Eccrine angiomatous hamartoma with verrucous hemangioma-like features – an unusual combination

Sir,

A 25-year-old male presented with a history of asymptomatic reddish-purple patches on the dorsal right hand since birth. On examination, there were several clustered, erythematous-violaceous and hyperkeratotic plaques on the dorsal right hand [Figure 1a]. Dermoscopy revealed a prominent blue-white background, with hyperkeratosis in the center of the lesions, surrounded by purple-brown round lacunae indicative of underlying dilated vessels [Figure 1b]. Doppler ultrasonography of the lesions detected dotted blood flow signals in the dermis and subcutis. Magnetic resonance imaging demonstrated mixed long T1 and T2 signals of lesions involving dermal and subcutaneous tissue without involving tendons and muscles, which was suggestive of a hemangioma.

Skin biopsy showed hyperkeratosis, acanthosis and papillomatosis in the epidermis. The papillary dermis had numerous thin-walled, ectatic and irregular vessels [Figures 2a and 2b]. In the reticular dermis and subcutis, proliferation of eccrine glands associated with thin walled vessels was observed [Figure 2c]. On immunohistochemistry, the vascular endothelial cells were positive for CD31 [Figure 2d] and weakly positive for GLUT-1 [Figure 2e]. Positive attaining for αSMA was seen in the yoepithelial layer of secretory coils and vascular smooth muscle cells [Figure 2f]. We speculated that the diagnosis could be eccrine angiomatous hamartoma with verrucous hemangioma-like features. Intralesional Nd: YAG laser (energy 130 mJ, frequency 60 Hz) was utilized to treat the lesions which improved satisfactorily after two treatments [Figure 3].

In a study of 26 cases of eccrine angiomatous hamartoma, seven demonstrated mild hyperkeratosis in the epidermis; another study involving 15 cases reported that one patient had verrucous changes in the epidermis and abundant capillaries in the papillary dermis.[1,2] Wang *et al*. analyzed 74 cases of verrucous hemangioma and found hyperplasia of the eccrine glands around abnormal vessels in four cases. They opined that these were verrucous hemangiomas with features of eccrine angiomatous hamartoma.[3] However, a literature search could find only two reported cases with the

Figure 1a: Multiple reddish-purple hyperkeratotic plaques on the dorsal right hand

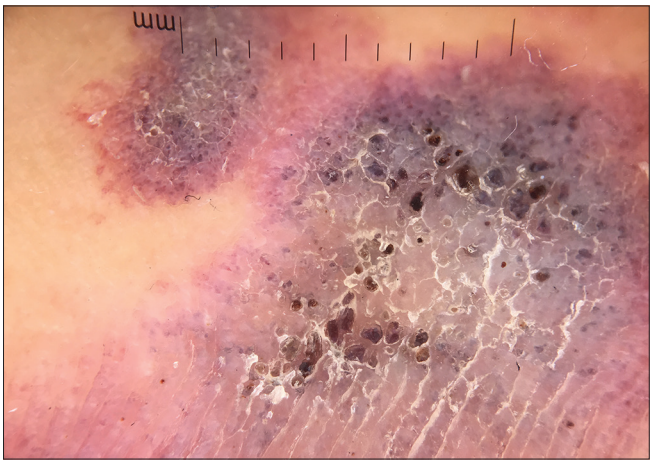

Figure 1b: Dermoscopic examination: blue-white background, hyperkeratosis in the middle of the lesion, surrounded by purple-brown round lacunae

