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Angiolymphoid hyperplasia with eosinophilia involving the common digital artery of the hand: A case report and classification of upper limb lesions

Mohammad M. Al-qattan [a,*], Maha Arafah [b], Felwa Al-Marshad [a]

[a] Division of Plastic Surgery, King Saud University, Riyadh, Saudi Arabia
[b] Department of Pathology, King Saud University, Riyadh, Saudi Arabia

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ABSTRACT

INTRODUCTION: Angiolymphoid hyperplasia with eosinophilia (ALHE) is a rare benign reactive inflammatory lesion. The usual presentation is a single or multiple skin nodules of the head and neck. Involvement of the hand is very rare and there have been no comprehensive reviews on ALHE of the upper limb. In this paper, we report on a case involving the common digital artery of the hand. We also review the literature and offer two classification systems for upper limb lesions: One according to the primary structure involved, and the other according to the presentation with either single or multiple lesions.
PRESENTATION OF CASE: A 32-year old female presented with a slowly growing subcutaneous mass at the second web space of the left hand. The mass was neither tender or mobile. An MRI showed a tri-lobed soft tissue lesion. At the time of surgery, the lesion was found to be within the common digital artery of the second web space. Complete excision was done. Histopathology confirmed the diagnosis of ALHE There has been no recurrence or cold tolerance at final follow-up 1 year later.
DISCUSSION: We offer two classification systems for upper limb lesions: One according to the primary structure involved, and the other according to the presentation with either a single or multiple lesions.
CONCLUSION: A rare case of ALHE of the hand is presented. The literature is reviewed and two classification systems for upper limb lesions are offered and their implications are discussed.

1. Introduction

Angiolymphoid hyperplasia with eosinophilia (ALHE) is a rare proliferative lesion which usually presents with a single or multiple skin nodules in the head and neck. Involvement of the upper limb is extremely rare with only 24 previously reported cases in the world literature [1–24]. In the upper limb, various sites for involvement have been reported such as the skin, the subungual region, the blood vessels, nerves, muscles, and bone [1–24]. As the name implies, the diagnosis is made histologically by the presence of vascular proliferation (angio-hyperplasia), lymphoid follicles (lymphoid hyperplasia) and a prominent eosinophilic infiltrate (eosinophilia). There have been no comprehensive reviews of upper limb lesions.

In this communication, we report on a case of ALHE of the common digital artery of the hand and review previously reported cases. Two classification systems are offered: one according to the primary structure involved and the other according to the presentation with either single or multiple lesions. The latter classification will have an impact on the risk of recurrence. The work has been reported in line with the SCARE criteria [25].

2. Case report

A 32-year old female presented with a slowly growing (over period of 6 months) subcutaneous mass at the second web space of the left hand (Fig. 1). The patient was otherwise healthy with negative family history and was free from medical diseases. The mass was neither tender or mobile. An MRI showed a tri-lobed soft tissue lesion with no tendon or bony involvement (Fig. 2). Surgery was performed by the senior author (MMA). At the time of surgery, the lesion was found to be within the common digital artery of the second web space (Fig. 3). Using a Doppler, there was no flow within the artery. Total excision was done (Fig. 4) by transecting the common digital artery 5 mm proximal to the lesion; and by transecting the radial digital artery of the middle finger and the ulnar digital artery of the index finger 5 mm distal to the lesion. The blood supply to the fingers was adequate and hence no vein graft reconstruction was done. The postoperative recovery was uneventful. Histopathology confirmed the diagnosis of ALHE with vascular proliferation

* Corresponding author at: PO Box 18097, Riyadh 11415, Saudi Arabia.
E-mail address: moqattan@hotmail.com (M.M. Al-qattan).

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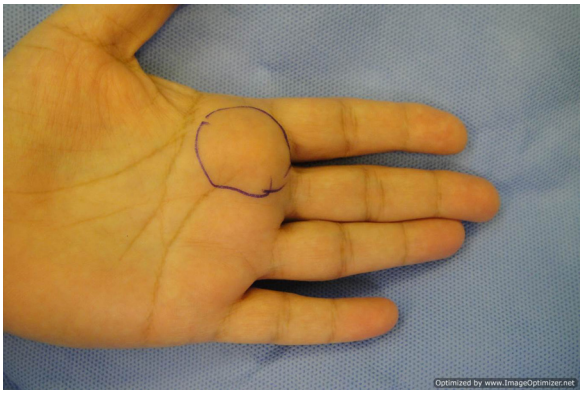

Fig. 1. The lesion.

