

*Case Report***Radial Artery Aneurysm in a Case of Angiolymphoid Hyperplasia with Eosinophilia**Bhalla Mitesh¹, Shrotri Harshvardhan², Patil Abhijit², Garg Sahil³, Kuber Rajesh⁴, Alawadhi Akshay¹¹Second Year Resident, ²Associate Professor, ³Third Year Resident, ⁴Professor,
Department of Radiodiagnosis, Padmashree DY Patil Medical College and Research Centre, Pimpri, Pune.

Corresponding Author: Bhalla Mitesh

*Received: 27/08/2015**Revised: 02/01/2016**Accepted: 06/01/2016***ABSTRACT**

We report a known case of angiolymphoid hyperplasia diagnosed radiologically as radial artery aneurysm. A 26 year old female from Pune, Maharashtra, presented in Surgery OPD with history of swelling over right wrist since 2 months. There was history of trauma to the hand 2 months back when the patient had first noticed the swelling.

A case of aneurysm in angiolymphoid hyperplasia with eosinophilia involving large arteries is extremely rare but what make it more interesting is, in review of the literature only few reports of ALHE affecting artery in the upper extremity. In our literature search, we have not found any case of ALHE with primary localization in the distal radial artery. We report a case of ALHE presenting as aneurysm of the distal radial artery.

Keywords: Angiolymphoid hyperplasia with eosinophilia, USG, MRI, CT Angiography.

INTRODUCTION

When a weak area of a blood vessel expands or bulges significantly, we call it an aneurysm. Objectively an aneurysm is a local and permanent artery dilatation with 50% greater diameter than the normal diameter. ⁽¹⁾ Angiolymphoid hyperplasia present with a localised swelling with no other symptoms. Usually they are expansile and pulsatile swellings with a thrill and bruit as in case of large aneurysms. These can also be painful if compressing adjacent soft tissues. We report a case of aneurysm in angiolymphoid hyperplasia with eosinophilia of distal radial artery. The patient developed a pulsatile swelling of the distal radial artery without any trauma to the volar aspect of her left wrist. The purpose of this case is to understand imaging findings of swelling in patient of

with angiolymphoid hyperplasia with eosinophilia. This will aid the physician in proper diagnosis for definitive management because it can be misdiagnosed as a non vascular swelling around the wrist as ganglionic cyst, simple hematoma or vascular swelling as simple aneurysm, epithelioid haemangioma or inflammatory angiomatous nodule. ⁽²⁾

CASE REPORT

A 26 year old female from Pune, Maharashtra, presented in Surgery OPD with history of swelling over left wrist since 2 months. The patient had first noticed the swelling after a trivial trauma. The swelling has gradually increased in size since then. The swelling was not painful with no restriction in wrist movement. Physical examination showed a swelling which was non-tender,

compressible and pulsating in nature. There were no associated dermal or intradermal papules, plaques, or nodules. Past medical history was non-contributory. There was no history of previous surgery to the left forearm. There was no lymphadenopathy.

USG examination was carried out using high frequency linear probe. The probe was placed over left wrist at the radial styloid process directly over the swelling to evaluate the underlying tendons of the first compartment and the

radial artery which is situated beneath. (Figure 1A and 1B)

USG showed hypoechoic mass around the distal radial artery measuring approximately 30 x 15 mm with an eccentric patent lumen showing flow. The lumen measured approximately 3 mm in diameter. A relatively hyperechoic area measuring 4 mm in diameter was seen towards the antero-lateral aspect of the lumen s/o thrombosis. The cortex of the underlying bone was intact. (Figure 2A and 2B)

