

Cutaneous Epithelioid Angiomatous Nodule with Unusual Localization

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Abstract: Brenn and Fletcher described cutaneous epithelioid angiomatous nodule (CEAN) as a benign vascular proliferation that was in 2004. It is very rare condition. It is characterized by endothelial cells that have histiocytoid appearance. It is generally found in the dermis or sub cutis and rarely, in visceral organs. Despite the etiology of CEAN is not clear, it is considered as a reactive entity and no association with either infection, trauma or immunosuppression has been described yet. A 76-year-old woman visited hospital for routine examination. The physician noticed an erythematous nodule on the upper part of her right labium minus which appears like a cystic lesion. Histopathological examination revealed a relatively circumscribed, unencapsulated, unilobular vascular lesion which is in the superficial dermis. There were also moderately lymphoplasmacytic infiltration on the background. There were also scattered extravasated erythrocytes around. Positive stained for CD34, CD31 and negative stained for HHV-8 (performed to rule out a possibility of Kaposi sarcoma), pancytokeratin and s-100 were prominent in epithelioid cells. The overall histopathological appearance was consistent with CEAN and there was not any previous report CEAN occurring at this area.

Keywords: Cutaneous epithelioid angiomatous nodule, Labium minus, trauma, Cystic lesion, Hemorrhagic, Nuclear atypia.

INTRODUCTION

Brenn and Fletcher described Cutaneous Epithelioid Angiomatous Nodule (CEAN) as a benign vascular proliferation that was in 2004 [1]. It is characterized by endothelial cells that have histiocytoid appearance. It is generally found in the dermis or subcutis and rarely, in visceral organs [2, 3]. It is generally seen on head and neck region and trunk; the extremities, mucosal surfaces, vulva and penis are the rare places that evolve [4]. It must be differentiated from malignant vascular tumors especially at older ages. Despite the aetiology of CEAN is not clear, it is considered as a reactive entity and no association with either infection, trauma, nor immunosuppression has been described yet [5].

Our case is a woman, who is 76-year old, with an erythematous nodule at labium minus, which is an unusual localization for CEAN.

CASE REPORT

A 76-year-old woman visited surgery department for routine examination. The doctor noticed a erythematous nodule on the upper part of her right labium minus which appears like a cystic lesion. The lesion completely excised. Grossly, a well-circumscribed nodule was identified measuring 0.8 cm. The cut surface of the nodule was solid and hemorrhagic. Histopathological examination revealed unilobular nodule in the superficial dermis with a well-circumscribed architecture. The nodule was composed of epithelioid cells that have abundant cytoplasm and vesicular nucle. The epithelioid cells have solid or sheet-like pattern (Fig. 1). There were also moderate-

ly lymphoplasmacytic infiltration on the background. The nuclear atypia was absent. There was endothelial-lined channels and those were lined by a monolayer of variably epithelioid endothelium (Fig. 2). There were also scattered extravasated erythrocytes around. Three mitotic figures were present and they weren't atypical (Fig. 3). There were also scattered extravasated erythrocytes around. positive stained for CD34, CD31 and negative stained for HHV-8 (performed to rule out a possibility of kaposi sarcoma), pancytokeratin and s-100 were prominent in epithelioid cells (Fig. 4). The overall histopathological appearance was consistent with CEAN. The patient had been followed up without any recurrence, till one year following excision.

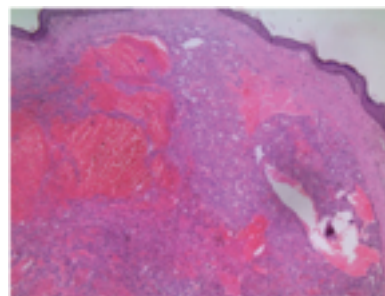


Fig. (1). Unencapsulated Vascular Lesion Located at Superficial Dermis (H&Ex40).

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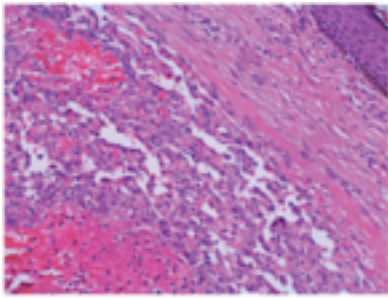


Fig. (2). Intralésional Endothelial-lined Channels (H&Ex200).

