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Case Report

A rare adnexal tumor: desmoplastic trichilemmoma

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Abstract

Background: Desmoplastic trichilemmoma (DT) is a rare benign cutaneous adnexal tumor originating from the outer root sheath of hair follicles. Histologically, it is chacularly basal cell carcinoma, making accurate diagnosis is essential to avoid overtreatment. DT most frequently arises in sun-exposed areas of elderly population and is rarely found on the eyelid.
Case Illustration: A 66-year-old male with a 1 cm, skin-colored, dome-shaped nodule on the left upper eyelid, present for four years. Histopathological examination revealed a well-circumscribed dermal tumor composed of clear cells arranged in nests and cords with peripheracterized by a prominent desmoplastic stroma that may resemble invasive malignancies, partiral palisading, embedded in dense, hyalinized stroma. No atypia or mitotic activity was noted. The diagnosis of DT was confirmed.
Discussion: DT is a variant of trichilemmoma with unique histologic features that can resemble infiltrative neoplasms. Eyelid involvement is particularly challenging due to cosmetic and functional considerations. Immunohistochemistry may assist in distinguishing DT from basal cell carcinoma, with CD34 positivity and BerEP4 negativity supporting the diagnosis.
Conclusions: DT should be included in the differential diagnosis of benign-appearing eyelid lesions that demonstrate histologic features suggestive of malignancy. Proper identification is crucial to ensure appropriate surgical management and to prevent unnecessary aggressive treatment.

Keywords: *desmoplastic, eyelid tumor, trichilemmoma*

Abstrak

Latar Belakang: Desmoplastic trichilemmoma (DT) merupakan tumor adneksa kulit jinak yang jarang, berasal dari selubung akar luar folikel rambut. Secara histologis, lesi ini ditandai oleh stroma desmoplastik yang menonjol yang dapat menyerupai keganasan invasif, terutama karsinoma sel basal, sehingga diagnosis yang akurat sangat penting. DT paling sering muncul pada area kulit yang terpapar sinar matahari pada populasi usia lanjut dan jarang ditemukan pada kelopak mata.
Ilustrasi Kasus: Seorang laki-laki berusia 66 tahun dengan nodul sewarna kulit, berbentuk kubah, berukuran 1 cm pada kelopak mata atas kiri, yang telah ada selama empat tahun. Pemeriksaan histopatologi menunjukkan tumor dermal berbatas tegas yang tersusun atas sel-sel jernih yang membentuk sarang dan kord, dengan palisade perifer, tertanam dalam stroma padat yang mengalami hialinisasi. Tidak ditemukan atipia maupun aktivitas mitosis. Diagnosis DT pun ditegakkan.
Diskusi: DT merupakan varian dari trichilemmoma dengan karakteristik histologis khas yang dapat menyerupai neoplasma infiltratif. Keterlibatan kelopak mata menjadi tantangan tersendiri karena pertimbangan kosmetik dan fungsional. Pemeriksaan imunohistokimia dapat membantu membedakannya dari karsinoma sel basal, di mana ekspresi CD34 positif dan BerEP4 negatif mendukung diagnosis DT.
Kesimpulan: DT perlu dipertimbangkan pada lesi kelopak mata yang tampak jinak namun menunjukkan gambaran histologis yang menyerupai keganasan. Identifikasi yang tepat sangat penting untuk memastikan tata laksana bedah yang sesuai serta mencegah tata laksana yang tidak diperlukan.

Kata kunci: *desmoplastik, trichilemmoma, tumor kelopak mata*

Background

Desmoplastic trichilemmoma (DT) is a rare cutaneous adnexal tumor originating from the outer root sheath of the pilosebaceous unit. It was first characterized as a distinct histopathologic entity by Hunt et al. in 1990. DT is considered a histologic variant of trichilemmoma, distinguished by a prominent desmoplastic stroma that can lead to diagnostic confusion with invasive malignancies such as basal cell carcinoma (BCC) or desmoplastic squamous cell carcinoma (SCC). The tumor most frequently arises in sun-exposed areas of elderly individuals, particularly the face, and rarely, the eyelids.[1,2]

Given the overlapping clinical and histopathological features with malignant neoplasms, identifying this rare tumor is essential to avoid misdiagnosis and overtreatment. The rarity of DT on the eyelid poses a diagnostic challenge for clinicians and dermato-logists.[2,3] In this report, we present a case of DT arising on the left upper eyelid in a 66-year-old male, along with a review of its diagnostic features and clinical significance.

