

Rapidly growing giant pilomatrixoma in 57-year-old Hispanic male

Aman Sahrawat, Touraj Kormi, Abha Soni

ABSTRACT

Introduction: Pilomatrixomas are benign adnexal tumors arising from hair follicle matrical and cortical cells that predominantly affect the head, neck, and upper extremities in younger adults. While typically benign, challenges in treatment and management arise with atypical features such as rapid growth, size (>5 cm), and gross complexity of the tumor. Distinguishing between benign, atypical, and malignant pilomatrixomas can be challenging both clinically and pathologically, but it is crucial for ensuring appropriate surgical management.

Case Report: We present the case of a 57-year-old male with a malignant pilomatrixoma located on his forearm, clinically resembling an aggressive neoplasm, such as a dermatofibrosarcoma sarcoma, due to its large size, rapid growth, and necrotic surface. Histology revealed an adnexal neoplasm, typically benign, but with atypical features such as expansile basaloid cell proliferation and an elevated Ki-67 index, rarely reported in the literature. A thorough histopathologic evaluation confirmed the diagnosis of atypical giant pilomatrixoma.

Conclusion: This case highlights the importance of both clinical and histopathologic evaluation in accurately diagnosing and treating neoplastic processes. While the clinical presentation suggested an aggressive neoplasm, prompting rapid surgical removal, the initial histologic evaluation resembled a benign pilomatrixoma. Careful integration of clinical and pathological findings led to the diagnosis of an atypical pilomatrixoma. This case also

provides insights into the surgical management of other similar lesions in future clinical scenarios.

Keywords: Adnexal tumor, Atypical pilomatrixoma, Calcifying epithelioma of Malherbe, Ghost cells, Hair follicle, Pilomatricoma, Pilomatrixoma

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INTRODUCTION

A pilomatrixoma is a benign adnexal tumor that arises from the matrical and cortical cells of the hair follicle. This neoplasm is primarily localized to the head and neck and upper extremities and is most prevalent in younger adults (1st and 2nd decades of life). It presents as a slow growing (0.5–3 cm) nodule that takes root in the dermal or subcutaneous layer of the skin [1].

Although, a pilomatrixoma is a generally well-recognized benign condition, with a favorable outcome, there are certain atypical clinical and diagnostic criteria that make its identification and treatment challenging. One such atypical clinical finding is the size of the neoplasm; a clinical size larger than 5 cm is classified as a giant pilomatrixoma [2–4]. Another atypical feature is the irregularity and gross complexity of the tumor itself (i.e., multi-nodularity or necrosis) [5].

A malignant pilomatrixoma is a rare, locally aggressive, low-grade malignant form of a benign pilomatrixoma with propensity of distant metastasis. It has been observed to spread to the lungs, bones, and lymph nodes [3]. This condition typically affects individuals in their sixth and seventh decades of life, with a male predominance and is primarily found in the head,

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neck, and back. These tumors can vary in size, ranging from 1 to 10 cm and also occur in the dermis and subcutis. Clinically, it can be challenging to distinguish between a benign and malignant pilomatrixoma. Therefore, the diagnosis requires a complete and thorough histologic evaluation [6].

The histological assessment should encompass factors such as the depth of invasion, extent of basaloid proliferation, presence of necrosis, atypical mitotic figures, and signs of perineural or vascular invasion. Poorly differentiated tumors with extensive invasion into nearby soft tissues are related to poorer prognosis. The diagnosis of malignant pilomatrixomas can be misdiagnosed if the tumor is partially sampled due to some overlapping histologic features of benign pilomatrixomas [7].

In this case report, we discuss the unique clinical presentation, surgical management, and outcome of a 57-year-old male with an atypical giant pilomatrixoma on his forearm.

CASE REPORT

A 57-year-old Hispanic male presented to a primary care physician with a skin lesion on his right forearm. The patient had noticed the growth on the outer aspect of his forearm for several years but had not sought medical attention. However, over the past 2–3 months, the lesion rapidly enlarged without any history of trauma to the area, causing tenderness, localized swelling, and intermittent bleeding. The patient also reported a loss of sensation around the lesion, due to its growing size. After being evaluated by multiple dermatologists without a clear diagnosis, he was referred to by our clinical team with a suspected diagnosis of a potential malignant neoplasm, such as a dermatofibrosarcoma protuberans (DFSP).