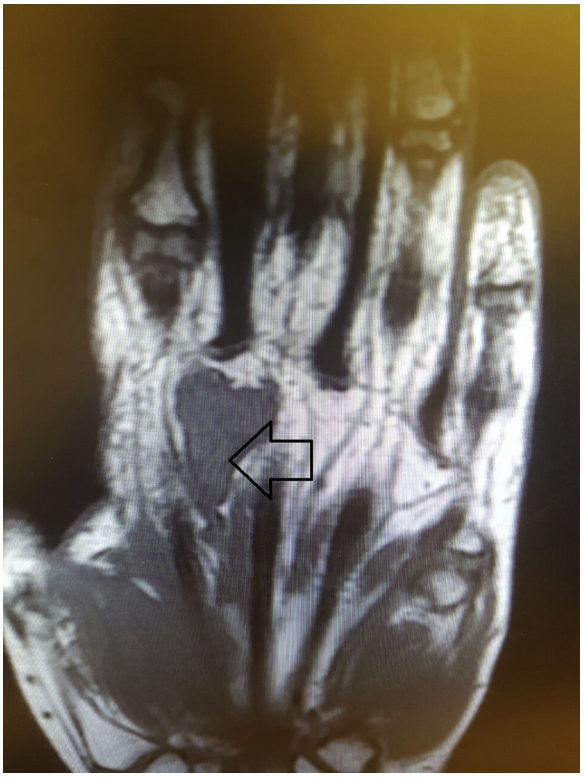

Fig. 2. MRI showing the tri-lobed lesion (arrow) at the second web.

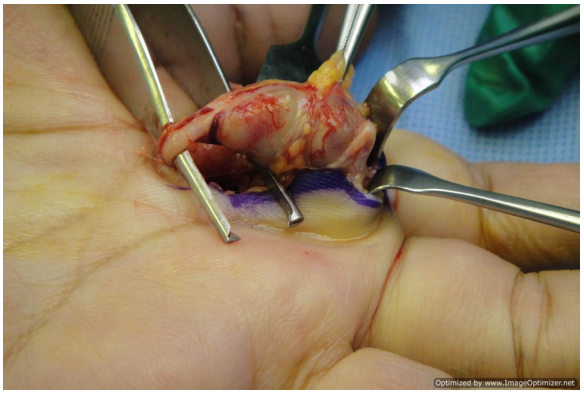

Fig. 3. Intraoperative appearance. The forceps is showing that the normal lumen of the common digital artery has become the bulge of the tumor. In retrospect, the proximal lobe the tumor is the origin from the common digital artery, while the 2 distal lobes represent the extension of the tumor into the digital arteries of the index and middle fingers.

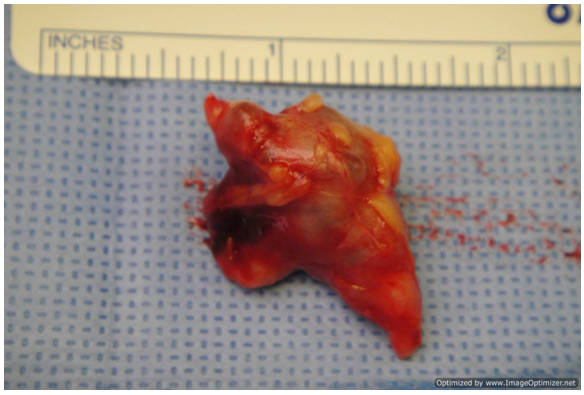

Fig. 4. The excised lesion.

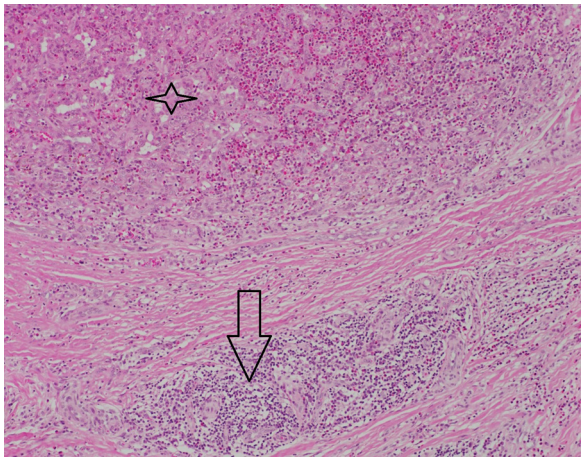

Fig. 5. Low power (x10) showing the vascular proliferation (star) and the lymphoid follicles (arrow). H & E stain.