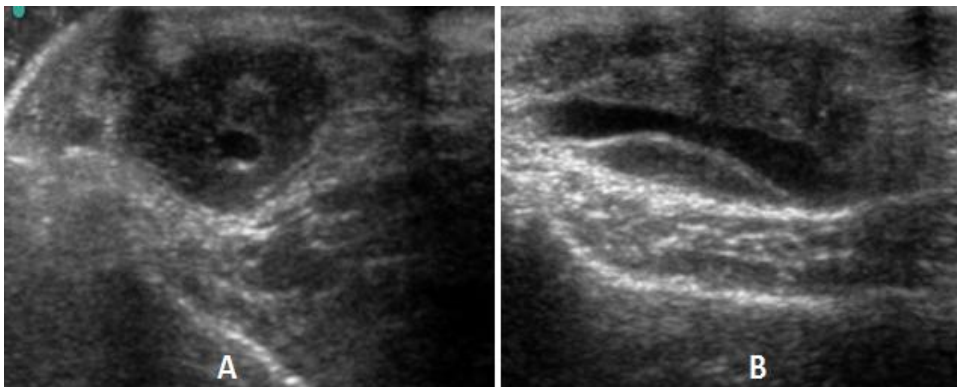


Figure: 1(A and B): B-mode ultrasounds of radial artery in transverse and longitudinal view demonstrate a patent lumen of the radial artery with relatively hyperechoic area representing the blood clot with the peripheral hypoechoic mass.

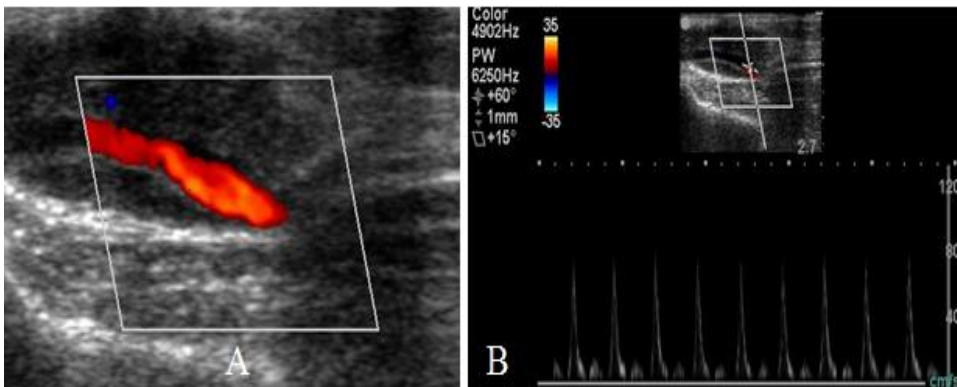


Figure 2(A and B): Colour and spectral doppler show flow in the patent lumen of the radial artery with non-turbulent triphasic waveform.



Figure-3(A and B): MRI T1W axial and coronal show the eccentric flow-void of the radial artery. Relatively hyperintense area surrounding the flow-void is representing blood clot in the aneurysm. Also noted is the surrounding lesion appearing isointense to the muscles.

MRI of left forearm with wrist and CT Angiography of the left upper limb were performed for further evaluation. MRI revealed a well circumscribed soft tissue lesion with smooth and round borders surrounding the distal radial artery. It was measuring approximately 14 (AP) x 13 (Tr) x 27 (CC) mm and appeared isointense to the muscles on T1WI (Figure 3A and 3B), heterogeneously hyperintense on T2WI (Figure 4A) and PDfs (Figure 5A and 5B).

A flow-void of the radial artery was seen eccentrically on all sequences measuring approximately 2 to 3 mm in diameter. A surrounding relatively hyperintense area on T1 (Figure 3A and 3B), T2 (Figure 4A and 4B) and PDfs (Figure 5A and 5B) was noted surrounding the flow-void representing a thrombosis. It was measuring approximately 4 mm in diameter.

CT- Angiogram showed aneurysmal dilatation of the distal radial artery just proximal to the wrist measuring approximately 20 mm in length and 6 to 7 mm in maximum diameter. Surrounding soft tissue density was also seen (Figure 6A and 6B). The flow in the ulnar artery was normal. Palmar arches show normal flow. (Figure 7A and 7B)

Based on the above findings diagnosis of an aneurysm of the radial artery with a surrounding soft tissue mass was made. On complete blood count investigation her Eosinophil count was 550 cells per microliter (cells/mcL).