On the initial examination, the patient had a large protruding mass on the posterior right forearm that measured 5 by 6 cm in maximum dimension (Figure 1). The central aspect of the tumor appeared to be necrotic with a rough surface and multiple indentations. There was no exudate, and it did not appear to be infected. After obtaining standard preoperative lab values and workup, the patient was scheduled for primary excision with a wide margin to the depth of the forearm fascia.

Microscopic evaluation revealed a neoplasm situated primarily in the reticular dermis (Figure 2) with extension into the subcutis. The tumor displayed an expansile proliferation of basaloid cells containing vesicular nuclei and eosinophilic cells, known as “shadow cells.” Shadow cells are degenerated keratinized cells which often appear in pilomatrixomas and contribute to the tumor’s distinctive histological appearance (Figure 2). These cells are thought to result from the differentiation of basal cells into keratinized material, reflecting the tumor’s adnexal etiology. In addition to the basaloid and shadow cells, calcifications were also present within the lesion, also a common histologic feature of benign pilomatrixomas.



Figure 1: Clinical examination revealed a large protruding mass on the posterior right forearm that measured 5 by 6 cm in maximum dimension. The central aspect of the tumor appeared to be necrotic with a rough surface and multiple indentations.

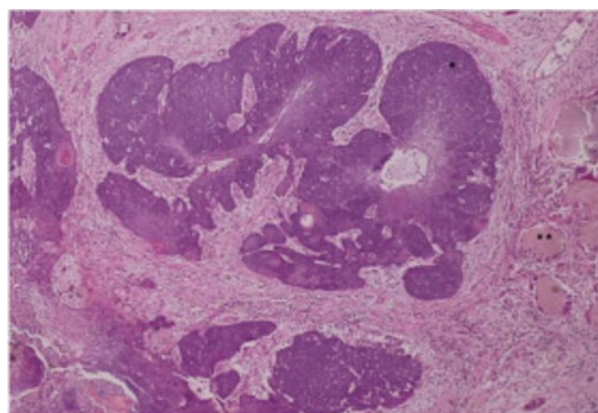


Figure 2: Microscopic evaluation (H&E with 10× magnification) revealed an expansile neoplasm characterized by a proliferation of basaloid cells (*) containing vesicular nuclei and eosinophilic shadow cells (**).

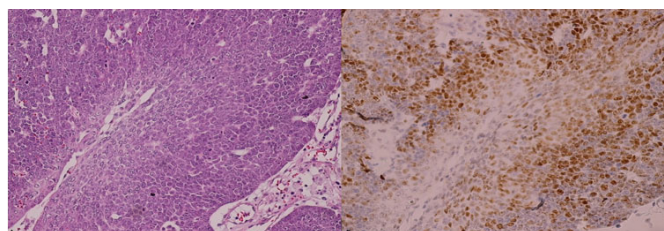


Figure 3: H&E with 20× magnification (A). Proliferative index was assessed using Ki-67 (B), which was only moderately increased (30–45%).



Figure 4: The neoplasm (A) was entirely excised. The patient received a full thickness skin graft from the groin (B) to cover the excisional defect. Post-surgery, the graft (C) and groin (D) healed well.

The tumor did not exhibit definitive malignant features such as an infiltrative growth pattern or increased mitotic activity and the lack of these histologic features initially suggested a benign lesion. However, on histology, the basaloid component of the tumor was overgrown and this in conjunction with the general size/expansile nature of the tumor, warranted additional workup. Immunohistochemical analysis was performed using p53 and Ki-67 (Figure 3). Ki-67, a marker of cell proliferation, showed a moderately increased proliferative index, while p53 did not demonstrate aberrant expression. Although a moderately increased Ki-67 index is an unusual finding, the absence of abnormal p53 expression makes a malignant tumor unlikely, supporting the diagnosis of an atypical pilomatrixoma.

Given this diagnosis, there was a risk of local recurrence and potential malignant transformation if the tumor was incompletely excised. The size of the pilomatrixoma presented initial challenges in wound closure. Due to the larger surface area of the lesion, primary closure (such as suturing) was not sufficient and posed a higher risk of infection. Therefore, a full-thickness skin graft from the patient's groin was used to cover the excisional defect, ensuring full coverage of the wound and adequate protection. The graft also helped restore a more natural skin appearance and minimized scar tissue formation. Post-surgery, the graft healed with nearly 100% take, and the patient was closely monitored in the office with follow-up visits every 4–6 weeks for up to one year. Complete healing was achieved without complications (Figure 4).

