

Single Case

Amelanotic Melanoma Arising in Surgical Cicatrix: A Case Report

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Keywords

Amelanotic melanoma · Cicatrix · Neoplasm · Marjolin's ulcer · Case report

Abstract

Introduction: Amelanotic melanoma (AM) is a rare subtype of melanoma characterized by the absence of pigmentation, making it difficult to diagnose. This case report describes AM arising in a cicatrix following cardiac surgery, highlighting diagnostic challenges and the importance of reassessment when the clinical findings do not fit the initial diagnosis. **Case Presentation:** A 76-year-old male underwent coronary artery bypass grafting in March 2022. Nine months later, he noticed a mass with ulceration in the sternal cicatrix. A punch biopsy showed inflammation and fibrosis, leading to a diagnosis of suture granuloma. Despite the lesion's progression over 8 months, no further biopsies or treatments were performed. In September 2023, the patient was referred to the Department of Plastic Surgery at Copenhagen University Hospital, Rigshospitalet, where an excision biopsy revealed an AM with a thickness of 20 mm. Preoperative PET-CT showed no evidence of metastasis, and re-excision with sentinel lymph node biopsy was performed. Adjuvant oncologic treatment was administered due to a positive sentinel node. **Conclusion:** This case emphasizes the diagnostic challenges of AM, especially when arising in a cicatrix. When the clinical picture does not align with the initial diagnosis, it is crucial to reconsider and conduct further evaluations to avoid delays in treatment.

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Introduction

Amelanotic melanoma (AM) is a rare and diagnostically challenging subtype of melanoma, primarily due to its lack of pigmentation and atypical presentation. Unlike conventional melanomas, AM often does not exhibit the hallmark ABCDE criteria (Asymmetry, Border irregularity, Colour variation, Diameter over 6 mm, and Evolution), making early detection difficult [1]. As a result, AM is often diagnosed at a more advanced stage compared to other types of melanoma [2], which leads to a poorer prognosis [3].

Malignant transformation within scar tissue is a recognized but uncommon phenomenon, called Marjolin's ulcer. It predominantly involves squamous cell carcinoma or basal cell carcinoma. While Marjolin's ulcers typically develop over a prolonged period, they can, in rare instances, present within a year [4].

The occurrence of melanoma within scar tissue is very rare [4, 5], with most cases reported in burn scars. This report describes a unique case of AM developing in a sternal cicatrix following coronary artery bypass grafting (CABG), emphasizing the diagnostic challenges.

Case Presentation

We present a 76-year-old well-preserved male, with a Fitzpatrick skin type 1 and frequent sunburns during childhood and adulthood. The patient had no prior malignancies or family history of melanoma.

In March 2022, the patient underwent CABG due to an acute myocardial infarction. Nine months post-surgery, the patient noticed a mass with ulceration in the sternal scar and sought medical attention. A punch biopsy performed by a dermatologist revealed inflammation and fibrosis without evidence of malignancy. The lesion was diagnosed as a suture granuloma.

During the subsequent 8 months, the patient had 16 consultations with seven different physicians from various medical specialties (general practitioner, dermatologist, orthopedic surgeon, plastic surgeon, and cardiothoracic surgeon). Despite the progression of the lesion, no further biopsies or treatment with any topical medication were performed. At one point, the patient was treated with the peroral antibiotic dicloxacillin due to a local infection with *Staphylococcus aureus*.

In September 2023, the patient was referred to the Department of Plastic Surgery at Rigshospitalet, Copenhagen, Denmark, for further evaluation. Clinical examination revealed a 40 × 40 × 20 mm ulcerated and nonpigmented tumor with central necrosis in the caudal part of the sternal cicatrix shown in Figure 1. An excisional biopsy was performed, and the histological examination revealed an AM with a thickness of 20 mm, with mitoses and ulceration. The histological image and further description are shown in Figure 2. Preoperative PET-CT showed no evidence of metastasis. According to standard guidelines, re-excision and sentinel lymph node biopsy were performed, with the sentinel node being positive with a melanoma metastasis of 1.6 mm staging the melanoma as T4bN1M0 according to the UICC/AJCC TNM classification.

The patient received adjuvant oncologic treatment with immunotherapy consisting of eight cycles of pembrolizumab without any major adverse effects. At 1-year follow-up, clinical examination of the skin and PET-CT scan showed no evidence of recurrence.

