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Extraocular Sebaceous Carcinoma of the Toe in a Young Adult: A Case Report.

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ABSTRACT

Background: Sebaceous carcinoma is a rare and potentially aggressive malignant adnexal tumor, most commonly occurring in the periocular region. Extraocular presentations, particularly involving the extremities, are uncommon and may pose diagnostic and management challenges due to their non-specific clinical appearance.

Case Presentation: We report the case of a 22-year-old female who presented with a chronic non-healing lesion over the medial aspect of the right big toe. The lesion had been present for two years with progressive enlargement. Initial excision and skin grafting were performed at an outside hospital; however, histopathology revealed positive surgical margins, and the wound failed to heal. Further evaluation confirmed sebaceous carcinoma with deep margin involvement. The patient was referred to a tertiary care center, where a definitive wide local re-excision achieved clear margins. The surgical defect entirely healed by secondary intention without the need for further reconstruction.

Conclusion: This case highlights an unusual presentation of extraocular sebaceous carcinoma in a young patient and underscores the importance of early suspicion, accurate histopathological diagnosis, and complete surgical excision with clear margins. Adequate oncologic management can result in favorable outcomes even in rare and atypical presentations..

Keywords: *Sebaceous carcinoma, Extraocular sebaceous carcinoma, Toe neoplasm, Wide local excision, Case report*

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1. INTRODUCTION

Sebaceous carcinoma is a rare, malignant adnexal tumor originating from sebaceous glands, accounting for less than 1% of all cutaneous malignancies and approximately 1–5% of eyelid cancers worldwide [1–3]. It is classically described as an aggressive neoplasm with a propensity for local recurrence, regional lymph node involvement, and distant metastasis if not adequately treated. Clinically, sebaceous carcinoma is broadly classified into periocular and extraocular forms, with periocular tumors constituting nearly 70–75% of reported cases due to the abundance of sebaceous glands in the eyelids, particularly the Meibomian glands [4,5].

Extraocular sebaceous carcinoma is considerably less common and may arise in areas with relatively sparse sebaceous glands, including the head and neck, trunk, genital region, and, rarely, the extremities [5–7]. Because of its rarity and nonspecific clinical presentation, extraocular sebaceous carcinoma frequently mimics benign lesions or other cutaneous malignancies such as squamous cell carcinoma, basal cell carcinoma, verrucous carcinoma, or chronic non-healing ulcers.

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As a result, diagnosis is often delayed, and initial surgical management may be inadequate, increasing the risk of recurrence [3,5].

The disease typically affects elderly individuals, with a peak incidence in the sixth to seventh decades of life, and a slight male predominance has been reported in large population-based studies [3]. Occurrence in young adults is distinctly uncommon and may raise consideration of underlying genetic syndromes such as Muir–Torre syndrome, a variant of Lynch syndrome associated with sebaceous neoplasms and visceral malignancies [3,5]. Although most extraocular sebaceous carcinomas arise sporadically, increasing evidence highlights the importance of careful clinical and pathological evaluation to exclude syndromic associations, particularly in younger patients.

Sebaceous carcinoma of the foot is exceptionally rare, with only a limited number of cases reported in the literature. Lesions in this location may present as chronic non-healing ulcers or nodular masses subjected to repeated trauma and pressure, further obscuring the diagnosis. This case report describes an unusual presentation of extraocular sebaceous carcinoma involving the toe in a young adult female, emphasizing the diagnostic challenges, importance of histopathological confirmation, and necessity of complete surgical excision with clear margins to achieve favorable outcomes.

CASE PRESENTATION

A 22-year-old female patient, with no known medical comorbidities, first presented to the Plastic and Reconstructive Surgery outpatient clinic on 30 April 2023 with a non-healing lesion over the medial aspect of the right big toe (**Figure 1).**

Figure 1. Preoperative clinical photograph showing the initial presentation of a non-healing lesion over the medial aspect of the right big toe.

