Case Report

Multifocal adenomatous oncocytic hyperplasia of the parotid gland: A report of two cases with review of the literature

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ABSTRACT

Multifocal adenomatous oncocytic hyperplasia (MAOH) is a rare non-neoplastic condition of the parotid gland. It is also referred to as "Oncocytosis." It accounts for approximately 0.1% of all the salivary gland lesions. The existence of this entity needs more understanding compared to its counterpart oncocytoma due to limited identification and studies. Here, we report a case series of two cases of MAOH in a 63-year-old man and in a 54-year-old woman over the left and right parotid region, respectively, both clinically operated for malignant parotid tumors. Fine-needle aspiration of both the lesions was reported with differentials of oncocytic rich lesions oncocytoma, Warthin's tumor, and an oncocytic variant of mucoepidermoid carcinoma. Histology was proven as MAOH as it showed multiple unencapsulated nodules composed of oncocytes. Cytology alone is not sufficient for its diagnosis as they overlap with other oncocytic rich lesions. Awareness of this entity is essential for surgeons and practicing pathologists as it is a benign condition, thereby avoiding extended surgeries.

Key words: Multifocal adenomatous oncocytic hyperplasia, Oncocytosis, Parotid gland

Greek work "Onco" refers to bulk, is the transformed epithelial cells seen in various organs such as kidneys, thyroid, salivary glands, ovary, adrenals, liver, breast, and many more. These cells are characterized by the presence of an excessive number of mitochondria giving them an intense eosinophilic granular cytoplasm. They are called by various names such as Hurthle cells, oxyphilic cells, and Askanazy cells. Oncocytes were first described in the oncocytoma of salivary glands by Jaffe in 1932 [1]. In salivary glands, the oncocytes are commonly seen in conditions such as Warthin's tumor, oncocytoma, oncocytic carcinoma, and oncocytosis. They can also be seen in various other conditions such as pleomorphic adenoma and mucoepidermoid carcinoma. Oncocytosis, also referred to as multifocal adenomatous oncocytic hyperplasia, is a rare entity and accounts for about 0.1% of all parotid tumors [2].

CASE REPORT

Case 1

A 63-year-old man presented to the ENT clinic with complaints of swelling over the left parotid region for 3 months. The swelling was associated with a gradual increase in size and was not associated with pain, fever, ulcer, discharge, localized, or cervical lymphadenopathy.

The general examination revealed that the vitals were stable. On local examination, the swelling was about 3 cm × 2.5 cm in size with well-defined margins, soft to firm in consistency, and non- tender. It was freely mobile and not associated with a local rise of temperature. The overlying skin appeared stretched out. Computed tomography (CT) findings done in the gentleman showed the left parotid swelling with multiple solid masses and cysts with well-defined margins, the largest measuring 1.5 cm in diameter. Few nodules were also mentioned in the tail of the parotid as possible lymph node metastasis.

Case 2

A 54-year-old woman presented to the ENT department attached to a hospital with complaints of swelling over the right parotid region for 4 months. It was gradually progressive in size and not associated with pain, fever, localized, or cervical lymphadenopathy.

General examination and vitals were within normal clinical limits. On local examination, the swelling was about 2.5 cm × 2 cm in size with well-defined margins, soft in consistency, and non-tender. It was freely mobile and not associated with a local rise of temperature. No enlarged cervical lymph nodes were

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identified. CT findings done showed the right parotid swelling with multiple solid masses and well-defined margins, the largest measuring 1.3 cm in diameter.

Investigation and treatment done for both cases

Fine-needle aspiration was performed in both the cases and showed smears rich in oncocytes on a hemorrhagic background. Differential diagnosis of oncocytoma, Warthin's tumor, and an oncocytic variant of mucoepidermoid carcinoma were given.

Superficial parotidectomy was performed in both cases. The resected specimen consisted of tissue measuring about 4.5 cm × 4 cm × 1 cm and 3.5 cm × 1.5 cm × 1 cm, respectively. Cut section in both the specimens showed multiple, well-circumscribed mahogany brown nodules which grossly resembled that of a normal lymph node (Fig. 1). Case 1 lesion measured about 3.2 cm × 2.5 cm × 0.8 cm. Case 2 lesion measured about 2.8 cm × 1.2 cm × 0.6 cm. Few cystic spaces were also noted amidst these nodules. Microscopically, both the lesions showed multiple circumscribed nodules composed of sheets, nests, and trabecular arrangement of bland appearing oncocytic cells separated by normal-appearing salivary ducts and acini (Fig. 2). These nodules were unencapsulated and seen amidst the salivary acinar cells. The cystic spaces were lined by bilayered oncocytic cells. No pleomorphism, mitosis, or necrosis were noted. Sections from the tail of the parotid also showed similar histological findings. Both the patients are on routine follow-up for 6 months and are disease free.

DISCUSSION

The World Health Organization has classified oncocytic lesions into three distinct histological types, namely, oncocytosis, oncocytoma, and oncocytic carcinoma [2]. Various studies on oncocytoma and oncocytic carcinoma of the parotid gland have been reported. In this context, oncocytosis of the parotid gland has been rarely studied and reported. To the best of our knowledge, only eight cases have been previously reported in the literature, and in our subcontinent, this is the second case report on oncocytosis of the parotid gland followed by Sah and Toran.

