

*Case Report***Clear Cell Hidradenoma of the Ear Lobe and Its Management:  
A Rare Case Report**K. Sampath Kumar Singh<sup>1\*</sup>, A. Rajesh Kumar<sup>2\*</sup>, M. Neeraja<sup>3\*\*</sup>, P. Venkata Ramana Babu<sup>4\*\*</sup><sup>1,4</sup>Associate Professor/ <sup>1</sup>Incharge HOD, <sup>2</sup>Assistant Professor, <sup>3</sup>Professor & HOD,  
\*Dept. of E.N.T., \*\*Dept. of Pathology,  
Govt. Medical College & Govt. General Hospital, Anantapuramu, Andhra Pradesh.

Corresponding Author: A. Rajesh Kumar

*Received: 20/08/2015**Revised: 11/09/2015**Accepted: 21/09/2015***ABSTRACT**

Clear cell hidradenoma is a benign skin appendage tumour, these lesions have a wide histological spectrum and many different terms are often used to describe the same tumour. These tumours grow slowly and present in their fourth decade or older. Wide local excision is the treatment option for hidradenoma. We report a rare case of a patient with clear cell hidradenoma of ear lobe, who underwent wide surgical excision under local anaesthesia. Its surgical repair places a challenge due to the difficulty of natural appearing and durable outcome. Reconstruction of ear lobe had done in a single stage procedure by using the limberg flaps. This produces a good looking ear lobe in our 6 months follow up period.

**Key Words:** clear cell hidradenoma, Hidradenocarcinoma, Limberg flap.

**INTRODUCTION**

Hidradenoma is a benign skin appendage tumour, which is usually present as a solitary skin coloured lesion. The patients with clear cell hidradenoma and clear cell hidradenocarcinoma are present in their fourth decade or older. Apocrine lesions of the skin are rare and are found mainly in body folds including the axillary, groin and anogenital regions, where apocrine glands are most concentrated, as well as in the umbilicus, eyelid (Moll's glands), areola and in external auditory meatus. Hidradenoma papilliferum rare variety has been reported in males, <sup>[1]</sup> and in extra-anogenital locations, particularly in the head and neck. <sup>[2]</sup> The tumours are solitary,

and are usually small and asymptomatic. Occasionally, the tumours can be large and elevated, forming a reddish-brown mass with an ulcerated and bleeding surface. <sup>[3]</sup> The tumour grows slowly and can be present for several years without apparent change. Wide local excision is the treatment option for hidradenoma, but it has a recurrence rate of approximately 12% if not fully excised, and 50% recurrence rate despite of aggressive surgery in case of clear cell hidradenocarcinoma (CCHC). Hidradenoma have variable histomorphological patterns.  
(a) hidradenoma–solid cystic variant,  
(b) hidradenoma–solid variant,

(c) hidradenoma–tubular/ ductal differentiation and clear cell change [clear cell hidradenoma], (d) hidradenocarcinoma – cellular type. In this case we report on the clinical, histological findings of a skin tumour, management of this tumour and reconstructive method for ear lobule defect by using limberg flap (transposition flap).

## CASE REPORT

Here we report a case of 44 years old male, farmer by occupation, presented to E.N.T. OP, with chief complaints of swelling in the left ear lobule since 6 months [Fig 1], and progressing continuously, not relieved by medication given by the general practitioner. Swelling is not associated with pain. On examination of the ear the painless swelling is limited to the left ear lobule 2x2 cm in size, at the center of the swelling 0.5 cm size ulcer noticed with irregular margin & profuse bleeds on touch. The pinna, external auditory canal and tympanic membrane are normal. Wedge biopsy taken and histopathological examination reported as clear cell hidradenoma.

**Operation:** we planed wide local excision of the tumour and the resected part of the pinna and ear lobule was reconstructed with local pedicled flaps in one stage procedure. Under local anaesthesia wide local excision of the tumour and part of the cartilage with skin also removed [Fig 2,3] and sent for histopathological examination. The defected earlobule was reconstructed with posterior auricular and anterior neck skin from under ear lobule tissue, suture removal done on 6<sup>th</sup> post operative day [Fig 4]. The size & shape of the pinna are appeared near to the opposite ear post operatively. We discharge the patient on 12<sup>th</sup> post – op day and regular checkup done weekly once for 1 month [Fig 5], and monthly once for 6 months.

**Gross appearance:** recieved skin covered grey brown soft tissue mass measuring

4x3x1 cm, external surface of the mass shows an ulcer measuring 0.5x 0.5cm. cut section of the mass shows a great white lesion with a cyst, filled with serous fluid is present within the lesion.