Case Illustration

A 66-year-old male presented to the dermato-oncology clinic with a skin-colored, dome-shaped nodule on his left upper eyelid, which had progressively enlarged over the past four years. The lesion was asymptomatic, non-pruritic, and non-tender. Physical examination revealed a 1 cm soft, flesh-colored nodule with a smooth surface and well-defined borders (Figure 1)

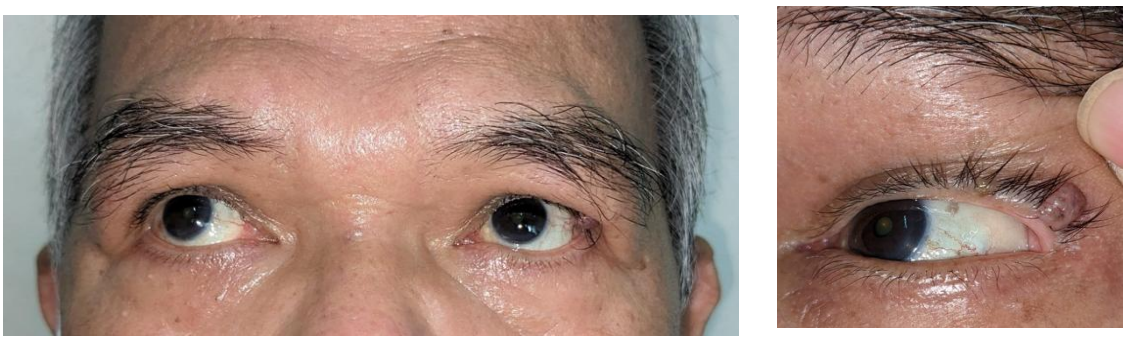

Figure 1. A mass lesion on the left upper eyelid of a 66-year-old man

There were no signs of ulceration, telangiectasia, or pigmentation (Figure 2). The differential diagnosis included basal cell carcinoma, epidermal inclusion cyst, and adnexal tumor.

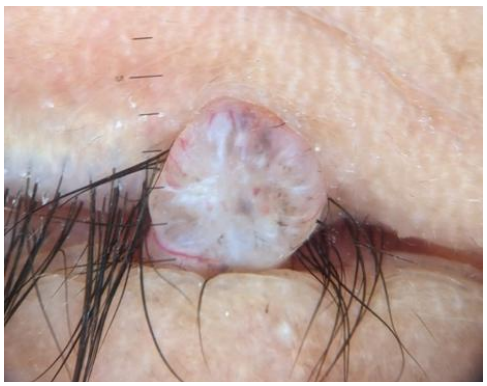

Figure 2. Dermoscopic feature of the mass show blue ovoid dots, multiple linear branching vessels

The patient's comorbidities included type 2 diabetes mellitus, stage 3 chronic kidney disease (CKD), coronary artery disease (CAD) post-percutaneous coronary intervention (PCI), hypertension, and dyslipidemia. Excisional biopsy was performed (Figure 3). Histopathological examination showed a well-circumscribed lobulated tumor in the dermis, partially connected to the epidermis and extending from the outer root sheath of a hair follicle. The tumor was composed of pale-staining cells with clear cytoplasm arranged in nests and cords. Peripheral palisading of nuclei was evident. The surrounding stroma was prominently desmoplastic, hyalinized, and showed dense collagenization. No significant nuclear atypia, necrosis, or abnormal mitotic activity was observed (Figure 4).

Clinicopathological conference was conducted with dermatopathology and pathology department, histopathological reading revealed that given the lack of significant nuclear atypia and the absence of mitotic activity, the differential diagnosis of basal cell carcinoma was conveniently excluded. The histopathology revealed a tumor composed of palisading basaloid cells with a central sclerotic stroma containing interspersed nests of tumor cells, leading to a diagnosis of desmoplastic trichilemmoma.

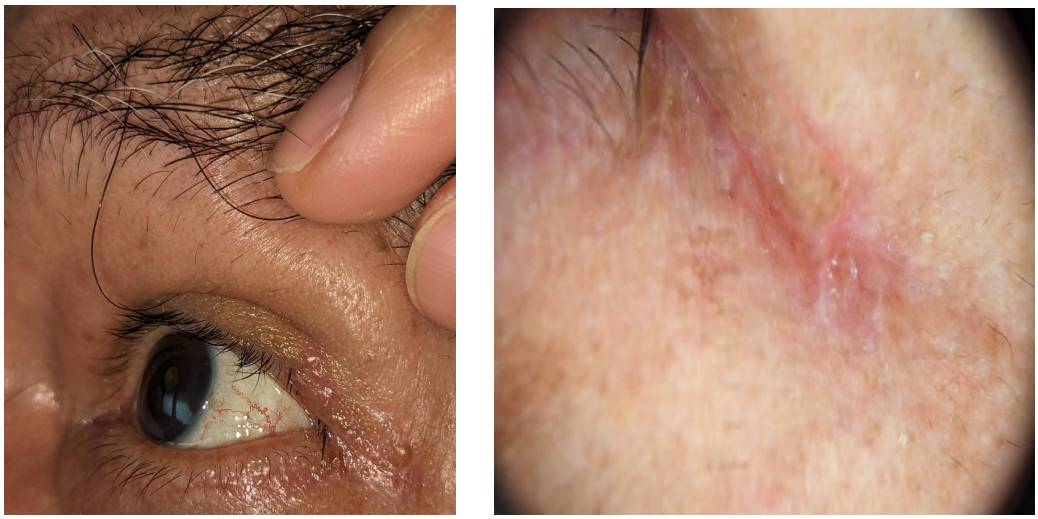

Figure 3. Skin lesion 1 month post excisional biopsy, show linear scar with erythematous background

