CASE REPORT:

Eccrine Angiomatous Hamartoma in A 13-Year-Old Girl

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ABSTRACT

Eccrine angiomatous hamartoma (EAH) is a rare benign malformation characterized by a proliferation of eccrine glands and capillary vessels. Occurrence is mostly congenital and adolescent with few adult presentations. Clinically, it is characterized by plaques and nodules, which could be symptomatic. The diagnosis of EAH is typically made after a histopathological evaluation. Treatment is primarily surgical. We report the case of eccrine angiomatous hamartoma in a thirteen-year-old girl.

Keywords: Eccrine angiomatous hamartoma, haemangioma, dermatopathology, arteriovenous malformation

Cas clinique: Hamartome Angiomateux Eccrine Chez une Fille de 13 ans.

Résumé

L'hamartome angiomateux eccrine (HAE) est une malformation bénigne rare caractérisée par une prolifération de glandes eccrines et de vaisseaux capillaires. La survenue est le plus souvent congénitale et adolescente avec quelques présentations adultes. Cliniquement, il est caractérisé par des plaques et des nodules, qui peuvent être symptomatiques. Le diagnostic d'HAE est généralement posé après une évaluation histopathologique. Le traitement est principalement chirurgical. Nous rapportons le cas d'hamartome angiomateux eccrine chez une fille de treize ans.

Mots clés: hamartome angiomateux eccrine, hémangiome, dermatopathologie, malformation artérioveineuse.

INTRODUCTION

Eccrine angiomatous hamartoma (EAH) is a rare benign malformation characterized by a proliferation of eccrine glands and capillary vessels.[1,2] Although congenital and adolescent occurrence is mostly reported, EAH does occur in adults.[3-5] The diagnosis of EAH is typically made after a histopathological evaluation.[1,2] We report the case of eccrine angiomatous hamartoma in a thirteen-year-old girl.

CASE REPORT

A 13-year-old girl was referred to the dermatology clinic for a skin biopsy. She had been diagnosed at the referral clinic with dystrophic calciphylaxis on account of painful plaques and patches associated with hyperhidrosis on the left leg for a year. There was a progressive increase in the size of the patches and plaques. Hyperhidrosis was spontaneous and limited to the site of the patches and plaques. The patient was reported to have had surgery on the same leg at the age of two years for an unknown indication by the mother. This was followed by poor healing, contracture, limb length discrepancy and the use of crutches for ambulation.

On examination, she had multiple hyperpigmented, firm, tender plagues and patches with an erythematous hue on the left leg (lateral calf region) and sweat droplets. Figure 1. Other examinations were essentially normal. A provisional diagnosis of

a Haemangioma with a differential of an adnexal tumour was made. A skin biopsy was done. Dermatopathology revealed a hyperkeratotic, acanthotic epidermis overlying a dermal proliferation of eccrine glands admixed with blood vessels, haemosiderin, sclerosed dermis and fibroblasts. The eccrine glands were ectatic, thick-walled and the blood vessels were telangiectatic, thin-walled: some were filled with red blood cells. Figures 2A to D. A diagnosis of Eccrine angiomatous hamartoma was made. The patient was referred to the surgeons for excision of the plaques.

DISCUSSION

Eccrine angiomatous hamartoma (EAH) occurs as plaques, patches and macules of various colours (bluish, brownish, yellowish, reddish).[4,6-8] Eccrine angiomatous hamartoma can occur as a single or multiple lesions with no sex predilection.[3,7] Our patient had hyperpigmented plaques with an erythematous hue. In the skin of colour individuals, erythema may appear hyperpigmented. EAH appears to have a predilection for the extremities, although occurrences on the neck and face have been documented.[3,6,8] Common symptoms of EAH are pain and hyperhidrosis.[3,6] The hyperhidrosis is attributed to the increased proliferation of eccrine glands and pain due to the involvement of superficial nerves.[6] Our patient reported hyperhidrosis and pain. Due to its rarity, Eccrine angiomatous hamartoma is frequently misdiagnosed clinically as angiokeratoma, arteriovenous malformation and haemangioma.[3,6] Clinically, our patient was diagnosed to have a haemangioma or an adnexal tumour. Dermoscopy patterns reported in EAH include a white centre surrounded by a purple-brown round lacuna,[4] Spitzoid patterns,[7] and pop-corn appearances.[8]

