Eccrine Angiomatous Hamartoma: A Retrospective Study of 15 Cases

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Background: Eccrine angiomatous hamartoma (EAH) comprises a rare nevoid proliferation of normal eccrine glands and small blood vessels and occasionally other elements in the middle and deep dermis with variable clinical manifestations. Case series have rarely been published except for case reports and literature reviews. The aims of this article were to investigate the clinical and pathologic features of patients with EAH in Taiwan and to compare our results with the results of previous studies.

Methods: A retrospective review of medical records and histopathological findings was performed on patients diagnosed with EAH in a medical center in Taiwan between 1994 and 2010.

Results: Fifteen patients with pathologically diagnosed EAH were collected. The mean age at the time of diagnosis was 38.6 years (range, birth to 67 years). The male to female ratio was 3 to 2. In most cases, EAH arose as a single lesion on a lower extremity. The symptoms and signs most commonly associated with EAH were pain (60%), hypertrichosis (13.3%), itching (13.3%) and hyperhidrosis (6.7%). Additional pathological findings included hemangioma (13.3%), verrucous hemangioma (6.7%), arteriovenous malformation (6.7%), and angiokeratoma (6.7%). None of the patients experienced spontaneous regression of the lesions before excision. Excisions were done in one patient under general anesthesia, and ten patients with local anesthesia. Four patients were kept under observation. Tumor recurrences were noted in two out of the eleven patients whose lesions were excised.

Conclusion: Compared with cases in the literature, we found additional histopathological findings and an increased tumor recurrence risk in our cohort. EAH remains a benign and uncommon hamartomatous condition. Further multi-center, retrospective studies with larger case numbers are needed to better characterize the disease presentation in Asian populations.

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Key words: eccrine angiomatous hamartoma, sweat gland tumor, hamartoma, hemangioma, ultrasonography

Eccrine angiomatous hamartoma (EAH) comprises a rare nevoid proliferation of normal eccrine glands and small blood vessels, and occasionally other elements such as fatty tissue and pilar struc-

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tures in the middle and deep dermis with variable clinical manifestations. Some authors have proposed a pathophysiological model which considers it a biochemical fault in the interactions between differentiating epithelium and subjacent mesenchyme that gives rise to an abnormal proliferation of adnexal and vascular structures.[1]

Clinically, it often presents as one or several nodules or a solitary large plaque localized to the distal extremities before puberty, and may be red, violet, blue, yellow, brown or skin-colored. Hyperhidrosis, pain, or hypertrichosis may be apparent, but not in all cases. Enlargement typically occurs commensurate with the growth of the patient.[2] Although it is a benign hamartoma, it is often excised because of pain or enlargement. The majority of reports in the literature are based on Western populations, and very few reports offer information specifically about Asian populations. We reviewed the presentations and courses of fifteen patients with EAH at a medical center in Taiwan over a 16-year period to compare the natural history, management and outcomes between Taiwanese and Western populations.

METHODS

Fifteen patients (9 male, 6 female, age range, birth to 67 years) with EAH whose pathological diagnoses were made at our institution from 1994 to 2010 were identified from the institution's computer database. A retrospective review of the medical records from the database was performed, along with a review of available photographic documentation of the cutaneous lesions. The following parameters were evaluated: sex, age at initial symptoms/signs, age at diagnosis, site involvement, therapy administered, clinical course and outcome. Routine histopathological and immunohistochemical analyses were performed by experienced pathologists. Cases were included in our review if lesions met the histologic criteria for EAH, which we defined as (1) proliferation of normal or dilated eccrine glands. (2) close association of the eccrine structures with capillary angiomatous foci, and (3) the variable presence of pilar, lipomatous, mucinous, and/or lymphatic structures. For immunohistochemistry, the antibodies used were carcinoembryonic antigen (CEA), S-100 protein, epithelial membrane antigen (EMA) and cytokeratins (CK1, Cam 5.2). All patients underwent further surveys after the pathological diagnosis of EAH, including a comprehensive history, physical examination and ultrasonography performed in one patient (case 4).

