

CASE REPORT

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Angiolymphoid hyperplasia with eosinophilia in a hand treated with a reverse digital island flap and artificial skin: a case report

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Abstract

Background: Angiolymphoid hyperplasia with eosinophilia is a rare nodular skin tumor characterized by eosinophilic invasion and vascular proliferation. Previous reports suggested that irritation and inflammation are the causative factors of this disease. Most cases of angiolymphoid hyperplasia with eosinophilia occur around the auricle, forehead, and scalp; the hand is rarely affected. Moreover, the tumor seldom presents as multiple nodules.

Case presentation: A 67-year-old Japanese woman presented with a complaint of skin masses on her left thumb and index finger, which had gradually grown in size over the past few months. A biopsy was performed confirming a diagnosis of angiolymphoid hyperplasia with eosinophilia. The aponeurosis on her index finger was resected and tissue was reconstructed using a reverse palmar digital island flap harvested from the base of her index finger. The thumb lesion was also resected and covered with collagen-based artificial skin. Gradual progression of skin epithelialization followed by healing was noted 2 months after the surgery.

Conclusion: Angiolymphoid hyperplasia with eosinophilia is a rare tumor; it is seldom seen in the hands. It is generally treated by surgical resection. It is important to resect a sufficiently large area of the tissue due to the possibility of relapse in some cases. Furthermore, appropriate reconstruction is mandatory after wide margin tumor resection.

Keywords: Angiolymphoid hyperplasia, Eosinophilia, Reverse palmar digital island flap, Collagen-based artificial skin

Introduction

Angiolymphoid hyperplasia with eosinophilia (ALHE) was first reported by Wells and Whimster in 1969 [1]. It is a rare skin tumor that occurs around the auricle, forehead, scalp, shoulder, chest, oral mucosa, and scrotum; the hand is rarely affected by this disease. ALHE is a benign tumor that presents with mild symptoms such as itching during the early stages. However, as the tumor grows, severe symptoms such as bleeding or pain may appear. Several studies indicated that the first choice of treatment of ALHE is surgical resection. However, the recurrence rate of this disease is reported to be as high as 41% [2], thus, necessitating the resection of a sufficiently large area in

the affected region to ensure surgical success. Unfortunately, wide resection margins also result in large skin deformities, thereby reducing the daily living activities of the patient. Therefore, reconstruction is an additional important component of ALHE treatment.

Here we present a rare case of ALHE of the hand in a woman.

Case presentation

A 67-year-old Japanese woman presented with complaints of a mass of skin on her left thumb and index finger that had been gradually increasing in size over the preceding few months. Her general condition was good (height, 147 cm; weight, 65 kg; heart rate, 62/minute; blood pressure, 136/72 mmHg, and body temperature, 36.3 °C). No abnormal breath or heart sounds were

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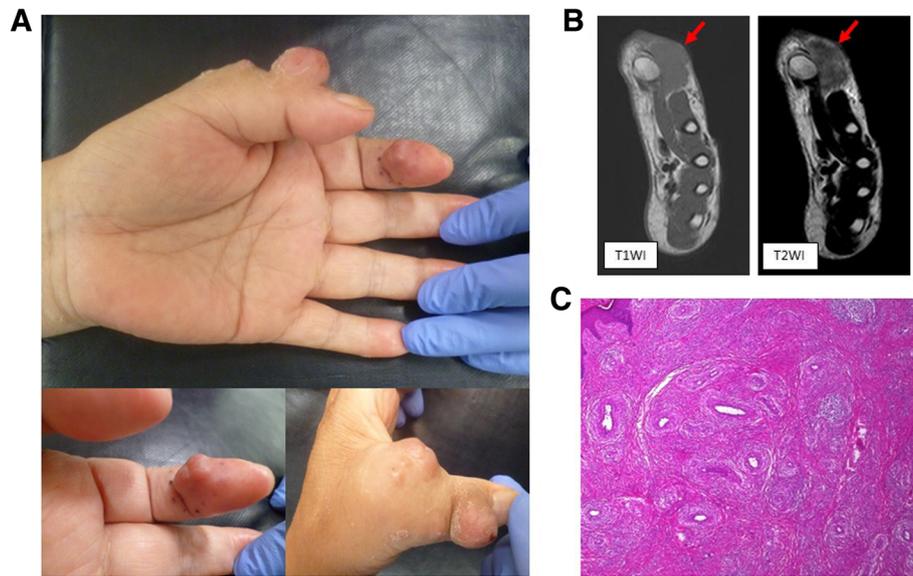


Fig. 1 **a** Skin masses observed on the base of the left thumb and on the volar side of the index finger. **b** Magnetic resonance imaging of the lesion (red arrows) on the dorsal side of the thumb showed isointensity in T1-weighted images and high intensity in T2-weighted images. The tumor was confined to the skin. **c** Increase in the number of blood vessels in the dermal layer and eosinophilic infiltrates were noted around vascular endothelial cells. *T1WI* T1-weighted image, *T2WI* T2-weighted image

