Eccrine spiradenoma on the anterior forearm: a case report

Zeina M. Habib¹, Moatasem Hussein Al-janabi¹, Azhar Sayed Ahmad ¹, Roula Baddour¹, and Rana Issa²

September 9, 2023

1.Introduction

Eccrine spiradenoma is a rare adnexal neoplasm originating from sweat glands. It typically presents as a small solitary nodule covered by a normal or bluish skin [1]. It can appear in nearly any part of the body, with 1/5 of cases occurring in extremities [2]. The tumor is most frequent in young adults, with no sexual or racial predilection [3,4]. Although it is considered benign, malignant cases have been reported [5]. Early diagnosis is essential to prevent chances of recurrence and malignant transformation [6].

2.Case report

A 12- year old female presented with a painful swelling in the anterior aspect of the left forearm. The lesion had first been noticed from one year and increased in size gradually over this period. On examination, there was a single movable intradermal nodule measuring about 1 cm, covered by a purple-coloured skin. There was no skin ulceration nor satellite nodules [Figure 1].

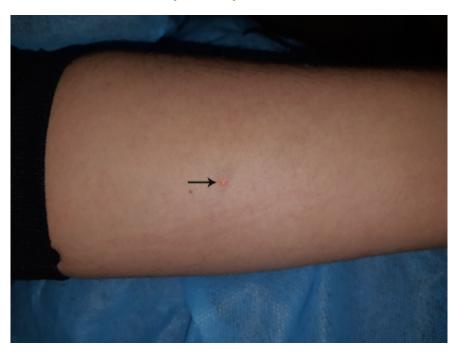


Figure 1: clinical image purple-coloured swelling in the anterior aspect of the left forearm, measuring

¹Tishreen University Hospital

²Tishreen University Faculty of Medicine

around 1 cm.

Complete excision was performed and the specimen was sent to the pathology department. Histopathological examination of the specimen revealed a circumscribed multinodular intradermal tumor [Figure 2A] [Figure 2B] . The tumor nodules separated by edematous and hyalinized stroma and consisted of small dark basaloid cells at the periphery with centrally located large pale cells and intratumoral lymphocytes [Figure 2C] [Figure 2D] . In addition, small ductal structures were identified [Figure 2D] . There was no connection to the overlying epidermis. Based on histopathological features, a diagnosis of eccrine spiradenoma was made.

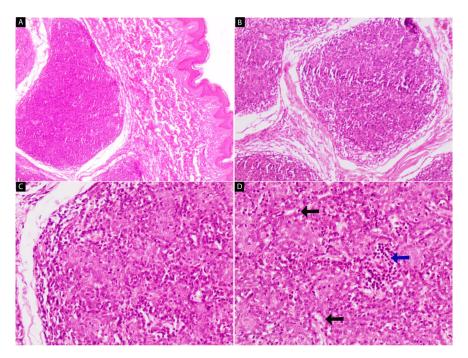


Figure 2: Hematoxylin and eosin-stain (A-D) Microscopic images of the biopsy. (A) The low-power view shows a circumscribed multinodular proliferation within the dermis (40x). (B) The stroma between the nests is edematous and hyalinized (100x). (C) Large pale cells are centrally located and small dark basaloid cells are at the periphery (200x). (D) Ductal structures (black arrows) and intratumoral lymphocytes (blue arrow) were identified (200x).

The patient underwent routine follow-up. After 18 months, she presented with a subcutaneous painful nodule at the same location of the previous tumor. The nodule was completely excised and histopathological examination showed a recurrent eccrine spiradenoma.

3.Discussion

Eccrine spiradenoma is a rare benign tumor deriving from eccrine sweat glands [5]. The precise etiology is still unclear, some evidences indicate the development of a follicle sebaceous-apocrine lineage rather than eccrine differentiation [2,7]. Little is known about the genetic alteration in this tumor, a mutation in the tumor suppressor CYLD gene on chromosome 16 is found in Brooke-Spiegler syndrome, which features multiple spiradenomas, cylindromas, and trichoepitheliomas [1]. Eccrine spiradenoma characteristically presents as a dermal or subcutaneous nodular lesion, with a gray, blue or purple hue. The lesions are usually small, though they may reach a diameter of several centimeters, and are occasionally associated with paroxysmal pain and tenderness [1,3]. The tumor is mainly located in the head, neck and ventral aspect of the trunk, and less often in the extremities. It is most commonly seen in patients between 15 and 35,

though any age group can be affected [3,5]. Various clinical forms have been reported such as multiple linear, zosteriform, blaschkoid, and nevoid forms [1]. Malignant transformation is extremely rare, and it can occur especially in patients older than 50 with long - standing lesions. The rate of metastasis is about 50% and that can be lethal if not diagnosed [1,6]. Although this tumor is considered as one of the rare reported cases in literature [5], it should be taken in consideration in a patient with a painful mass, in addition to other causes of painful dermal tumors such as leiomyoma, neuroma, dermatofibroma, angiolipoma and glomus tumor [2,7]. Therefore, clinical suspicion is not sufficient to make the diagnosis, and a skin biopsy is needed as the definitive diagnosis is based on histopathological findings. Our patient presented with classical features of eccrine spiradenoma, but in an atypical location as the tumor rarely arises on the anterior portion of the extremities. Surgical excision is the gold standard in the management of eccrine spiradenoma with low rates of recurrence [5-8]. In this case, the tumor recurred after 18 months of follow-up.

4. Conclusion:

Eccrine spiradenoma is a rare tumor that should be kept in mind in case of a patient with a painful nodule. Early diagnosis by mean of excisional biopsy is beneficial by providing quick and simple treatment and avoiding malignant transformation. Long-term follow-up is required in these cases.

Conflict of interest statement

The authors declare that they have no conflict of interest.

Consent

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

References:

- 1. Miceli A, Ferrer-Bruker SJ. Spiradenoma. StatPearls publishing; 2022.
- 2. Zheng Y, Tian Q, Wang J, Dong X, Jing H, Wang X, Feng Y, and Xioa S. Differential diagnosis of eccrine spiradenoma: A case report. Experemental and therapeutic medicine. 2014.
- 3. Saad S, Ghariani Fetoui N, Amri F, Aounallah A, Mokni S, Ben Hamouda M, et al. A case of eccrine spiradenoma presenting as a painless, reddish nodule on the forearm. Our Dermatol Online. 2022;13(3):338-339.
- 4. Son JH, Choi YW, Cho YS, Byun YS, Chung BY, Cho HJ, et al. A case of Eccine Spiradenoma: A rarely seen soft tissue tumor on the extensor surface of arm. Ann Dermatol. 2017;29 519-22.
- 5. Syauqi MS, Marliza H and BHK NG. A Rare Adnexal Tumor of Head & Neck: Eccrine Spiradenoma. Journal of Dermatology Research and Therapy. 2021.
- 6. Kanwaljeet S, Chatterjee T. Eccrine Spiradenoma: A rare adnexal tumor. Indian J cancer. 2017;54:695-6.
- 7. Chedid R, Nagel J, Barreto TM, Plata GT, Dutra ALB, Azevedo TP. Eccrine spiradenoma: a case report. Surg Cosmet Dermatol. 2015;7(2):171-3.
- 8. Miceli, A. (2022, October 13). Spiradenoma. StatPearls NCBI Bookshelf.https://www.ncbi.nlm.nih.gov/books/NBK448150/#_ncbi_dlg_citbx_NBK448150.