RESULTS

The detailed clinical and pathological features are listed in Table 1.

Clinical findings

Nine of the fifteen patients were male and 6 were female (male to female ratio = 3:2). The mean age at the time of diagnosis was 38.6 years (range, birth to 67 years), and 3 cases had early onset before 18 years. The associated symptoms and signs, if any, were pain (9/15, 60%), hypertrichosis (2/15, 13.3%), itching (2/15, 13.3%) and hyperhidrosis (1/15, 6.7%). One patient (case 4) had both hyperhidrosis and hypertrichosis, and another both pain and hypertrichosis (case 13). The lesions mostly manifested as a solitary nodule or plaque (Fig. 1). Two or more skin lesions were found in three patients (cases 2, 13 and 14). The lesions were present on the lower limbs (9/15, 60%), face (3/15, 20%), chest (1/15, 6.7%), neck (1/15, 6.7%) and upper limbs (1/15, 6.7%). Misdiagnosis of the lesions before biopsy or excision was noted in all patients. Ultrasonography performed in one infant revealed a strong increase in blood flow (case 4).

Histopathology

Microscopically, all cases showed typical features of EAH (Fig. 2). The lesions were well demarcated but not encapsulated and in the middle or deep dermis. They showed a proliferation of fully developed eccrine sweat glands of normal appearance with duct formation and abnormally abundant vascular (generally capillary) structures that were occasionally dilated. Additional pathological findings included hemangioma (proliferation of endothelial cells and capillaries grouped in lobules in the upper and deep dermis) (case 4, Fig. 2A and case 13) (2/15, 13.3%), verrucous hemangioma (verrucous changes in the epidermis and numerous dilated capillaries in the papillary dermis) (case 9, Fig. 2D) (1/15, 6.7%), arteriovenous malformation (thin-walled vessels and large abnormally thickened vessels in the dermis and subcutis) (case 14) (1/15, 6.7%), and angiokeratoma

Table 1A. Clinicopathologic Data of Fifteen Patients with Eccrine Angiomatous Hamartoma

Pt No.	Sex	age at diagnosis	duration of lesion	location	size (cm)	symptom(s)	No. of lesions	clinical features	clinical diagnosis	pathological diagnosis	treatment	treatment response
1	M	34	1 y	right thigh	1.5 x 1	itching	1	violaceous indurated plaque	dermatofibroma	EAH	excision	recurrence after 4 months
2	M	32	years	right shin	0.7 x 0.7	asymptomatic	multiple lesions	Brown to blackish nodule	skin appendage tumor	EAH with angiokeratoma	excision	NR
3	F	67	years	buttock	1 x 2	pain	1	large brownish plaque	deep fungal infection	EAH	excision	NR
4	F	1	since birth	left thigh	12 x 11	Hypertrichosis, hyperhidrosis, itching	1	gradual enlarging palm sized reddish infiltrative plaque with prominent superficial telangiectasias	infantile hemangioma	EAH with capillary hemangioma	excision	echo showed strong blood flow; NR
5	M	1	since birth	left forearm	2.5 x 2	asymptomatic	1	red to violaceous dermal plaque	skin appendage tumor	EAH	observation	
6	M	48	years	forehead	4 x 3	pain	1	subcutaneous mass with superficial telangiectasias	fibrolipoma	EAH	excision	NR
7	M	39	years	right neck	1.5 x 1	pain	1	excoriated erythematous plaque	chronic dermatitis	EAH with chronic dermatitis	excision	NR
8	F	62	6 mo	Left cheek	1.5 x 1	pain	1	Erythematous to violaceous nodule	skin appandageal tumor	EAH	observation	
9	F	54	years	buttock	1.5 x 1	pain	1	erythematous hyperkeratotic plaques	verrucous hemangioma	EAH with verrucous hemangioma	excision	recurrence after 2 years
10	M	50	3 mo	chest wall	1.5 x 1	pain	1	indurated plaque	dermatofibroma	EAH	excision	NR
11	M	15	years	right knee	1.5 x 1.5	asymptomatic	1	brownish plaque	lipoma	EAH	excision	NR
12	F	49	years	nose	1.5 x 1.0	asymptomatic	1	dull reddish plaque	discoid lupus erythematosus	EAH	observation	
13	M	3	1 y	right leg	1 x 1	hypertrichosis pain	2	firm bluish subcutaneous nodules, formed after trauma	pilomatricoma	EAH with hemangioma	observation	
14	M	49	2 yrs	left ankle	0.7 x 0.5	pain	multiple	several blanchable dermal papules	arteriovenous hemangioma	EAH with arteriovenous malformation	excision	NR
15	F	75	years	left buttock	2.5 x 1	pain	1	hyperkeratotic brownish plaque	chronic eczema	EAH	excision	NR