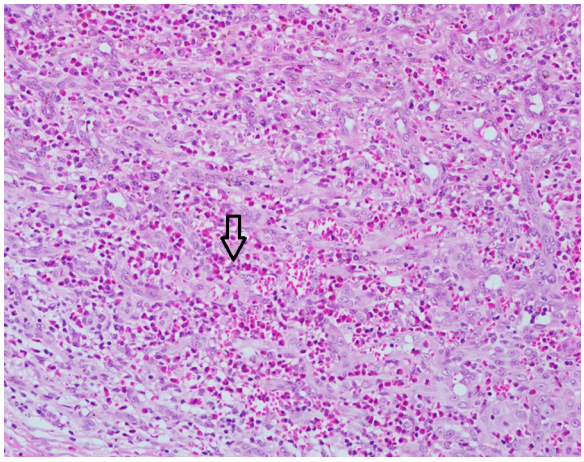

Fig. 6. High power (x40) showing the intense eosinophilic infiltrate (arrow). H & E stain.

within the lumen of the artery, multiple lymphoid follicles, and prominent eosinophilic infiltrate (Figs. 5 and 6). After knowing the diagnosis, we checked the preoperative complete blood count; and there was eosinophilia (10.1%). A repeat postoperative blood test showed resolution of the eosinophilia (1%). Serology testing showed evidence of an old Epstein-Barr virus (EBV) infection (positive IgG viral capsid antigen and positive IgG Epstein-Barr nuclear antigen). The patient recalled being sick with high fever about one

Table 1
Differentiating features between ALHE and Kimura disease.

	ALHE	KIMURA'S DISEASE
Median Age	3rd decade	2nd decade
Sex RaceThe most common presentation	More in females Any race Single or multiple skin lesions of the head and neck (1–2 cm nodules)	More in males More in Orientals poorly-defined skin masses (3 cm) in the head and neck (skin around the ear is the most common site)
Other presentations	Primary involvement of muscle, nerve, bone, and blood vessels have been reported	Salivary gland involvement in 40% of cases, and lymph node involvement in 70%. Nephrotic syndrome may be present
Peripheral Eosinophilia	20% of cases	90% of cases
Serum IgE	Normal	Usually elevated
Histology	Vascular proliferation, lymphoid follicular hyperplasia, prominent eosinophilic infiltrate	Vascular proliferation and eosinophilic infiltrates are minimal. Numerus lymphoid follicles are the main histological features.

month prior to the appearance of the hand lesion. There has been no recurrence or cold tolerance at final follow-up 1 year later.

3. Discussion

ALHE should be considered as benign reactive inflammatory lesion rather than a tumor [2]. In the older literature, the lesion used to be known as epitheloid hemangioma [26] and histiocytoid hemangioma [4,5]. The actual etiology is unknown; although two viral causes have been proposed [2]. The first is herpes virus type 8 (HHV-8) which is also known to cause Kaposi's sarcoma. The other virus is the EBV which is also known to cause infectious mononucleosis, Hodgkin's lymphomas, Burkitt's lymphomas, and nasopharyngeal carcinoma [2]. In our patient, serological testing showed evidence of a previous EBV infection. Another interesting feature in patients with ALHE is the occasional occurrence of peripheral eosinophilia which was also present in our case. The main differential diagnosis is a similar but district disease known as Kimura's disease [2,26]; and the differences between the two entities are summarized in Table 1.

