

CASE REPORT

Oncocytoma of minor salivary gland involving the retromolar region: A rare entity

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ABSTRACT

Oncocytic neoplasms comprise a group of rare tumors of salivary glands. Histologically, World Health Organization (WHO) (1991) classified them into three distinct types: Oncocytosis, oncocytoma and oncocytic carcinoma. Oncocytoma is also known by the more descriptive and less confusing terms of oxyphilic adenoma and oxyphilic granular cell adenoma. Oncocytoma is a rare benign salivary gland neoplasm composed of large epithelial cells with characteristic bright eosinophilic granular cytoplasm (oncocytic cells). It accounts for approximately 0.4-1% of all salivary gland neoplasms, occurring primarily in parotid glands, with only a small percentage occurring in minor salivary glands of palate, tonsillar fossi, larynx, nasal cavity, maxillary sinus and the lacrimal gland. It occurs primarily in persons older than 50 years of age. According to some authors, the biologic behavior of oncocytomas arising from minor salivary glands is that of an aggressive nature and should be considered as low-grade malignancies. Only 17 cases of histologically verified oncocytoma of an intraoral minor salivary gland are reported in literature, the current case being the 18th. Owing to the lack of large series, assiduous study of the cases reported in the literature may lead to better understanding of this rare entity. We herein describe a rare and probably the first case of minor salivary gland oncocytoma involving retromolar area in a 32-year-old female patient with a brief review of literature.

Key words: Minor salivary gland, oncocytoma, retromolar

INTRODUCTION

Oncocytic neoplasms comprise a group of rare benign neoplasm of salivary glands accounting for approximately 0.4-1% of all salivary gland neoplasms, occurring primarily in parotid glands in persons older than 50 years of age.^[1] Oncocytomas are designated as oxyphilic granular adenoma, oxyphilic adenoma and eosinophilic adenoma.^[2] Oncocytomas arise very rarely from the minor glands of tonsillar fossi, larynx, nasal cavity, maxillary sinus and the lacrimal gland.^[3] Reports of well-documented intraoral lesions arising from minor salivary glands are very rare.

In this report, a case of oncocytoma arising in intraoral minor salivary glands in the right retromolar area of a 32-year-old female patient is presented.

CASE REPORT

A 32-year-old female patient reported with a chief complaint of a large swelling in her right back teeth region since several months. Clinical examination revealed a 2 × 1.5 cm² in dimensions, well-circumscribed, lobulated and pinkish mass on the right retromolar area [Figure 1]. The swelling was soft in consistency and nontender on palpation. There was no paresthesia, facial asymmetry or regional lymphadenopathy noted. Normal complement of dentition was seen. Provisional diagnosis of irritational fibroma was made. Differential diagnosis included peripheral ossifying fibroma, peripheral giant cell granuloma, pyogenic granuloma, mucocele, lipoma, other benign mesenchymal tumors and benign salivary gland neoplasms. Following written consent from the patient, the growth including the normal surrounding tissue was excised under local anesthesia. Postoperative period was uneventful.

Excisional biopsy specimen was fixed in 10% formalin and sent to the Department of Oral Pathology and Microbiology for histopathological examination. Histopathologically, hematoxylin and eosin stained sections revealed a well-circumscribed tumor composed of granular cells that were arranged in solid

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sheets, alveoli, nests, columns or cords with scattered focal clear cells throughout the tumor tissue, separated by a delicate fibrovascular network [Figure 2]. On higher magnification, the tumor cells were polyhedral to round with distinct cell borders. Abundant cytoplasm, with centrally located nucleus and little mitotic activity were seen [Figure 3]. Features were in favor of oncocytoma rather than fibroma. To confirm the diagnosis, phosphotungstic acid-hematoxylin (PTAH) staining was carried out, which revealed abundant coarse cytoplasmic granules that correspond to abundance of mitochondria [Figure 4]. The final diagnosis was determined as oncocytoma of minor salivary glands of the retromolar area.