Abbreviations: F: female; M:male; EAH: eccrine angiomatous hamartoma; NR: non-recurrence

Table 1B. Comparison of Clinical Findings between Fifteen Patients with Eccrine Angiomatous Hamartoma

case no.	Age/ sex	Symptoms/ Signs					Location		Comments
		itching	pain	hypertrichosis	hyperhidrosis	extremities*	trunk	head and neck	
1	34/M	Y				Y (lower)			Single lesion; excised; recurrence after 4 months
2	32/M					Y (lower)			Multiple lesions; excised; histologically showed association with angiokeratoma
3	67/F	Y				Y (lower)			Single lesion; excised
4	1/F	Y		Y	Y	Y (lower)			Single lesion; enlarged during growth; excised; histologically showed association with capillary hemangioma
5	1/M					Y (upper)			Single lesion; observed
6	48/M	Y						Y	Single lesion; excised
7	39/M	Y						Y	Single lesion; excised
8	62/F	Y						Y	Single lesion; observed
9	54/F	Y				Y (lower)			Single lesion; excised; recurrence after 2 years; histologically showed association with verrucous hemangioma
10	50/M	Y					Y		Single lesion; excised
11	15/M					Y (lower)			Single lesion; excised
12	49/F							Y	Single lesion; observed
13	3/M		Y	Y		Y (lower)			2 lesions; observed; histologically showed association with hemangioma
14	49/M		Y			Y (lower)			Multiple lesions; excised; histologically showed association with arteriovenous malformation
15	75/F		Y	Y (lower)					Single lesion; excised
	(M:F = 3:2)	2/15 (13.3%)	9/15 (60%)	2/15 (13.3%)	1/15 (6.7%)	10/15 (66.7%)	1/15 (6.7%)	4/15 (26.7%)	

Abbreviations: Y: yes; *: Lower Extremities, defined here to include buttocks, legs and feet.

