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Case Report

Polypoid Malignant Melanoma with Spontaneous Decapitation Showing Favorable Prognosis: A Case Report

Yasuaki Ikuno Hiraku Kokubu Miwa Kato Kazuma Okada
Akihiko Yamaguchi Toshifumi Takahashi Takeshi Kato Noriki Fujimoto

Department of Dermatology, Shiga University of Medical Science, Setatsukinowa, Otsu, Japan

Keywords
Malignant melanoma · Spontaneous decapitation · Favorable prognosis

Abstract
Introduction: Regression of malignant melanoma (MM) is a commonly observed phenomenon, which refers to disappearance or loss of all or part of MM. It can be identified both clinically and histologically, and high probability of histological regression of MM (10–58%) has been reported. However, the decapitation of skin tumor has rarely been described in the English literature, and decapitation in MM has not been reported. Here, we report the case of polypoid MM with spontaneous decapitation. ***Case Presentation:*** An 81-year-old Japanese woman was referred to our hospital due to a polypoid nodule on her cheek. She was diagnosed with MM at stage IIC (T4bN0M0) by histological examinations. Three weeks after the biopsy, the nodule decapitated spontaneously without bleeding, and an ulcer developed on the base of the polypoid nodule. The histological examination of the additionally resected ulcerative lesion under the nodule revealed infiltration of T cells mainly composed of $CD8^+$ natural killer T cells. No recurrence or metastasis has been observed for 4 years. ***Conclusion:*** This is the first case report of polypoid MM with spontaneous decapitation, which may be attributed to natural killer T cells. Moreover, this case shows favorable prognosis, while it is said that the regression in thick MM does not have prognostic advantage and polypoid topography has been reported to be related to extremely poor prognosis. Further investigations are needed to evaluate the prognostic advantage of decapitation in MM and other skin tumors.

Correspondence to:
Hiraku Kokubu, kokubu@belle.shiga-med.ac.jp

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Introduction

Malignant melanoma (MM) is the most aggressive and deadly of skin cancers. Several observations point to melanoma as an immune-responsive tumor [1], immune checkpoint inhibitors are the cornerstone of cancer therapy, and a wide amount of practice-changing clinical trials led to the approval of immunotherapy in the treatment algorithm of MM [2–4].

As a consequence of a host immunological response directed against the tumor cells, regression in MM is commonly observed. Regression in MM refers to disappearance or loss of all or part of MM. It can be identified both clinically (macroscopically) and histologically (microscopically), and is sometimes apparent to the naked eye. Macroscopic regression can be appreciated using a dermatoscope to examine a pigmented lesion. The presence of regression is commonly observed not only in melanoma, but also in benign nevi and other tumors [5, 6].

In contrast, the decapitation is the occasional phenomenon often reported in gastrointestinal tumor [7]. However, the decapitation of skin tumor has rarely been described in the English literature, and decapitation in MM has not been reported. Here, we report the case of polypoid MM with spontaneous decapitation. After the biopsy, the polypoid MM decapitated spontaneously, and no recurrence or metastasis has been observed for 4 years. While it is known that regression in MM is sometimes observed, this is the first case report of polypoid MM with spontaneous decapitation. Moreover, this case shows favorable prognosis, while it is said that the regression in thick MM does not have prognostic advantage and polypoid topography has been reported to be related to extremely poor prognosis. The CARE Checklist has been completed by the authors for this case report, attached as supplementary material (for all online suppl. material, see https://doi.org/10.1159/000535194).