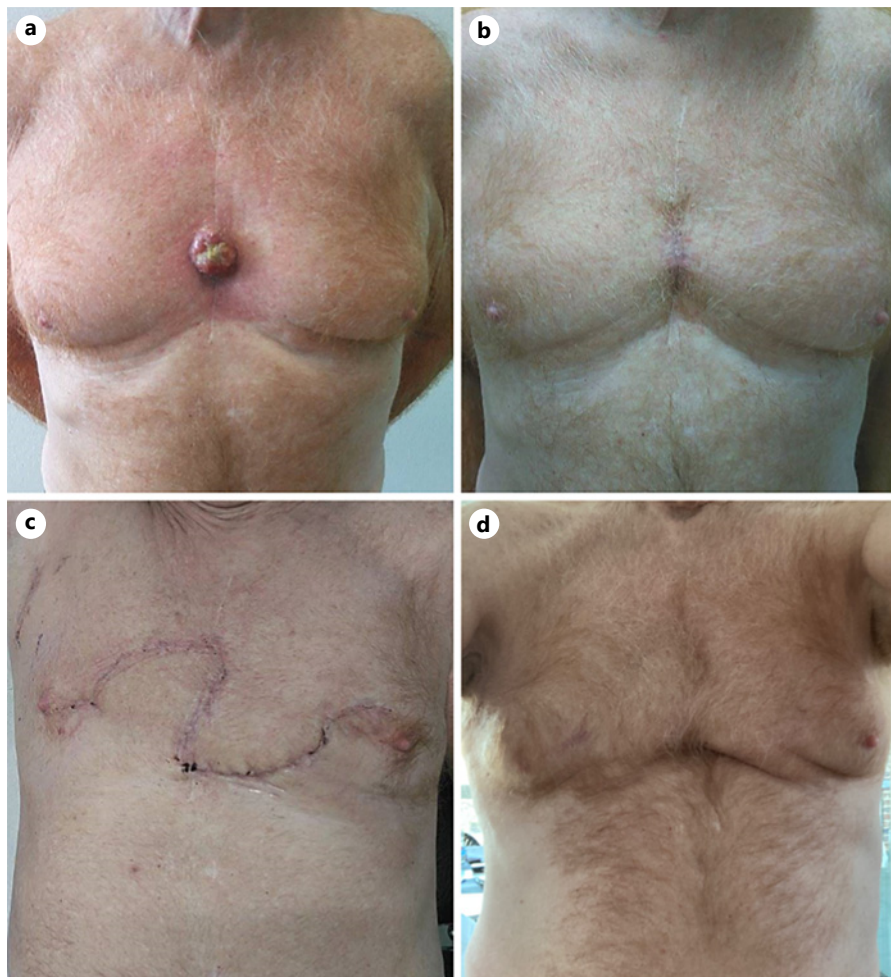


Fig. 1. Clinical photograph showing the presentation of the AM prior to excision. **a** The lesion, located at the sternal cicatrix following coronary artery bypass grafting (CABG), lacks pigmentation and exhibits an atypical appearance of melanoma. **b** Clinical photograph of the cicatrix 1 month after complete removal of the melanoma with the initial excisional biopsy. **c** Clinical photograph taken 2 weeks after re-excision to a 2 cm margin and reconstruction of the defect using bilateral hatchet flaps. **d** Clinical photograph 1 year and 4 months after the re-excision and reconstruction and 3 months after end treatment with pembrolizumab, showing a satisfactory esthetic outcome and no signs of recurrence.

Discussion

This case highlights the significant diagnostic challenges associated with AM [1]. Due to its atypical presentation, AM is often misidentified as a benign or less aggressive condition, as seen in this case. It arose in cicatrix and the initial biopsy, on histology, demonstrated fibrosis and inflammation only, and was subsequently diagnosed as a suture granuloma.

The literature reports that these diagnostic difficulties contribute to delayed diagnosis and an increased risk of metastasis [2]. In this case, the lack of pigmentation and unusual presentation delayed accurate diagnosis and appropriate treatment. It may have contributed to the patient having metastasis in the sentinel lymph node at the time of diagnosis.