The pathogenesis of EAH is not known.[3,4,6] Theories of pathogenesis include radiation therapy, frequent trauma, and abnormal interaction of epithelial and mesenchymal elements resulting in the atypical development of eccrine and vascular structures.[4-6] Our patient had surgery at the age of 2 years and walks with crutches with daily micro-trauma. The authors opine that trauma may be a factor in the occurrence of EAH in this patient. The diagnosis of EAH is histopathological and based on the criteria proposed by Pelle *et al*.[1,2] Histopathological features are a dermal proliferation of mature, normal or dilated eccrine glands, intimate relationship of the eccrine structures with benign thin-walled vascular proliferation and varying occurrence of pilar, lipomatous, mucinous, and/or lymphatic structures.[1-4,6] Our patient had all the features (figures 2A-E). Therapeutic options in EAH include Lasers, botulinum toxin injection and surgical excision.[4,7,9]

The prognosis for EAH is good: no malignant transformation, although spontaneous regression does not occur.[10] Our limitations were the inability to follow up with this patient concerning the surgical outcome and the inability to perform immunohistochemistry for reasons of unavailability and finance. We have reported this case for the following reasons. Eccrine angiomatous hamartoma is a rare congenital tumour. It is only correctly diagnosed following histology. This case report highlights the importance of histopathology in diagnosing skin diseases.

Conflict of interest: None

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Declaration

This manuscript is original and has not been submitted to another journal.

Conflict of interest

The authors have no conflict of interest.

Legends

1. Figure 1. Left leg: multiple hyperpigmented plaques and patches, sweat droplets
2. Figure 2A. The proliferation of eccrine glands, thin-walled vessels and haemosiderin. H&E x 10
3. Figure 2B. Ectatic thin-walled vessels, haemosiderin, nested ectatic eccrine glands. H&E x 20
4. Figure 2C. Telangiectatic vessels filled with red blood cells, proliferation of fibroblasts and haemosiderin. H&E x 20
5. Figure 2D. Thick-walled ectatic eccrine glands surrounded by haemosiderin. H&E x 20
6. Figure 2E. Ectatic thick-walled eccrine glands. H&E x 20

