

Cutaneous lymphadenoma: a rare case and brief review of a diagnostic pitfall

Ryan Yu, Samih Salama, Salem Alowami
Department of Pathology and Molecular Medicine, McMaster University, Hamilton, ON, Canada

Abstract

We report a case of cutaneous lymphadenoma on the posterior left ear of a 67-year-old woman. Although it is benign, recognition of cutaneous lymphadenoma is important as it presents a diagnostic pitfall to the unsuspecting dermatologist and general surgical pathologist, who may readily misdiagnose the lesion because it is not only very rare, but also clinically and histologically resembles the far more common and locally destructive basal cell carcinoma.

Introduction

Cutaneous lymphadenoma is a rare epithelial neoplasm. It was originally described by Santa Cruz and Barr in 1987 as *lymphoepithelial tumor of the skin*.¹ Since then, cutaneous lymphadenoma has been reported in the medical literature under various designations, including epithelio-lympho-histiocytic tumor and lymphotropic adamantinoid trichoblastoma.^{2,3} Currently, cutaneous lymphadenoma is considered a variant of nodular trichoblastoma with adamantinoid features.

Although it exhibits benign biologic behavior, recognition of cutaneous lymphadenoma is important because it presents a diagnostic pitfall to the unsuspecting dermatologist and general surgical pathologist by close clinical and histologic resemblance to basal cell carcinoma.

Case Report

A 67-year-old woman with no significant medical history presented to her primary care physician with a posterior left ear nodule of unknown duration. It was suspicious for a nevus or basal cell carcinoma and subsequently excised. Microscopic examination showed a well circumscribed lesion with irregularly-shaped nests of basaloid cells in a collagenous stroma (Figure 1A). The nests showed absence of peripheral palisading and retraction artifact (Figure 1B). The deep margin was rimmed by

a prominent mature lymphocytic infiltrate (Figure 1C), which was also present throughout the stroma and within the nests (Figure 1D).

Results and Discussion

Cutaneous lymphadenoma is a rare neoplasm with 56 cases reported in the English literature. Patients range in age from 14 to 87 years (median 45 years). Males are affected more often than females (about 1.5:1). Most cases occur on the head, especially the cheek, forehead, eyelid, and temple. It occurs rarely at other sites, such as the extremities.^{4,6} The tumor frequently presents as a solitary, small (from less than 1 cm to 2.5 cm), flesh-colored, non-ulcerated, asymptomatic nodule of many months to years duration. The clinical impression is usually that of a basal cell carcinoma,⁷ appendageal tumor, nevus, or cyst.

Histologically, cutaneous lymphadenoma is characterized by a well-circumscribed, unencapsulated, intradermal nodule of variably-sized, round-to-irregularly shaped epithelial lobules embedded in a fibrous stroma. The lobules are usually separate, but may show interconnection. They may involve the full thickness of the dermis and border, or extend into the subcutaneous fat. There is variable connection to the epidermis. The lobules are rimmed by 1 or more layers of small, bland, flat-to-cuboidal basaloid cells which do not invariably show peripheral palisading. Retraction artifact is not observed. Rudimentary follicular papillae/hair germs (papillary mesenchymal bodies) may be present focally. The lobules are occupied centrally by large cells admixed with a variably dense infiltrate of small mature lymphocytes without plasma cells. The large cells may be dissociated by extracellular mucinous material.⁸ They exhibit vesicular nuclei, prominent nucleoli, and abundant clear-to-faintly eosinophilic cytoplasm. Mitoses are rare. No atypical mitotic figures are seen. Occasionally, the lobules contain large, multinucleated Reed-Sternberg-like cells or lacunar cells.⁴ Some lobules may contain aggregates of compact, eosinophilic keratin or duct-like structures lined by a glassy cuticle.⁹ The stroma surrounding the tumor lobules consists of dense collagen with variable clefts and numerous scattered lymphocytes as seen within the lobules. In some areas, stromal lymphocytes may appear to penetrate the tumor lobules and obscure the stroma-lobule border. Heavier lymphocytic infiltrate may be found at the dermal subcutaneous junction. Occasionally, keratin foreign body granulomas,⁴ or stromal lymphocytes arranged in germinal center-like nodules,¹⁰ may be seen at the periphery of the tumor.

