Localized eruptive acquired multiple angiokeratomas with spontaneous resolution in a healthy child

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Key words: angiokeratoma; child; hand.

INTRODUCTION

Angiokeratomas are rare vascular skin lesions caused by the dilatation of blood vessels and skin hyperkeratosis. There are following 5 types of angiokeratomas: (1) solitary angiokeratomas; (2) angiokeratomas of the scrotum and vulva; (3) angiokeratoma corporis diffusum; (4) angiokeratoma of Mibelli; and (5) angiokeratoma circumscriptum. Angiokeratomas are caused by the dilatation of preexisting papillary blood vessels, except for the angiokeratoma circumscriptum subtype, which is caused by the malformation of capillary or capillary and lymphatic vessels.[1] We report a case of acquired eruptive angiokeratomas in a previously healthy child.

CASE REPORT

A 7-year-old healthy girl presented to the dermatology outpatient clinic with a 2-week history of asymptomatic skin lesions over her right hand and wrist, with rapid progression in the number and size of the lesions. She had no previous medical problems, no fever or other symptoms, no history of physical trauma, and no contact with animals.

A dermatologic examination revealed multiple hyperkeratotic erythematous and dark blue nodules of varying sizes. The lesions were distributed over the palmar and dorsal aspects of the right hand, extending to the right wrist. The size of the lesions ranged from 2 × 2 mm to a maximum size of 1 × 1 cm in diameter (Fig 1).

Our differential diagnoses at presentation included acquired angiokeratoma or other vascular tumors, acral pseudolymphomatous angiokeratoma of children, bacillary angiomatosis, Kaposi sarcoma, or giant molluscum contagiosum. A skin biopsy was performed on one of the well-formed lesions during the initial visit. This biopsy showed vascular ectasia of the papillary dermis extending into the epidermis with hyperkeratosis, acanthosis, and elongation of the rete ridges, with the epidermis encircling the dilated vascular spaces as shown in Fig 2. The periodic acid—Schiff and Grocott-Gomori's methenamine silver stains were negative for microorganisms. These findings were consistent with angiokeratomas.

Blood investigations included a complete blood cell count, liver function test, renal function test, coagulation profile, and HIV screening, all of which were normal. Initially, the condition progressed in lesion number and the size of the lesions, with bleeding secondary to the avulsion of one lesion with regrowth at the same site. She felt tenderness at the base of the thumb, involving the first metacarpophalangeal joint, which was relieved by the intake of oral analgesics.

At subsequent follow-up visits, she still had the same previous lesions without progression. Cryotherapy was offered, but her parent preferred observation with no intervention, given the stability of the disease. Almost 95% of the lesions resolved spontaneously within 3 months of presentation. One

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Funding sources: None.

IRB approval status: Not applicable.

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JAAD Case Reports 2022;28:90-3.
2352-5126

https://doi.org/10.1016/j.jdcr.2022.07.046

Fig 1. Multiple angiokeratomas over the palmar (**A**) and (**B**) dorsal aspects of the right hand.

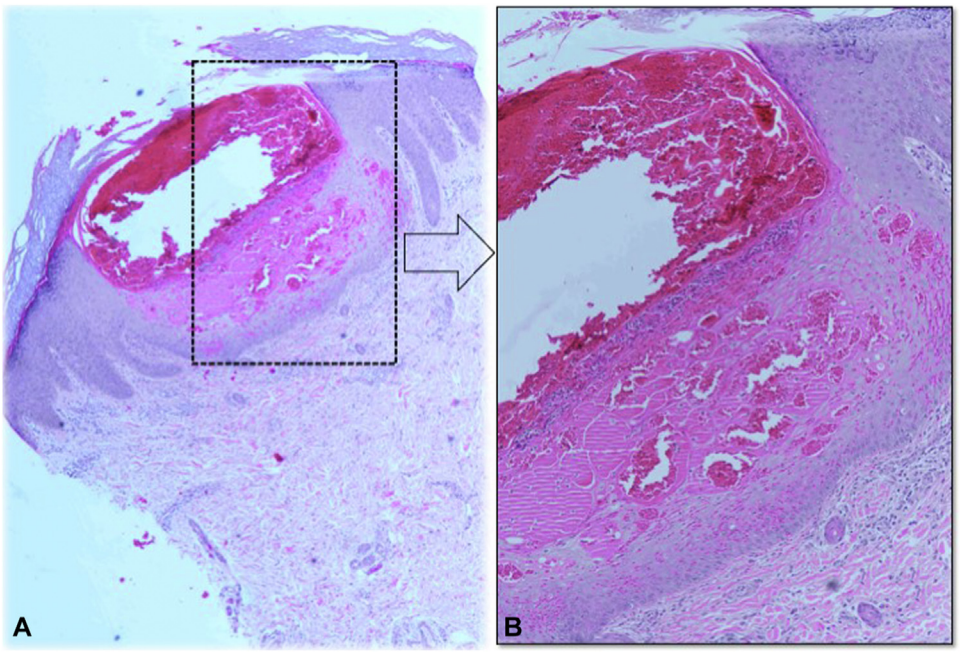

Fig 2. Histopathology (hematoxylin-eosin) showing vascular ectasia of the papillary dermis, extending into the epidermis with hyperkeratosis, acanthosis, and elongation of the rete ridges with epidermis encircling the dilated vascular spaces.

small papule remained, which was treated with one session of cryotherapy. The lesions resolved completely with no recurrence after a 1-year follow-up, as shown in Fig 3.