Resection with great saphenous vein interposition graft. Histopathological examination revealed plump epithelioid-appearing endothelial cells (Figure 8). These findings were consistent with angiolymphoid hyperplasia with peripheral eosinophilia. Post-operative scan did not reveal any recurrent / residual mass.

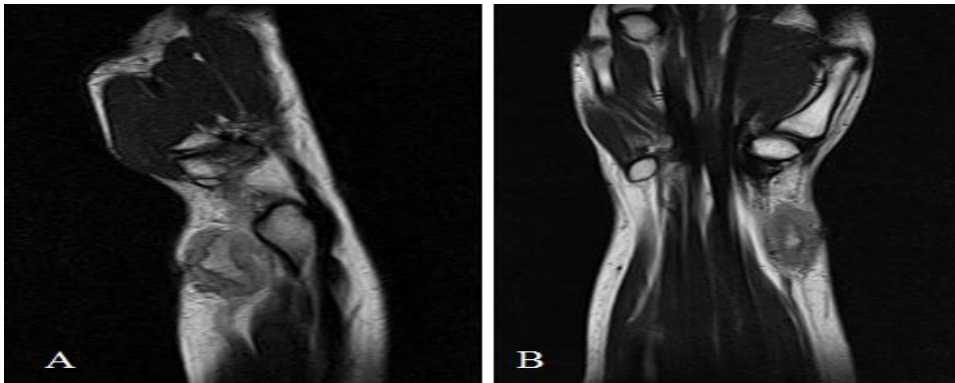


Figure-4(A and B): MRI T2W sagittal and coronal images show hyperintense area surrounding the artery representing an organized blood clot in the aneurysm. The peripheral iso to hyperintensity represents the lesion.



Figure-5(A and B): MRI Axial and coronal PDfs (proton density fat saturated) images showed the patent and thrombosed lumen of the radial artery as patent flow-void and surrounding thrombosed portion of the aneurysm as hyperintensity. The heterogeneously hyperintensity represents the surrounding lesion.



Figure: 6(A and B): CT angiogram axial and coronal reformatted images show aneurysm in the distal radial artery.

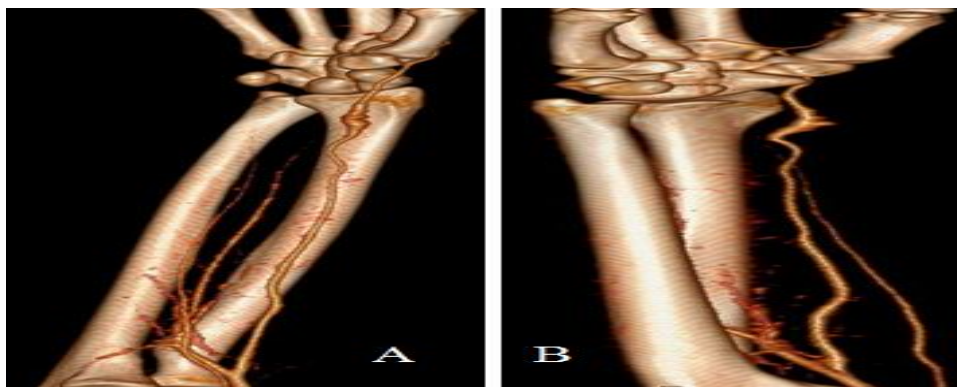


Figure-7(A and B): A3D volume rendered reconstruction of a CT-Angiogram showing the aneurysm of the radial artery.

