CASE ARTICLE

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Eccrine spiradenoma presenting as leg swelling: A case report of rare presentation

G. Singh[1], N. Nargotra[2], S. Singh[2]*, P. Sahni[3]

[1]Consultant Pathologist, Saral Diagnostics, H.No-2 and 3, Shakti Vihar, Pitampura, New Delhi, India, [2]Department of Pathology, NDMC Medical College and Hindu Rao Hospital, Malka Ganj, New Delhi, India, [3]Senior Consultant Pathologist, Saral Diagnostics, H.No-2 and 3, Shakti Vihar, Pitampura, New Delhi, India

ABSTRACT

Eccrine spiradenoma (ES) is an uncommon benign adnexal tumor of the skin. The highest rate of incidence is observed among young adults, without any sex predilection. The most commont site of presentation is the head and neck and ventral aspect of the trunk. We report a case of 80-year-old male who presented with leg swelling for the past 25 years. Cytosmear showed tight clusters of multilayered, uniform, cuboidal cells arranged at places around pink hyaline material. Cells were uniform with scant cytoplasm, round-to-ovoid nuclei, and inconspicuous nucleoli. A differential diagnosis of skin adnexal tumor and adenoid cystic carcinoma was considered. The histologic findings confirmed ES. Cytological features of ES are sparsely reported in the literature. Although histopathological examination is considered to be the gold standard with characteristic diagnostic features, cytology can also play an important role in the management of such rare adnexal tumors.

Key words: Adnexal tumors, eccrine, spiradenoma

INTRODUCTION

Eccrine spiradenoma (ES) is a rare benign adnexal tumor which arises from duct of eccrine sweat gland.[1] Most commonly involved patient age group is the second to fourth decade. Most common site of presentation is the ventral aspect of the body, proximal extremities, and occasionally on the ear, eyelid, lip, and hand.[2]

ES can present either as single, tender, well-circumscribed intradermal nodule, or as multiple lesions. Solitary ES comprises more than 97% of the cases and has similar incidence in men and women, whereas multiple ES is more common in females (M:F ratio of 1:3).[1,3] Rare malignant transformation has been reported in the literature, mostly after 20-30 years of chronicity.[3]

Histopathological diagnostic criteria of various adnexal tumors are well documented in the literature, whereas cytological criteria are sparsely reported. Furthermore, exact cytological diagnosis of adnexal tumors is very difficult due to multiple lines of differentiation of various adnexal tumors. We hereby report a case with cytohistological correlation of ES in an elderly male patient.

CASE REPORT

An 80-year-old male presented to cytology clinic with a mildly tender slow-growing swelling for the past 25 years. On physical examination, the swelling was firm, mobile, slightly tender, measuring 3 × 2 cm in diameter.

Fine-needle aspiration yielded blood mixed aspirate. Cytosmears were moderately cellular and showed uniformly sized cuboid cells with scanty cytoplasm, round-to-oval nuclei, and inconspicuous nucleoli. At places, these cells were arranged around hyaline material. Sheets of these epithelial cells also showed interspersed spindle-shaped myoepithelial cells with regular nuclear contour and scanty cytoplasm. Background shows few scattered lymphocytes. [Figure 1a-c] On the basis of above cytological features, two differential diagnosis, skin adnexal tumor and adenoid cystic carcinoma, were considered.

Excision was advised for exact categorization. Resected gross specimen showed a skin covered tissue measuring 4 × 4 × 2 cm. Cut surface showed multiple grayish-white nodules ranging in size from 0.5 to 1.5 cm in diameter, in the dermal region with no connection to overlying epidermis. Histopathological sections showed multiple basophilic lobules separated by hyalinized stroma in the dermis. Lobules showed interwining cords around edematous connective tissue, trabecular arrangement, and hyaline globules of varying sizes. These cords were lined by small cells having dark nuclei, and in the center, there were few large cells with scant cytoplasm and pale nuclei [Figure 1d]. No mitosis or areas of cystic changes was seen. Final diagnosis of ES was given. The patient is on follow-up and has not reported any recurrence.

DISCUSSION

Eccrine sweat glands are the skin appendages and are composed of three segments - acrosyringium (intraepidermal duct), intradermal duct, and secretory coil lying in reticular dermis.[4] ES are uncommon benign adnexal tumors which rarely involve

Address for correspondence:
N. Nargotra, NDMC Medical College and Hindu Rao Hospital, Malka Ganj, New Delhi, India. E-mail: drgarimasingh2011@gmail.com

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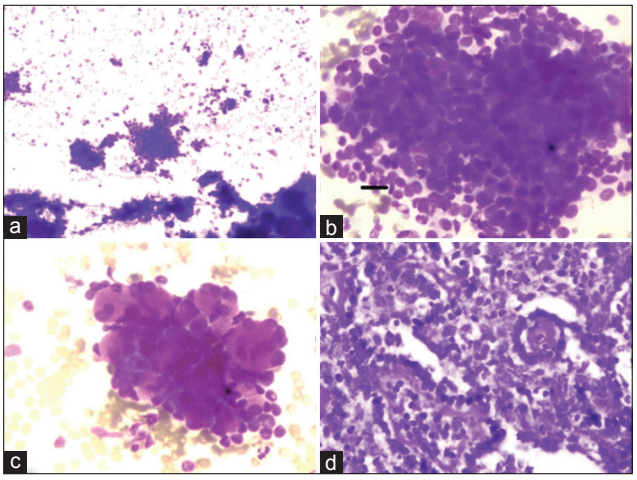