The patient reported that the lesion had been present for approximately two years, initially appearing as a small swelling, with progressive enlargement over the preceding year. The lesion was associated with persistent non-healing ulceration, but there was no history of active bleeding, discharge, or systemic symptoms such as fever, weight loss, or night sweats.

Prior to presentation at our institution, the patient sought medical attention at a private hospital, where an initial biopsy was performed. The external histopathology report suggested a skin adnexal neoplasm, prompting referral for further evaluation.

She had no personal history of malignancy, and no family history of cancer was reported. There was no history of trauma to the affected area. Psychosocial history was unremarkable, and there was no known exposure to radiation, immunosuppression, or genetic syndromes associated with adnexal tumors.

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As part of management at the outside hospital, the patient underwent lesion excision with skin graft coverage on 8 August 2023. Postoperatively, the wound showed delayed healing, and the histopathology report demonstrated positive surgical margins. She subsequently re-presented to our service on 29 August 2023 with a grafted but non-healing wound, along with documentation of positive margins, for further evaluation and management (**Figure 2).**

Figure 2. Postoperative clinical photograph following initial lesion excision and skin grafting at an outside hospital, demonstrating a non-healing grafted wound on re-presentation.

On initial examination at our clinic, local inspection of the right foot revealed a non-healed wound over the medial eminence of the right big toe, measuring approximately 3 cm in greatest dimension. The lesion involved the plantar–medial aspect of the toe and extended to the skin surface.

There were no signs of active bleeding, purulent discharge, or surrounding cellulitis. The wound bed appeared clean, and there were no clinical features suggestive of acute infection. The distal neurovascular status of the foot was intact. No palpable regional lymphadenopathy was detected on clinical examination. At follow-up after the external excision, the wound was noted to be covered by a skin graft, with incomplete healing but no acute inflammatory signs.

Magnetic resonance imaging (MRI) of the right foot demonstrated a well-defined subcutaneous soft tissue mass located at the medial plantar aspect of the right big toe, at the level of the proximal phalanx, reaching the skin surface. The lesion measured approximately 2.6 × 1.5 × 2.0 cm.

On MRI, the mass showed low signal intensity on T1-weighted images, intermediate signal on T2-weighted images, and high signal on proton density fat-suppressed sequences, with intense enhancement following intravenous contrast administration. Importantly, there was no evidence of invasion of the underlying tendons, bones, or adjacent joints, and no extension into the metatarsophalangeal or interphalangeal joints (**Figure 3).** A CT scan of the head and neck with intravenous contrast was performed as part of the staging workup and showed no evidence of regional or distant disease,

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with an unremarkable study.

Figure 3. Magnetic resonance imaging of the right foot demonstrating a subcutaneous soft tissue mass at the medial plantar aspect of the right big toe without bone or tendon invasion.

Serum tumor markers, including CA 19-9, CA-125, CA 15-3, carcinoembryonic antigen (CEA), and alpha-fetoprotein (AFP), were all within normal limits. Routine laboratory investigations were unremarkable.

Definitive histopathological evaluation of the excised right big toe lesion revealed a malignant epithelial neoplasm involving the dermis and subcutaneous tissue. Microscopic examination demonstrated infiltrating epithelioid tumor cells arranged in irregular nests and sheets, associated with moderate cytologic atypia and scattered mitotic figures (**Figure 5A-C).**