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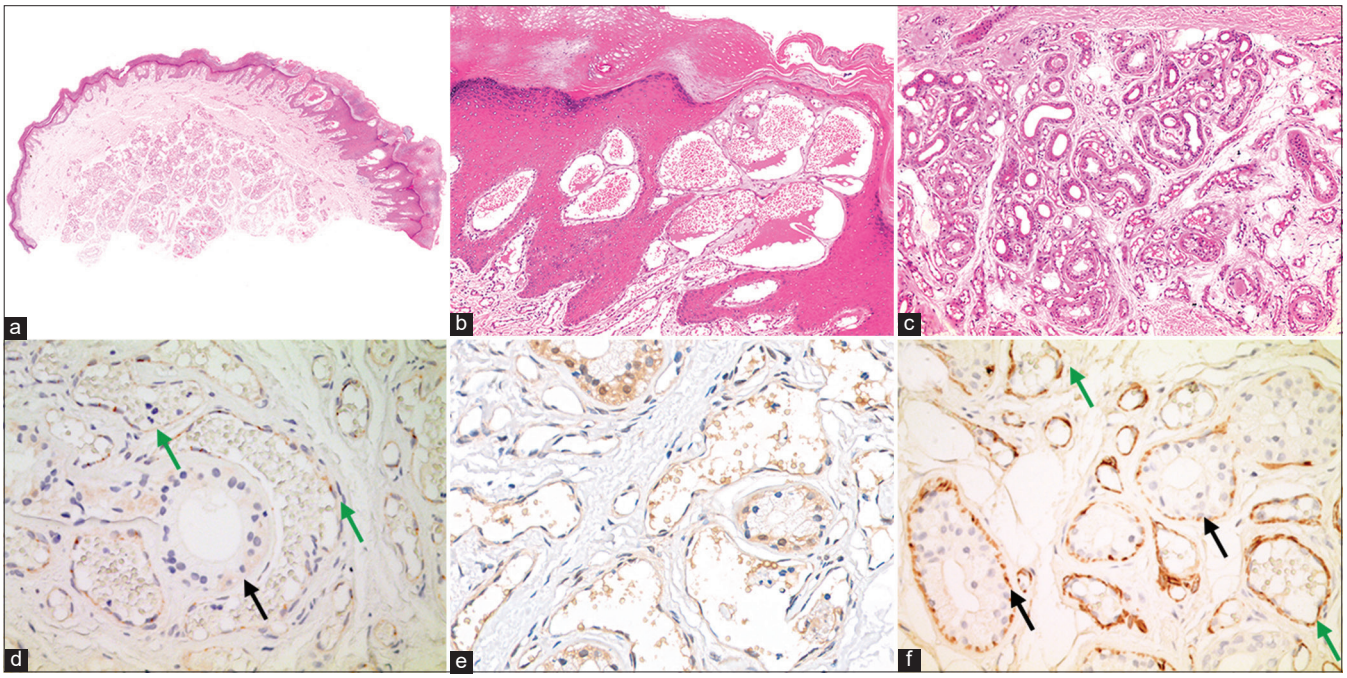

Figure 2: Histopathology examinations of lesions from dorsal aspect of the right hand. (a) Low-power view of biopsy (hematoxylin–eosin[HE],original magnification ×10). (b) High-power view showing hyperkeratosis, acanthosis and papillomatosis in epidermis,thin-walled, ectatic, vessels in papillary dermis (×100). (c) Proliferation of eccrine glands associated with thin-walled vessels in reticular dermis (×100). (d) Immunohistochemical staining showing that the vascular endothelial cells (green arrow) were positive for CD31 and eccrine glands (black arrow) were negative (×400). (e) Vessels were weakly positive for GLUT-1 (×400). (f) Myoepithelial layer of secretory coils (black arrow) and vessels (green arrow) were positive for αSMA (×400)

Figure 3: Apparent improvement of the plaques (blue arrow) after treatment with Nd:YAG laser

diagnosis of eccrine angiomatous hamartoma with verrucous hemangioma-like features. In Table 1 we have summarized the characteristics of eccrine angiomatous hamartoma and verrucous hemangioma so as to highlight their salient differences.].

Both eccrine angiomatous hamartoma and verrucous hemangioma present mostly at birth or childhood with lesions principally localized to the extremities. Eccrine angiomatous hamartoma is mostly isolated papules, while verrucous hemangioma is characterized as multiple and clustered plaques or nodules. The dermoscopic features of eccrine angiomatous hamartoma are spitzoid or popcorn patterns, whereas those of verrucous hemangioma are reddish-blue or bluish lacunae or dermis with a bluish-white hue. Clinically and dermoscopically the lesions seen in our patient were more suggestive of verrucous hemangioma. The symptoms of eccrine angiomatous hamartoma include pain and hyperhidrosis, whereas those of verrucous hemangioma are itch, oozing and bleeding. However, our patient did not exhibit typical symptoms of either entity.

