International Journal of Research in Medical Sciences
Reddy S et al. Int J Res Med Sci. 2023 Jan;11(1):369-371
www.msjonline.org
pISSN 2320-6071 | eISSN 2320-6012

DOI: https://dx.doi.org/10.18203/2320-6012.ijrms20223665

Case Report

Rare benign tumour at an unusual site-angiomyoma of the ear: a case report and literature review

Sridhar Reddy, Vinod Kumar Gonuru*, Sri Vishnupriya Ravipati

[1]Department of Otorhinolaryngology, Apollo Institute of Medical Sciences and Research, Hyderabad, Telangana, India

Received: 31 October 2022
Revised: 30 November 2022
Accepted: 14 December 2022

***Correspondence:**
Dr. Vinod Kumar Gonuru,
E-mail: drvinodkumar_g@apolloimsr.edu.in

ABSTRACT

The objective was to demonstrate the occurrence of the rare benign tumour angiomyoma or angioleiomyoma in the ear canal. Angiomyoma, known as vascular leiomyoma or angioleiomyoma, is a benign tumour of smooth muscle arising from the muscular wall of veins and comprises less than 5% of all benign smooth muscle tumours. Peak incidence is observed in the fourth to sixth decades with the site of predilection being the subcutaneous tissue in the lower limbs, followed by the upper limbs. This is in contrast to our present case, where the angiomyoma was found in the ear canal which is very rare. A 60-year-old female, an otherwise healthy patient presented with fullness in the left ear. Excision of the swelling was done without any complications.

Keywords: Angiomyoma, Angioleiomyoma, External auditory canal, Benign tumour

INTRODUCTION

Cutaneous angioleiomyomas (ALMs) are a part of leiomyomas, that is, benign tumours originating from smooth muscle cells present in the skin. Angioleiomyomas (also known as vascular leiomyomas or angiomyomas) are derived from the muscle layer of dermal blood vessels.[1] They manifest typically as solitary, deep dermal, or subcutaneous flesh-coloured, well-circumscribed nodules usually located on the lower legs of adult women. Minor trauma, venous stasis and hormonal changes, especially estrogen could be some of the reasons for its occurrence.[2] They may be identified clinically because they are usually painful to pressure or even spontaneously, especially when they develop on the lower limbs, but are usually misdiagnosed clinically as epidermoid cysts. In less than 10% of cases, ALMs may develop in the head/neck area, and in that case, they are usually painless, rendering the clinical diagnosis even more difficult.[3,4] Most of the time preoperative diagnosis is not particular, microscopic examination is needed for accurate diagnosis and complete excision of the swelling.

CASE REPORT

A 60-year-old woman came to the ENT outpatient department with a complaint of fullness in the left ear for three months. The patient was apparently asymptomatic three months ago when she developed fullness in the left ear which was insidious in onset and progressive in nature. On examination a diffuse soft swelling was observed in the external auditory canal in the left ear, the tympanic membrane was intact and there was no tenderness. The patient was planned for excision of the swelling under general anaesthesia. The preoperative workup was normal. Surgery was performed under general anaesthesia with the patient in the supine position with the left ear facing upwards. 2% xylocaine with adrenaline was infiltrated locally. An endaural incision was given and the tympanomeatal flap was elevated. The soft cystic swelling measuring 1×0.5 cm was excised and

sent for histopathological examination with a probable diagnosis of hemangioma. Hemostasis was secured and the wound was closed in layers with 3-0 vicryl. A gauze pack was placed in the external auditory canal. The rest of the stay was uneventful. Microscopic examination showed a well-circumscribed mass and fibro-adipose tissue. Well-circumscribed mass is composed of numerous vascular channels containing RBCs in the lumina showing marked medial hyperplasia. Some of the vessels show narrowing of the lumina due to medial hyperplasia (Figure 1). These findings were diagnostic of angiomyoma or angioleiomyoma. The patient followed up after six months and on examination, the external ear canal was patent and normal without any growth.

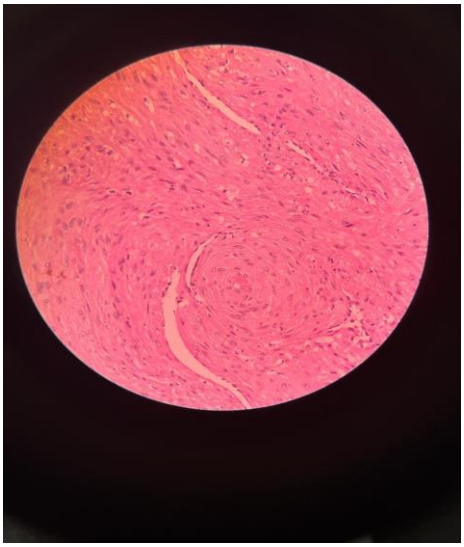

Figure 1: Well circumscribed mass with vascular channels showing medial hyperplasia.