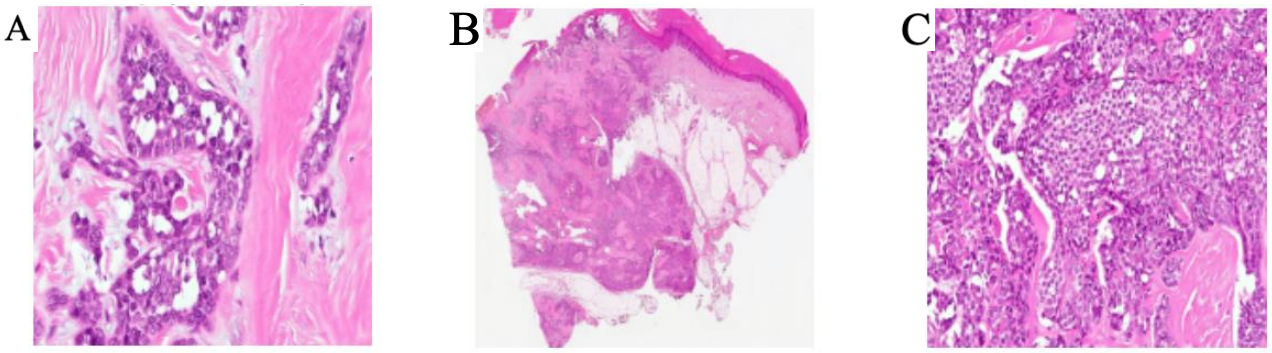

Figure 5. Histopathological findinds (hematoxylin and eosin stain) demonstrating malignant adnexal tumor morphology consistent with sebaceous carcinoma

The lesion was ulcerated and extended into the subcutaneous fat, with tumor involvement reaching the proximal and deep surgical margins, while peripheral margins were free of tumor. No lymphovascular or perineural invasion was identified. The tumor measured approximately 2.9 cm in greatest dimension.

Based on the histomorphologic features and subsequent expert pathology review, the findings were consistent with sebaceous carcinoma, confirming incomplete excision at the initial surgery and necessitating definitive re-excision.

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Based on the clinical presentation, imaging findings, and histopathology, the differential diagnosis included: Sebaceous carcinoma; other malignant adnexal tumors (e.g., porocarcinoma, hidradenocarcinoma); squamous cell carcinoma of the foot; verrucous carcinoma; and chronic non-healing ulcer with malignant transformation (Marjolin ulcer). Histopathological features and expert review favored a diagnosis of sebaceous carcinoma.

Given the diagnosis of sebaceous carcinoma with positive deep margins and evidence of local invasion, the patient was referred to a higher-specialty tertiary hospital for definitive oncologic management.

At the tertiary center, the patient underwent a second surgical procedure on 15 October 2023, consisting of wide local re-excision of the lesion. Histopathological assessment following re-excision confirmed clear (free) surgical margins.

Given satisfactory local wound conditions and adequate oncologic clearance, the surgical defect was managed by secondary intention healing, and no skin graft or additional reconstructive procedure was required.

Following definitive re-excision, the wound demonstrated progressive and complete healing by secondary intention. No postoperative complications were reported, and no further surgical intervention was required.

At the time of last follow-up, the patient had achieved complete wound healing, with no clinical evidence of residual or recurrent disease. She continues follow-up under specialized care for ongoing surveillance (**Figure 6).**

Figure 6. Final follow-up clinical photograph showing complete wound healing by secondary intention without the need for skin grafting.

2. DISCUSSION

Sebaceous carcinoma is an uncommon but clinically significant cutaneous malignancy characterized by aggressive local behavior and potential for regional and distant metastasis. While periocular sebaceous carcinoma has been extensively described, extraocular disease remains poorly characterized due to its rarity and heterogeneous clinical presentation [3,5]. The present case is notable for several reasons, including its occurrence in a young adult, its rare anatomical location involving the toe, and the initial incomplete excision that necessitated definitive oncologic re-intervention.

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Extraocular sebaceous carcinoma most frequently arises in the head and neck region, followed by the trunk and genital area, reflecting the distribution of sebaceous glands [5–7]. Involvement of the distal extremities is exceptionally rare, and lesions arising in the foot are particularly prone to misdiagnosis due to chronic irritation, pressure-related changes, and resemblance to benign conditions such as plantar warts, calluses, or traumatic ulcers [8]. In this case, the lesion persisted for nearly two years before definitive diagnosis, illustrating the diagnostic delay commonly reported in extraocular sebaceous carcinoma [3,5].