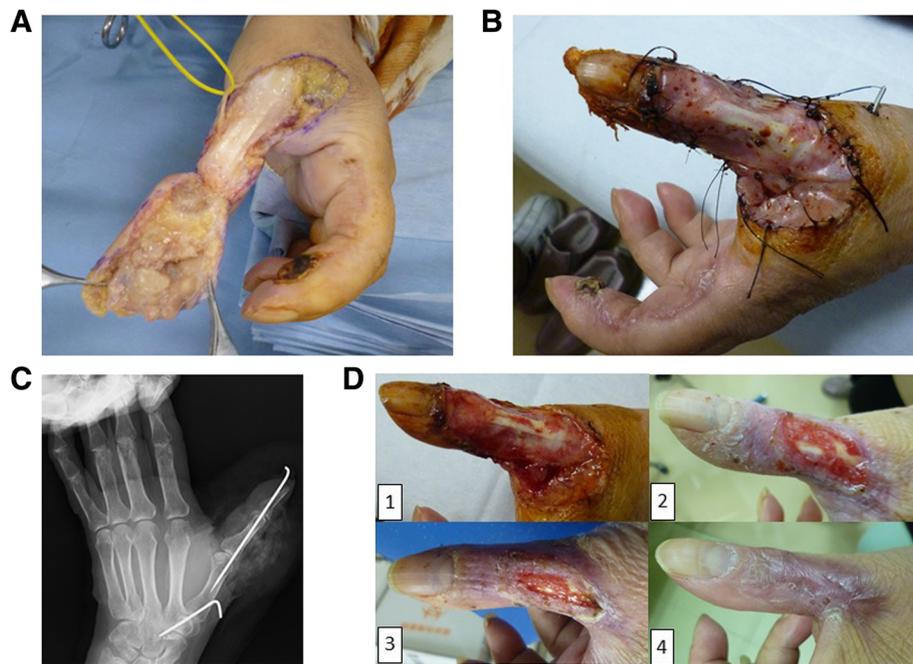


Fig. 2 **a** The thumb lesion was excised along the margin of the tumor along with the aponeurosis. **b** After resection, the defect was covered with artificial skin using collagen. **c** Arthrodesis of the interphalangeal, metacarpophalangeal, and carpometacarpal joints of the thumb were performed using a Kirschner wire. **d** Gradually, epithelialization was noted after a few months

thumb and index finger were surgically excised using the tumor margins and aponeuroses as the resection range (Fig. 2a). As shown in Fig. 2b, the skin defect was covered with collagen-based artificial skin (Pelnac®, Gunze Co. Ltd., Ayabe, Japan). Subsequently, the interphalangeal and metacarpophalangeal joints of her thumb and the carpometacarpal joint were fixed using a Kirschner wire (Fig. 2c). She was prescribed loxoprofen sodium (180 mg/day) for approximately 10 days. The Kirschner wire was removed 2 weeks after the surgical procedure. A gradual progression of the epithelialization of the artificial skin was noted along with healing 2 months post-surgery (Fig. 2d). Mild contracture of the dorsal side of her thumb finger was observed after surgery; however, no hindrances in daily life activities were reported. Importantly, no tumor recurrence was noted at the 12-month follow-up.

Discussion

ALHE is a nodular skin tumor associated with eosinophilia and vascular proliferation. Most cases occur on the auricle, forehead, and scalp, whereas rare cases, such as those involving the hand, account for only 4% of all ALHEs [2]. Table 2 describes the documented cases of ALHE involving hands. ALHE is difficult to cure [3]; the gold standard for treatment of ALHE is complete surgical resection. Due to the possibility of recurrence in some cases, complete resection of the tumor is necessary. Furthermore, functional reconstruction in conjunction with complete resection is essential for the treatment of ALHEs of the hand.

On clinical examination, it is important to differentiate between ALHE and Kimura's disease, using both laboratory and histological tests. Similar to AHLE, Kimura's disease is classified as a type of eosinophilic dermatitis. Although eosinophilia was observed in the blood of the patient in the current report, systemic eosinophilia is generally rare. In addition, ALHE exhibits a slow growth rate and rarely presents with lymphadenopathy (5–20% of cases) [4]. Moreover, unlike in Kimura's disease, the itching sensation is strong in AHLE. In many cases, immunoglobulin levels, such as that of immunoglobulin E, are normal. ALHE often involves neoangiogenesis and, accordingly, presents with plump, epithelioid endothelial cells and arteriovenous shunts [4]. Often, ALHE lesions are located in the dermis. Kimura's disease is probably an allergic or autoimmune response, whereas ALHE is the result of a benign neoplasm of endothelial cells caused by inflammation or stimulation [5]. The cause of ALHE is unknown [6]. Some clinicians speculate that it develops in response to various infections, such as herpes virus 8 or human papillomavirus-6, or injury, such as excessive contact with the rim of eye glasses [7, 8]. Meanwhile, tumor growth is reported to occur in response to changes in hormone levels during pregnancy; however, there are no clear differences between men and women [9–12]. In the present case report, our patient had numerous skin injuries on her fingers other than that affected by the lesion. She reported that she frequently fed wild cats near her home and had received several scratches from the animals in the past, which may be related to the onset of ALHE in this case.