Figure 1: (a) Cytosmear showing cohesive multilayered clusters (Giemsa, X100), (b) cytosmear showing round-to-oval pale epithelial cells, spindle-to-oval myoepithelial cells having darker nuclei, and scanty cytoplasm (black arrow) (Giemsa, X400), (c) cytosmear showing tumor cells arranged around hyaline globules (Giemsa, X400), (d) microphotograph showing two cell population, small basaloid cells at the periphery, and large pale cells at the center (H and E, ×400)

extremities with only one case of knee swelling and only few cases of thigh swelling reported in literature till date.[5] Involvement of distal extremities is very rare with no case published in the English literature, to the best of our knowledge. ES may present as multiple nodules either in a zosteriform pattern or in a linear arrangement.[5] Due to very few cases reported in the literature, exact cytological diagnostic criteria are still lacking.

Cytological smears of ES show tight clusters of multilayered uniform cuboidal bland epithelial cells, which at places surround the hyalinized material. Other key components are spindle-shaped myoepithelial cells, scattered bare nuclei, and small lymphocytes along with tubules and pseudorosette formation around hyaline material.[2,6]

Most closest cytological differential diagnosis of ES is adenoid cystic carcinoma, glomus tumor, and other adnexal tumors such as cylindroma, hidradenoma, chondroid syringoma, and malignant counterpart of ES.[7] On cytology, adenoid cystic carcinoma can also have cell balls, clusters, and rosette-like arrangement around the stromal matrix cores as seen in ES, but single type of cell population as compared to three types of cell population in ES is the key to differentiation.[8]

In the literature, glomus tumor has been cytologically misinterpreted as ES.[2] Cytomorphology of glomus tumor comprises of cohesive clusters of uniform round cells with scant cytoplasm along with basement membrane-like material. Arrangement of glomus cells around blood vessel, lack of three types of cells, and absence of acini formation are distinguishing points in favor of glomus tumor.[7] Hidradenoma cytologically shows cohesive clusters of polygonal/eosinophilic cells along with large clear cells. Nuclei have smooth nuclear contour and distinct nucleoli. Cylindroma shows palisaded arrangement of small basaloid cells, few light staining cells, and hyaline globules on cytological smears. Chondroid syringoma is synonymous to pleomorphic adenoma of salivary gland with epithelial and myoepithelial cells in a chondromyxoid stroma.[6]

On histopathological examination, ES shows single to multiple sharply demarcated lobules in the dermis without any epidermal connection. These lobules appear to be deeply basophilic due to close aggregation of the nuclei. The tumor lobules are comprised of two types of cells arranged in interwining cords around islands of edematous connective tissue. Both types of cells have scant cytoplasm. The smaller one with dark nuclei is located at the periphery of cellular aggregates, and the second type with large pale nuclei is present toward center. Rosettes may be formed in the absence of luminae. In between the tumor cells, hyaline droplets may be present.[4]

Malignant transformation is rare and takes place in longstanding lesions. Surgical excision is the mainstay of treatment and as recurrence is reported to close follow-up of the patient is required.

CONCLUSION

ES is a rare benign adnexal tumor which can very rarely present as a leg swelling. Histopathological examination is considered to be the gold standard for diagnosis; however, with characteristic diagnostic features, cytology can also play an important role in the management of such rare adnexal tumors.

REFERENCES

1. Yoshida A, Takahashi K, Maeda F, Akasaka T. Multiple vascular eccrine spiradenomas: A case report and published work review of multiple eccrine spiradenomas. J Dermatol 2010;37:990-4.
2. Vidyavathi K, Udayakumar M, Prasad CB, Harendra KM. Glomus tumor mimicking eccrine spiradenoma on fine needle aspiration. J Cytol 2009;26:46-8.
3. Englander L, Emer JJ, McClain D, Amin B, Turner RB. A rare case of multiple segmental eccrine spiradenomas. J Clin Aesthet Dermatol 2011;4:38-44.
4. Murphy GF. Tumors of the epidermal appendages. In: Elder D, Elenitsas R, Johnson B, editors. Lever Histopathology of Skin. 9th ed. Philadelphia, PA: Lippincott-Raven; 1997. p. 867-926.
5. Sharma A, Sengupta P, Das AK, Nigam MK, Chattopadhya S. Eccrine spiradenoma in knee. Indian J Dermatol 2014;59:513-5.
6. Devanand B, Vadiraj P. Fine needle aspiration cytology of eccrine skin adnexal tumours. J Cytol Histol 2011;2:129.
7. Sinha A, Pal S, Phukan JP. Fine needle aspiration cytology of eccrine spiradenoma of back: Report of a rare case. J Lab Physicians 2014;6:130-2.
8. Kolda TF, Ardaman TD, Schwartz MR. Eccrine spiradenoma mimicking adenoid cystic carcinoma on fine needle aspiration. A case report. Acta Cytol 1997;41:852-8.

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