Case Report

DOI: 10.7860/JCDR/2016/20472.8538

# Angiomatous Hamartoma - A Rare Presentation

Ear, Nose and Throat Section

SHARAD HERNOT[1], RAMAN WADHERA[2], MADHURI KAINTURA[3], SANDEEP BHUKAR[4], DHEERAJ SHASHIKUMAR PILLAI[5]

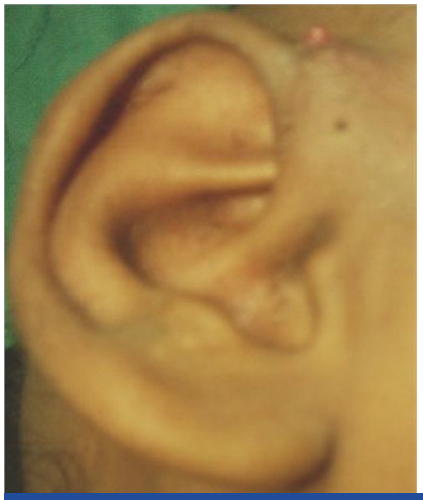
## ABSTRACT

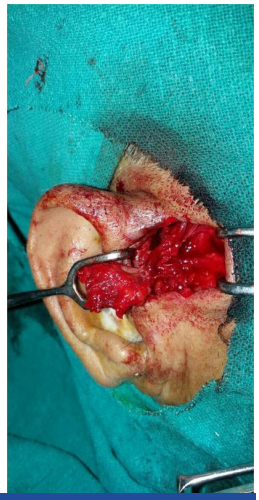
Eccrine Angiomatous Hamartoma (EAH) is a benign rare skin neoplasm characterised histologically by abnormal proliferation of sweat glands and surrounding capillaries and other dermal elements like fatty lobules and hair. It usually presents at birth or in early childhood in the form of solitary nodules mostly affecting the extremities. Here, we report a case of angiomatous hamartoma over the face which presented as a cystic swelling in preauricular region in a 55-year-old man. The late onset and a rare site for presentation of EAH prompted us to report the case. There is not even a single case of EAH arising in the "preauricular" region, reported.

**Keywords:** Eccrine Angiomatous Hamartoma (EAH), Late onset, Preauricular region

## CASE REPORT

A 55-year-old male presented to the ENT out-patient department with complaints of swelling in right preauricular region for a duration of 2 months [Table/Fig-1]. It was insidious in onset, gradually progressive in size and was occasionally painful. There was no history of local trauma or associated features like hypertrichosis or hyperhidrosis. However he gave history of undergoing surgery for the same complaint 10 years back, but patient was never fully symptomless even after surgery, and suffered intermittent complaints like pain and swelling in the right preauricular region. There was no family history of similar complaints. Examination of his right pinna showed an ill-defined swelling of approximate size 3cm x 3cm at the root of helix pushing the pinna outwards. It was soft, fluctuant, non-tender and non-transilluminant. The color and temperature of the overlying skin was normal. The swelling was not attached to the underlying structures or the overlying skin. There was no similar swelling in any other part of the body. His routine blood examinations were normal. An ultrasonogram of the swelling revealed an ill-defined hypoechoic lesion with thick septations and debris in the pre and retro auricular region near the upper part of helix. Fine Needle Aspiration Cytology (FNAC) of the swelling was done which was suggestive of cystic lesion with haemorrhagic changes, suggestive of haematoma, infected preauricular sinus, angiomatous polyp, or EAH. The cyst was excised under local anaesthesia. An elliptical incision was given over the cyst in the preauricular region and blunt dissection was done gently to separate the cyst from the surrounding tissue. During the procedure the cyst was found to be surrounded by fibrous tissue. The superficial temporal artery was attached to the surface of the cyst and surrounding tissue by dense adhesions [Table/Fig-2]. The cyst was carefully dissected from the artery, and removed in toto. The excised cyst was sent for histopathological examination which confirmed it to be EAH. The incision was stitched and a drain was put for 24 hours. The patient was started on intravenous antibiotics and analgesics in the ward where the postoperative period was uneventful. The drain was removed the next day and the patient was discharged from the ward on 2nd postoperative day.