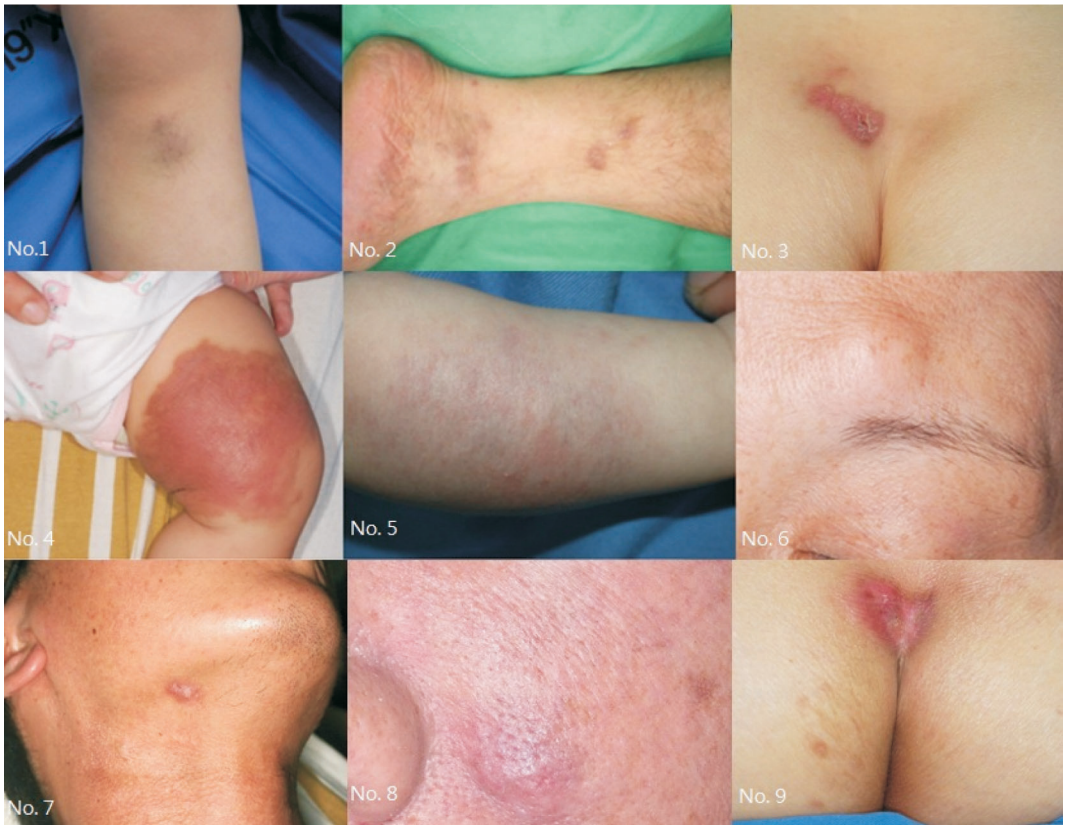

Fig. 1 Clinical photos of eccrine angiomatous hamartomas.

(ectatic, thin-walled vascular spaces in the papillary dermis associated with a hyperplastic epidermis) (case 2) (1/15, 6.7%). In all patients, the epidermis appeared unremarkable except for mild hyperkeratosis and acanthosis, whereas the pilosebaceous follicles were unaffected. No cytological atypia or atypical mitosis was noted.

Immunohistochemistry

The immunohistochemical findings were similar to those observed in normal eccrine glands; thus, anti-CEA antiserum labeled both the secretory and ductal parts of the glands, anti-S-100 antiserum labeled the secretory part only, anti-EMA mAb labeled the secretory part intensely and the ductal part weakly, anti-cytokeratin CK1 labeled the ductal part only, and Cam 5.2 labeled the secretory part only.

Treatment and clinical response

Treatment and clinical responses are summarized in Table 1A. No spontaneous remission was noted in any of our patients who did not have treatment. One patient (case 4, one-year-old girl) had an excision under general anesthesia, because of progressive enlargement of her lesion and her young age. Ten patients (cases 1-3, 6, 7, 9-11, 14, 15) received excision of their lesions with local anesthesia. The surgical technique applied in our study was simple excision of the tumor with a clinically visible tumor-free margin. No other adjuvant therapy was applied after the operation. Tumor recurrences were noted in two out of the eleven patients after excision. The remaining four patients (cases 5, 8, 12, 13) chose to be kept under observation.

