

Cutaneous Lymphadenoma Presenting as a Solitary Forehead Papule: A Clinicopathological Correlation

Keywords: Cutaneous Lymphadenoma; Trichoblastoma Variant; Adnexal Tumor; Forehead Papule; Basaloid Nests

Abstract

Cutaneous lymphadenoma represents an uncommon follicular adnexal neoplasm characterized histologically by epithelial lobules associated with dense lymphocytic infiltrates. We describe a young woman with a solitary forehead papule whose biopsy demonstrated basaloid epithelial islands admixed with prominent intratumoral lymphocytes, leading to the diagnosis. Complete excision resulted in an excellent outcome with no recurrence. Awareness of this rare entity is essential to differentiate it from basal cell carcinoma and other adnexal tumors.

Introduction

Cutaneous lymphadenoma, currently regarded within the spectrum of trichoblastoma, is an exceptionally uncommon adnexal neoplasm. It usually affects the head and neck region and may clinically mimic basal cell carcinoma because of its pearly papular appearance. Histologically, the lesion is composed of epithelial nests embedded within a lymphocyte-rich stroma.

Cutaneous lymphadenoma- or Adamantinoid Trichoblastoma was first described by Santa Cruz and Barr in 1987 as "lymphoepithelial tumor of skin" which was later renamed as cutaneous lymphadenoma in 1991. The lesions are seen mainly in the head and neck and present as nonspecific papules and nodules. usually occurs in young adults. Both sexes are equally affected. It is a rare benign adnexal tumor with follicular, sebaceous or ductal differentiation therefore regarded as a variant of Trichoblastoma, a neoplasm with folliculosebaceous and apocrine germ differentiation. The tumor consists of nests and lobules of basaloid cells in the reticular dermis with no epidermal connection. The aggregates of cells are embedded in fibrous stroma. Tumor cells are bland and display peripheral palisading. Presence of prominent infiltration by T lymphocytes noted in the centre of tumor lobules and nests. Focal areas of keratinisation are seen in some cases. No cellular atypia seen and mitotic figures are rare. The pattern of cytokeratin 17 expression is patchy and peripheral in cutaneous lymphadenoma as opposed to diffuse expression in Basal cell Merkel cells may be found in cutaneous lymphadenoma, CEA and EMA expression may be seen. Less than 50 cases have been reported in the world literature

Case Report

A 24-year-old lady presented with a firm nodular slowly progressive swelling in the forehead for last 1 year duration.

Punch biopsy specimen of 4 mm was taken from the lesion over the forehead. Section showed unremarkable epidermis. Dermis shows nests of basaloid cells with peripheral palisading of basaloid



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cells. These enclose larger epithelial cells with eosinophilic cytoplasm, vesicular nuclei and prominent Many lymphocytes are admixed with these large epithelial cells . Hair follicular differentiation is noted in one focus. lymphocytic infiltrate is seen in the stroma. No evidence of malignancy seen. Reed Sternberg like cells also seen

Cutaneous lymphadenoma is a rare benign adnexal neoplasm that poses a significant diagnostic challenge because of its close resemblance to basal cell carcinoma (BCC) and other follicular tumors. In the present case, the lesion clinically presented as a small, dome-shaped, pink papule over the forehead without ulceration or telangiectasia, leading to consideration of benign adnexal tumors, intradermal nevus, and BCC in the differential diagnosis.

Histopathological examination was decisive in establishing the diagnosis. Microscopy demonstrated sharply demarcated epithelial lobules situated within the dermis. The lobules were composed predominantly of basaloid cells arranged along the periphery, enclosing larger pale-staining epithelial cells amid dense lymphocytic infiltrates.

Trichoblastoma was another important differential diagnosis because of the follicular germinative differentiation seen in both entities. However, trichoblastoma typically lacks the prominent intraepithelial lymphocytic infiltrate characteristic of cutaneous lymphadenoma. Likewise, lymphoepithelioma-like carcinoma of the skin was excluded due to the absence of cellular atypia, invasive growth pattern, and malignant cytological features.

The intimate admixture of reactive lymphocytes within epithelial tumor islands is considered a hallmark of cutaneous lymphadenoma and remains the most useful diagnostic clue distinguishing it from other basaloid adnexal neoplasms.

Diagnosis- Nodular Adamantinoid Trichoblastoma

Cutaneous lymphadenoma (variant of trichoblastoma).