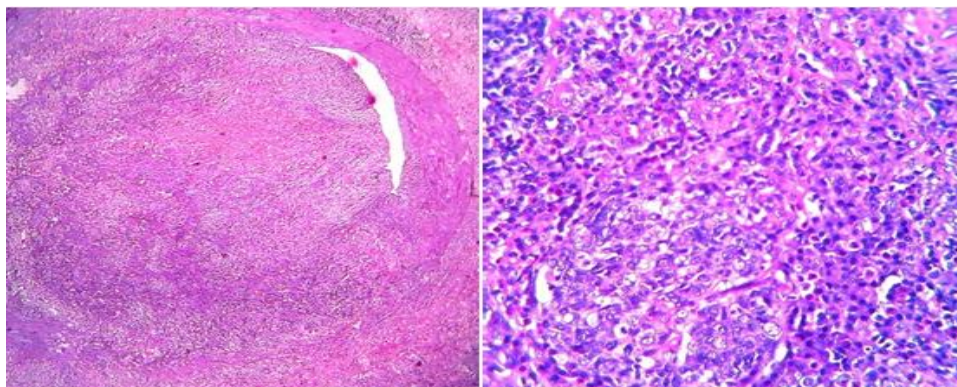


Figure-8: Histo-pathology findings of low power and high power views shows evidence of nodular proliferation of inflammatory cells separated by fibrous tissue. Lymphoid cell proliferation was predominantly seen with prominent lymphoid follicles. Sections also show predominance of eosinophilic infiltrate with a few giant cells.

DISCUSSION

Angiolymphoid hyperplasia with eosinophilia (ALHE) is a rare vascular proliferative disorder, which most commonly involves the skin of the head and neck regions. Noncutaneous localization of this pathology is unusual, and its primary localization in large arteries presenting as a pulsatile mass is extremely rare. It has been debated whether ALHE is a reactive or neoplastic lesion, as is reflected by the various names given to the condition, including

epithelioid haemangioma, pseudopyogenic granuloma, and inflammatory angiomatous nodules.⁽²⁾

Patients usually present with a painless swelling of large vessels including axillary artery and popliteal artery. As these swellings are pulsatile, imaging is done to confirm the nature and origin of the swelling and also its relation with the vessels before any intervention is carried out.

USG can be the initial radiological technique to confirm the aneurysmal

nature of the lesion and to detect the presence of a mass lesion or thrombus. It is also easily reproducible and cost effective with no radiation hazards.

Plain radiographs are unlikely to detect any abnormality. CT and MRI may show a nodular heterogeneous mass around the lumen which help in suspecting the possibility of ALHE. ⁽⁸⁾ Histopathological examination is necessary for confirmation of the same.

Similarly in our case physical examination confirming the presence of a palpable mass, imaging consistent with the mass being of vascular etiology, and a histologic specimen showing plump epithelioid-appearing endothelial cells with peripheral eosinophilia and raised IgE, a diagnosis of distal radial artery aneurysm associated with angiolymphoid hyperplasia with eosinophilia (ALHE) was made.

It is important to differentiate a mass associated with ALHE from isolated aneurysms – true / false. An aneurysm can be a true aneurysm with surrounding hematoma. Literature however says true aneurysms of the distal radial artery are rarer than aneurysms of the proximal radial artery. They are usually associated with trauma, usually penetrating or iatrogenic. ⁽³⁾ True aneurysms are frequently associated with non-penetrating trauma such as contusions. ⁽⁴⁾ False / pseudo aneurysm is a breach in the vessel wall such that blood leaks through the wall but is contained by the adventitia or surrounding perivascular soft tissue. Pseudo aneurysms are well documented complication after surgery, arterial puncture or trauma, and develop after any procedure that causes partial disruption of vessel wall. ⁽⁵⁾

Other potential differential diagnosis in this case includes Kimura's disease. Kimura's differs from ALHE in one or more distinct ways. Kimura's disease is a chronic inflammatory condition that typically presents as a swelling, usually in the head or neck

region. It often is confused with ALHE largely because of a similar histologic appearance. ⁽⁶⁾

But Kimura's disease is a chronic inflammatory condition that typically presents as a swelling, usually in the head or neck region. How ALHE is unique in that its endothelial cells are characteristically plump; the endothelial cells in Kimura's disease are much flatter. A second key difference between ALHE and Kimura's disease is that the latter typically affects deeper tissues, such as lymph nodes and salivary glands, than the former. ⁽⁷⁾

Patients with ALHE usually present with a painful papule or nodule. It is typically dome-shaped and appears erythematous to brown.