DISCUSSION

The term EAH was coined by Hyman and coworkers in 1968,[3] although EAH was first described by Lotzbeck in 1859 as an angiomatous-appearing lesion on the cheek of a child.[4] Clinically,

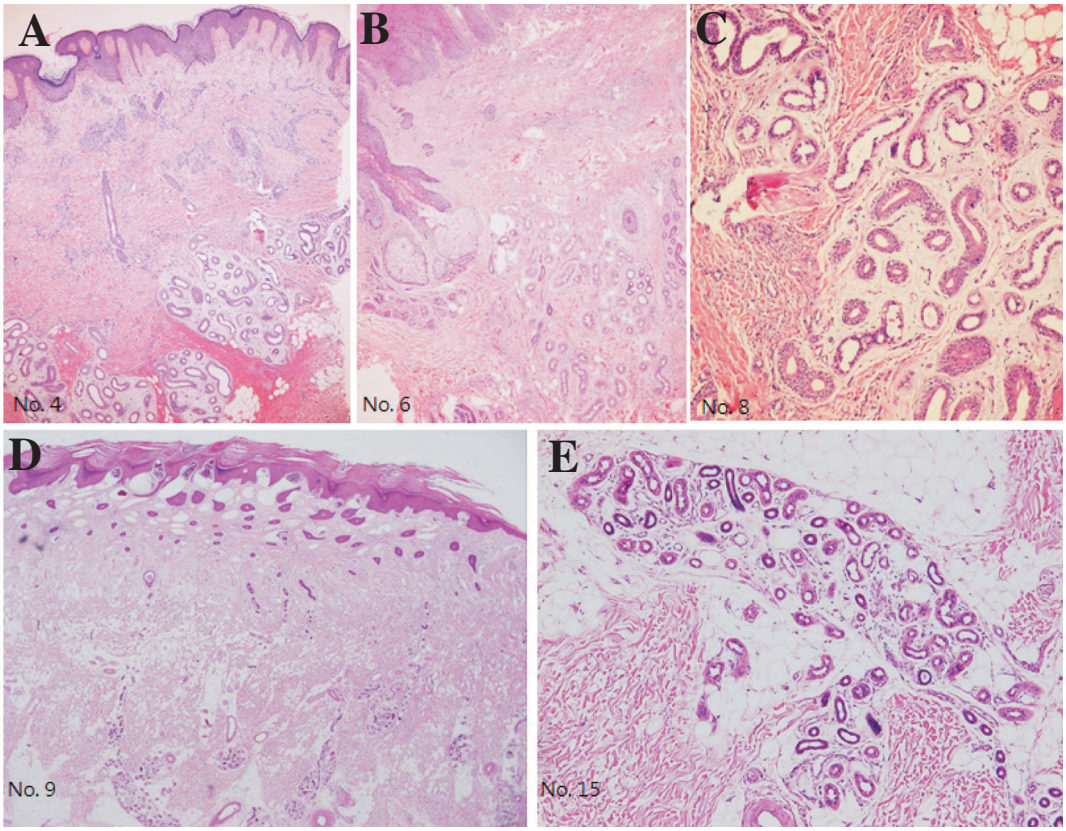

Fig. 2 Representative histopathological features of skin -biopsy specimens from the lesions. (A): Proliferation of endothelial cells and capillaries in the superficial and deep dermis. Blood vessels vary in size from small lumina lined with endothelial cells to dilated vessels lined with flattened endothelium. (case 4) (H & E, x 40). (B): Increased numbers of large eccrine sweat coils and adjacent clusters of vessels in the mid and deep dermis (case 6) (H & E, x 40). (C): The vascular component is clearly predominant, with knots of proliferating blood vessels interspersed with glandular structures (case 8) (H & E, x 200). (D): The overlying epidermis shows acanthosis, papillomatosis and hyperkeratosis. An increased number of dilated capillaries in the upper and dermis, and eccrine structures with intermingled vascular channels in the deep dermis (case 9) (H & E, x 40). (E): Proliferation of fully developed eccrine sweat glands of normal appearance, together with duct formation and abnormally abundant vascular structures (case 15) (H & E, x 100).