Differential diagnosis

Basal cell carcinoma was considered an important differential



Figure 1: Solitary dome-shaped pink papule over the forehead.

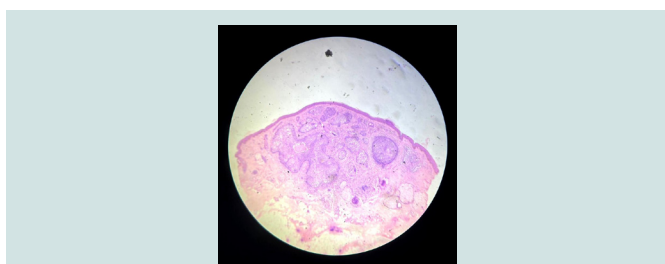


Figure 2: Low-power view showing well-circumscribed dermal tumor nests (H&E).

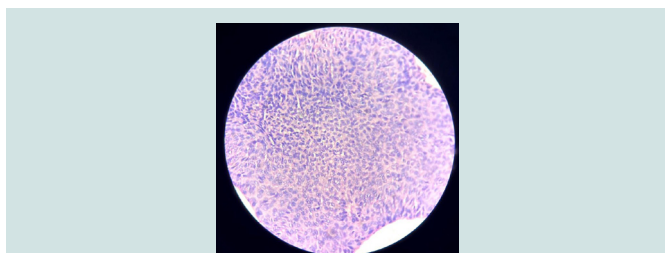


Figure 3: Basaloid nests with peripheral palisading (H&E).

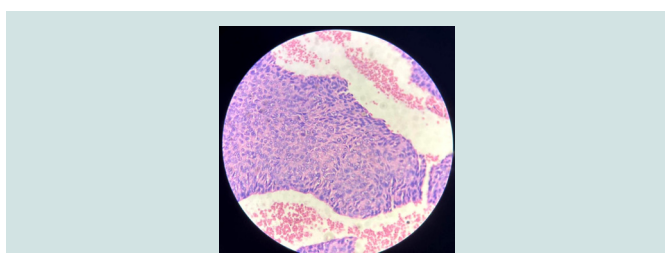


Figure 4: Central pale epithelial cells with vesicular nuclei (H&E).

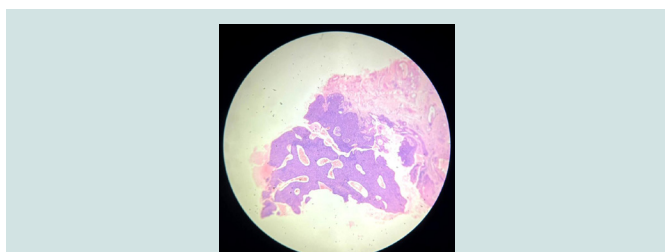


Figure 5: Dense lymphocytic infiltrate within tumor islands (H&E).

diagnosis because both lesions may present as solitary pearly papules over sun-exposed facial skin. However, the absence of stromal retraction clefts, mucinous stromal alteration, significant cytological atypia, and increased mitotic activity argued against basal cell carcinoma in the present case.

Trichoblastoma was another close histological mimic due to the presence of basaloid follicular germinative cells. Nevertheless, conventional trichoblastoma generally lacks the dense intratumoral lymphocytic infiltrate that represents a defining feature of cutaneous lymphadenoma.

Lymphoepithelioma-like carcinoma of the skin was also excluded because the present lesion did not demonstrate infiltrative growth, marked nuclear pleomorphism, or overt malignant cytological features.

Clear cell syringoma may occasionally enter the differential diagnosis because of the presence of epithelial nests; however, ductal eccrine differentiation and characteristic tadpole-shaped structures were absent in our specimen.

Pilomatricoma was ruled out due to the lack of matrical differentiation, ghost cells, and calcification. Similarly, dermatofibroma was excluded because the lesion lacked a fibrohistiocytic proliferation and instead showed distinct epithelial lobules with peripheral basaloid arrangement.

Overall, the combination of epithelial islands, prominent lymphocytic infiltrates, follicular differentiation, and bland cytology strongly favored a diagnosis of cutaneous lymphadenoma.

Basal cell carcinoma – present usually in sun exposed area, usually single, painless pearly or waxy papule or nodule often with central ulceration. (Rodent ulcer)

Presence of telangiectasia, often bleeding or crusting

Presence of retraction artefact, fibromyxoid stroma, peripheral palisading with atypia.