Case Report

An 81-year-old Japanese woman was referred to our hospital due to a polypoid nodule on her cheek in 2018. Physical examination showed a polypoid erythematosus nodule measuring 25 × 20 mm with crusts on her left cheek (Fig. 1a). Because she had mild dementia, it was uncertain when the nodule grew to be polypoid. Laboratory examinations were all within normal limits. Computed tomography scans of the entire body revealed no lymphadenopathies or metastasis. Histological examination of a biopsy specimen from the nodule showed a proliferation of atypical spindle cells and infiltrating lymphocytes on the dermis (Fig. 1b, c). Immunohistochemical staining revealed that spindle cells were positive for SOX10, partially positive for melan-A and tyrosinase, and negative for CK-AE1/3, CK-34βE12, α-SMA, and desmin. We diagnosed her with MM at stage IIC (T4bN0M0) and planned to excise it. However, 3 weeks after the biopsy, the nodule decapitated spontaneously without bleeding, and an ulcer developed on the base of the polypoid nodule (Fig. 2a). Her family brought the decapitated tumor. Histological and immunohistochemical examination of the decapitated nodule showed SOX10, melan-A, and tyrosinase-positive proliferating atypical spindle cells covered by the epidermis and necrotic cells around the peduncle of this nodule (Fig. 2b, c). We additionally resected the ulcerative lesion with a 10 mm surgical margin. Histological examination of the resected specimen showed no atypical cells with infiltration of lymphocytes. The immunohistochemical examination revealed that these lymphocytes were dominantly positive for CD3 and CD8, and partially positive for CD4, around the ulcer (Fig. 2d, e). After that, no recurrence or metastasis has been observed for 4 years without any therapies (Fig. 3).

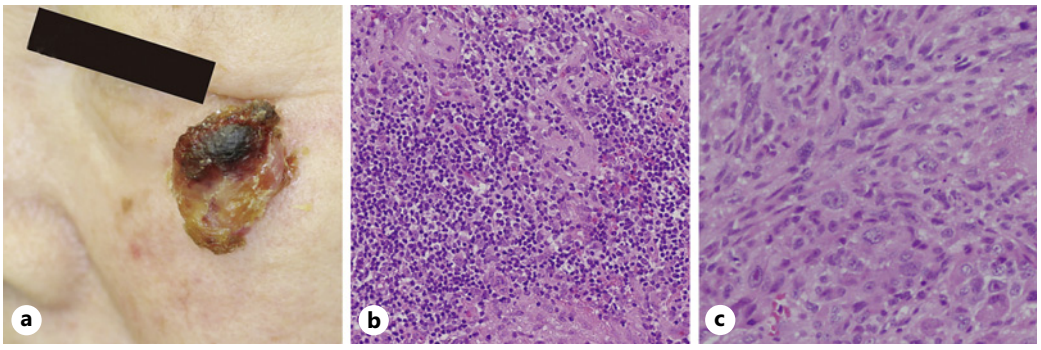

Fig. 1. a Clinical presentation at the first visit. A polypoid erythematosus nodule measuring 25 × 20 mm with scales, crusts, and ulcers on her left cheek. **b**, **c** Histological examination of a biopsy specimen from the nodule showed a proliferation of atypical spindle cells and infiltrating lymphocytes on the dermis [hematoxylin-and-eosin staining; original magnification, ×100 (**b**) and ×200 (**c**)].

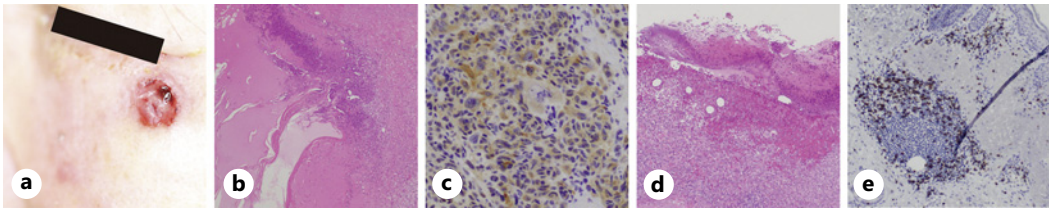

Fig. 2. a Clinical presentation after the decapitation of a nodule. An ulcer measuring 12 × 8 mm developed on the base of the polypoid nodule. **b** Histological examination of the decapitated nodule showed proliferating atypical spindle cells covered by the epidermis and necrotic cells around the peduncle of this nodule (hematoxylin-and-eosin staining; original magnification, ×40). **c** Immunohistochemical staining showed that proliferating cells were positive for tyrosinase (original magnification, ×100). **d** Histological examination of the resected specimen of the ulcerative lesion showed no atypical cells with infiltration of lymphocytes around the ulcer (hematoxylin-and-eosin staining; original magnification, ×40). **e** Immunohistochemical staining showed that infiltrating lymphocytes around the ulcer were positive for CD8 (original magnification, ×40).