Fig. 1: Pre - op clinical photograph



Fig. 2: Per - op photograph showing defective area after excision of the tumor

**Microscopic features:** haematoxylin & eosin stained section studies shown structures of skin with epidermis and dermis. Epidermis is unremarkable, dermis shows an intradermal lobule, tumour composed of two layers of cells - predominantly cells are round to oval with clear cytoplasm, round bland nucleus and some cells with pale eosinophilic granular cytoplasm [Fig 6,7]. There is no atypical mitotic activity. Septae with thin blood



vessels are seen along with occasional duct like spaces with eosinophilic secretion [Fig. 8]. Features are suggestive of benign adenexal tumour – clear cell hidradenoma.



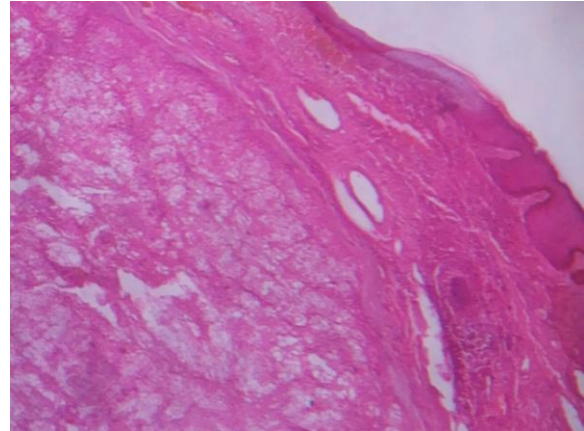
**Fig. 3:** Excised part of the tumour with skin & cartilage



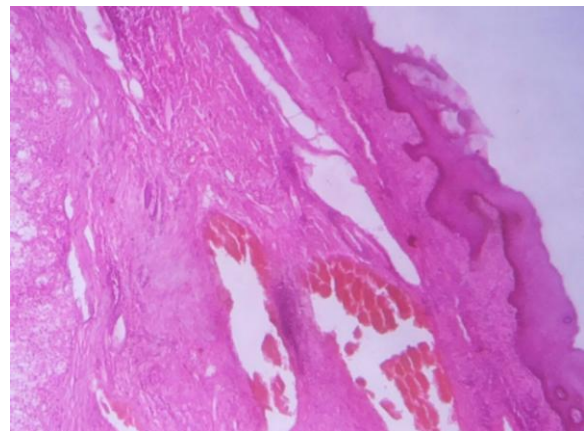
**Fig.4:** Post- op photograph after 1<sup>st</sup> week



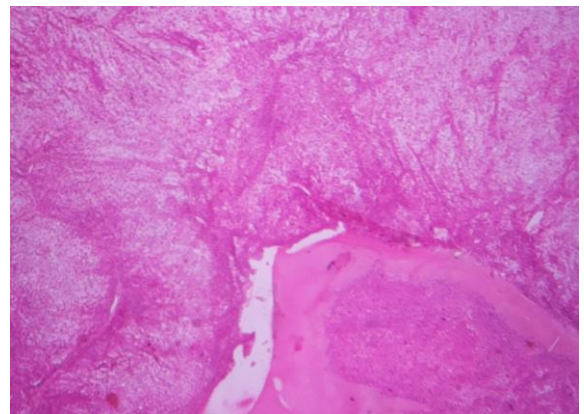
**Fig. 5 :** post – op photo after 3 weeks



**Fig. 6:** Low power (10x) view of H & E stain showing epidermis with underlying well circumscribed lobulated dermal nodule



**Fig. 7:** low power (10x) view of H & E stain showing epidermis with underlying dermal tumour along with dilated congested blood vessels.



**Fig. 8:** High power (40x) view of H & E stain showing lobules of tumour with cystic space filled with eosinophilic secretion. Tumour cells are round to polygonal with clear cytoplasm and uniform nuclei, stroma is eosinophilic & hyalinized.

## DISCUSSION

Definition Hidradenoma is a form of benign adnexal neoplasm that is a close

relative of poroma, but is usually characterized by cells with ample cytoplasm. Some authorities use the broad designation *acrospiroma* to refer to both hidradenoma and poroma jointly. Although commonly classified as an “eccrine” neoplasm by authorities of the past, hidradenomas are currently thought to be of either apocrine or eccrine lineage. [4,5] Hidradenoma is probably of apocrine lineage, most of the time. [6]