Intradermal naevus- papules or macules tan brown uniformly pigmented and small (less than 0.6 cm), usually adjacent to hair follicle. Often erosion and ulcerations are present.

Presence of naevus cell aggregates in the dermis. Abundant eosinophilic to amphophilic cytoplasm containing coarse melanin granules.

Clear cell syringoma – Multiple small (1-4 mm) firm flesh coloured lesion typically bilaterally symmetrical Common in female(eyelid). Pruritus exacerbates during summer months and menstruation. Presence of tadpole-like structures of eccrine differentiation, absence of intratubular lymphocytic infiltrate.

Lymphoepithelial tumor of skin-This is a malignant tumor that also features basaloid nests with prominent lymphocytic infiltration (lymphotropic). However, LELC shows more cellular atypia and higher mitotic figures compared to the bland cytology of AT.

Trichoblastic carcinoma -Malignant, highly infiltrative counterpart that shows much greater cytologic atypia than the benign AT.

Pilomatricoma -Hair matrix tumor that can also contain basaloid cells, but it usually shows distinct ghost cell keratinization

Dermatofibroma: A superficial dermal tumor that can be excluded by its characteristic fibrotic histology and lack of distinct basaloid nests.

The patient underwent complete surgical excision of the lesion. The postoperative period was uneventful, with satisfactory cosmetic healing. No evidence of local recurrence or development of new lesions was observed during follow up.

Discussion

Adamantinoid Trichoblastoma/ Cutaneous lymphadenoma is a rare benign adnexal tumor commonly presenting as asymptomatic dome shaped, flesh coloured papule or nodule in the head and They are commonly seen in young to middle aged patients with male dominance. Due to the rarity of Adamantinoid Trichoblastoma, it is often misinterpreted as Basal cell carcinoma, adnexal tumor, naevus or dermatofibroma. The present case was clinically thought to be intradermal naevus.[2]

Microscopic examination revealed a triphasic tumor with cell nests with peripheral palisading of basaloid cells (epithelial component), lymphoid infiltration and desmoplastic stroma (mesenchymal components). There can be large Reed Sternberg like cells within the lobules. Focal ductal, follicular and sebaceous differentiation and central keratinisation and stromal mucinosis have been documented [3]. The tumor is well circumscribed and unencapsulated, but presence of infiltrating outlines makes tumor recurrence possible.[2]

The Histological differential diagnosis includes clear cell Basal cell carcinoma, clear cell syringoma, trichoepithelioma and malignant lymphoepithelioma like carcinoma. In younger patients, it has to be differentiated from dermal thymus, which is an Aberrant location of thymic tissue in the skin due to defective migration and present as linear symmetric ulcerated scar Like lesions in the neck in patients with other facial branchial anomalies.[2]

Several proposals regarding the possible histogenesis of the tumour have been reported. Santa cruz Barr told it to be immature pilosebaceous differentiation. [5,6]

Others suggested eccrine or lymphotropic solid syringoma, lymphotropic eccrine benign tumors.[7,8]. Dahil and Seywright reported synchronous occurrence of cutaneous lymphadenoma and eccrine syringoid carcinoma in a single person providing additional evidence in support of eccrine differentiation. Filipo et al [10] argued that cutaneous lymphadenoma/ Adamantinoid Trichoblastoma is a distinct entity that may mimic Basal cell carcinoma with pilar or eccrine differentiation. Mc Niff et al [11] evaluated the immunohistochemical staining pattern of CK 20 Merkel cells, BCL2 epithelial cells, S100 & CD1a (langerhans cell), CD34 (stromal cells) in cutaneous lymphadenoma, Trichoblastoma and nodular Basal cell carcinoma. They observed similar staining pattern of first three markers in cutaneous lymphadenoma and Trichoblastoma (importantly peripheral staining of BCL2 in tumor lobules) and were of the opinion that cutaneous lymphadenoma is a variant of Trichoblastoma. Most recently these tumors have been classified as Adamantinoid Trichoblastoma considering its resemblance to dental

Adamantinomas. (Epithelial and mesenchymal tumor of oral cavity). [2,12,13].

The marked lymphocytic infiltrate in these tumors is thought to be due to defective lymphocytes – epithelial interaction or exuberant host response to tumour cells. [14,15].