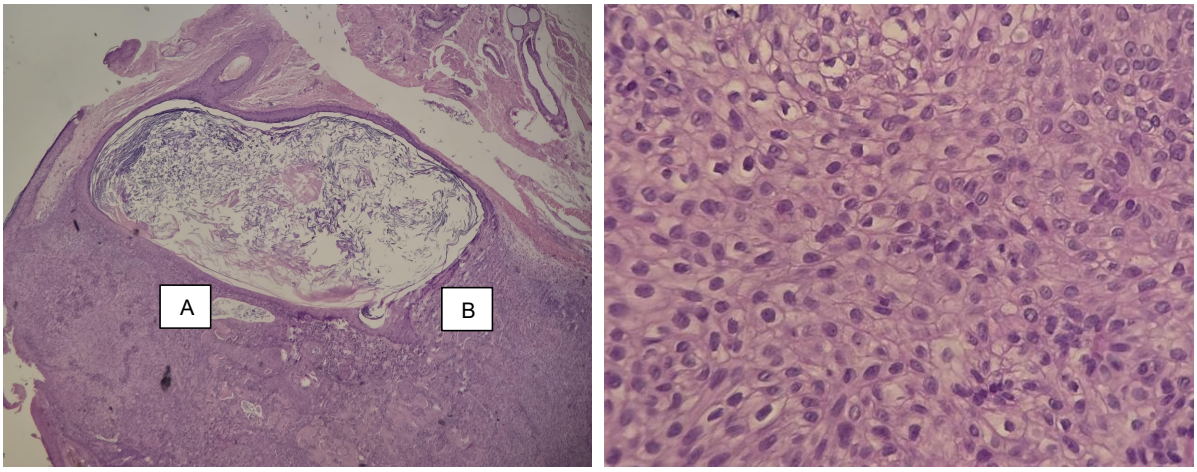

Figure 4. A) Well-circumscribed lobulated tumor in the dermis, partially connected to the epidermis and extending from the outer root sheath of a hair follicle, B) Pale-staining cells with clear cytoplasm arranged in nests and cords

Discussion

Trichilemmoma is a benign adnexal neoplasm that differentiates toward the outer root sheath of the hair follicle. The desmoplastic variant is histologically characterized by lobules or cords of glycogen-rich clear cells embedded within a dense, sclerotic stroma. Unlike classic trichilemmoma, the desmoplastic variant exhibits a fibrous background that may simulate the appearance of infiltrative carcinomas, notably BCC and desmoplastic SCC.[1,4]

The eyelid, abundant in pilosebaceous units, is a recognized but uncommon location for DT. The differential diagnosis is particularly important in this area due to the functional and cosmetic implications of surgical intervention. The desmoplastic stroma and structural irregularities can resemble invasive malignancies, potentially leading to overly aggressive treatment if not accurately diagnosed.[2,3]

Histopathological features of DT over malignancy include symmetry, circumscription, peripheral palisading, and the absence of cytologic atypia or perineural invasion. Immunohistochemical studies may aid in diagnosis; CD34 is often positive in trichilemmoma, especially at the periphery, while BerEP4 is usually negative, distinguishing it from

BCC. PAS-positive, diastase-sensitive staining of the cytoplasm supports the presence of intracellular glycogen, a characteristic of outer root sheath differentiation.[3,4]

Treatment of DT is primarily surgical. Complete excision is both diagnostic and curative. Recurrency is rare if the excision is adequate. No malignant transformation has been reliably documented in DT, although rare cases of trichilemmal carcinoma may share overlapping features and must be considered in ambiguous cases.[5]

This case underlines the importance of considering DT in the differential diagnosis of eyelid tumors, particularly when the lesion exhibits benign clinical behavior but alarming histologic features. Awareness of this entity can prevent misdiagnosis and avoid unnecessary radical procedures.

Conclusion

Desmoplastic trichilemmoma is a rare, benign neoplasm of follicular origin that may clinically and histologically mimic malignant cutaneous tumors. It poses a diagnostic challenge, particularly when located on the eyelid. Accurate histopathological evaluation, supported by immunohistochemistry, when necessary, is essential to establish the correct diagnosis and avoid unnecessary aggressive treatment. Recognition of DT is crucial in dermatologic practice to ensure appropriate patient management and favorable outcomes.

References

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