EAH is a heterogeneous entity. It consists of a benign skin hamartoma defined by the histologic presence of proliferative eccrine and vascular structures, generally capillaries, located at the dermal-subcutaneous level. Although the origin of EAH is uncertain, the congenital form is thought to involve a defective biochemical interaction between the differentiated epithelium and the underlying mesenchyme during early organogenesis, leading to a proliferation of abnormal adnexal and vascular structures.[2] Late onset lesions are related to recurrent trauma.[5,6]

EAH is an uncommon entity, but its exact incidence is unknown. We herein described fifteen cases of EAH diagnosed over a period of sixteen years in a medical center in Taiwan.

EAH typically manifests as a single, flesh-colored, blue-brown, or reddish papule, plaque, or nodule, although uncommonly, multiple lesions or hyperkeratotic, verrucous variants are also seen.[7-9] It may be located in any part of the body, although it is more frequently found on the extremities, especially localized to the palmoplantar regions or the lower limbs. The face, neck, abdomen, sacral region and back may also be involved.[7,9-19] In accordance with literature findings, twelve of our fifteen patients presented with single lesions (80%), and there was also

a clear predilection of EAH localized to the extremities (66.7%, 10/15 patients).

No gender predilection has been noted in the literature, however a male predominance was noted in our study (nine males and six females, M:F = 3:2). Lesions typically occur at birth (such as cases 4 and 5) and later in childhood or in young adults (such as cases 11 and 13).[2,10,12-21] The occurrence of EAH lesions reported in previous studies was less common in older adults.[18,22,23] However, in our study, eleven out of the fifteen patients were 32 years old or older.

Individual EAH lesions have been reported to range from 3 mm to 11 cm.[1,2,18,19] Thirteen of our fifteen patients had red to brownish, violaceous or blue-reddish round papules or plaques from 0.5 cm to 2.5 cm in diameter, while the other two patients (cases 4 and 6) presented with an uncommon appearance of flesh-colored or large blue-reddish tumors from 4 cm to 12 cm in diameter with the surfaces covered by prominent telangiectasias. Ultrasonography was performed in patient 4, who had the largest lesion (12 x 11 cm in size) in our case series, and the image revealed a strong increase in blood flow. Of note, Larralde et al. reported that such tumoral EAH patients presented with locally increased temperature at palpation that was absent in other patients.[1] Local hypertrichosis is another feature of EAH, and it was found in two children in our study (13.3%, cases 4 and 13).

EAH is usually asymptomatic, although pain and hyperhidrosis have been reported to occur in 42% and 34% of cases, respectively.[19] The discomfort of EAH is thought to be due to small infiltrating nerves, as revealed by electron microscopy, and local hyperhidrosis as a manifestation of the eccrine component.[1,2,19,24,25] Four of our fifteen patients presented with asymptomatic EAH (26.7%), while nine patients manifested mild to severe pain or tenderness (60%). Patient 4 had sweating at the lesion site (6.7%), and patients 1 and 4 complained of itching at the lesion site (13.3%). The associated findings of EAH such as enlargement of the lesion, hypertrichosis, hyperhidrosis, pain or itching can provide valuable diagnostic clues that help differentiate EAH clinically from vascular malformations, hamartomas, hemangiomas, tufted angiomas, smooth muscle hamartomas, glomus tumor, and blue rubber bleb nevus.

Our study revealed several clinical characteristics of EAH which were different from those in the study by Martinelli *et al*., which had the largest number of cases (n = 42) in our literature review (Table 2).[19] Consistent with previous studies, there were no differences in clinical features between genders and onset of age in our study. The novel findings of our study were fewer congenital cases, more late onset adult cases, a male predilection, a higher rate on the head and neck area (4/15, 26.7%) and itching as a symptom. Whether these discrepancies occurred because of our small sample size or ethnic differences between study subjects remains to be elucidated. We also found that all of our patients were misdi-

Table 2. Clinical Features of Our Case Series and Reported Cases of Eccrine Angiomatous Hamartoma