DISCUSSION

Angiomyoma is a rare benign tumour originating from vascular smooth muscle cells, the tunica media, and the characteristic feature is the detection of numerous blood vessels together with spindle-shaped smooth muscle cells. In most cases, it occurs in the subcutaneous tissue of the limbs, especially in the lower limbs, but it is very rare to occur in the head and neck area. In the head and neck area, it develops most frequently in the larynx and the turbinates, and in addition, its occurrence in the oral cavity (lip, hard palate, tonsil), nose, ear, cheek, parotid gland, and the submandibular region has been reported.5 The most common locations for angioleiomyomas are the lower limbs in 67% of cases, the upper limbs in 22%, the head/neck region in 8.5%, the trunk in 2.5%, and the ear in 2.8% of cases.3 The clinical appearance of ALMs in the ears is variable. Auricular angioleiomyomas may be located on the helix,6,7 the antihelix, 8,9 the lobule, 10,11 or the external auditory canal.12 Most cases appear during adult life, typically during the fifth or sixth life decade, as in our patient. Although rare cases of painful ALMs on the ears have been reported,9 head/neck ALMs are usually painless, contrasting with ALMs of the limbs, which are usually painful upon pressure or even spontaneously.[4-8,11,13-15] ALMs of the external auditory canal can be one of the causes of hearing loss.12 Clinically, ALMs must be differentiated from other benign tumours of the auricle, namely, hemangiomas, glomus tumours, epidermoid cysts, auricular pseudocysts, angiolymphoid hyperplasia with eosinophilia, and neurofibromas. The diagnosis of this tumour is generally made by histological examination. The histological picture shows a well-demarcated dermal tumour consisting of smooth muscle cells, in which a variable amount of vascular channels is seen. Three subtypes of angioleiomyomas have been recorded, including a solid type (smooth muscle bundles surround numerous small slit-like vessels), a cavernous type (dilated vessels with a thick muscular wall merging with the intervascular smooth muscle cells), and a venous type (thick-walled vessels easily distinguished from the intervascular smooth muscles).1 The commonest type is the solid type, followed by the venous and the cavernous type. Rarer variants consist of the epithelioid and the pleomorphic type.1 The pathological diagnosis of ALMs is generally easy, but sometimes the lesions may be mimicked by other spindle-cell proliferation (such as dermato(myo)fibromas, glomus tumours, myopericytomas, hemangiomas, or keloids). The course of ALMs of the ear is usually uneventful and simple surgical excision is curative, the rate of recurrences of ALMs, in general, being less than 0.4%.3

CONCLUSION

Although ALMs only rarely develop on the ear, they should be included in the clinical differential diagnosis of tumours developing in this anatomic zone as misdiagnosis of an angioleiomyoma is very often as seen in the case above. Hence, a histopathological examination of the biopsy (or excision) specimen should be taken for a definitive diagnosis of the case.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

REFERENCES

1. Kanitakis J. Angioleiomyoma of the auricle: an unusual tumor on a rare location Case Rep Otolaryngol. 2017;2017:8289710.
2. Aljarbou O, Sangueza OP. Soft tissue pericytic (perivascular) angioleiomyoma. In: Mantilla JG, eds. Pathology Outlines Inc; 2021.
3. Hachisuga T, Hashimoto H, Enjoji M. Angioleiomyoma: a clinicopathological reappraisal of 562 cases. Cancer. 1984;54(1):126-30.
4. Yoon TM, Yang HC, Choi YD, Lee DH, Lee JK, Lim SC. Vascular leiomyoma in the head and neck region: 11 years' experience in one institution. Clin Experiment Otorhinolaryngol. 2013;6(3):171-5.

5. Kim H, Jung S, Kwon H, Sohn W, Moon S. Angiomyoma in the Buccal Space. J Craniofac Surg. 2010;21(5):1634-5.
6. Kim HU, Mun JH, Park SH, Yun SK, Ihm CW. Angioleiomyoma on the helix of the ear. J Dermatol. 2004;31(10):858-9.
7. Citil R, Ciralik H, Gül A, Sayar H. Auricular angioleiomyoma: a case report. Türk Patholoji Dernegi. 2011;27(3):268-70.
8. Chen YA, Chuang WY, Hsueh S, Chan KC. Solitary nodule on the auricle. Int J Dermatol. 2012;51(12):1427-8.
9. Palleschi GM, Cristofaro G, Maio V. Angioleiomyoma of the auricle: a rare finding. Giornale Italiano di Dermatologia e Venereologia. 2013;148:537-8.
10. Choe KS, Sclafani AP, McCormick SA. Angioleiomyoma of the auricle: a rare tumor. Otolaryngol Head Neck Surg. 2001;125(1):109-10.
11. Akkash L, Nilles M, Mayser P. Solitary blue nodule on the ear helix. Hautarzt. 2008;59(1):54-7.
12. Picciotti PM, Cantore I, LaGreca C, DiNardo W, Scarano E. Angioleiomyoma of the external auditory canal. Am J Otolaryngol. 2007;28(4):235-7.
13. Kim HI, Roh SG, Lee NH, Yang KM, Park HS. Angioleiomyoma of the auricle. Archives of Plastic Surgery. 2013;40(1):68-9.
14. Wirth GA, Sundine MJ, Kong AP, Carpenter PM. Auricular angioleiomyoma: a case report and review of the literature. Ear Nose Throat J. 2007;86(5):281-3.
15. Oh JH, Park JM, Choi H, Choi Y. Two cases of angioleiomyoma of the auricle. J Int Adv Otol. 2015;11(1):95-6.

Cite this article as: Reddy S, Gonuru VK, Ravipati SV. Rare benign tumour at an unusual site-angiomyoma of the ear: a case report and literature review. Int J Res Med Sci 2023;11:369-71.