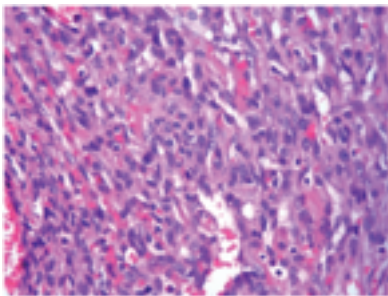


Fig. (3). Mitos and Extravasated Erythrocytes Around (H&Ex400).

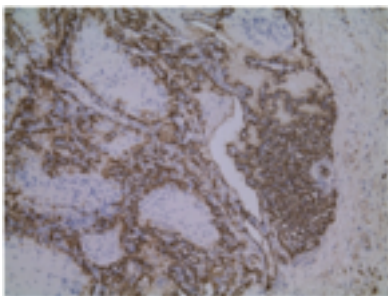


Fig. (4A). CD34 Positivity x100.

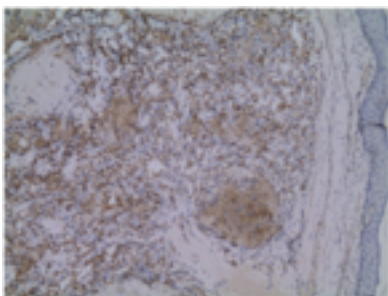


Fig. (4B). CD31 Positivity x100.

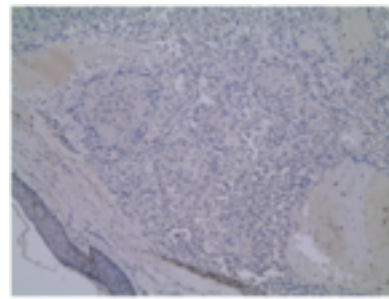


Fig. (4C). HHV-8 Negativity x100.

DISCUSSION

Brenn and Fletcher described cutaneous epithelioid angiomatous nodule (CEAN) as a benign vascular proliferation that was in 2004 [1]. From that day on about 60 cases have been described in literature [6]. CEAN has not any sex or age predilection, patients' ages were ranging 10 to 84 years old [2]. The lesions are commonly reddish to bluish, small nodules or papules that grows rapidly. They are nearly always seen alone [2, 6]. It is generally seen on head and neck region and trunk; the extremities, mucosal surfaces, vulva and penis are the rare places that evolve [4]. In our case CEAN was at labium minus and there was not any previous report of CEAN occurring at this area.

CEANs are histologically circumscribed, unencapsulated, unilobular vascular lesion that should be differentiated from other vascular lesions. Pyogenic granuloma (PG) can be easily distinguished because of its multilobular appearance and trauma history [1, 6, 7]. Also PG has an ulcerated epithelium overlying it. There was no trauma history, multilobulation or epithelial ulceration in our case.

Epithelioid hemangioendothelioma (EH) is a low-grade malignant neoplasm occurring in young females, often arises in solid organs and deep soft tissue. It is consisted of infiltrating cords and trabeculae. Unlike EH, CEAN is unilobular and it seen more densely cellular than vasoformative. It is rarely extends into the subcutaneous tissue [1, 8].

Epithelioid angiosarcoma (EA) is the malignant vascular tumor which is usually larger, show solid sheet-like pattern and infiltrating borders. Usually shows necrosis and atypical mitotic figures. CEAN usually does not shown prominent atypia of epithelioid cells or a destructive infiltrating pattern with necrosis [9]. The differential diagnosis can be very difficult especially those cases of CEAN presenting with moderate atypia and more than 5 mitotic figures/10 High-Power Field (HPF) [6,7]. In our case, we ruled out EH and EA with the absence of infiltrative growth pattern. Therefore absence of nuclear atypia, atypical mitotic figures and deep soft tissue involvement helped us. We also applied

HHV-8 for diferantiating from kaposi sarcoma and it was negative.

CONCLUSION

CEAN is a rare benign vascular tumor. It should be distinguished from malignant ones especially at older aged patients. Although malignancy was the first thing that came to mind in older ages, this case is a good example of a benign situation at older age and also with unusual localization.

CONFLICT OF INTEREST

Declared none.

ACKNOWLEDGEMENTS

Declared none.

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