Hidradenoma is a benign tumour, which usually presents as a solitary, skin-coloured lesion and occurs more commonly in females. [7] Apocrine lesions of the skin are rare and are found mainly in body folds including the axillary, groin and anogenital regions, where apocrine glands are most concentrated, as well as in the umbilicus, eyelid (Moll's glands), areola and in external auditory meatus. Hidradenoma may have variable histomorphological patterns, reflected by the various terms used to describe this entity: nodular hidradenoma, eccrine acrospiroma, solid-cystic hidradenoma, clear cell hidradenoma, and clear cell acrospiroma. It shows variably sized nests and nodules of neoplastic epithelial cells, with small ductular lumens, confined within the upper dermis. The tumour cells are small, monomorphous and polyhedral, resembling those of poroma cells. In fact, some tumours have epidermal attachment, and occasionally may also have features overlapping with those of typical poromas. [8] Clear cell change and/or squamous metaplasia may be prominent. However, squamoid change does not seem to denote a worse prognosis. Occasionally, focal apocrine components may also be present. The lesion is also characterised by its pushy, but well-circumscribed, peripheral border. Nodular hidradenoma should be fully excised, as malignant transformation may be present in other areas of the lesion.

Furthermore, hidradenoma has a recurrence rate of approximately 12% if not fully excised, [9] especially in lesions with irregular peripheral margins. [8]

**Clinical findings:** Hidradenoma lacks any distinctive clinical attributes and is only diagnosable by biopsy. The lesion presents as a solitary nodule; sometimes a cystic quality or a small amount of serous drainage can be detected.

**Histopathological findings:** Hidradenomas are mostly dermal neoplasms with a nodular, sharply circumscribed pattern when viewed at low magnification. Some hidradenomas may display a juxtaepidermal pattern superficially with multifocal attachment to the epidermis, much like poroma. Although the stroma of hidradenoma is commonly sclerotic and may contain ectatic vessels, usually the granulation tissue-like quality that typifies the stroma of a poroma is not present. The cells of hidradenoma are large with ample cytoplasm but uniform nuclei, which are generally somewhat larger than the nuclei of poroma. Overt clear cell change (“clear cell” hidradenoma) is common, as is cystic degeneration (“solid-cystic” hidradenoma). [10,11] Ductal differentiation can be found with scrutiny, although the degree of ductal differentiation is typically less conspicuous than poroma. Some lesions contain tubules lined by columnar cells with a “decapitation” pattern along their luminal border; such lesions can be designated as apocrine hidradenoma without question. Not surprisingly, hybrid lesions with features of both hidradenoma and poroma can be encountered, and thus a judgement regarding the “closest fit” must sometimes be made; the hybrid term *poroid hidradenoma* has also been applied to such lesions. [12]

The tumour cells stain positively for LMWK, and the ductal structures/luminal surfaces are highlighted by EMA and CEA. Although these rare tumours do not always

behave aggressively, they may have an aggressive course with metastasis and/or local recurrence. The primary treatment is wide local excision with or without lymph node dissection. [13,14]

Although most cases of hidradenocarcinoma arise de novo, the tumour may also arise in pre-existing hidradenoma. [8] Hidradenocarcinoma is also often referred to as malignant nodular/clear cell hidradenoma, malignant clear cell acrospiroma, clear cell eccrine carcinoma or primary mucoepidermoid cutaneous carcinoma.

Interestingly, clear cell hidradenoma and hidradenocarcinoma may occasionally mimic metastatic clear cell carcinomas including thyroid, lung or renal cell carcinomas. However, the first two are usually distinguished by their positivity to thyroid transcription factor-1 (TTF-1), and the latter by its prominent vascularity, and the presence of haemorrhage and focal granular necrosis within the lesion. [15] Renal cell carcinoma also expresses both EMA and CD10.

The topographic shape of the auricle the convexities and concavities of the cartilage and auricle's blood supply, present a surgical challenge in reconstruction. After primary surgical excision the goal is to recreate an ear that is symmetric to the opposite ear and as normal as possible. The ear must be reconstructed in three dimensions. The location, size and vascularity of the defect dictate the reconstruction plan. [16] After GAVELLOS method description in 1970's, up to now several techniques has been described to restore natural appearance of the ear lobule, but often requires a two stage procedure. [17] Pre and post auricular flaps, [18] super imposition of two opposed or paired flaps, [19] various techniques came into limelight for reconstruction. SLEILATI proposed total ear lobe reconstruction with double crossed

skin flaps. [20] One from pre auricular area and the other from the retro auricular/ or neck skin. In our case we used a doubled over limberg flap (transposition flap). [21] One flap from post auricular skin and another skin flap harvested from under ear lobule tissue. By using limberg flaps procedure good looking ear lobule reconstructed, there is no necrosis at the tip of folded flaps and no ear lobe shrinkage in our follow-up period.

## CONCLUSION

Clear cell hidradenoma of the ear lobule are rare and mostly asymptomatic. These skin tumours are only diagnosable by biopsy, after wide surgical excision reconstruction of ear lobe had done in one stage procedure by using the limberg flaps. This procedure gives cosmetically better ear.