According to the patient, there was no evidence of a lesion before surgery. However, it is uncertain whether the melanoma developed in the incision line before and was coincidentally found within the cicatrix or whether a chronic inflammation, such as a suture granuloma,

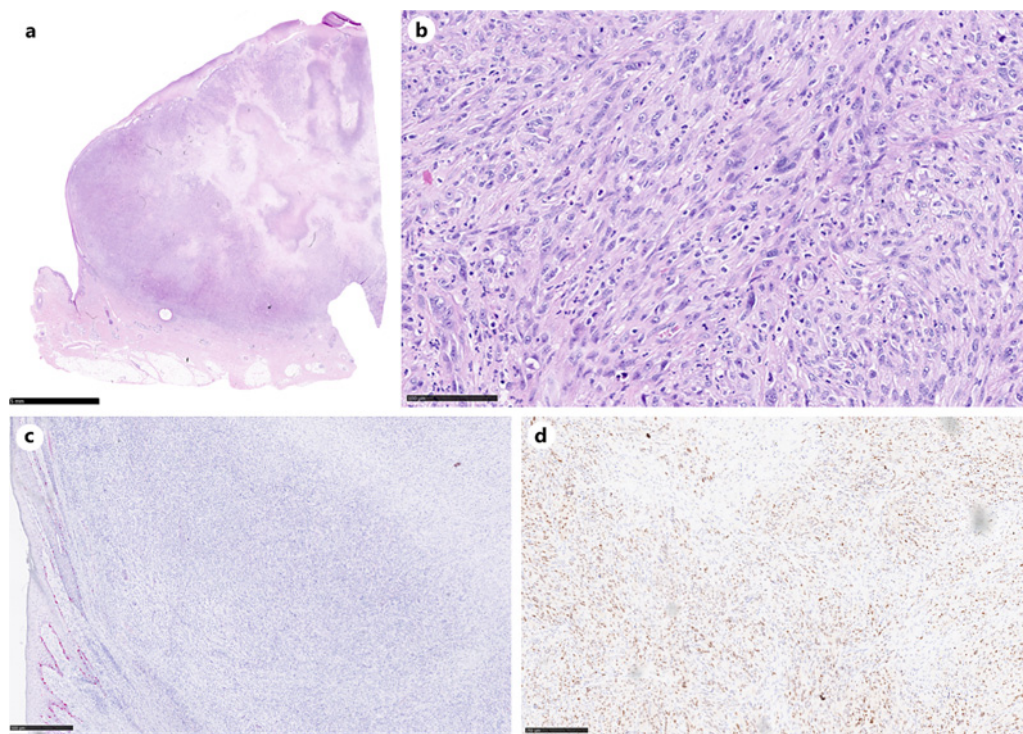


Fig. 2. **a** Histological sections through skin excision showing large tumor with ulcerated surface epithelium. **b** Higher magnification shows spindle-shaped tumor cells with pleomorphic and elongated nuclei, while in other areas, they are more rounded. The cytoplasm is abundantly light eosinophilic with no pigmentation. Numerous mitoses are present in the tumor tissue, including several abnormal ones. **c** Immunohistochemically, there is loss of Melan-A except for normally appearing single-layered melanocytes located basally in the reactive epithelium. **d** The tumor cells are positive for S-100, SOX10 (not shown), and PRAME.

could have played a role in the development of the malignancy, categorizing it as a rare instance of Marjolin's ulcer [4, 5].

Due to the mechanisms of chronic inflammation and association with thermal burns in the development of Marjolin's ulcer, theoretically, the use of electrocautery, such as diathermy, near the skin during the CABG, might have played a role in the development of the neoplasm, but to our knowledge, no literature suggests a connection between diathermy and local malignancies. To our knowledge, AM arising in a cicatrix within a year has not previously been described in the literature, regardless if it is categorized as a Marjolin's ulcer or not.

The patient expressed frustration with the prolonged diagnostic process and felt unheard by multiple specialists who deemed the lesion benign. He emphasized the importance of increased awareness of this tumor, as he had to persistently advocate for a more thorough evaluation.

Conclusion

The progression of the lesion without further biopsy underscores the need for heightened vigilance and the importance of reconsidering the diagnosis when clinical findings do not align with the initial diagnosis. Early recognition and timely intervention are essential in managing this aggressive type of melanoma to prevent metastasis and delays in treatment.

The CARE Checklist has been completed by the authors for this case report, attached as online supplementary material (for all online suppl. material, see <https://doi.org/10.1159/000545287>).

Statement of Ethics

Ethical approval is not required for this study in accordance with local or national guidelines. A written informed consent was obtained from the patient for publication of the details of their medical case and the accompanying image.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

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Author Contributions

A.L.N. has drafted the manuscript and made a final approval of the manuscript. C.B.B. has performed the pathology evaluation and description and made a final approval of the manuscript. L.L. has reviewed and made a final approval of the manuscript.

Data Availability Statement

The data used for this case report are the patient's medical journal in an IT program with restricted access to the medical team treating this patient and are therefore not publicly available. Further inquiries can be directed to the corresponding author.

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