A benign clinical course has been reported in the literature. Complete surgical excision is the definitive treatment of choice. To conclude

Adamantinoid Trichoblastoma is a benign adnexal tumor that clinically masquerade as a variety of benign and malignant. Due to the presence of infiltrating lymphocytes within tumor lobules, we feel that “Lymphotropic Adamantinoma” would be the appropriate terminology to describe this rare entity.

Cutaneous lymphadenoma remains an exceptionally uncommon adnexal tumor, with relatively few cases documented in literature. Most reported cases occur in middle-aged or elderly individuals and predominantly involve the head and neck region, especially the cheek, forehead, eyelids, and nose. The present case is noteworthy because the lesion occurred in a young 24-year-old female, which is relatively uncommon for this entity.

The tumor is currently regarded as a variant of trichoblastoma with follicular germinative differentiation. Some authors propose derivation from hair follicle stem cells because of its morphological overlap with follicular adnexal tumors. Despite its benign nature, cutaneous lymphadenoma continues to be clinically important because it is frequently mistaken for BCC, both clinically and histologically.

Recent dermatopathology literature emphasizes the importance of careful recognition of its characteristic lymphocyte-rich epithelial islands and the absence of stromal retraction artifact. Misdiagnosis as BCC may lead to unnecessarily wide excision or aggressive management, particularly when lesions occur on cosmetically sensitive facial sites.

Immunohistochemistry may occasionally assist in diagnostically difficult cases. Cytokeratin positivity confirms epithelial differentiation, while CD3 and CD20 highlight the reactive T- and B-lymphocytic infiltrate. BerEP4 positivity may overlap with BCC and therefore should be interpreted cautiously in conjunction with morphology.

Overall prognosis is excellent following complete surgical excision, and recurrence is extremely uncommon. No definite malignant transformation has been convincingly established in the literature to date.

Skin coloured semicircular nodule with telangiectasia, absence of ulceration or crusting

Although dermoscopy was not formally documented in the present case, previously reported dermoscopic features of cutaneous lymphadenoma include pink-white structureless areas, fine arborizing vessels, and homogeneous erythematous background. These findings may overlap with basal cell carcinoma, further emphasizing the importance of histopathological confirmation.

Cutaneous lymphadenoma is an uncommon benign adnexal neoplasm currently regarded within the spectrum of follicular tumors, particularly trichoblastoma. Owing to its rarity and nonspecific clinical appearance, the lesion is frequently mistaken for more common cutaneous neoplasms, especially basal cell carcinoma. Most reported lesions arise on the head and neck region as slowly enlarging asymptomatic papules or nodules.

The present case is notable because the lesion occurred in a young adult female and clinically simulated a benign papular lesion of the forehead. Histopathological examination was essential for establishing the diagnosis. The biopsy demonstrated sharply circumscribed dermal epithelial lobules composed of peripheral basaloid cells surrounding larger pale epithelial cells within a dense lymphocyte-rich stroma. Importantly, there was no evidence of stromal retraction, necrosis, significant atypia, or brisk mitotic activity.

The characteristic admixture of epithelial nests and abundant reactive lymphocytes remains the most valuable diagnostic clue in distinguishing cutaneous lymphadenoma from other basaloid adnexal tumors. Some lesions may additionally demonstrate follicular or sebaceous differentiation. Occasional Reed–Sternberg-like cells and focal keratinization have also been described in previous reports.

Although the exact histogenesis remains uncertain, most authors support follicular germinative differentiation because of the morphological overlap with trichoblastoma. Immunohistochemical studies described in the literature have demonstrated expression patterns supporting epithelial differentiation along with reactive T- and B-lymphocytic infiltrates. However, routine histopathology is usually sufficient for diagnosis in typical cases.

Recognition of this entity is clinically important because misinterpretation as basal cell carcinoma may lead to unnecessarily aggressive treatment. Complete surgical excision remains curative in the majority of cases, and recurrence is distinctly uncommon.

In conclusion, cutaneous lymphadenoma should be considered in the differential diagnosis of solitary facial papules showing basaloid epithelial nests with dense lymphoid infiltrates. Careful clinicopathological correlation is essential to avoid diagnostic pitfalls and overtreatment.

Conclusion

Awareness of this uncommon follicular adnexal neoplasm is important because its clinicopathological resemblance to basal cell

carcinoma may result in diagnostic confusion and unnecessary aggressive treatment.

Patient Consent

Written informed consent was obtained from the patient for publication of clinical images and data.

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