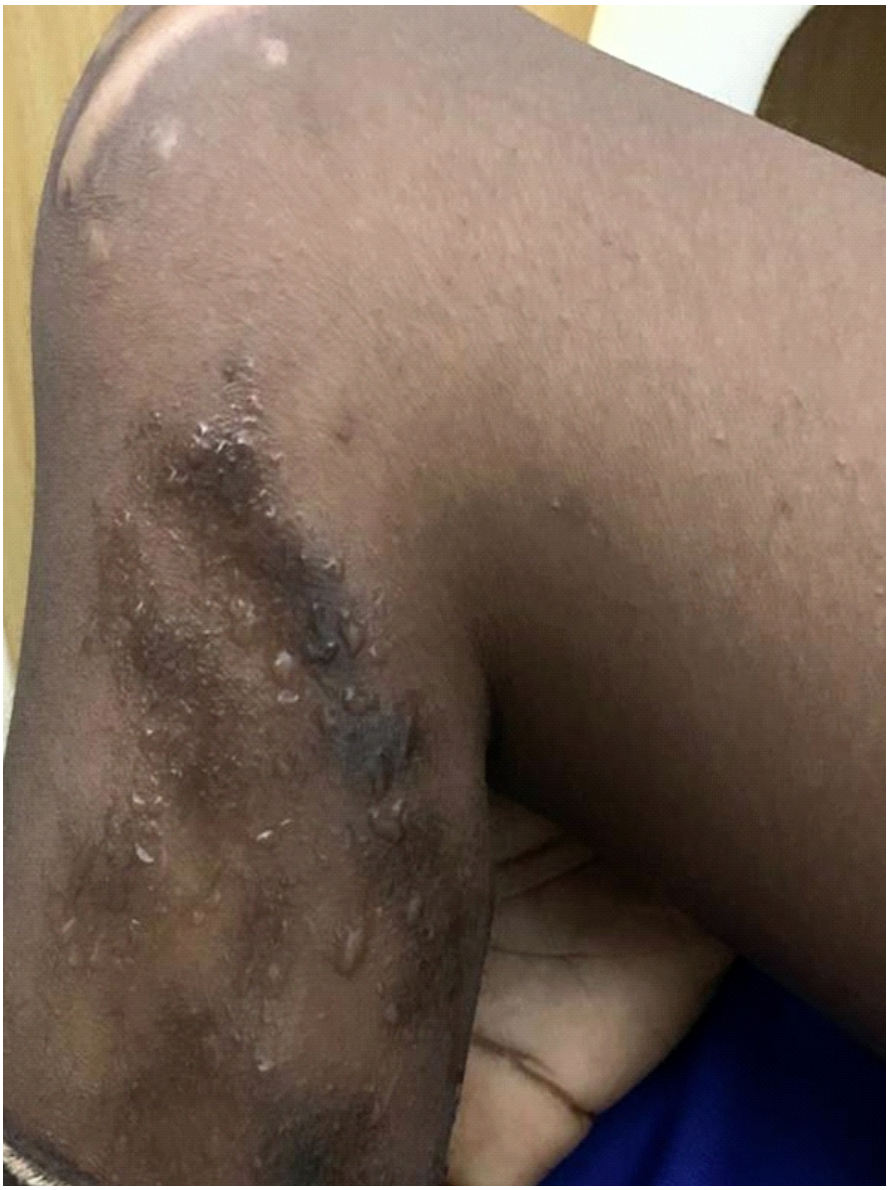

Figure 1: Left leg: multiple hyperpigmented plaques and patches, sweat droplets

