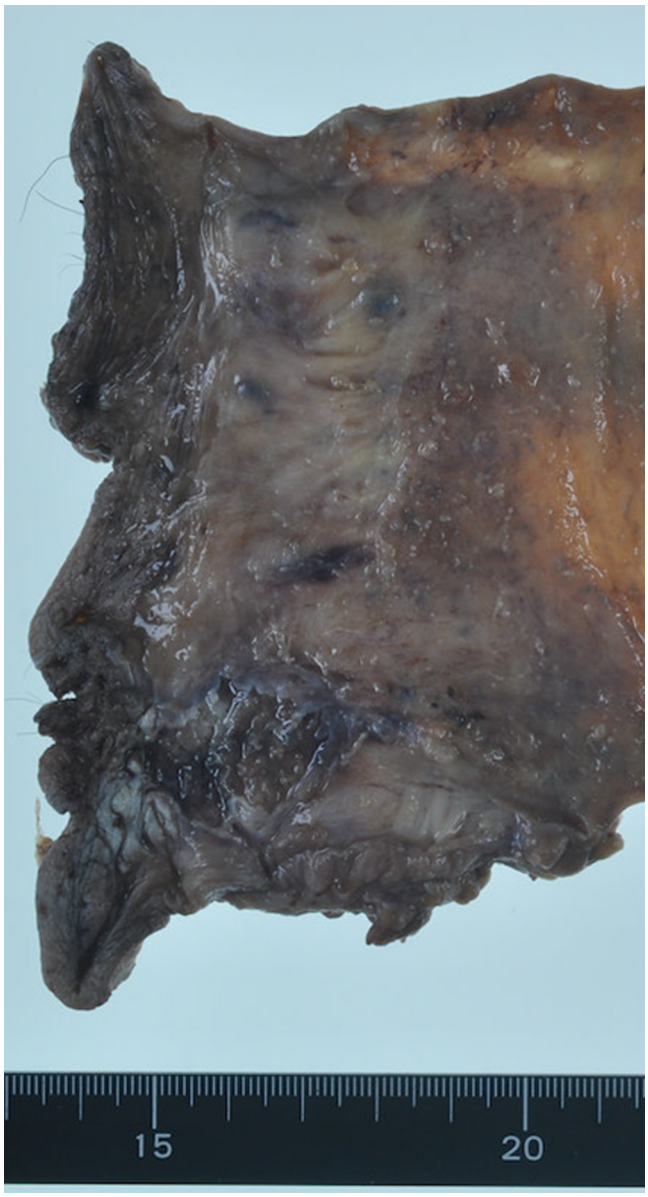

Fig. 4. The surgical specimen: black changes were found at the anal canal.

melanoma and atypical junctional activity in esophageal malignant melanoma; these are considered to be the earliest forms of tumor that can be diagnosed pathologically [14–16]. Because the tumor cells have not yet reached the blood vessels, a tumor detected at this stage can be completely excised. Search results of the national database for mucosal spreading of anorectal malignant melanoma showed that, although there were four previously reported cases, none showed extensions spreading to a 7.0 × 2.3 cm area as was observed in the present case. There are few reported cases of skipped lesions in anorectal malignant melanoma, and it is well known that these lesions represent the earliest form of pathologically diagnosable tumors in cutaneous and esophageal malignant melanoma. Hence, examinations and surgical procedures should be carefully performed in order to detect possible skipped lesions along with the main lesion. The surrounding black spots could not be detected prior to total resection in the present case; this was considered to be due to contact with the anal verge, which made it difficult to recognize the black spots during preoperative endoscopic observation.

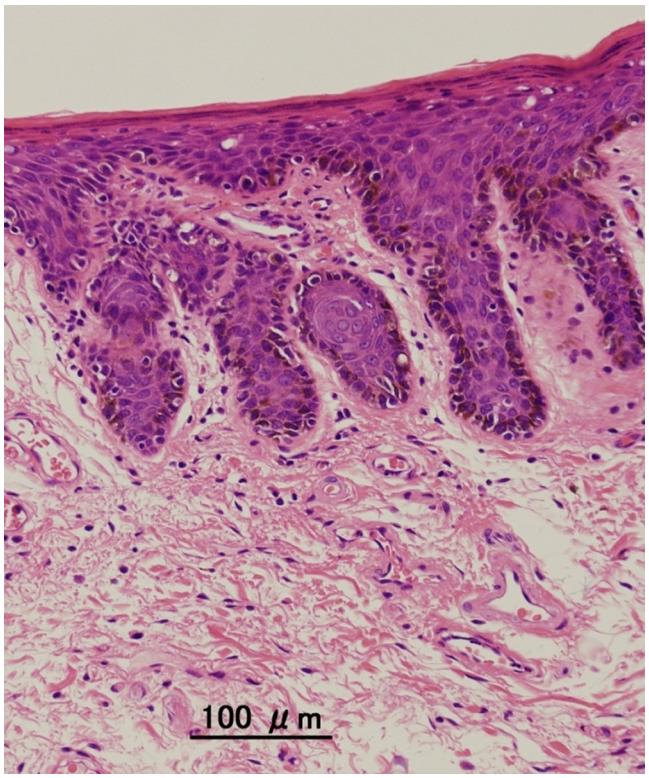

Fig. 5. Surgical pathology: intraepithelial basolateral proliferation of large atypical melanocytes with increased chromatin, intraepithelial basolateral proliferation, nuclear irregularity, and partial cancer cell nest formation were observed. These melanocytes were found in an area of approximately 7.0 × 2.3 cm around the main lesion, although interstitial infiltration was not evident in the specimen.

4. Conclusion

The take-home messages of this case are 1) The possibility exists of an extended tumorous area of malignant melanoma occurring at a site that is macroscopically remote from the main lesion, 2) such tumors can spread horizontally in the mucosa, and 3) it is therefore of utmost importance to carefully observe pigment changes in the surrounding area before and during surgery, as well as to verify that the anorectal area has been properly examined. This will ensure detection of all lesions and will be critical for determining the type of surgical procedure as well as the extent of resection.

Conflicts of interest

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Ethical approval

This submission is case report and we does not need any ethical approval.

Consent

We obtained consent to publish a case report from the patient.

Author's contribution

All authors contributed to study concept or design at this submission.

Guarantor

Corresponding author; Keiichi Arakawa.

Care criteria compliance

The case described in this manuscript was reported in line with the CARE criteria as outlined in the CARE statement (http://www.care-statement.org/), as applicable to this type of case.

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