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Multiple painful brownish plaques associated with local hyperhidrosis

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ABSTRACT Eccrine angiomatous hamartoma is a rare hamartomatous lesion characterized by proliferation of eccrine glands and small blood vessels, and occasionally other elements. It generally arises congenitally or later in childhood, as solitary or multiple lesions on the distal extremities. Adult-onset multiple lesions are very rare. Herein we describe a 33-year-old male with symptomatic multiple eccrine angiomatous hamartoma lesions.

Case presentation

A 33-year old male presented with a 6-year history of brownish plaques on both legs. His medical and family histories were unremarkable. The patient reported that the plaques were occasionally painful and that the lesions produced more sweat than the surrounding skin. The patient also reported that there was no trauma preceding the appearance of the lesions. Dermatological examination showed brownish indurated plaques at the medial sides of the bilateral popliteal fossa and posterior of the right leg (Figure 1). The plaques were painful upon palpation. The starch-iodine test was performed to determine the presence of hyperhidrosis. To perform this test the plaque on the patient's right leg was painted with iodine solution and allowed to dry thoroughly. Then, a starch powder was sprinkled on the surface and the appearance of purple-black speckling was observed, indicating presence of hyperhidrosis. Next, an incisional biopsy specimen was obtained from the same lesion. Hematoxylin-eosin (H&E) staining of the biopsy specimen showed lobules with mucinous stroma in the deep dermis and subcutaneous tissue (Figure 2). The lobules were composed of hyperplastic and proliferated eccrine glands, and numerous small blood vessels (Figure 3). With the clinical and histopathological findings, a diagnosis of eccrine angiomatous hamartoma (EAH) was made.

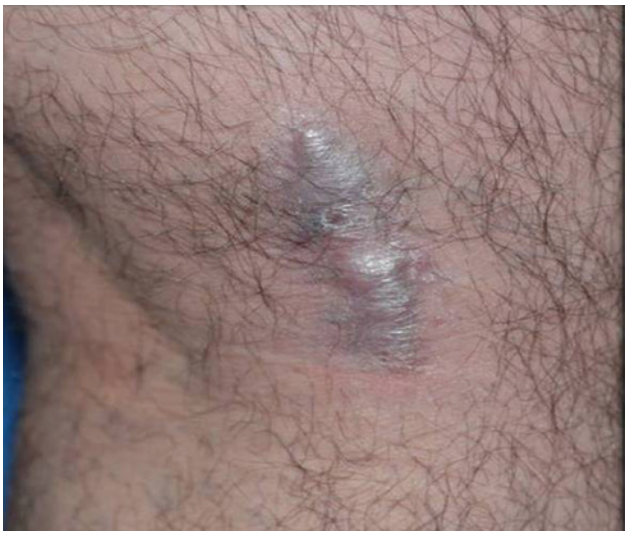

Figure 1. Brownish indurated plaque at the medial side of the popliteal fossa. (Copyright: ©2015 Duman et al.)

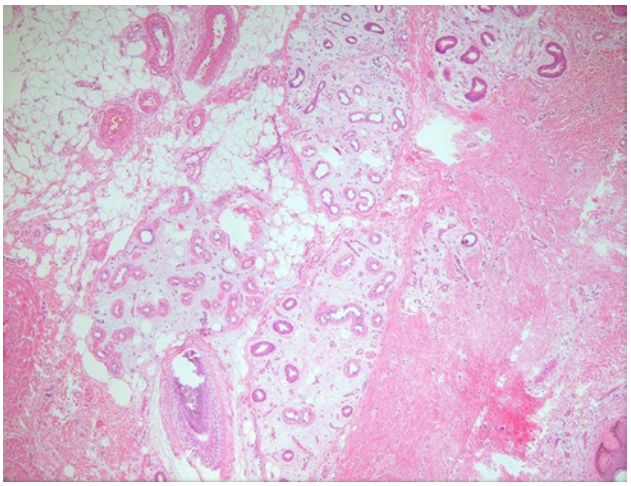

Figure 2. Proliferative lobules with mucinous stroma in the deep dermis and subcutaneous tissue (H&E, x10 original magnification). (Copyright: ©2015 Duman et al.)

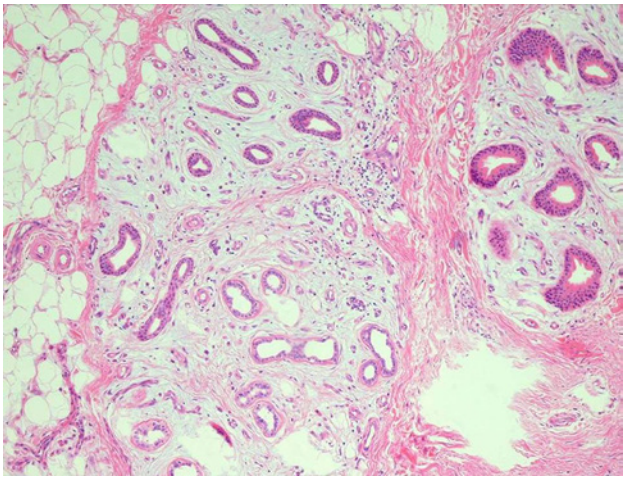

Figure 3. Proliferative eccrine glands and small blood vessels within the mucinous stroma (H&E, x40 original magnification). (Copyright: ©2015 Duman et al.)

Conclusion

EAH is a rare hamartomatous lesion characterized by proliferation of eccrine glands and small blood vessels, and occasionally other elements, such as adipose tissue, hair, and epidermis [1,2]. EAH generally occurs congenitally or later in childhood as solitary or multiple lesions on the distal extremities, especially the legs; however, pubertal- and adult-onset lesions, and trunk and head involvement were also reported [3,4]. To date, a gender predilection has not been reported. The clinical presentation of EAH ranges from a simple angiomatous nodule to erythematous-purpuric plaques. It is generally asymptomatic, but occasionally can be associated with pain and hyperhidrosis, and less frequently hypertrichosis [1,4].

The etiopathogenesis of EAH remains unknown. Some researchers have suggested that faulty interaction between differentiating epithelium and the underlying mesenchyme gives rise to abnormal proliferation of adnexal and vascular structures in congenital forms. Late onset lesions have been associated with recurrent trauma [2]. Histopathological examination of EAH is usually characterized by a circumscribed, non-encapsulated lobular lesion in the mid to deep dermis that is composed of mature-looking and occasionally dilated eccrine glands, and associated benign vascular proliferation. The vascular component is composed of small blood vessels exhibiting variable dilatation. Occasionally, mucinous change, a lipomatous component, or pilar structures are observed [4]. The epidermis is usually normal, but occasionally verrucous changes are present.

EAH must be differentiated from vascular malformations, tufted angioma, smooth muscle hamartoma, glomus tumor, blue rubber bleb nevus, and macular telangiectatic mastocytosis [3]. Definitive diagnosis of EAH is based on clinicopathological correlation.

As EAH is a benign, slow-growing lesion, aggressive treatment is not indicated. Simple excision is currently the treatment of choice in patients with undesirable symptoms, such as pain or hyperhidrosis, those with cosmetic concerns, and in cases of progressive enlargement of the lesions [2]. Botulinum toxin may be considered to treat lesions associated with hyperhidrosis [2]. As the lesions in the presented patient were symptomatic, they were excised.

In conclusion, EAH is a rare entity with a unique clinical and histopathological presentation that should be considered in the differential diagnosis of vascular and hamartomatous lesions.

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