INTRODUCTION

Clear cell hidradenoma (CCH) is an uncommon benign tumor of eccrine glands and a variant of hidradenoma first described by Liu in 1949. They are usually seen as solitary, well-defined, non-encapsulated, slowly growing tumor in the dermal layer of head, face, and extremities; rarely show aggressive clinical behavior and malignant transformation. The histopathologic features of CCH is often confused with the salivary gland tumors. Very few cases of oral CCH is documented in the literature and we present a rare case of CCH in a 67-year-old female patient in the left buccal mucosa.

CASE REPORT

A 67-year-old, well-oriented, otherwise normal, female patient presented with an asymptomatic swelling in her left cheek of 5-month duration. Intra-oral examination revealed a solitary, well-defined, submucosal nodule measuring around 2 × 2 cm, with intact mucosa unattached to the lesion. This led to the clinical diagnosis of a lipoma. Other differential diagnoses included fibroma and cysticercus cellulose owing to the details of regular use of ham in diet history.

A firm nodule was surgically excised under local anaesthesia in toto. Macroscopic examination showed, an oval shaped solitary lesion, whitish in color, firm in consistency, measuring around 1.5 × 1 cm. The lesion was cystic and filled with yellow, viscous, odorless fluid with white flecks [Figure 1]. Cystic fluid was smeared on glass slide and stained with H and E, which revealed epidermoid cells on a background of eosinophillic fibrillar material similar to keratin. Microscopic evaluation of H and E stained slides revealed cystic lesion showing well defined tumor lobule composed of cellular masses separated by collagenous stroma. The tumor was made up of two type of cell population [Figure 2]. The predominant type of cell was epidermoid with faintly cosinophillic granular cytoplasm and round nucleus. The other type of cell was a large round clear cell with small dark nuclei, found along with duct like spaces [Figure 3]. Cystic areas at few places were lined by 1-3 layer of flat to cuboidal epithelium [Figure 4]. Few areas in the stroma showed chronic inflammatory cells and melanin pigment [Figure 5]. The sections stained with Periodic Acid Schiff (PAS) showed clear cells positive for the stain. All these above features were suggestive of a benign CCH.

DISCUSSION

Eccrine glands are sweat glands present everywhere on the skin except on vermillion zone of lip and nail bed; whereas apocrine glands occur only in the axilla and anogenital region. Eccrine sweat gland has larger lumen than apocrine; whereas apocrine gland produces less-quantity of viscous fluid than eccrine glands. Embryonic stratum germinativum provides...
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Basal cells of epidermis and also primary germ cells for the formation of hair, sebaceous, apocrine, and eccrine glands. Eccrine glands are first seen in 12-13 weeks embryo on palms and soles. By 16 weeks both intra-epidermal and intra-dermal tubular lumina begins to form. At this stage, double layer of epithelial cells are seen in both secretory and ductal components. The double layer is made up of inner luminal and outer basal cells that at later stage, in the secretory region, differentiate into tall columnar luminal cells and myoepithelial cells; myoepithelial cell contraction facilitates excretion of substances by luminal cells. Secretory epithelial cells along with myoepithelial cells are the functional components of salivary and sweat glands. At birth, eccrine glands show nearly equal number of clear cells containing glycogen and darker cells containing PAS positive diastase resistant neutral mucopolysaccharide.

CCH is a benign tumor of skin appendages that arises from the distal excretory duct of eccrine sweat glands on the skin of head, face, and upper extremities.
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Liu in 1949 first described and termed it as eccrine acrospiroma, where “spiroma” means adenoma of sweat glands and “acro” means the topmost or end. This tumor resembles the cells and structures of ductal segment of the eccrine sweat gland.[1]

Other terminologies such as CCH, nodular hidradenoma or solid and cystic hidradenoma are also preferred.[6]

CCH occurs at any age but usually seen in 4th-8th decade of life and are twice more common in females.[7] The tumors are asymptomatic, usually solitary, 0.5-1 cm sized, skin colored intra-dermal nodule that is slightly elevated above the surrounding skin. They are seen as well defined, non-encapsulated, slowly growing tumor in the dermal layer of head, face, and extremities. The lesion may rarely show aggressive clinical behavior along with rapid growth, pigmentation, and ulceration.