	Clinical feature of the reported cases* (n = 42)	Our case series (n = 15)
Congenital, No. (%)	18† (45.0)	2 (13.3)
Mean age in noncongenital cases (Range)	21.3 y (2 mo-73 y)	44.38 y (2 y-75 y)
Gender		
Male, No. (%)	20 (47.6)	9 (60)
Female, No. (%)	22 (52.4)	6 (40)
M: F ratio	1:1.1	3:2
No. of lesions, No. (%)		
Single	31 (73.8)	12 (80)
Multiple	11 (26.2)	3 (20)
Location of lesions, No. (%)		
Limited to extremities	31 (73.8)	10 (66.7)
Limited to trunk	5 (11.9)	1 (6.7)
Limited to head and neck	0	4 (26.7)
Extremities and trunk or neck	6 (14.3)	0
Symptoms, No. (%)		
Pain	17‡ (41.5)	9 (60)
Hyperhidrosis	13‡ (31.7)	1 (6.7)
Pain and hyperhidrosis	7† (17.1)	0
Hypertrichosis	0	2 (13.3)
Itching	0	2 (13.3)

*: Information from reference 17; †: Information of two cases were not provided in reference 17; N = 40; ‡: Information of one case was not provided in reference 17; N = 41.

agnosed initially, which was likely due to the rarity of EAH and consequentially, a lack of familiarity with this lesion by physicians.

Microscopically, EAH is generally seated in the middle or deep dermis. It is well demarcated but not encapsulated. Histologic criteria for EAH are the proliferation of normal to enlarged or dilated eccrine glandular structures, interspersing vasculature (generally thin-walled capillaries), and the variable presence of pilar, lipomatous, mucinous, and lymphatic structures, apocrine glands and rarely, bony structures.[1,2,25-28] In our study, patients were only included if their lesions fulfilled the histologic criteria for EAH (Fig. 2). No cytological atypia or atypical mitosis was disclosed. Smith *et al*. proposed the existence of three histological variants of EAH, namely follicular, lipomatous and mucinous.[28] Some authors have also reported epidermal alterations, but in the present study there were none except for mild hyperkeratosis and hyperplasia. Immunohistochemically, the labeling of EAH lesions in this study with antibodies to CEA, S-100 protein, EMA and cytokeratins was similar to that observed in normal eccrine glands, as has been reported previously.[2,14,18,28] Eccrine nevus and sudoriparous angioma may show similar histopathological findings. In the former, hyperplastic eccrine glands are not associated with a proliferation of capillaries. In the latter, there is a predominant angiomatous component, large caliber capillaries, and dilated but not hyperplastic eccrine elements. EAH has been reported in combination with other vascular tumors in only a few instances, such as arteriovenous malformation, spindle cell hemangioma, verrucous hemangioma, or overlying verrucous hemangioma-like features.[5,14,29,30-33] It is interesting that in our cases we found additional pathological findings including hemangioma (13.3%), verrucous hemangioma (6.7%), arteriovenous malformation (6.7%), and angiokeratoma (6.7%). To the best of our knowledge, no previous studies have reported the coexistence of EAH and other vascular tumors. These cases exhibited additional components of vascular tumors or arteriovenous malformation that added to the spectrum of histologic findings seen in this entity and also corroborated its hamartomatous nature.

The natural course of EAH is that of enlargement commensurate with the growth of the patient.[2] Rapid growth or increase in lesional pain may occur, and some authors have regarded this as being most likely in response to hormonal stimulation because of onset or exacerbation during puberty or pregnancy.[1,2,9,14,29,34] Others have suggested that lesional pain may be caused by fluid retention associated with menstruation and pregnancy.[34] Only one instance of multiple EAHs with partial spontaneous regression of one of the lesions has been reported.[35] In our study, there was no spontaneous regression of lesions before excision. In general, EAH has no malignant potential and no tendency to regress spontaneously. Thus, although it usually does not warrant treatment, surgical excision is the current treatment of choice for patients with cosmetic concerns, progressive enlargement of the lesions, or undesirable symptoms, such as disabling pain or excessive hyperhidrosis. Deep excision with full-thickness grafting, or amputation of a finger or toe may be required for symptom control in those with larger lesions on acral parts.[34,36] Pulsed-dye laser and Nd: YAG laser treatment have been performed without much success.[16,37] Botulinum toxin might be considered in hyperhidrotic cases.[38]