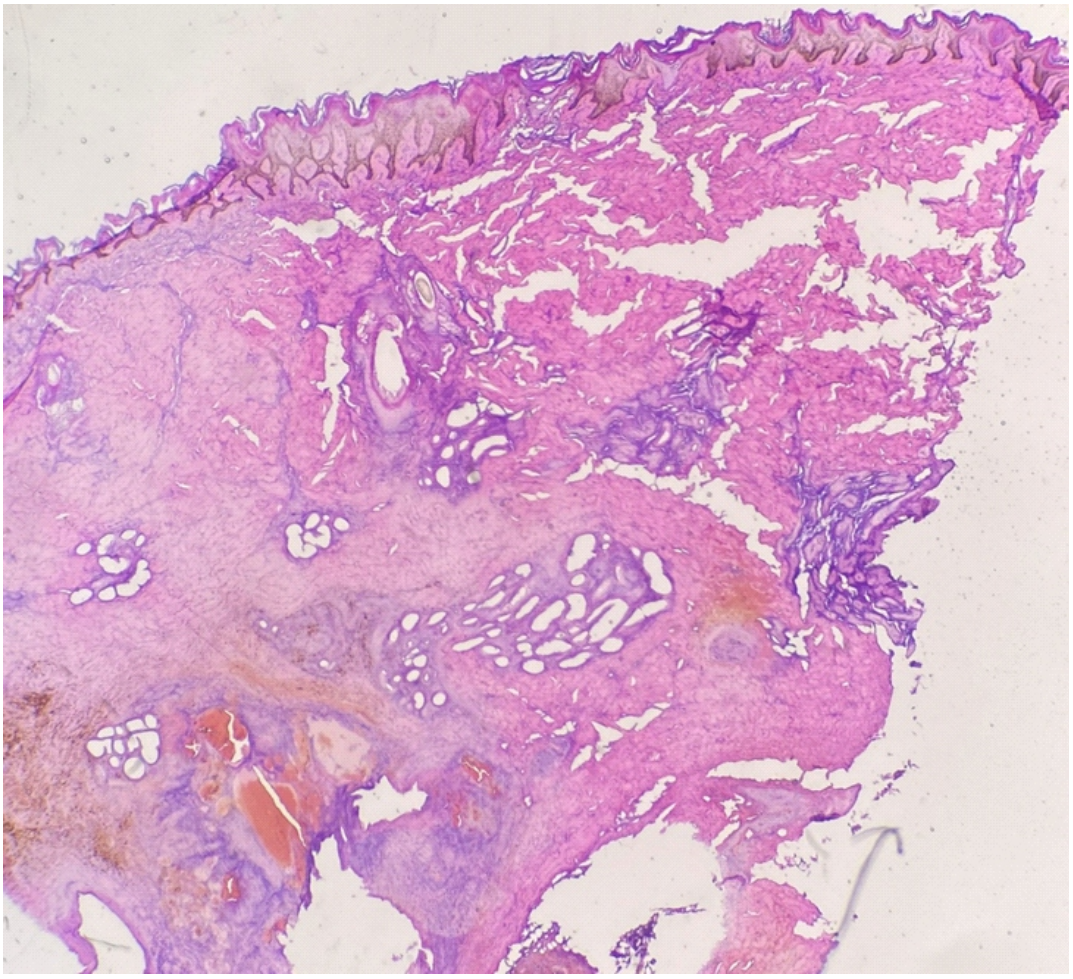

Figure 2A: Proliferation of eccrine glands, thin-walled vessels and haemosiderin. H&E X10