## ACKNOWLEDGEMENT

Authors wish to extend their deepest gratitude to Dr. Rajakullayappa plastic surgeon helped in surgery and follow up period.

### Abbreviations:

LMWK: Low Molecular Weight Keratin  
EMA: Epithelial Membrane Antigen  
CEA: Carcino Embryonic Antigen

## REFERENCES

1. Ishida-Yamamoto A, Sato K, Wada T. *et al* Syringocystadenocarcinoma papilliferum: case report and immunochemical comparison with its benign counterpart. J Am Acad Dermatol 2001;45:755–759.
2. Loane J, Kealy W F, Mulcahy G. Perianal hidradenoma papilliferum occurring in a male: a case report. Irish J Med Sci 1998, 167:26–27.
3. Vang R, Cohen P R. Ectopic hidradenoma papilliferum: a case report and review of the literature. J Am Acad Dermatol 1999, 41:115–118.
4. Hashimoto, K. and W.F. Lever, Histogenesis of skin appendage tumors. Arch Dermatol, 1969. 100(3): p. 356-69.



5. Hashimoto, K. and W.F. Lever, Skin appendage tumors. *Arch Dermatol*, 1970. 101(2): p. 252-3.
6. Gianotti, R. and E. Alessi, Clear cell hidradenoma associated with the folliculosebaceous- apocrine unit. Histologic study of five cases. *Am J Dermatopathol*, 1997.19(4): p. 351-7.
7. Kakinuma H, Miyamoto R, Iwasawa U. *et al* Three subtypes of poroid neoplasia in a single lesion: eccrine poroma, hidroacanthoma simplex, and dermal duct tumor. Histologic, histochemical, and ultrastructural findings. *Am J Dermatopathol* 1994 1666–72.72
8. Crowson A N, Magro C M, Mihm M C. Malignant adnexal neoplasms. *Mod Pathol* 200619(Suppl 2) S93–126.126
9. Patterson J W, Wick M R. Neoplasms and pseudoneoplastic proliferations of the sweat glands. In: Silverbeg SG, Sobin LH, eds. *AFIP Atlas of tumor pathology. Non-melanocytic tumors of the skin*. Washington, DC: Armed Forces Institute of Pathology, 2006137–199.199
10. Wolff, K., R.K. Winkelmann, and R.H. Decker, Solid-cystic hidradenoma: an enzyme histochemical, biochemical, and electron microscopic study. *Acta Dermatol Kyoto Engl Ed*, 1968. 63(4): p. 309-22.
11. Ohnishi, T. and S. Watanabe, Histogenesis of clear cell hidradenoma: immunohistochemical study of keratin expression. *J Cutan Pathol*, 1997. 24(1): p. 30-6.
12. Requena, L. and M. Sanchez, Poroid hidradenoma: a light microscopic and immunohistochemical study. *Cutis*, 1992. 50(1): p. 43-6.
13. Headington J T, Niederhuber J E, Beals T F. Malignant clear cell acrospiroma. *Cancer* 197841641-647.647
14. Helwig E B. Eccrine acrospiroma. *J Cutan Pathol* 198411415–420.420
15. Maeda T, Mori H, Matsuo T. *et al* Malignant eccrine poroma with multiple visceral metastases: report of a case with autopsy findings. *J Cutan Pathol* 199623566–570.570
16. Bob B Armin M.D; Robert O Ruder and Babak Aziza Deh. *Semin Plast Surg*. 2011 Nov; 25 (4) :249–256.
17. Okada.E.; Maruyamma.Y. *Plast Reconstr Surg* 1998, 101; 162 – 166.
18. Khemami.S.; Rannard.F; Kenyon. G; Bilobar post auricular skin flap for reconstruction of ear lobe. *J. Laryngol Otol* 2007; 121; 1094 – 1095.
19. Alconchel. M.D.; Rodrigo. J.; Cimorruga. A combined flap technique for ear lobe reconstruction in one stage. *Br J Plast Surg* 1996; 49; 242 – 244.
20. Sleilati F. Immediate earlobe reconstruction with double-crossed skin flaps. *J Plast Reconstr Aesthet Surg* 2006; 59:1003–1005.
21. Singh A, Singh G. Earlobe reconstruction using Limberg flaps in six ears. *Br J Plast Surg*. 2003; 56:33–36.

How to cite this article: Singh SK, Kumar AR, Neeraja M et al. Clear cell hidradenoma of the ear lobe and its management: a rare case report. *Int J Health Sci Res*. 2015; 5(10):362-367.

\*\*\*\*\*