DISCUSSION

The rarity of malignant pilomatrixomas underscores the complexity of their clinical recognition and surgical

management. Our case report highlights the significance of atypical features, such as large size, rapid growth, and an expansile histologic pattern with a moderately increased proliferative index, which deviate from the typical characteristics of benign pilomatrixomas and suggest the presence of a potentially locally aggressive subtype.

Upon initial clinical review, given the tumor's size and appearance, DFSP was considered in the differential diagnosis. DFSP, a low-grade malignancy, is known for its slow, infiltrative growth and its characteristic storiform pattern of spindle cells on histology [8]. However, DFSP typically exhibits a more infiltrative nature, which was not seen in our case. The presence of eosinophilic shadow cells, basaloid cell proliferation, and calcifications, along with the non-infiltrative nature of the tumor, pointed strongly toward pilomatrixoma, leading us to favor this diagnosis over DFSP.

Long-term management of DFSP typically involves more aggressive and ongoing surveillance due to its higher recurrence rate and potential for transformation into a more aggressive fibrosarcomatous form [8], whereas atypical pilomatrixomas generally requires less intensive follow-up, as it is a rarer and more indolent tumor with a lower risk of recurrence and metastasis after complete excision [9].

This case also challenges the traditional understanding that pilomatrixomas are typically painless and slow-growing, as our patient presented with a rapidly growing, painful mass. This highlights the need for heightened clinical suspicion when encountering atypical presentations of pilomatrixomas [5]. Recognizing subtle variations in clinical presentation plays a critical role in accurately identifying and diagnosing different types of pilomatrixomas. Although no specific marker exists for diagnosing atypical pilomatrixomas, immunohistochemical markers such as Ki-67 and p53 can be useful for detecting rapidly proliferating cells and possible mutations in tumor cells. A high proliferative index indicated by Ki-67 [10], along with strong expression of the tumor suppressor gene p53, can aid in differentiating malignant pilomatrixomas from benign ones.

The management of malignant pilomatrixomas is further complicated by the lack of established treatment guidelines for the rare malignant variant. While benign pilomatrixomas typically warrant conservative management—ranging from observation to surgical excision—the limited number of documented cases makes it difficult to establish definitive management protocols [10]. The absence of clear treatment guidelines for atypical or malignant pilomatrixomas in the existing literature highlights the need for further research and reporting in this area.

Thus far, our patient has shown no signs of local recurrence or distant complications. This case highlights the importance of careful clinical and histological evaluation in the management of atypical

pilomatrixomas. It also underscores the need for ongoing research and development of more defined treatment protocols, especially for rare and atypical variants of this neoplasm. Regular follow-up (at one year intervals) and tailored management are crucial in ensuring favorable long-term outcomes and advancing our understanding of these uncommon tumors.

CONCLUSION

In conclusion, the unique challenges associated with malignant pilomatrixoma necessitate heightened clinicopathologic awareness. Early recognition and precise diagnosis are paramount to guide appropriate management strategies and mitigate the risk of recurrence or malignant transformation.

The present case contributes to the evolving understanding and diagnosis of this rare low-grade malignancy, emphasizing the need for further similar case presentations to elucidate optimal diagnostic and surgical management approaches.

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Author Contributions

Aman Sahrawat – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Touraj Kormi – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Abha Soni – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.

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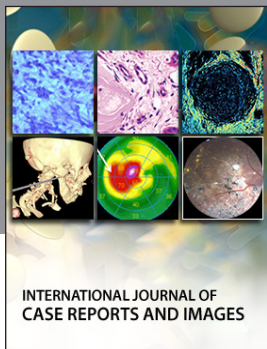
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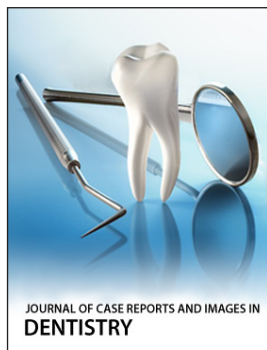
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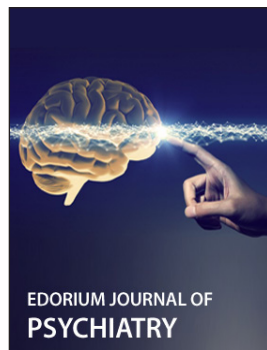
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