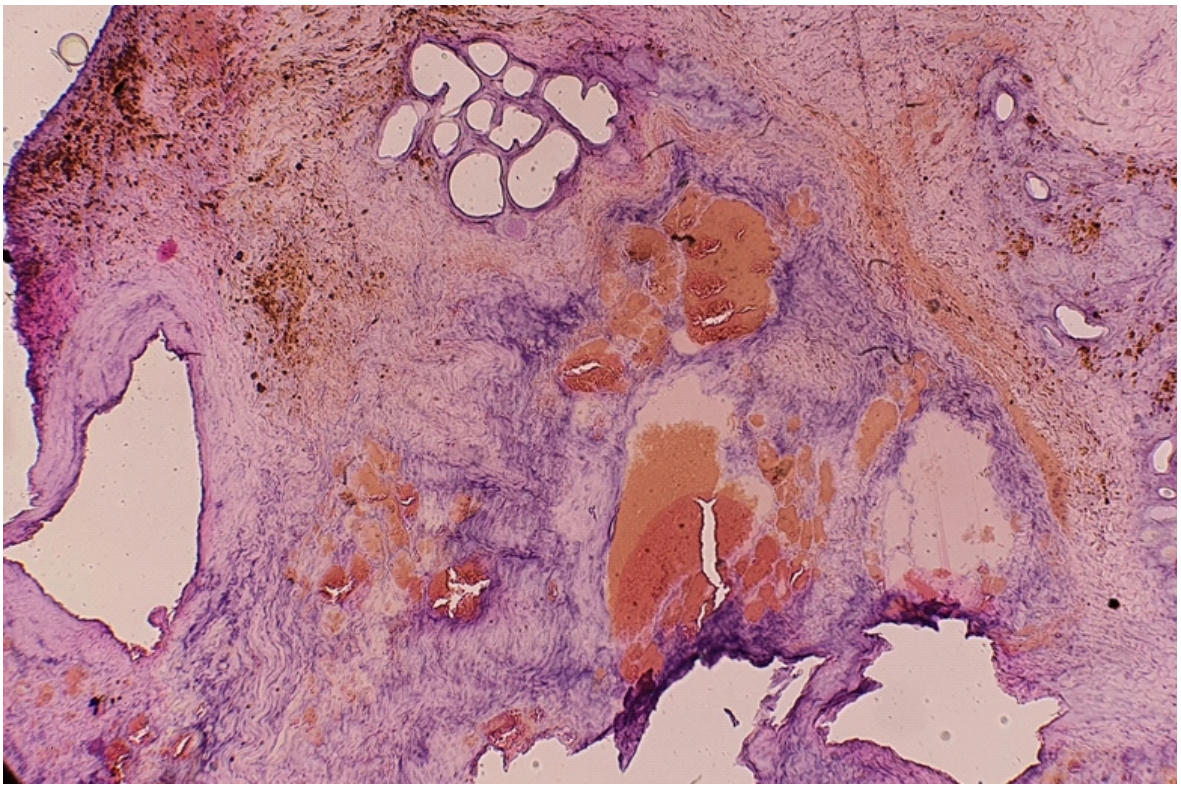

Figure 2B: Ectatic thin-walled vessels, haemosiderin, nested ectatic eccrine glands. H & E X20

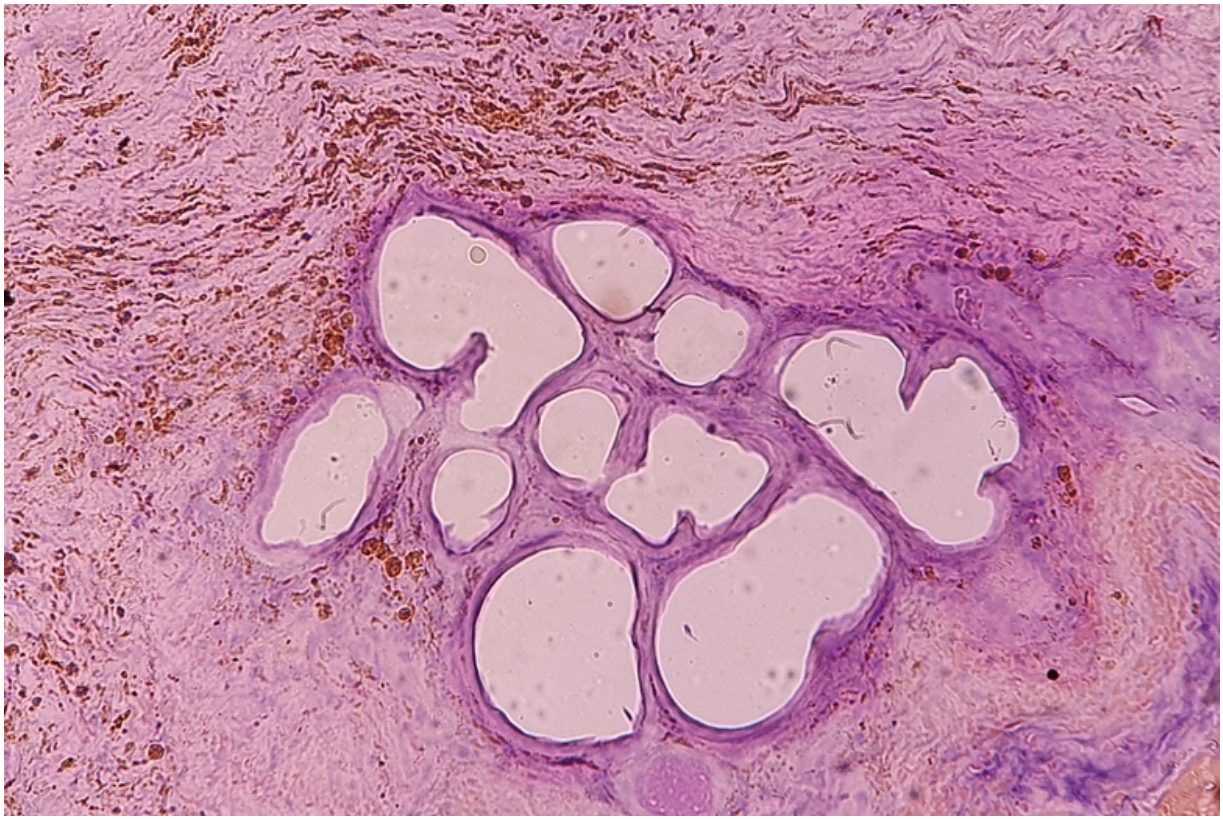

Figure 2C. Thick-walled ectatic eccrine glands surrounded by haemosiderin. H & E X2O

