

OXYPHILIC ADENOMA INVOLVING FLOOR OF THE MOUTH: A CASE REPORT

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ABSTRACT:

Oxyphilic adenomas are unusual neoplasm of the head and neck region affecting the glandular epithelial tissue. They were first called as oncocytoma, which is composed of oncocytes and were first described by Schaffer and Hamperl. Oncocytes are polyhedral cells and contain abundant cytoplasm filled with eosinophilic granules. Oxyphilic adenomas are benign neoplasm and represent approximately less than 1% of all salivary gland tumors. They are found generally in the parotid gland, affecting parotid gland region extraorally and rarely encountered in various other sites. Involvement of floor of the mouth is very rare. Presented in this article is a case report of patient suffering from neoplasm, which was histopathologically diagnosed as oxyphilic adenoma.

KEYWORDS: oxyphilic adenoma, floor of the mouth, granular swollen cells, oncocytes, mitochondria.

INTRODUCTION

Salivary gland tumors constitute a heterogeneous group of lesions of a great morphological variation. Although uncommon, they are not so rare. The most common location of the salivary gland tumors are the parotid gland and rarely involving other salivary glands^{1,2,3}.

Oxyphilic adenoma is a benign neoplasm that affects the salivary gland and other glandular tissues and some cases have also been described in the kidneys, thyroid, hypophysis, ovary and bronchi. They manifest as, a well circumscribed tumor with swollen eosinophilic cells known as oncocytes².

Oncocytes are the cells with increased number of mitochondria in their cytoplasm and they resemble the intercalated duct cells. They are thought to be result of degeneration of the salivary gland parenchyma or due to metaplastic process seen in hyperplasia of parenchyma⁴.

Oxyphilic adenomas are commonly encountered in major salivary glands, It is a neoplasm of old age group and more commonly noticed affecting women comparing to men. The neoplasm usually measures 2 to 5 cm in diameter and appears as a discrete encapsulated mass which may also be rarely nodular⁵.

Case report:

A 42-years-old women, reported at Oral and Maxillofacial surgery, Rama dental college, Hospital and Research center, Kanpur. Patient presented with an

asymptomatic growth located in the left side of the floor of the mouth, elevating the tongue, leading to discomfort.

(Fig.1) The duration of the growth was since 2 years. History given by the patient was, the growth started as small swelling which gradually attained the present size and remained same from past one year.

Clinically growth was firm in consistency with smooth, shiny surface texture and normal pinkish in color. Growth showed well defined margins ranging from lingual alveolar mucosa to retromandibular region anterioposterly and mesiodistally from lateral to midline of alveolar mucosa measuring approximately about 4 cm X 1.5 cm. Palpation of the mass revealed firm consistency which was non tender with submental and submandibular lymph nodes palpable. Based on these features, clinical differential diagnosis was given as plexiform neurofibroma and neoplasm of the minor salivary glands.

Incisional biopsy of the tumor was done. The specimen fixed in 10% formaldehyde solution was sent to the Oral Pathology Laboratory of Rama dental college, Hospital and Research center, Kanpur, where the histopathological analysis was carried out. Hematoxylin and Eosin stained section showed tumor cells arranged in sheets, rows and chords, which showed the presence of alveolar and lobular pattern(**Fig.2**). On higher magnification tumor cells were large with eosinophilic cytoplasm and distinct cell membrane. Few mitotic figures were noticed in the section. Included in the histological framework, there were some salivary gland



Fig.1.Swelling located in the left side of the floor of the mouth, elevating the tongue.

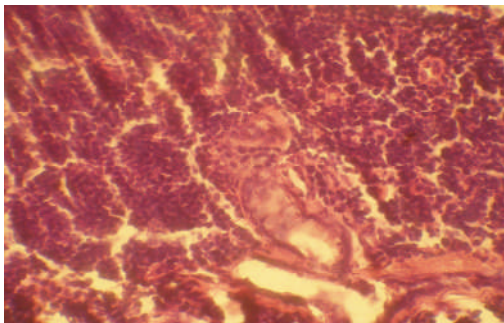


Fig.2. Tumor cells arranged in sheets and chords.

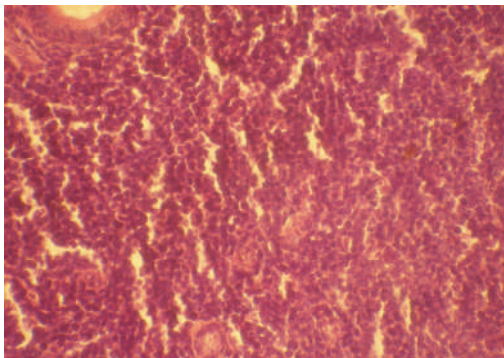


Fig.3.On higher magnification tumor cells were large with eosinophilic cytoplasm and distinct cell membrane.

acini with lymphocytes and hyperemic blood vessels. These histopathological findings are compatible with the diagnosis of oxyphilic adenoma (**Fig.3**).