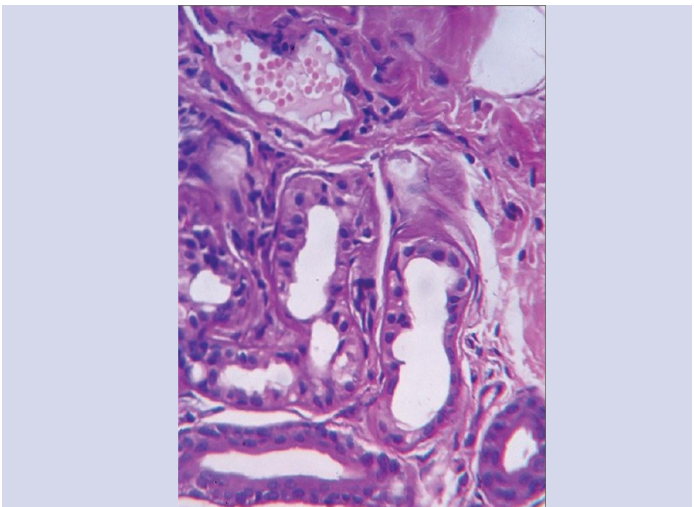




**[Table/Fig-1]:** Swelling in the right preauricular region. **[Table/Fig-2]:** Intraoperative view showing superficial temporal artery attached to the surface of the cyst and surrounding tissue by dense adhesions.

## DISCUSSION

EAH is a benign rare skin neoplasm characterised histologically by abnormal proliferation of sweat glands and surrounding capillaries and other dermal elements like fatty lobules and hair. It usually presents at birth or in early childhood in the form of solitary nodules mostly affecting the extremities. Nearly 80% of the cases are reported over the extremities, with a predilection for palms and



**[Table/Fig-3]:** Photomicrograph showing microscopic examination of EAH depicting dermal proliferation of well-differentiated eccrine glands that are closely associated with thin-walled angiomatous channels. (H & E, 40 X).

soles, and also rare sites like lower eyelids can be involved [1-3].

As mentioned, EAH is a rare cutaneous neoplasm appearing histologically as a proliferation of eccrine sweat glands with angiomatous vascular elements that are of capillary origin. EAH usually appears at birth or during childhood [2,4], although in our case it appeared at the age of 45 years. However, looking at the literature it was found that there are just a few recent reports of EAH cases presenting in adulthood for instance, in 2012, Sen et al., has reported EAH presenting over the dorsum of left hand in a 26-year-old female [5]; in 2012, Nowakowski reported a case with adult onset EAH on the vulva [6]; and in 2009, Natrajan et al., has also reported adult onset EAH in a 37-year-old female [7].

EAH presents mostly as a solitary plaque, nodule or, less commonly, as a macule, which is localized mainly to the extremities, particularly the palms and the soles. The colour of the lesions may be violet, brown, blue, red, pink. It can also be of the normal skin colour, as observed in our patient. Martinelli and Zeller have described about the atypical clinical variants with superficial changes resembling acanthosis nigricans, linear verrucous lesions, and hypertrichosis in their respective reports [8,9]. The lesions of EAH are usually asymptomatic, but associated symptoms of pain and hyperhidrosis have been reported in nearly one-third of reported cases; these were not detected in our patient. Associated features may include knuckle pads, nevus sebaceous, verrucous epidermal nevus, and neurofibromatosis [10,11], which were also not present in our case.

Microscopic examination of typical EAH as already mentioned shows dermal proliferation of well-differentiated eccrine glands that are closely associated with thin-walled angiomatous channels [Table/Fig-3]. However, the microscopic examination of atypical EAH may include unusual histopathologic variants like pilar structures, lipomatous foci, lymphatics, mucin, or dense collagen fibres [12,13].

As a rule, aggressive treatment has not been indicated for this benign disorder; and surgery has been the only definitive therapy for EAH [14]. In our case also, the swelling was excised surgically and sent for histopathological examination.

Immunohistochemical study was not performed in this case, but studies report that antigens frequently found in eccrine glands, such as the Carcinoembryonic Antigen (CEA) and protein S-100, are reduced Quantitatively in the eccrine glands of EAH [15].

## CONCLUSION

To conclude, a high index of suspicion should be kept for rare disorders of the skin of the head and neck region which can present at an unusual age with common or uncommon symptoms/ complaints.