DISCUSSION

Angiokeratoma is a vascular tumor that usually occurs as a solitary lesion.[2] The pathogenesis of angiokeratoma remains unclear. Various factors have

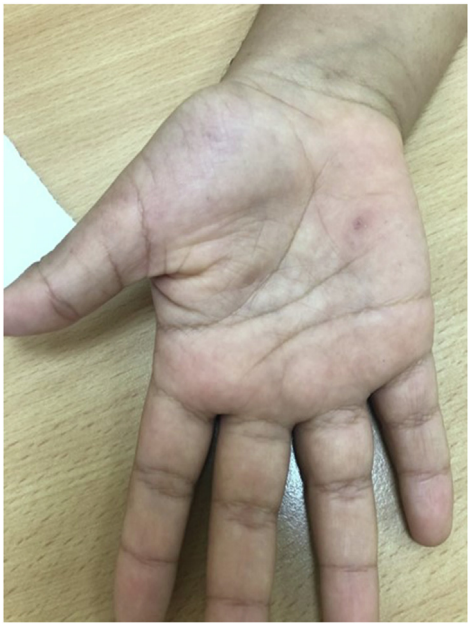

Fig 3. Complete clearance of lesions over the right hand after 1 year.

been considered to be the cause of angiokeratoma, such as congenital factors, nevoid or vascular malformation, tissue asphyxia, acute or chronic trauma, high venous pressure, chilblains, or pregnancy.[3,4] For the patient, in this case, multiple localized angiokeratomas erupted rapidly over a short period with an unknown trigger. Acral pseudolymphomatous angiokeratoma of children may appear similarly to eruptive angiokeratomas, but the 2 entities are distinguished by histopathology. Acral pseudolymphomatous angiokeratoma presents histologically with dense inflammatory infiltrates that must be differentiated from true lymphoma by the immunohistochemical study.[5] Here, acral pseudolymphomatous angiokeratoma of children was excluded by the typical changes of angiokeratoma, as seen in the histopathology.

Nearly all the known types of angiokeratomas are permanent and tend not to regress spontaneously.[2] Usually, they need surgical intervention, such as electrocautery, cryotherapy, surgical excision, or laser treatment.[1] The presented case is rare because the patient developed eruptive lesions without provoking factors and regressed spontaneously. The majority of reported cases with the same condition were adults who had preceding potential triggers, such as trauma, surgery, radiation, or drug-induced lesions, and nearly all of the previously reported cases required an intervention (Table I).[2,6-9]

This case report is an interesting occurrence of rapidly eruptive multiple angiokeratomas, localized

Table I. Summary of reported cases of multiple eruptive angiokeratomas

Ref.	Age (y)	Sex	Clinical presentation	Location	Triggering factor	Speed of onset	Treatment	Resolution
6	56	M	Multiple cutaneous and cerebral hemangiomas with angiokeratomas	Legs, thighs, abdomen, and chest	Unknown	Eruptive	Infrared coagulator, pulsed dye laser, curettage and cautery, intralesional interferon α-2a.	Not much benefit with reoccurrence
7	51	F	Multiple angiokeratomas	Left hand	Unknown	Eruptive	Electrocautery and laser therapy	Complete resolution
2	84	F	Eruptive overspread angiokeratomas	Hands, legs, oral mucosa	Drug, enoxaparin	Eruptive, 10 days after enoxaparin	Cessation of enoxaparin, prednisone 50 mg for 3 d	Complete resolution
8	56	M	Localized angiokeratoma	Left malar region	Unknown	Eruptive	Offered electrocoagulation	-
9	21	F	Multiple angiokeratomas	Flanks	Unknown	Eruptive	Oral and topical β-blockers	Angiokeratomas maintained at a smaller size

over the right hand in a previously healthy child. To the best of our knowledge, there are no previous reports of localized acquired multiple angiokeratomas in a healthy child with spontaneous resolution. The other interesting finding in this patient was that the lesions resolved completely over 3 months without intervention. There is great importance to the early recognition and diagnosis of unusual skin lesions, to rule out a more serious differential diagnosis, such as Kaposi sarcoma, malignant melanoma, and bacillary angiomatosis, which all require early diagnosis and intervention to avoid serious consequences which could be avoided through early identification.

Conflicts of interest

None disclosed.

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