Yonago Acta Medica 2023;66(3):380–381 doi: 10.33160/yam.2023.08.004

Eccrine Angiomatous Hamartoma Occurring on the Nail Bed

Hiroshi Notsu,* Hiroyuki Goto,* Naomi Tani* and Kazunari Sugita*†

**Division of Dermatology, Department of Sensory and Motor Organs, School of Medicine, Faculty of Medicine, Tottori University, Yonago 683-8503, Japan, and †Division of Dermatology, Department of Internal Medicine, Faculty of Medicine, Saga University, Saga 849-0937, Japan*

ABSTRACT

A 71-year-old woman was referred to our clinic with a 2-month history of pain and exudate from her right first toe. Physical examination revealed a subungual nodule that elevated the nail plate and produced distal onycholysis. We performed onychectomy and removed the nodule. A histopathological specimen from the nodule showed hyperkeratosis and acanthosis in the epidermis, papillomatosis and dilated vessels in the superficial dermis, and aggregation of eccrine glands in the middermis. Based on these findings, a diagnosis of eccrine angiomatous hamartoma (EAH) was made. EAH is a rare cutaneous hamartoma that is histologically characterized by proliferation of eccrine glands and vascular components. To our knowledge, EAH occurring in the subungual region has not been reported. The pathogenesis of acquired EAH has not been completely elucidated, but several cases have been reported to be caused by external stimuli. Although the subungual area generally has no sweat glands, our case suggests that a traumatic stimulus can induce EAH anywhere including regions where sweat glands are normally not found.

Key words cutaneous hamartoma; eccrine angiomatous hamartoma; nail bed; onycholysis

Eccrine angiomatous hamartoma (EAH) is a rare benign hamartoma that is histologically characterized by proliferation of eccrine glands and vascular components. EAH usually occurs on the extremities, and EAH occurring in the subungual region has not been reported. Here, to our knowledge, we present the first case of EAH occurring on the nail bed and discuss its underlying pathogenic mechanisms.

PATIENT REPORT

A 71-year-old woman was referred to our clinic with a 2-month history of pain and exudate from her right first toe. Physical examination revealed a subungual nodule that elevated the nail plate and produced distal onycholysis (Fig. 1a). Differential diagnoses included subungual exostosis, subungual fibrokeratoma and glomus tumor. Since an X-ray examination showed that the bone fragment was separated from the distal phalanx of the thumb, we excluded the possibility of exostosis (Fig. 1b). We performed onychectomy, and intraoperative findings showed a skin-colored nodule and central cleft. In addition, there was a piece of isolated bone in the central cleft (Fig. 1c). We removed the isolated bone and the nodule. A histopathological specimen from the nodule showed hyperkeratosis and acanthosis in the epidermis, papillomatosis and dilated vessels in the superficial dermis, and aggregation of eccrine glands in the middermis (Figs. 1d and e). Immunohistochemistry revealed that the dilated vessels were positive for CD31 and negative for D2-40 (Fig. 1f). Alcian-blue staining for mucin was strongly positive around the eccrine glands (data not shown). Based on these findings, a diagnosis of EAH was made. The patient's symptoms were greatly improved after removal of the bone fragment.

DISCUSSION

EAH was first described by Lotzbeck in 1859 and the term was first coined by Heyman et al. in 1968. Approximately 200 cases of "EAH" have been reported since 1968. Among 99 patients with available information, 43 patients (43.4%) were male and 56 patients (56.6%) were female. Fifty-six cases (56.6%) developed before the age of 12 months. The lesions were commonly located on lower extremities (44 patients, 44.4%), upper extremities (38 patients, 38.4%), trunk (20 patients, 20.2%), and head and neck area (8 patients, 8.1%). Therefore, our case was the first case in the context of its location. The histopathological characteristic of EAH is proliferation of eccrine glands and vascular components. It was previously reported that EAH is a lymphatic proliferation and that it should be named eccrine lymphangiomatous hamartoma.[1] According to that report, some cases of EAH were positive for D2-40 in the vessels and it was suggested that it could have originated from lymphatic vessels.[1] EAH may have two possibilities of vascular and lymphatic origins. In the present case, D2-40 was negative and CD31 was

Corresponding author: Naomi Tani, MD
taninaomi@tottori-u.ac.jp
Received 2023 April 14
Accepted 2023 June 2
Online published 2023 July 18
Abbreviation: EAH, eccrine angiomatous hamartoma