Patterson *et al*. developed the histopathological criteria for eccrine angiomatous hamartoma including: (a) hyperplasia of normal or dilated eccrine glands; (b) intermixing of eccrine glands with abundant capillaries; (c) variable presence of apocrine, lymphatic or mucinous structures.[4] Histology of verrucous hemangioma demonstrates hyperkeratosis, papillomatosis and acanthosis in the epidermis with numerous dilated vessels extending into the subcutaneous tissues. Our case showed histological characteristics of both eccrine angiomatous hamartoma and verrucous hemangioma.

GLUT-1 and WT-1 are markers that differentiate vascular neoplasms from vascular malformations. Trindade *et al*. reported that verrucous hemangioma was positive for GLUT-1 in 13 cases (100%), suggesting that verrucous hemangioma may be categorized as a form of vascular

Table 1: Comparison between eccrine angiomatous hamartoma and verrucous hemangioma

Features	Eccrine angiomatous hamartoma	Verrucous hemangioma
Onset	Congenital or later in childhood	Congenital or in early infancy
Location	Distal extremities	Distal extremities
Distribution	Mostly solitary papules	Grouped of plaques or nodules
Symptom	Pain, hyperhidrosis	Itch, oozing, bleeding
Dermoscopy	A spitzoid pattern or a popcorn pattern	Bluish-white hue (hyperkeratosis), reddish-blue or bluish lacunae
Histopathology	Eccrine sweat glands associated with thin-walled, aggregated vessels in the middle and lower dermis	Hyperkeratosis, acanthosis and papillomatosis in epidermis, vascular component in dermis and subcutaneous tissue
GLUT-1 expression	Negative	Mostly positive
Therapy	Surgery, laser, botulinum toxin	Surgery, laser, topical steroid with salicylic acid ointment

tumor.[5] However, Wang *et al*. found that vessels in verrucous hemangioma were positive for GLUT-1 in 49 cases (66%), focally positive for Prox1 in 69 (93%) cases, while negative for WT-1 in 60 cases (81%).[3] They proposed that verrucous hemangioma is a vascular malformation and an incomplete lymphatic immunophenotype. The reason why for GLUT-1 is positive in verrucous hemangioma is not clear, but the staining is less intense than in infantile hemangiomas.

In conclusion, the mixed histological features comprising both eccrine gland proliferation as well as vascular proliferation and weak GLUT-1 positivity support the diagnosis of eccrine angiomatous hamartoma with verrucous hemangioma-like features in our case. To the best of our knowledge, this is the first reported case of eccrine angiomatous hamartoma with verrucous hemangioma-like features, to be characterized in such great detail using a combination of histopathology, immunohistochemistry and dermoscopy.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

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References

1. Sanusi T, Li Y, Sun L, Wang C, Zhou Y, Huang C. Eccrine angiomatous hamartoma: A clinicopathological study of 26 cases. Dermatology 2015;231:63-9.
2. Lin YT, Chen CM, Yang CH, Chuang YH. Eccrine angiomatous hamartoma: A retrospective study of 15 cases. Chang Gung Med J 2012;35:167-77.
3. Wang L, Gao T, Wang G. Verrucous hemangioma: A clinicopathological and immunohistochemical analysis of 74 cases. J Cutan Pathol 2014;41:823-30.
4. Patterson AT, Kumar MG, Bayliss SJ, Witman PM, Dehner LP, Gru AA. Eccrine angiomatous hamartoma: A clinicopathologic review of 18 cases. Am J Dermatopathol 2016;38:413-7.
5. Trindade F, Torrelo A, Requena L, Tellechea O, Del Pozo J, Sacristán F, *et al*. An immunohistochemical study of verrucous hemangiomas. J Cutan Pathol 2013;40:472-6.

Malnutrition dermatosis following adrenal insufficiency in an adult

Sir,

A 26-year-old man presented to the dermatology outpatient department of the All India Institute of Medical Sciences, Bhubaneswar, with redness and peeling of skin on the trunk and buttocks, blisters on the legs along with weakness, head reeling, vomiting and decreased appetite for 1 month. General examination revealed severe pallor, pitting acral edema and low blood pressure (90/60 mmHg). On cutaneous

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