DISCUSSION

Oncocytic lesions were described nearly a century ago by Schaffer when he described “granular swollen cells” in the ductal and acinar elements of salivary glands. Although in 1927, McFarland described a tumor as an “adenoma”, without specifically calling it an oncocytoma, the description and illustrations certainly appeared to demonstrate an oncocytoma

of the parotid gland.^[4]

Hamperl has designated “oncocyte” (from Greek; onkousthai = 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. Oncocytes can be found in normal organs, as in tumors, like major and minor salivary glands, the thyroid, parathyroid, pituitary gland, adrenal gland, gall bladder, uterus, testicle, fallopian tube, pancreas, liver, stomach, kidney, lung, pharynx, trachea and esophagus.^[4] The term “oncocytoma” was first introduced by Jaffe in 1932, referring to the lesion currently called adenolymphoma or Warthin’s tumor.^[5]

The World Health Organization (WHO) (1991) 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 Armed Forces Institute



Figure 1: Intraoral picture revealing a solitary swelling in the retromolar region

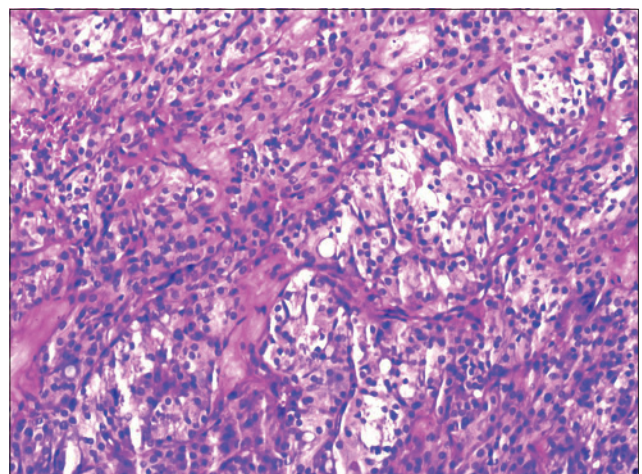


Figure 2: Photomicrograph showing neoplastic cells arranged in solid sheets, nests, columns/cords and separated by fibrovascular network (H&E stain, ×100)

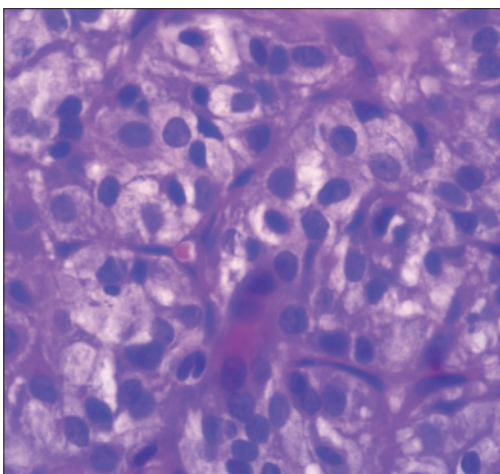


Figure 3: Photomicrograph showing polygonal tumor cells with centrally placed nuclei and abundant dense granular eosinophilic cytoplasm (H&E stain, ×400)

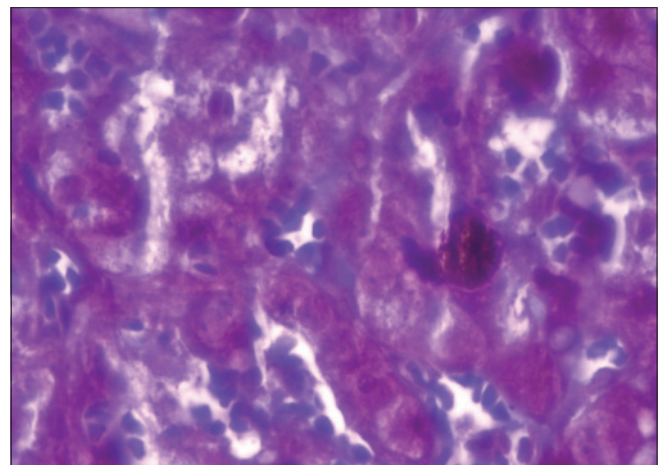


Figure 4: Photomicrograph showing abundant cytoplasmic granules (PTAH stain, ×400). PTAH = Phosphotungstic acid-hematoxylin

of Pathology (AFIP) classification, oxyphilic adenoma is considered a benign neoplasia of secretory epithelial origin.^[4]