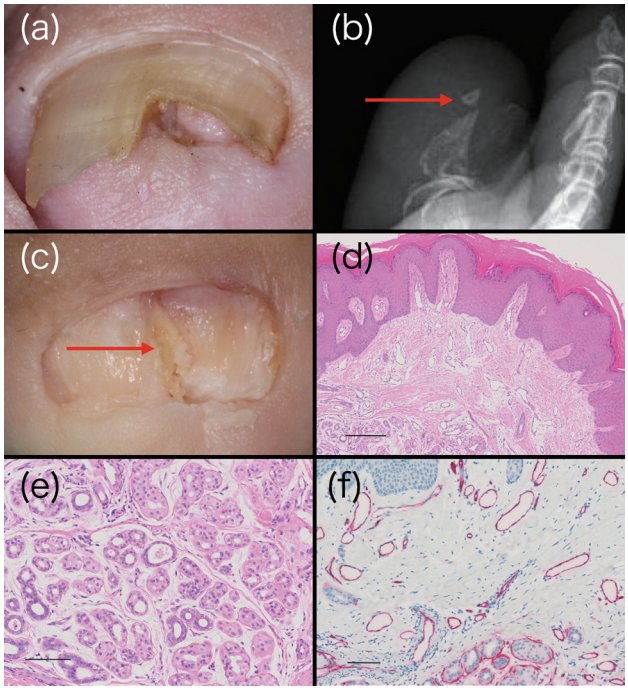

Fig. 1. (**a**) Physical examination revealed onycholysis in the distal nail bed and a subungual nodule. (**b**) X-ray examination revealed an isolated bone (red arrow) in the distal part of the distal phalanx of the thumb. (**c**) Intraoperative findings of onychectomy showed a skin-colored nodule and central cleft. In addition, an isolated bone was seen in the cleft (red arrow). (**d**) A histopathological specimen from the nodule showed mild hyperkeratosis, acanthosis and papillomatosis in the epidermis and dilated vessels in the superficial dermis (H&E staining, bar = 250 μm). (**e**) In the mid-dermis, eccrine glands were aggregated (H&E staining, bar = 100 μm). (**f**) CD31 staining was positive for the dilated vessels in the superficial dermis (CD31 staining, bar = 80 μm).

positive for the vessels. Therefore, we figured that the present case originated from vascular components.

The pathogenesis of acquired EAH has not been completely elucidated, but several cases have been reported to be caused by external stimuli.[2] In this case, we speculated from the X-ray findings that the bone fragment indicated bone fracture caused by an external injury. There have been some cases of EAH in which apocrine glands, hair follicles and adipose tissue were observed around the lesion of EAH.[3–6] Although the subungual area generally has no sweat glands, unusual elements such as an adnexal structure and fat tissue can be included in EAH.

In conclusion, our case suggests that a traumatic stimulus can induce EAH anywhere including regions where sweat glands are normally not found.

The authors declare no conflicts of interest.

REFERENCES

1 Wang L, Wang S, Gao T, Wang G. Eccrine angiomatous hamartoma is a lymphatic proliferation. Eur J Dermatol. 2013;23:614-7. DOI: 10.1684/ejd.2013.2152, PMID: 24200985
2 Hawryluk EB, Schmidt B, Maguiness S. Enlargement of eccrine angiomatous hamartoma following trauma. Pediatr Dermatol. 2015;32:e92-4. DOI: 10.1111/pde.12564, PMID: 25781494
3 Donati P, Amantea A, Balus L. Eccrine angiomatous hamartoma: a lipomatous variant. J Cutan Pathol. 1989;16:227-9. DOI: 10.1111/j.1600-0560.1989.tb00046.x, PMID: 2794166
4 Chien AJ, Asgari M, Argenyi ZB. Eccrine angiomatous hamartoma with elements of an arterio-venous malformation: a newly recognized variant. J Cutan Pathol. 2006;33:433-6. DOI: 10.1111/j.0303-6987.2006.00272.x, PMID: 16776719
5 Zeller DJ, Goldman RL. Eccrine-pilar angiomatous hamartoma. Report of a unique case. Dermatology. 1971;143:100-4. DOI: 10.1159/000252176, PMID: 5138948
6 Smith VC, Montesinos E, Revert A, Ramon D, Molina I, Jorda E. Eccrine angiomatous hamartoma: report of three patients. Pediatr Dermatol. 1996;13:139-42. DOI: 10.1111/j.1525-1470.1996.tb01421.x, PMID: 9122072