The mainstay of treatment for ALHE is surgical resection of the entire mass. Topical or intralesional steroids, radiation, and cryotherapy also have been used with mixed results. ⁽⁹⁾

Our patient underwent surgical resection and great saphenous vein interposition graft repair of right radial artery aneurysm. She is currently 8 months postoperative and doing well with full hand and wrist function and no physical / radiological signs of disease recurrence. Our case report is unique in that it describes a rare tumor in an even rarer location. There are only a handful of case reports describing ALHE in the hand presenting with radial artery aneurysm. Although it may be rare, ALHE is a diagnosis that every radiologist should keep on his or her differential.

REFERENCES

1. Ho P K, Weiland A J, Mc Clinton MA, Wilg is EF. Aneurysms of the upper extremity. *J Hand Surg Am.* 1987 Jan; 12(1):39-46. PMID: 3805642.
2. Vandy F, Izquierdo L, Liu J, Criado E. Angiolymphoid hyperplasia involving large arteries. *J Vasc Surg.* 2008 May; 47(5):1086-9. PMID: 18455650.

3. Morton K, Robertson AJ, Hadden W. Angiolymphoid hyperplasia with eosinophilia: report of a case arising from the radial artery. *Histopathology*. 1987 Sep; 11(9):963-9. PMID: 3666679.
4. Pero T, Herrick J. Pseudoaneurysm of the radial artery diagnosed by bedside ultrasound. *West J Emerg Med*. 2009 May;10(2):89-91.PMID: 19561825.
5. Kerr CD, Duffey TP. Traumatic false aneurysm of the radial artery. *J Trauma*. 1988 Nov; 28(11):1603-4.PMID: 3184228.
6. Behar JM, Winston JS, Knowles J, Myint F. Radial artery aneurysm resulting from repetitive occupational injury: Tailor's thumb. *Eur J VascEndovasc Surg*. 2007 Sep; 34(3):299-301 PMID: 17572115.
7. Pappas N, Deirmengian GK, Jian B, Bozentka DA. Orthopaedic radiology pathology conference: hand mass in an 18-year-old woman. *Clin Orthop Relat Res*. 2010 Jun; 468(6):1722-5.PMID: 19609628.
8. Googe PB, Harris NL, Mihm MC Jr. Kimura's disease and angiolymphoid hyperplasia with eosinophilia: two distinct histopathological entities. *J Cutan Pathol*. 1987 Oct; 14(5):263-71.PMID: 3680714.
9. Wolff K, Goldsmith LA, Katz SI, Gilchrest BA, Paller AS, Leffell DJ. Angiolymphoid hyperplasia with eosinophilia and Kimura disease. *Fitzpatrick's Dermatology in General Medicine*. V Vol 1. Ed 7. New York, NY: McGraw-Hill Professional; 2007:313–314.

How to cite this article: Bhalla M, Shrotri H, Patil A et al. Radial artery aneurysm in a case of angiolymphoid hyperplasia with eosinophilia. *Int J Health Sci Res*. 2016; 6(2):427-432.

International Journal of Health Sciences & Research (IJHSR)

Publish your work in this journal

The International Journal of Health Sciences & Research is a multidisciplinary indexed open access double-blind peer-reviewed international journal that publishes original research articles from all areas of health sciences and allied branches. This monthly journal is characterised by rapid publication of reviews, original research and case reports across all the fields of health sciences. The details of journal are available on its official website (www.ijhsr.org).

Submit your manuscript by email: editor.ijhsr@gmail.com OR editor.ijhsr@yahoo.com