Histopathology reveals an encapsulated tumor with both solid and cystic components in varying proportions. The tumor shows tubular lumina of ducts lined by 1-3 layers of cuboidal or columnar cells with or without cystic spaces. Solid portions of tumor show two types of cell population; epidermoid cells are the predominant type of cells, which shows faintly eosinophilic granular cytoplasm and round to oval nucleus. The other type is a pale or clear cell with a round nucleus along with duct like spaces. These clear cells contain glycogen and periodic acid-Schiff–positive, diastase-resistant material. Oncocytic, epidermoid, and pigmented variants of hidradenoma have also been reported.[8] Lund suggested that eccrine tumors of the perioral region could be influenced by “tissue factors” of the oral cavity, thus, consequently displaying some of the histologic components of salivary glands. The secretary and myoepithelial cells of salivary glands and eccrine sweat glands are similar; therefore, tumors arising from these glandular structures will also be similar. However, eccrine tumor shows areas of apocrine decapitation secretory activity, primitive hair follicles and melanin pigmentation.[5] These features signify cutaneous origin of the tumor rather than salivary origin.

Melanocytes are not found in sweat glands so adnexal tumors of skin do not show melanin pigmentation. However, melanocytes are component of sweat duct germs in 14 weeks embryo. Hence, melanogenesis is seen in adnexal tumors of skin.[4]

Malignant transformation of CCH is rare and is marked clinically by a rapid growth with cutaneous ulceration; and histologically by nuclear atypia, increased number of mitotic figures, infiltrative pattern, angiolympathic or perineural invasion with areas of necrosis.[7,9]

Routine haematoxylin and eosin stained sections are usually sufficient for diagnosis and immunohistochemical analysis is not required. Immunohistochemical analysis may become essential in distinguishing CCH from its mimics that may include both benign and malignant lesions. Biernat et al. showed cytokeratin (CK) expression in CCH. Most keratin expression such as CK6/18, CK7, and CK8/18 was noted in squamoid cells and tubule lining cells. Clear cells were consistently positive for epithelial membrane antigen and CK10/17/18.[10]

In conclusion, oral CCH are rare benign tumors. The diagnosis of these tumors is based on routine microscopy and an awareness of similarity of tumors arising from salivary and eccrine sweat glands. CCH can be treated by simple surgical excision and recurrence is rare, while lesion recur after incomplete excision.

REFERENCES

1. Bagga PK, Shahi M, Mahajan N. Clear cell hidradenoma: A case report. Internet J Pathol 2009;8: ISSN: 1528-8307.
2. Shelley WB, Hurley HJ Jr. The physiology of human axillary apocrine sweat gland. J Invest Dermatol 1953;20:285-97.
3. Hashimoto K, Gross BG, Lever WF. The ultrastructure of the skin of human embryos. I. The intraepidermal eccrine sweat duct. J Invest Dermatol 1965;45:139-51.
4. Hashimoto K, Gross BG, Lever WF. The ultrastructure of human embryos skin. II. The formation of the intradermal portion of the eccrine sweat duct and secretory segment during the first half of embryonic life. J Invest Dermatol 1966;46:513-29.
5. Vincent SD, Hammond HL. Nodular hidradenoma: An eccrine sweat gland analog of pleomorphic adenomas of salivary gland origin. J Oral Maxillofac Surg 1987;45:80-3.
6. Elder D, Ellenitsas R, Ragsdale BD. Tumors of the epidermal appendages. In: Lever’s Histopathology of the Skin. 8th ed. Philadelphia, PA: Lippincott-Raven; 1997. p. 747-804.
7. Kaur S, Amanjeeet, Thami GP, Mohan H. Intralesional steroid induced histological changes in the skin. Indian J Dermatol Venereol Leprol 2003;69:232-4.
8. Roth MJ, Stern JB, Hijazi Y, Haupt HM, Kumar A. Oncocytic nodular hidradenoma. Am J Dermatopathol 1996;18:314-16.
9. Biddelstone LR, Mclaren KM, Tidman MJ. Malignant hidradenoma: A case report demonstrating insidious histological and clinical progression. Clin Exp Dermatol 1999;116:474-7.
10. Volmar KE, Cummings TJ, Wang WH, Creager AJ, Tyler DS, Xie HB. Clear cell hidradenoma: A mimic of metastatic clear cell tumors. Arch Pathol Lab Med 2005;129:e113-6.

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