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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

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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

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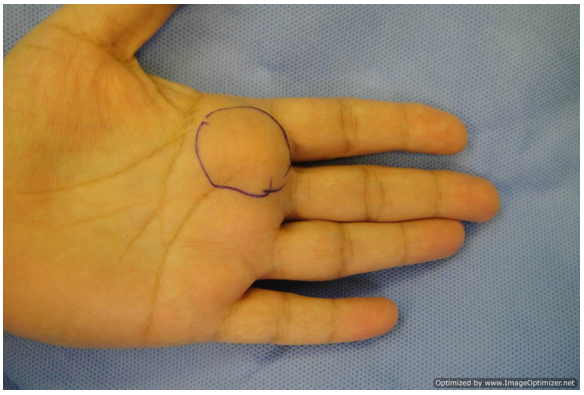

Fig. 1. The lesion.

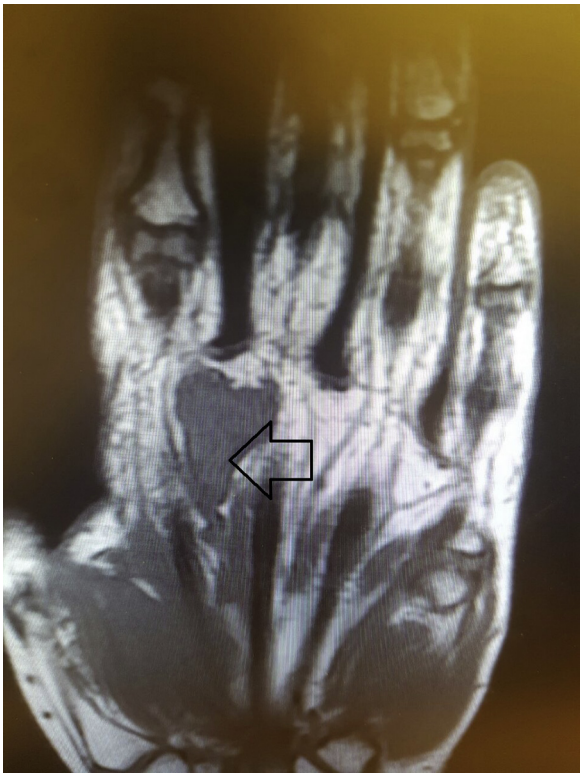

Fig. 2. MRI showing the tri-lobed lesion (arrow) at the second web.

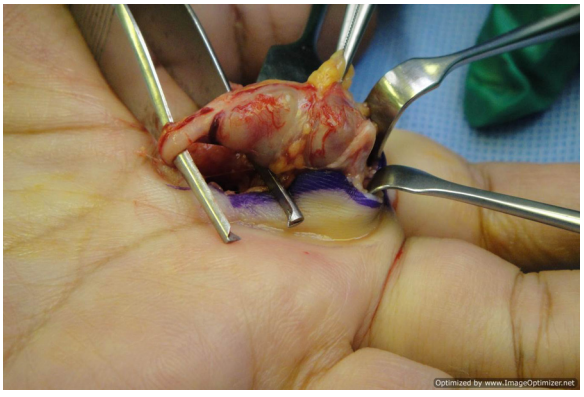

Fig. 3. Intraoperative appearance. The forceps is showing that the normal lumen of the common digital artery has become the bulge of the tumor. In retrospect, the proximal lobe the tumor is the origin from the common digital artery, while the 2 distal lobes represent the extension of the tumor into the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

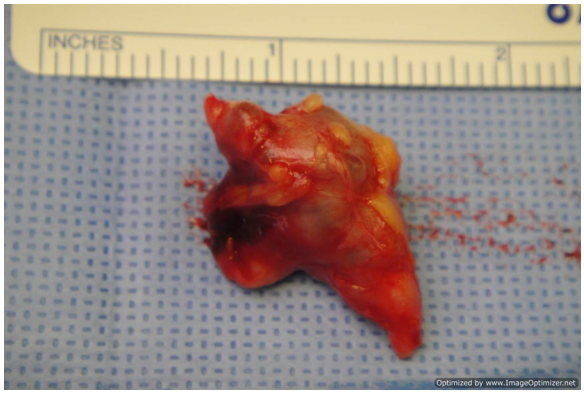

Fig. 4. The excised lesion.

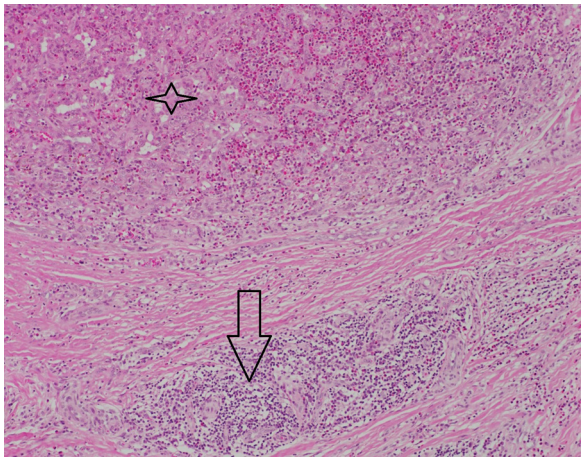

Fig. 5. Low power (x10) showing the vascular proliferation (star) and the lymphoid follicles (arrow). H & E stain.

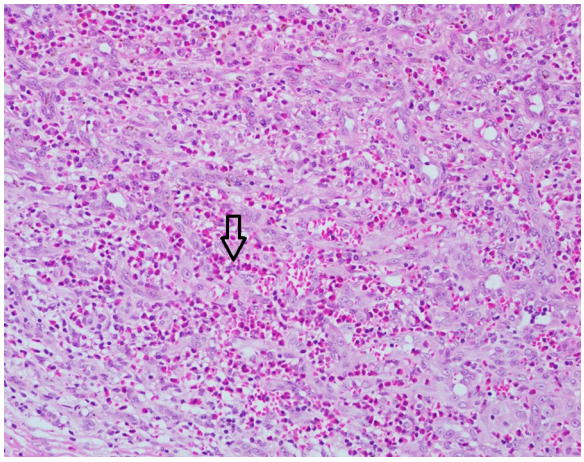

Fig. 6. High power (x40) showing the intense eosinophilic infiltrate (arrow). H & E stain.

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