Discussion:

Oncocytic lesions were described nearly a century ago by Schaffer where he described "granular swollen cells" in the ductal and acinar elements of salivary glands. McFarland in 1927 described a tumor as an "adenoma," but the description and illustrations certainly appeared to demonstrate an oncocytoma of the parotid gland².

Hamperl is considered to be the "father" of oncocytes, originally referred to as "onkocytes." He has designated "oncocyte" from Greek word onkosthai (swollen) and cytos (cell) as a special type of epithelial cell characterized by a larger than the original cell, with a mitochondria-rich considerably dense cytoplasm containing acidophilic granules. Hamperl described oncocytes in many organs, including parotid, submaxillary, sublingual, and minor salivary glands, the thyroid, parathyroid, pituitary gland, adrenal gland, gallbladder, uterus, testicle, fallopian tube, pancreas, liver, stomach, kidney, lung, pharynx, trachea, and esophagus⁶.

Oncocytes are also been called oxyphilic cells, Askanazy cells and Hurthle cells in the thyroid gland. The parotid gland is the most common site where oncocytic changes may occur, usually at the ductal or acinar cell level⁷. They are one to two times the size of normal acinar cells, display abundant granular eosinophilic cytoplasm and a central pyknotic nucleus. The cytoplasmic granularity is due to the accumulation of mitochondria that may occupy up to 60% of the cytoplasm. In contrast, mitochondria occupy only 5.2% of the cytoplasm of normal acinar cells⁸.

The World Health Organization (WHO) classification of Salivary gland neoplasms recognizes three oncocytic entities: oncocytosis, oncocytoma and oncocytic carcinoma. Oncocytosis is considered to be a hyperplastic change and may present with generalized enlargement of the glands whereas oncocytoma and oncocytic carcinoma represent neoplastic processes and in agreement with the AFIP classification, oxyphilic adenoma is considered a benign neoplasia of epithelial origin⁹.

Oncocytic changes are noted with increasing age and are almost a universal finding in individuals around the seventh decade of life. Oncocytic changes of secretory epithelia are thought to be metaplastic, a protective phenomenon against adverse change. Hamperl considered oncocytes to be burnt out cells as they lost their original specialization and increased in number with age¹⁰.

Bonikos DS et al suggested that oncocytic change may be the result of compensatory mitochondrial hyperplasia in normal cells caused by mitochondrial damage or the exhaustion of one or more mitochondrial enzymes. Linnane et al advocated that aging caused the accumulation of mitochondrial DNA errors leading to "mitochondrial respiratory failure" and multisystem degeneration¹¹.

Sunmunn et al stated that oncocytic change could be a regressive alteration of previously hypertrophic or hyperplastic ductal epithelium with the appearance of a mitochondriopathy⁷.

Oxyphilic adenoma of salivary glands most commonly involves the parotid gland (82%) and the rest are located at the submandibular gland and minor salivary glands. They are typically tumors of older adults. However, they may rarely present in children and middle age. A slight female predominance is noted for the conventional Oxyphilic adenoma and a more marked preponderance for females in the clear cell variant of Oxyphilic adenomas^{1,9,7}.

Oncocytomas most often occur as asymptomatic, well-circumscribed, solitary, painless masses usually measuring 3 to 4 cm in size, but may reach up to 7 cm in diameter. Rarely, they may present with pain or discomfort. They may also occur as multifocal or bilateral neoplasms. The differential diagnosis for oncocytic lesions in the salivary gland include; papillary cystadenoma lymphomatosum, acinic cell carcinoma, clear cell carcinoma, oncocytic carcinoma and mucoepidermoid carcinoma^{1,5}.

Microscopically the tumor is seen as solid cords or clusters of tightly packed oncocytes. A thin strand of fibrovascular stroma is present separating the cells. Scattered lumina of variable sizes, of which some with associated eosinophilic intraluminal secretions may be prominent. The cells are large cuboidal to columnar in shape and arranged in an organoid pattern with prominent eosinophilic, finely granular cytoplasm and round nuclei. The eosinophilia is variable and hence there may be an admixture of light and dark stained cells. Occasionally cystic changes are noted in oxyphilic adenomas.

Treatment is done by surgical resection of the tumor. Probably, there is no need for chemotherapy and/or irradiation because of the benign nature and slow growth rate of the tumour. Recurrence is less than 20%, mainly because of incomplete surgical resection¹².

CONCLUSION:

Oxyphilic adenoma is a rare, benign tumors of glandular tissue origin. The neoplastic cells are filled with eosinophilic granules like mitochondria, which are easily demonstrated histopathologically. Complete surgical resection is adequate therapy, with less recurrence rate.

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