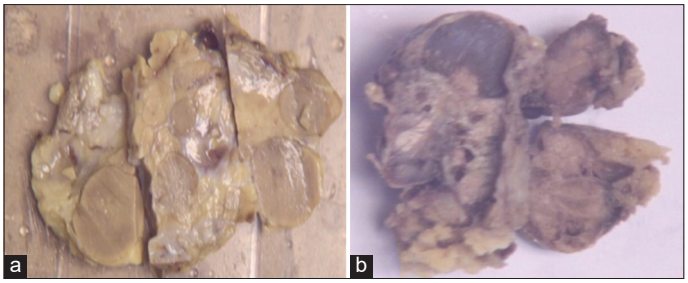

Figure 1: Parotidectomy showcasing multiple mahogany solid and cystic lesions in (a) case 1 and (b) case 2

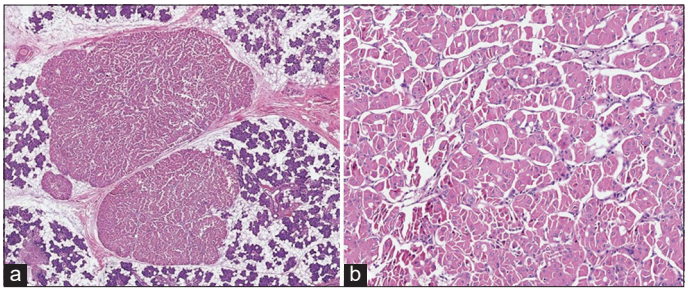

Figure 2: (a) Light microscopy showing multiple circumscribed unencapsulated nodules comprised of oncocytes (case 1); (b) high-power view showing bland appearing sheets of oncocytes (case 2)

Oncocytic cells are metaplastic cells formed in response to adverse changes within the cells or due to aging that cause functional exhaustion of mitochondrial enzymes, leading to compensatory hyperplasia of mitochondria, which, in turn, is responsible for the typical microscopic change. Indeed, microscopically, they can be incidentally found as single cells in aging salivary tissue, with a reported incidence of 80% presence in persons older than 70 years of age or as multiple macroscopic nodules [3,4].

The closest differentials are oncocytoma and oncocytic carcinoma. In addition, oncocytic change can be seen in a few of the other parotid tumors such as pleomorphic adenoma, Warthin's, basal cell adenoma, cystadenoma, acinic cell carcinoma, and mucoepidermoid carcinoma. In contrast, oncocytoma comprises bland oncocytes presenting as a single circumscribed encapsulated nodule in an organoid pattern with compression of adjacent structures [4-6]. Oncocytic carcinoma will show multiple infiltrative islands and nests of pleomorphic oncocytic cells with the presence of capsular invasion and vascular invasion. Young and Warfield reported that differentiating oncocytosis from other oncocytic lesions of the parotid gland on cytology is difficult, and on some occasions, even impossible. Therefore, it can only be diagnosed on histopathology [7].

The cytological findings in Warthin's will show the presence of a good number of lymphoid cells accompanied by oncocytes. The cytological findings in mucoepidermoid carcinoma will show the presence of mucin-secreting cells, atypical squamoid cells on a dirty background in addition to oncocytes [8]. Most cytological findings of multifocal adenomatous oncocytic hyperplasia (MAOH) are not reported. Goyal *et al*. described the cytological findings of MAOH as cells having: A low N/C ratio; central round nuclei; anisonucleosis; prominent nucleoli (in part of the cells); and abundant eosinophilic cytoplasm. In the present case, the tumor cells had similar findings to those of Goyal *et al*. [9]

Oncocytosis of the parotid gland was first described by Schwartz and Feldman in the year 1969 [10]. It is commonly seen in women and in the sixth decade [5]. The clinical features may include swelling and tenderness of the parotid region of 4 months–25 years duration [5,9]. The nodules, unlike oncocytoma, have incomplete capsules. The growth of the small nodules may cause their confluence. The multinodular growth may suggest malignancy, but the presence of oncocytic proliferation in intercalated ducts and ductules in the surrounding aids in recognizing the benign condition [5,6].

Shellenberger *et al*. found that oncocytic metaplastic nodules demonstrate an inhomogeneous mild degree of enhancement on CT findings, whereas the oncocytoma nodules showed a well-defined mildly enhancing tumor with compression of the

surrounding parenchyma [11]. Shellenberger *et al*. and Capone *et al*. have reported the presence of synchronous oncocytosis and oncocytoma in a single parotid specimen [11,12].

The standard treatment is parotidectomy with preservation of the facial nerve. The patients should be regularly followed up for any local recurrences arising from the residual tumor in cases of incomplete resection. A peripheral residual clearance of about 5 mm should be done during the parotidectomy procedure for such cases.

CONCLUSION

This is the first case series in our subcontinent highlighting two cases of MAOH. It recommends that when multiple small nodules are found in the parotid gland with a large solid or cystic mass evident on CT or magnetic resonance imaging, a diagnosis of nodular oncocytic hyperplasia and oncocytoma should be considered, particularly in patients 60 years of age or older. After parotidectomy for oncocytic hyperplasia, patients should be followed for recurrence arising from residual parotid tissue.

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