CASE REPORT

Multifocal adenomatous oncocytic hyperplasia of the parotid gland

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Summary Oncocytes appear as the principal component in some pathological conditions of the salivary glands including diffuse oncocytosis, oncocytic metaplasia, oncocytoma, oncocytic carcinoma, and multifocal adenomatous oncocytic hyperplasia. Oncocytic tumors rarely affect the major salivary glands, accounting less than 1% of all salivary gland tumours. Multifocal adenomatous oncocytic hyperplasia in the salivary glands is an extremely rare condition. The differential diagnosis of the clear cell variant includes many malignant salivary gland tumors.

We report a new case of multifocal adenomatous oncocytic hyperplasia of the parotid gland in a 64-year-old man. The differential histopathogenesis, diagnosis, and treatment of this rare entity are discussed.

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Introduction

Oncocytes represent a special class of transformed epithelial cells. They were first recognized by Shaffer in 1897,1 but later named and more fully characterized by Hamperl in 1931.2 The term oncocyte derives from the Greek word onkousthai, meaning “to swell”. Oncocytes arise principally from glandular and secretory epithelia, and they are characterized by their large size and abundant, finely granular eosinophilic cytoplasm which is rich in mitochondria.3 Because oncocytes are strongly eosinophilic, the alternate term oxyphilic cells has also been applied.

The term oncocytoma was first introduced by Jaffé in 1932 to designate tumors of the salivary glands that consist predominantly of oncocytes,4 although the lesion he originally referred to is actually called adenolymphoma (papillary cystadenoma lymphomatous or Warthin tumor).

Oncocytoma is similar to adenolymphoma of the salivary glands, except for the lack of lymphoid tissue. Oncocytomas exist in many different organs such as major and minor salivary glands, endocrine

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glands, kidneys, breasts, testes, buccal mucosa, the upper respiratory tract, and ocular annexa. Because of marked histologic similarities of oncocytoma arising from different organs, Hamperl referred to oncocytic transformation as a form of "convergent differentiation". He also depicted oncocytes as "burnt out" cells because they lost their original specialized function and increased in number with age.

Oncocytomas are benign tumours making up 0.5–1% of all parotid gland tumours. Multifocal adenomatous oncocytic hyperplasia (MAOH) is a different condition representing a multifocal oncocytic proliferation of the duct system occurring very rarely. MAOH makes up 0.1% of the parotid gland diseases.

Case report

A 64-year-old man was admitted to our Department with a small swelling in the left parotid region. This had appeared about eight months earlier and gradually increased in volume. There was no pain, dryness of mouth or swelling of the tumour related to meals. No signs of infection or general symptoms were present. Clinical examination revealed a non-well-circumscribed soft mass with a diffuse border, located behind the angle of the mandible (Figs. 1 and 2). There was no adhesion to the skin,

Figure 1  A small swelling in the area of the left parotid gland.

Figure 2  A small swelling in the area of the left parotid gland.

Figure 3  Intra-operative view during the dissection.
but its deep boundary and its relation to the parotid gland were difficult to determine. There were no palpable cervical lymph nodes, and no pathological changes were detected clinically in the other parotid gland.

Pre-operative sonography revealed multiple cystic structures, measuring 8–10 mm in diameter in the superficial lobe.

The general physical data and the admission laboratory tests revealed no abnormalities.

As the fine needle aspiration biopsy was insufficient, a superficial parotidectomy was performed (Fig. 3).

The surgical specimen (Fig. 4) consisted of multiple fragment which together measured 3 × 4 × 6 cm. The tissue appeared slightly indurated and included cysts up to 8 mm in diameter.

On microscopic examination, a slight decrease in the number of acinar structures was seen and some interstitial fat cells were haphazardly intermingled. Multiple cysts were found, with a two-layered epithelial lining of oncocytic cells. Multiple solid foci of oncocytic cells, arranged in tubulo-acinar structures, were also seen. The oncocytic cells appeared with abundant eosinophilic cytoplasm forming compact, non-encapsulated complexes. The tumor cells showed no pleomorphism or hyperchromatism of the nuclei, and no mitoses were seen. No haemorrhages or necroses were found (Figs. 5 and 6).