Correspondence: Ryan Yu, Department of Pathology and Molecular Medicine, McMaster University, HSC-2N10, 1280 Main Street West, Hamilton L8S 4K1, ON, Canada.
Tel. +1.289.339.5990.
E-mail: ryan.yu@medportal.ca

Key words: cutaneous lymphadenoma, lymphotropic adamantinoid trichoblastoma.

Contributions: RY, data acquisition, literature review, drafting of manuscript; SS, critical revision of manuscript for intellectual content; SA, concept, critical revision of manuscript for intellectual content.

Conflict of interests: the authors declare no potential conflict of interests.

Received for publication: 18 February 2014.

Accepted for publication: 14 April 2014.

This work is licensed under a Creative Commons Attribution NonCommercial 3.0 License (CC BY-NC 3.0).

©Copyright R. Yu et al., 2014
Licensee PAGEPress, Italy
Rare Tumors 2014; 6:5358
doi:10.4081/rt.2014.5358

Immunohistochemistry shows positive reactivity for cytokeratin AE1/AE3 (intralobular basaloid and clear cells), S100 protein (intralobular dendritic cells), and CD34 (stroma).^{3,4} The lymphocytic infiltrate is polymorphous.⁷ The morphologic features of cutaneous lymphadenoma are distinctive enough to allow accurate diagnosis on routine hematoxylin-eosin stain without use of ancillary immunohistochemistry. The main histologic differential diagnosis includes clear cell variants of basal cell carcinoma (BCC) and syringoma, dermal thymus, and lymphoepithelioma-like carcinoma of the skin (LELC). The clear cell variant of BCC usually shows features of conventional BCC that distinguish it from cutaneous lymphadenoma, including epithelial mitotic activity, apoptotic bodies, and clefting artifact with blue-tinged stromal mucin in the retraction space. Clear cell syringoma shows tadpole-shaped ducts and islands consisting largely of glycogen-rich clear cells embedded in a dense red sclerotic stroma. A prominent lymphocytic infiltrate is not found. Dermal thymus is characterized by aberrantly located thymic tissue in the skin with Hassall's corpuscles. LELC shows morphological similarity to undifferentiated nasopharyngeal carcinoma (lymphoepithelioma) with well-circumscribed lobules or nests of large, cohesive, epithelioid cells closely associated with a dense, mixed T and B lymphocytic infiltrate.