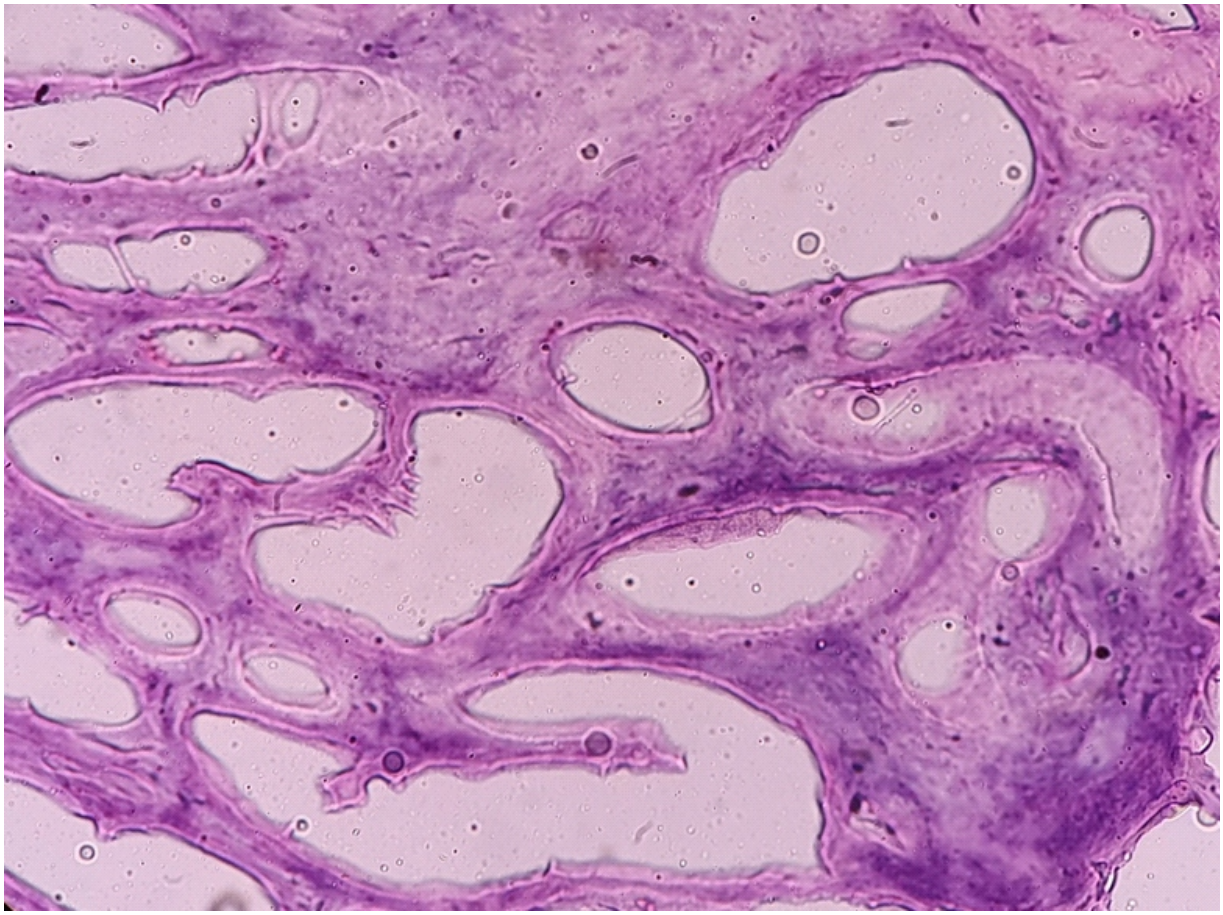

Figure 2D. Ectatic thick-walled eccrine glands