It is generally accepted that for a tumor to be called an oncocytoma, it must comprise exclusively of oncocytes. Even though oncocytes are considered to contain abundant eosinophilic cytoplasm, cases with clear cell changes have been described in the literature.^[6]

There are no definitive etiologic factors for this tumor, although there has been an association with radiation in some reports.^[6]

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 age.^[4]

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 deoxyribonucleic acid (DNA) errors leading to “mitochondrial respiratory failure and multisystem degeneration”.^[4]

Sunmunn *et al.*, stated that oncocytic change could be a regressive alteration of previously hypertrophied or hyperplastic ductal epithelium with the appearance of a mitochondriopathy.^[4]

Till date, the PubMed and the Medline search has revealed only 17 reported cases of histologically verified oncocytoma of an intraoral minor salivary gland. The current case is the 18th and the first case to be reported in the retromolar region. Their sex, age and site distribution, together with the current case is summarized in the Table 1.^[2,3,5,7]

Of the 17 cases reported, 58.8% (10) were women and the age distribution of the reviewed cases at diagnosis varied from 23 to 77 years, with a mean age of 53 years. However, studies of oncocytomas arising in major salivary glands have shown that there is an equal predilection for both sexes and predilection for older individuals, with a peak incidence in the seventh to ninth decades of life. The most frequent site of origin of minor salivary gland oncocytoma is the hard and soft palate (52.9%), followed by buccal mucosa (35.3%).

Microscopically, the tumor is seen as solid cords or clusters of tightly packed oncocytes and a thin strand of fibrovascular stroma is present separating these 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 are arranged in an

Table 1: Reported cases of minor salivary gland oncocytomas

Author	Age/sex	Site
Ahlbom (1935) ^[3]	59/F	Hard palate
Miyoshi <i>et al.</i> , (1963) ^[3]	53/F	Hard palate
Sato and Watanabe (1964) ^[3]	23/F	Hard palate
Jalisi (1968) ^[3]	42/F	Oropharynx
Crocker <i>et al.</i> , (1968) ^[3]	77/F	Palate
Te-i (1969) ^[3]	27/M	Hard palate
Mastuda <i>et al.</i> , (1974) ^[3]	53/F	Buccal mucosa
Kohno (1981) ^[3]	59/M	Hard palate
Hayashi <i>et al.</i> , (1985) ^[3]	62/M	Hard palate
Hayashi <i>et al.</i> , (1985) ^[3]	64/M	Soft palate
Regezi <i>et al.</i> , (1985) ^[3]	63/F	Buccal mucosa
Chau and Radden (1986) ^[3]	58/M	Buccal mucosa
Damm <i>et al.</i> , (1989) ^[3]	73/F	Buccal mucosa
Kochhar L (1990) ^[7]	45/F	Hard palate
Kanazawa <i>et al.</i> , (2000) ^[3]	32/F	Buccal mucosa
Camara AC (2005) ^[5]	71/M	Jugal (buccal) mucosa
Nergiz Yilmaz (2010) ^[2]	72/M	Maxillary posterior alveolus
Present case	32/F	Retromolar area

M: Male; F: Female

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.^[4] The histological features observed in the present case were satisfactory for the diagnosis as oncocytoma.