Figure 4  Macroscopic appearance of the resected tumor. The tissue appeared slightly indurated and included cysts.

Figure 5  Micrograph of the tumor. Multifocal adenomatous oncocytic hyperplasia showing nodules of clear cells. Although oncocyes appear infiltrative in some areas, the nodules still respect the normal lobular architecture of the parotid (hematoxilin–eosin stain, original magnification 40×).

Figure 6  Micrograph of the tumor. Oncocytic focus including microcysts. The constituent cells display oncocytic features with abundant eosinophilic cytoplasm (hematoxilin–eosin stain, original magnification 100×).
The post-operative course was uneventful and, at the most recent follow-up examination, 15 months post-operatively no signs of recurrence were found.

Discussion

Oncocytes can occur in both normal organs and in neoplasms. Their formation is a special degenerative metaplasia that does not prevent the cells from dividing.3

The histological distinguishing feature of an oncocyte is intensely eosinophilic granular cytoplasm, typically due to increased numbers of mitochondria. As other cytoplasmic structures can cause acidophilia, additional distinguishing criteria have been proposed.8 These include: high levels of oxidative activity, variation in the size and shape of mitochondria, absence of dense granules in the intra-mitochondrial matrix, and regression of electronmicroscopical features such as brush borders and basal-infoldings.

Oncocytes appear as the principal component in some pathological conditions of the salivary glands including diffuse oncocytosis, MAOH, oncocytoma and oncocytic carcinoma.7 This may occasionally raise differential diagnosis problems. All these lesions are predominantly seen in the parotid gland. Oncocytic cell complexes can also appear in pleomorphic adenoma. The most frequently seen oncocytic tumor of salivary glands is Warthin’s tumor (cystadenolymphoma).7,9

In the normal tissue of the salivary glands oncocytes occur sporadically among the normal ductal and acinar cells and more frequently with advancing age.10

MAOH in the salivary glands is an extremely rare condition.11 It is mainly diagnosed in women and in the sixth decade.7,12 The clinical features may include swelling and tenderness of the parotid region of 4 months to 25 years duration.12,13 Characteristically, MAOH is described as multinodular non-encapsulated oncocytic foci measuring 0.1–1 cm, containing inclusions of salivary gland remnants, occasionally with ductal oncocytic metaplasia. The nodules consist of bland, polyhedral cells with round, centrally located nuclei, eosinophilic, granular cytoplasm and conspicuous cell borders. Clear cells are rarely present. No mitoses are seen. The nodules, unlike oncocyte, have incomplete capsules. 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 surroundings aids in recognizing the benign condition.6,14

To differentiate oncocytic adenomatous hyperplasia from diffuse oncocytosis and oncocytic adenoma, the quantity of oncocytes serves as a discriminating feature. In contrast to these oncocytic lesions, oncocytic adenomatous hyperplasia is characterized by focal accumulation of only few oncocytes, without capsule formation. Oncocytic adenomatous hyperplasia shares the oncocytic epithelium and the formation of cystic spaces with Warthin’s tumor. The latter can be clearly distinguished from oncocytic adenomatous hyperplasia by the surrounding lymphoid tissue, which frequently forms germinal follicles, the frequent papillary projections of the oncocytic epithelium and the thin capsule.15

MOAH is often interpreted as invasive clear cell tumours, particularly clear cell variants of acinic cell carcinoma or secondary renal clear cell carcinomas.16,17 The differential diagnosis of the clear cell variant includes many malignant salivary gland tumors.18

MOAH should be regarded as a benign lesion, best treated by surgery. Recurrences may be expected in cases of partial resection. The lesion is not responsive to irradiation and the possibility of radio-induced malignant transformation should be considered.6,12

References