Simple surgical excision is sufficient for

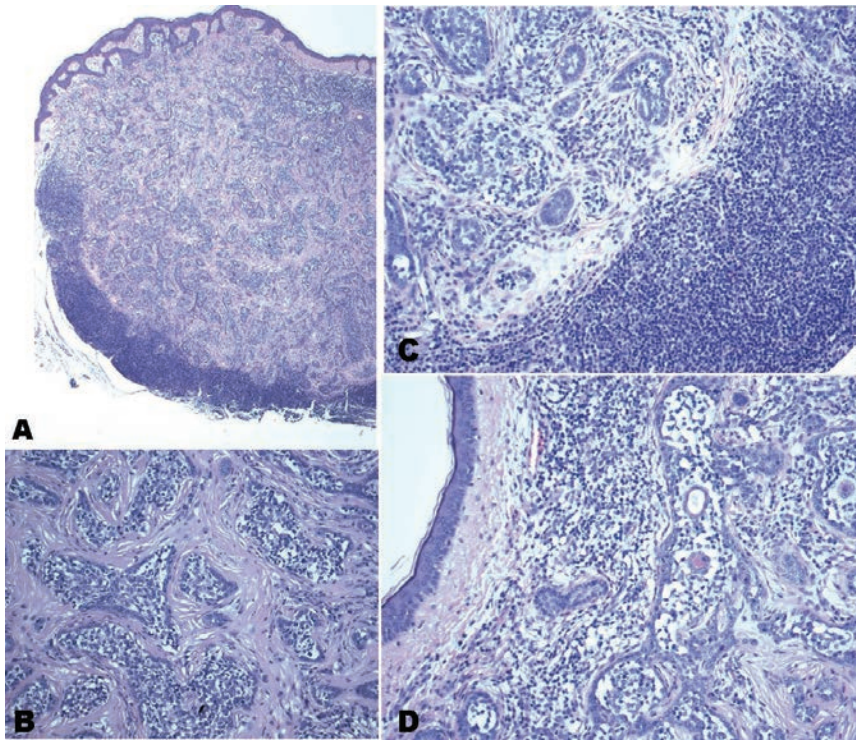


Figure 1. A) A well-circumscribed lesion with basaloid nests in the dermis, rimmed by prominent lymphocytic inflammation at the deep margin (Hematoxylin & Eosin, 20 \times). B) Irregularly-shaped nests of basaloid and clear cells, without peripheral palisading or retraction artifact, in a collagenous stroma (Hematoxylin & Eosin, 100 \times). C) A thick rim of mature lymphocytes is present at the deep margin (Hematoxylin & Eosin, 100 \times). D) Mature lymphocytes are seen in the nests and stroma (Hematoxylin & Eosin, 100 \times).

treatment of cutaneous lymphadenoma. Successful management by Mohs micrographic surgery (MMS) has been reported in several cases.¹¹ According to Lo Piccolo *et al.*,¹² MMS may allow for more definitive tumor removal for large cases of cutaneous lymphadenoma and greater tissue preservation at sites that are functionally and aesthetically-sensitive (*i.e.*, the face). Simple surgical re-excision may be sufficient and preferred in the case of microscopically positive surgical margins, given the resource-intensive and time-consuming nature of MMS. No case of recurrence or metastasis following complete excision of cutaneous lymphadenoma has been reported.

Conclusions

Cutaneous lymphadenoma is highly susceptible to misdiagnosis because it is very rare and closely resembles basal cell carcinoma, which is a very common epithelial neoplasm. Close attention to morphologic features on routine hematoxylin-eosin stain allows accurate diagnosis without use of ancillary studies.

References

1. Santa Cruz DJ, Barr RJ. Lymphoepithelial tumor of the skin. *J Cutan Pathol* 1987; 14:35.
2. Filosa G, Bugatti L, Ciattaglia G. Epitheliolympho-histiocytic tumor: a controversial entity. *Eur J Dermatol* 1994;4:376-8.
3. Murphy M, Brierley T, Pennoyer J, et al. Lymphotropic adamantinoid trichoblastoma. *Pediatr Dermatol* 2007;24:157-61.
4. Santa Cruz DJ, Barr RJ, Headington JT. Cutaneous lymphadenoma. *Am J Surg Pathol* 1991;15:101-10.
5. Diaz-Cascajo C, Borghi S, Rey-Lopez A, Carretero-Hernandez G. Cutaneous lymphadenoma. A peculiar variant of nodular trichoblastoma. *Am J Dermatopathol* 1996; 18:186-91.
6. Inalöz HS, Chowdhury MM, Knight AG. Cutaneous lymphadenoma. *J Eur Acad Dermatol Venereol* 2001;15:481-3.
7. Alsadhan A, Taher M, Shokravi M. Cutaneous lymphadenoma. *J Am Acad Dermatol* 2003;49:1115-6.
8. Wechsler J, Fromont G, André JM, Zafrani ES. Cutaneous lymphadenoma with focal mucinosis. *J Cutan Pathol* 1992;19:142-4.
9. Pardal-de-Oliveira F, Sanches A. Cutaneous lymphadenoma. *Histopathology* 1994;25:384-7.
10. Aloï F, Tomasini C, Pippione M. Cutaneous lymphadenoma: a basal cell carcinoma with unusual inflammatory reaction pattern? *Am J Dermatopathol* 1993;15:353-7.
11. Hanlon A, Clayton A, Stasko T. Mohs micrographic surgery for cutaneous lymphadenoma. *Dermatol Surg* 2012;38:1552-4.
12. LoPiccolo MC, Chaffins ML, Kouba DJ. Mohs micrographic surgery for the treatment of cutaneous lymphadenoma. *J Drugs Dermatol* 2011;10:1324-6.