Discussion

Regression in MM is sometimes apparent to the naked eye and is histologically characterized by dermal fibrosis, inflammatory infiltrate, melanophages, ecstatic blood vessels, epidermal attenuation, and/or apoptosis of keratinocytes or melanocytes [5]. The histological regression of MM is estimated to occur between 10% and 58% [8, 9]. Although many cases of MM showing regression have been reported, to our knowledge, this is the first case of polypoid MM that might decapitate spontaneously from the skin. Considering the histological features, including necrotic change around the peduncle and infiltrating lymphocytes, and the clinical feature that no bleeding was observed at decapitation, this decapitation was probably caused not accidentally but immunologically through histological regression mediated by lymphocytes and apoptosis of keratinocytes at the base of the peduncle mediated by lymphocytes. The triggers generating antitumor immunity during spontaneous tumor regression have been reported, and the initiating event might be related to the trauma from biopsy [10]; therefore, the skin biopsy might have acted as a trigger of inflammation leading to tumor decapitation in our case. The infiltrating lymphocytes, especially natural killer T cells, have been identified in melanoma lesions usually

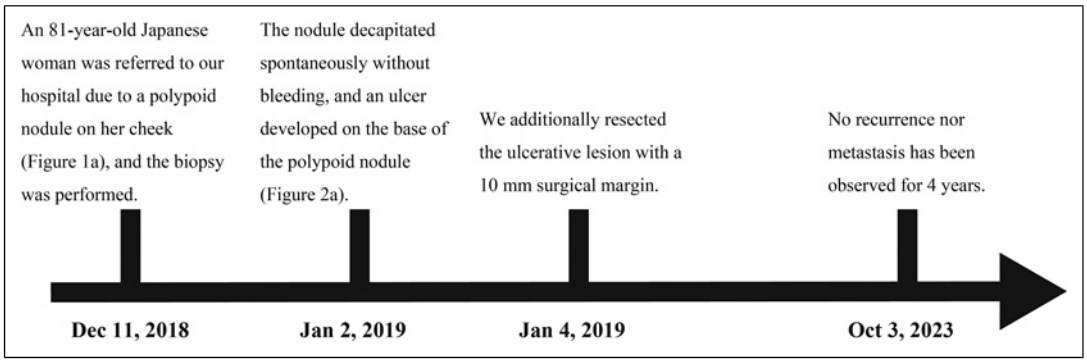

Fig. 3. A timeline summarizing the main events of this case.

associated with spontaneous tumor regression and favorable prognostic in primary melanoma [11]. In our case, dominantly infiltrating T cells, mainly composed of $CD8^+$ natural killer T cells, may contribute to MM decapitation and favorable prognosis. Further investigations are needed to elucidate the contribution of natural killer T cells to MM decapitation.

The recent cohort study reported that histologically confirmed regression is a favorable prognostic factor, especially in thin Breslow thickness MM. However, in thick MM, the regression lost its prognostic advantage [6]. In addition, polypoid topography has been reported to be related to extremely poor prognosis [12, 13]. In our case, the patient has been free from recurrence and metastasis even 4 years after the decapitation. Decapitation might be a favorable prognostic factor even in thick polypoid MM. Prognostic impact of decapitation has not been reported in any skin tumors, except for a case report of spontaneously decapitated DFSP with relapse-free survival 5 months after Mohs micrographic surgery [14]. Also, regression and decapitation of MM reflect host immunological response directed against the tumor cells; therefore, immune checkpoint inhibitors may enhance this response. However, the impact of regression or decapitation on efficacy of immune checkpoint inhibitor is largely unknown. Further investigations are needed to evaluate the prognostic advantage of decapitation and impact of decapitation on efficacy of immune checkpoint inhibitor in MM and other skin tumors.

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Statement of Ethics

Written informed consent was obtained from the patient for publication of this case report and any accompanying image. Ethical approval was not required for this study in accordance with local or national guidelines.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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Author Contributions

Y.I. and H.K. contributed to the design of the report and the pathology revision and drafted the manuscript. Y.I., H.K., M.K., K.O, A.Y, T.T, T.K., and N.F. contributed to the design of the report. H.K., A.Y., T.T., and N.F. critically revised the manuscript.

Data Availability Statement

All data generated or analyzed during this study are included in this article and its online supplementary material. Further inquiries can be directed to the corresponding author.

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