Oncocytoma’s clinical behavior is generally benign. From their clinical appearance, the lesions were often indistinguishable from other benign tumors as seen in the present case. Most reported cases were treated by surgical excision without recurrence. However, a recurrence rate of 20% has been reported due to incomplete surgical eradication or occult multifocality. For this reason, oncocytomas of the minor salivary glands should be carefully removed, together with a small margin of uninvolved healthy soft tissue, because these lesions occasionally have a thin incomplete capsule and rare malignant cases of oncocytic carcinoma have been reported.^[2]

Though the prognosis after tumor excision is excellent, Gray *et al.*, in their review of the literature reported six oncocytomas arising from the seromucous glands in the nasal mucosa or the palate that were more aggressive than the typical oncocytomas but were not malignant. They classified the group as “locally aggressive oncocytomas”, in which local infiltration of the capsule, scattered mitotic figures and focal cellular pleomorphism were seen. According to some authors, the biologic behavior of oncocytomas arising from minor salivary glands in the nasal cavity and maxillary sinuses have an aggressive nature and should be considered as low-grade malignancies.^[3]

Oncocytic cells are associated with other definable major salivary gland tumors. Oncocytic differentiation has been observed in pleomorphic adenoma, monomorphic adenoma,

Table 2: Histopathological differential diagnosis of oncocytoma

Lesion	Histopathological features
Oncocytoma	Well-circumscribed oncocyte arranged in solid sheets or in nests and cords, which form alveolar or organoid patterns. Fibrovascular septa with thin-walled blood capillaries. Centrally located nucleus. ^[10]
Warthin's tumor	Combination of lymphoid matrix and papillation lined by two layers of oncocytic epithelial cells forming cystic spaces. ^[10]
Oncocytic carcinoma	Overwhelming evidence of nuclear and cellular pleomorphism, increased or abnormal mitotic figures, perineural and vascular invasion, infiltration into connective tissue and regional lymph node metastasis. ^[10]
Mucoepidermoid carcinoma	Epidermoid, intermediate and mucous cell differentiation. Clear cells if predominant are often associated with areas that show epidermoid differentiation. Cystic growth pattern in low- and intermediate-grade tumors. ^[10]
Acinic cell carcinoma	Presence of acinar differentiation. Classic patterns of acinic cell carcinoma like solid, microcystic, papillary cystic, follicular and macrocystic. Neoplastic acinar cells contain abundant finely granular cytoplasm, which may be amphophilic, pale eosinophilic or basophilic with an eccentrically placed nuclei. ^[10]
Clear cell adenocarcinoma	Conspicuous hyalinized collagenous stroma. Poorly circumscribed with an infiltrative growth pattern with involvement of peripheral nerves. ^[10]
Metastatic renal cell carcinoma	Organoid pattern of arrangement. Blood lakes and prominent stromal vascularity. Nuclear pleomorphism in high-grade tumors. Infiltrative growth pattern. ^[11]
Granular cell ameloblastoma	Cells located in the central portion of the follicles have granular eosinophilic cytoplasm with eccentrically placed nuclei and the peripheral cells resemble ameloblasts. ^[12] Mostly found as an admixture with follicular subtype. The follicles (epithelial component) are embedded in a mature connective tissue stroma. ^[13]

basal cell adenoma, canalicular adenoma, myoepithelioma, cystadenoma (lipoadenoma), adenoid cystic carcinoma, acinic cell carcinoma, cystadenocarcinoma, adenocarcinoma and much less frequently in mucoepidermoid carcinoma.^[8] Synchronous oncocytoma and Warthin's tumor in the ipsilateral parotid gland has been reported in the literature.^[9]

There have been rare examples of malignant oncocytic tumors and the criteria for malignancy includes capsular invasion, destructive growth, necrosis, increased pleomorphism, lymphatic or distant metastasis, vascular/neural invasion and mitotic figures and so forth.^[1] Except for few mitotic figures, the above features were absent in the present case.

The histological differential diagnosis for oncocytic lesions in the parotid gland include papillary cystadenoma lymphomatosum (Warthin's tumor), acinic cell carcinoma, clear cell carcinoma, oncocytic carcinoma, mucoepidermoid carcinoma, metastatic renal cell carcinoma and granular cell ameloblastoma. The oncocytoma is differentiated from its mimickers by feature enlisted in the Table 2.^[10-13]

The clinical, histological and histochemical data from the reported case conform to that in the literature.

CONCLUSION

This oncocytoma of minor salivary gland origin was, to our knowledge, the 18th and the first case to be reported, arising in the right retromolar area. Owing to the lack of large series, industrious study of the cases reported in the literature may lead to better understanding of this rare entity.

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