Age is a critical epidemiological consideration. Most cases occur in older adults, and presentation in patients under 30 years of age is distinctly uncommon [3]. When sebaceous carcinoma occurs in young individuals, clinicians should consider the possibility of Muir–Torre syndrome, particularly if there is a personal or family history of visceral malignancies [5]. Although no such history was identified in the present case, the patient's age underscores the importance of maintaining vigilance for sebaceous carcinoma even outside the typical demographic profile.

Histopathologically, sebaceous carcinoma is characterized by lobules or nests of atypical epithelial cells with sebaceous differentiation, cytologic atypia, mitotic activity, and infiltrative growth patterns [4,5]. Immunohistochemistry, although not required in all cases, may aid in differentiating sebaceous carcinoma from other adnexal tumors, particularly poorly differentiated squamous cell carcinoma or basal cell carcinoma with sebaceous differentiation. Accurate histopathological diagnosis is essential, as misclassification may result in inadequate surgical margins and suboptimal treatment.

Surgical excision with histologically clear margins remains the cornerstone of management for both periocular and extraocular sebaceous carcinoma [5]. Wide local excision or Mohs micrographic surgery is recommended when feasible, particularly in anatomically sensitive areas [9]. In extraocular lesions, wide local excision with margin assessment is generally sufficient, provided that complete tumor clearance is achieved. In our case, initial excision at an outside institution resulted in positive deep margins, highlighting a common pitfall in the management of rare adnexal malignancies. Subsequent wide local re-excision at a tertiary center achieved clear margins and definitive disease control.

Reconstruction following excision of sebaceous carcinoma should be individualized based on oncologic safety, wound characteristics, and patient factors. Although skin grafting or flap reconstruction is often considered, healing by secondary intention may be appropriate in selected cases, particularly when margins are clear and the wound bed is favorable. In this patient, secondary intention healing resulted in complete wound closure without complications, demonstrating that complex reconstruction is not always necessary when oncologic principles are prioritized.

The role of sentinel lymph node biopsy and adjuvant therapy in extraocular sebaceous carcinoma remains controversial. While regional lymph node metastasis has been reported, especially in larger or poorly differentiated tumors, routine sentinel lymph node biopsy is not universally recommended and should be considered on a case-by-case basis [3,5]. Similarly, adjuvant radiotherapy is generally reserved for unresectable disease, recurrent tumors, or cases with perineural invasion or nodal involvement [10]. None of these features was present in the current case.

Prognosis in extraocular sebaceous carcinoma is generally more favorable than in periocular disease, with lower rates of metastasis and disease-specific mortality when complete excision is achieved [3,11]. Long-term follow-up remains essential, as local recurrence can occur years after initial treatment. Regular clinical surveillance of the surgical site and regional lymph nodes is therefore recommended.

This case adds to the limited literature on sebaceous carcinoma of the extremities and highlights several important clinical lessons: the need for heightened suspicion in chronic non-healing lesions, the importance of expert histopathological evaluation, and the central role of complete surgical excision in achieving durable disease control. Reporting such rare presentations contributes to improved awareness and may facilitate earlier diagnosis and optimal management in future cases.

3. CONCLUSION

Sebaceous carcinoma is a rare malignancy that may present in unusual extraocular locations and mimic benign or less aggressive lesions, leading to delayed diagnosis. This case highlights the importance of maintaining a high index of suspicion for sebaceous carcinoma in chronic non-healing lesions of the extremities. Complete surgical excision with clear margins remains the cornerstone of treatment and can result in excellent outcomes, even when secondary intention healing is employed.

4. PATIENT CONSENT

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. The patient was informed about the purpose of the publication, the nature of the information to be shared, and the measures taken to protect her identity and confidentiality. She was assured that no identifying personal information would be disclosed and that participation was voluntary.

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