Our review of the literature revealed 24 well documented cases [1–24]. We did not include cases with poor documentation [27]. Based on our review, we propose two classification systems of ALHE of the upper limb. Table 2 shows a classification according to the primary site of involvement. The skin and the arterial system remain to be the most two common sites of involvement. The ALHE lesion in our patient involved the common digital artery and there were two other previously reported cases with common digital artery involvement [1,23]. Involvement of the radial artery [10,20], ulnar artery [8,9,14], brachial artery [2,11], and axillary artery [12,13] have also been reported. Total excision of the involved artery is curative; and reconstruction with vein grafts is only mandatory after axillary and brachial artery excisions. One case involved the ulnar artery at the wrist and extended along the branches of the ulnar artery to the little and ring fingers. Total excision in that case required amputation of the ulnar two rays [8]. Involvement of the venous system is very rare and excision is curative [5,15]. The presentation of subungual lesions mimics the presentation of other subungual hand tumors [1,2,14] and may cause bony erosion of dorsal aspect of the distal phalanx [4,19,24]. One case involved the ulnar nerve requiring excision and nerve grafting without recurrence [21]. One case presented with multiple intramuscular nodules of the trapezius muscle [6]. Primary bone involvement (of the middle phalanx) was only seen in one case.

Table 2
A total of 25 cases of ALHE of the upper limb: Classification according to the site of involvement.

Primary Structure involved	Number of reported cases (specific sites)	References
Skin	10 in the hand, 3 in the forearm, 2 in the arm	1,2,3,4,7,14,16,18,22,23[a], 24
Subungual	a) Lesion under the nail bed without bony erosion (3 cases) b) Lesion under the nail bed with bony erosion (3 cases)	1,2,144,19,24
Artery	A total of 12 cases: axillary artery (n = 2), brachial artery (n = 2), ulnar artery (n = 3), radial artery (n = 2), common digital artery (n = 3)	1,2,8,9,10,11,12,13,14,20,23, current case
Vein	Two cases (antecubital vein, dorsal vein)	5,15
Nerve	One case (ulnar nerve at wrist)	21
Muscle	One case (Trapezius)	6
Bone	One case (middle phalanx)	17

Note that the reference numbers are repeated because several cases had involvement of multiple sites. Also note that skin and arteries are the two most common sites of involvement.

[a] Case 23 is the only case in which there was a concurrent ALHE lesion outside the upper limb (forehead lesion). In the remaining 24 cases, all ALHE lesions were confined to the upper limb.

Table 3
A total of 25 cases of ALHE of the upper limb: recurrence in patients with single versus multiple lesions.

17 patients with single lesions	8 patients with multiple lesions
A) No recurrences in 14 cases treated surgically (references #: 5,7,8,9,10,11,12,13,15,16,19,20,21, current case).	A) No recurrence in one case treated with radiotherapy (reference #24)
B) Spontaneous resolution of a skin lesion post-incisional biopsy (reference #18)	B) One case of failure of radiotherapy to completely treat the skin lesions (reference #4).
C) Two recurrences after surgical excision: One in bone (reference #17) and one in skin (reference #22).	C) One case treated surgically with no follow-up and hence recurrence is unknown (reference #3).
	D) The remaining 5 cases with multiple lesions had recurrence post-surgical excision (references #1,2,6,14,23).

Curettage resulted in a recurrence indicating that total excision is also required in primary bony lesions [17].

Table 3 classifies upper limb ALHE lesions according to whether the initial presentation was with single or multiple lesions. About two thirds of reported cases presented with single lesions. The majority of patients (14 out of 17 cases) with single lesions had no recurrence post-excision. One single skin lesion in an 11-year old girl resolved spontaneously [18]. only two patients presenting with single lesions experienced recurrences; and in both, the recurrences were attributed to incomplete excision [17,22]. In contrast, the majority of patients (five out of 8 cases) presenting with multiple lesions had recurrence after surgical excision [1,2,6,14,23]. Hence, some authors recommended radio-therapy for multiple lesions. In the upper limb, radiotherapy was used in two cases with multi-site involvement. Radiotherapy was curative in one case with skin and subungual lesions [24]. In the second case, radiotherapy was curative for fingertip and subungual lesions; but not for other palmar and dorsal skin lesions of the hand [4].

4. Conclusion

A rare case of ALHE of the hand is presented. The literature is reviewed and two classification systems for upper limb lesions are offered and their implications are discussed.

Conflict of interest

None

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Ethical approval

The study was approved by the Research Committee of National Hospital (Riyadh Care), Riyadh, Saudi Arabia.

Consent

Written informed consent was obtained from the patient for publication of this case report. A copy of the written consent is available for review by Editor-In-Chief of this journal on request.

Authors' contribution

All authors contributed significantly and in agreement with the content of the manuscript. The first author did the surgery, the second author did the histopathology, and the third author did the literature review. All authors participated in data collection and wrote the final draft.

Guarantor

M.M. Al-Qattan.

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