Laeng *et al*. emphasized the importance of preoperative imaging to evaluate the size, shape, and vascular supply of the lesion to be removed.[29] Ultrasonography allows detection of a vascular lesion (as in case 4), and magnetic resonance imaging sequences may provide more information about the precise dimensions and relationship with surrounding structures. Imaging may be an invaluably helpful tool, especially for large tumors.

Local surgical excision was successful in most cases reported in previous studies. In our study, 11 patients received surgical treatment and 4 patients continued to follow up at our department. However, tumor recurrence was noted in cases 1 and 9 (2/11, 18.2%). This might have been due to regrowth of residual tumor (residual tumor recurrence) or growth of a new independent tumor (new tumor recurrence). Since EAH is benign, aggressive treatment should be avoided.

In our study, symptoms such as pain or itching occurred more often, there were variable additional histopathological findings, the recurrence risk after surgical excision was increased, and acral distribution was less than reported previously in the literature, but the reasons remain elusive; Whether these represent true ethnic differences remains to be answered. The small number of cases with this

uncommon disease requires that collaborative research be conducted to better characterize the disease presentations in Asian populations.

In conclusion, we herein report fifteen cases of EAH and compared the baseline, clinical, and histopathological findings and outcomes with those reported previously in the English language literature, and emphasize the need for histopathologic evaluation of lesions with a similar appearance.

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小汗腺血管缺陷腫：15 個案例之回顧

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背 景： 小汗腺血管缺陷瘤爲一罕見良性之小汗腺及小血管之增生。主要位於中層至深層眞皮，其中偶見其他組織。其臨床表現多樣。迄今除少數病例報告及文獻回顧外，較無大規模之病例研究。在此，我們分析患有小汗腺血管缺陷瘤的台灣人之人口統計、臨床、病理發現及追蹤結果；同時比較以往之病例文獻。

方 法： 我們回顧過去 16 年來 (1994-2010) 在本院經切片證實爲小汗腺血管缺陷瘤之病患之病歷紀錄及病理切片變化。

結 果： 我們共蒐集了 15 名組織病理檢查診斷爲小汗腺血管缺陷瘤之病患。年齡分布廣泛，平均年齡 38.6 歲 (從出生～67 歲)；結果顯示男性病例較多，男女比 3:2。多數患者臨床呈現肉色、紅色、紫色、深藍色位於下肢之單一結節或斑塊；其中有三名病患有多於兩個以上的病灶。此外，同時合併之症狀如疼痛 (60%)、多汗 (6.7%)、多毛 (13.3%)、癢感 (13.3%)。病理變化除典型之小汗腺血管缺陷瘤樣變化外，另可見兩名病患同時有血管瘤 (13.3%)，一名病患有疣狀血管瘤 (6.7%)，一名病患有動靜脈畸形 (6.7%)，一名病患有血管角化瘤 (6.7%)。11 名患者接受手術切除治療，其中有兩名患者有復發之情形。這些患者術前或其他僅接受觀察追蹤之病患並未發現病灶有自行緩解之情形。

結 論： 小汗腺血管缺陷瘤臨床雖具多變的臨床症狀與表徵，仍爲一良性但少見之病灶，治療不宜過於侵略性，針對症狀難以忍受或單發性之病灶，通常予以切除完整病灶即可。與西方文獻相比，本研究中病人的病灶較常併有其他病理變化，且術後之再發機率較高，此發現仍須透過日後跨醫學中心、更大樣本數來證實。

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關鍵詞： 小汗腺血管缺陷瘤，汗腺腫瘤，錯誤瘤，血管瘤，超音波

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