Table 2 Reported instances of angiolymphoid hyperplasia with eosinophilia in the hand

Study	Age	Sex	Lesion site	Number of lesions	Size	Symptoms	Treatment	Outcome	Systemic eosinophilia	Progression period
James M. Swinehart et al., 1979 [20]	24	M	Lt.palm, subcutaneous	single	NA	NA	resection	local recurrence	+	8 months
Arnold M et al., 1999 [21]	20	F	left arm and hand subcutaneous	multiple	5-10 mm	bleeding, pain	resection	no local recurrence, but another site	+	24 months
B. D. Krapohl et al., 2003 [22]	33	F	Rt.palm and ring finger subcutaneous	multiple	NA	discoloration	resection	no recurrence in 3 months	NA	18 months
C Conill et al., 2004 [23]	32	F	Rt.index and middle fingers subcutaneous and bone	multiple	NA	deformation of the nail, pain	radiation therapy	no recurrence in 9 years	-	NA
A. Satpathy, 2005 [24]	11	F	Rt.dorsum of hand subcutaneous	single	20 mm	itching	spontaneous resolution	no recurrence in 12 months	-	a month
H Ozcanli et al., 2007 [25]	42	F	Rt.palm, middle and ring fingers subcutaneous	multiple	30-80 mm	itching, pain, disturbance of sensation	resection, laser treatment	no recurrence in 2 years	+	12 months
Nick Pappas et al., 2010 [26]	18	F	Rt.palm subcutaneous	single	15-10 mm	no pain	resection	no recurrence in 1 year	-	a month
Mohammad M. Al-qattan et al., 2017 [27]	32	F	Lt.palm subcutaneous	single	NA	NA	resection	no recurrence in 1 year	+	6 months

F female, Lt. left, M male, NA not available, Rt. right

Is it necessary to treat benign ALHE tumors?

ALHE does not develop complications during conservative therapy, and no cases of malignant transformation have been reported so far [5]. Yet, ALHE can be exacerbated by surgical stress; hence, refraining from excision without careful consideration is critical. Some cases of ALHE also recognize genetic mutation in T cell. According to Kempf *et al.*, ALHE is not a vascular lesion, but a type of CD4-positive T cell lymphoma [13–15]. Accordingly, careful observation is necessary, even in cases not involving surgical procedures. Early symptoms include itching and a throbbing sensation, but bleeding and pain are only observed as the tumor grows in size. Surgical resection may be indicated in the presence of more serious symptoms, such as pain, and if the patient wishes to undergo resection.

Is there a cure for ALHE other than surgical resection?

In some cases the outcomes of administration of local and systemic corticosteroids to patients with ALHE were unclear. Furthermore, laser treatment has also been provided with unsatisfactory results [16, 17]. These treatments may be effective in elderly people and in situations where surgical procedures are contraindicated (such as in individuals using anticoagulants). The primary treatment choice for ALHE is surgical resection. However, the recurrence rate following surgical excision is high.

What is critical for success in surgical resection?

Pathologically, ALHE involves vascular damage characterized by the presence of an unstable basement membrane and arteriovenous shunts [18, 19]. The removal of these abnormal vessels may be important during surgical resection. Furthermore, difficulties in identifying the margins of these lesions are believed to contribute to their high recurrence rate. Abnormal blood vessels related to ALHE often exist in the dermis; therefore, it is advisable to use natural anatomic barriers, such as the aponeurosis and fascia, as the resection margin. Several clinicians believe that ALHE resection should mirror the resection margin of malignant soft tissue tumors. It is important to examine the preoperative image before surgical treatment. In the present case study, both abnormal vessels and aponeurosis at the bottom of the tumor were excised. Recurrence is less likely when a wide resection with margins involving the dermis is performed. Reconstruction using a skin flap or artificial epithelium is recommended in cases where the deformity is large.

Conclusion

ALHE is a rare, benign skin tumor characterized by the presence of vascular proliferation and eosinophil invasion, and is often treated conservatively. However, when

accompanied by symptoms such as pain, surgical resection of the tumor is required. In such cases, removal of a sufficiently large area of resection is important to ensure a favorable outcome. In addition, adequate reconstruction is necessary to guarantee post-surgical functions.

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Availability of data and materials

All data and materials related to this report are accessible at any time upon request.

Authors' contributions

NO, SN, HT, KH, and RK were involved in patient care, review of literature, and writing of the manuscript. NO, RK, and MA prepared the manuscript. All authors read and approved the final manuscript.

Ethics approval and consent to participate

This case report does not involve any active intervention on patients; therefore, ethics approval is waived.

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Competing interests

The authors declare that they have no competing interests.

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