## REFERENCES


[1] Halder C, Naskar B, Biswas P, Mitra PK. Eccrine angiomatous hamartoma: Late onset facial presentation. *Indian J Dermatol*. 2014;59(4):403-05.
[2] Sanmartin O, Botella R, Alegre V, Martinez A, Aliaga A. Congenital eccrine angiomatous hamartoma. *Am J Dermatopathol*. 1992;14:161–64.
[3] Morrell DS, Ghali FE, Stahr BJ, McCauliffe DP. Eccrine angiomatous hamartoma: A report of symmetric and painful lesions of the wrists. *Pediatr Dermatol*. 2001;18:117–19.
[4] Pelle MT, Pride HB, Tyler WB. Eccrine angiomatous hamartoma. *J Am Acad Dermatol*. 2002;47(3):429- 35.
[5] Sen S, Chatterjee G, Mitra PK, Gangopadhyay A. Eccrine angiomatous naevus revisited. *Indian J Dermatol*. 2012;57:313–15.
[6] Nowakowski K, Dyduch G. Adult onset eccrine angiomatous hamartoma on the vulva. *Indian J Dermatol.* 2012;51(7):840-41.
[7] Natarajan K, Rai R, Sundararajan V, Venkatchala S. Eccrine angiomatous hamartoma in an adult. *Indian J Dermatol Venereaol Leprol*. 2009;75:193–94.
[8] Martinelli PT, Tschen JA. Eccrine angiomatous hamartoma: a case report and review of the literature. *Cutis.* 2003;71:449–55.
[9] Zeller DJ, Goldman RL. Eccrine-pilar angiomatous hamartoma. Report of a unique case. *Dermatologica*. 1971;143:100–04.
[10] Campen R, Zembowicz A, Liu V, Wrone D. Linear ectodermal cutaneous hamartoma. *Int J Dermatol*. 2003;42:376–79.
[11] Castilla EA, Schwimer CJ, Bergfeld WF, Skacel M, Ormsby A. Eccrine angiomatous hamartoma in a neurofibromatosis type-1 patient. *Pathology*.2002;34:378–80.
[12] Donati P, Amantea A, Balus L. Eccrine angiomatous hamartoma: a lipomatous variant. *J Cutan Pathol.* 1989;16:227–29.
[13] Sulica RL, Kao GF, Sulica VI, Penneys NS. Eccrine angiomatous hamartoma (nevus): immunohistochemical findings and review of the literature. *J Cutan Pathol*. 1994;21:71–5.
[14] Kirby EJ, Shereff MJ, Lewis MM. Soft-tissue tumors and tumor-like lesions of the foot. An analysis of eighty-three cases. *J Bone Joint Surg Am*. 1989;71:621–26.
[15] Laeng RH, Heilbrunner J, Itin PH. Late-onset eccrine angiomatous hamartoma: Clinical, histological and imaging findings. *Dermatology* 2001;203:70-74.



**PARTICULARS OF CONTRIBUTORS:**
1. Senior Resident, Department of ENT, Pandit Bhagwat Dayal Sharma Post Graduate Institute of Medical Sciences, Rohtak, Haryana, India.
2. Professor, Department of ENT, Pandit Bhagwat Dayal Sharma Post Graduate Institute of Medical Sciences, Rohtak, Haryana, India.
3. Junior Resident, Department of ENT, Pandit Bhagwat Dayal Sharma Post Graduate Institute of Medical Sciences, Rohtak, Haryana, India.
4. Junior Resident, Department of ENT, Pandit Bhagwat Dayal Sharma Post Graduate Institute of Medical Sciences, Rohtak, Haryana, India.
5. Junior Resident, Department of ENT, Pandit Bhagwat Dayal Sharma Post Graduate Institute of Medical Sciences, Rohtak, Haryana, India.

**NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:**
Dr. Sharad Hernot,
Pocket-Ap, 115-A, Virat Apartments, Pitampura, Delhi-110034, India.
E-mail: hernots@yahoo.com



**FINANCIAL OR OTHER COMPETING INTERESTS:** None.

Date of Submission: **Apr 02, 2016**
Date of Peer Review: **May 24, 2016**
Date of Acceptance: **Jul 18, 2016**
Date of Publishing: **Sep 01, 2016**