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CASE REPORT

Parotid gland oncocytic carcinoma: A rare entity in head and neck region

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Abstract

Oncocytic carcinoma of the salivary gland is an uncommon tumour in the head and neck region. Owing to its rarity, identifying the histopathological features of a malignant tumour can be difficult and challenging. We report a case of a 70-year-old man who presented with a left facial weakness for six months in a background history of left parotid swelling over the past 10 years. Clinical examination revealed a 3x3cm left parotid mass and grade 4 facial nerve palsy. Fine needle aspiration of the mass showed scattered cohesive, monolayered sheets of uniform oncocytic cells. Subsequently, a left total parotidectomy and selective neck dissection were performed. Histological examination showed sheets of small oncocytes with minimal nuclear atypia. Evidence of nerve entrapment, capsular invasion and perivascular permeation were identified in focal areas. Thus, a final diagnosis of oncocytic carcinoma was rendered.

Keywords: Oncocytic carcinoma, salivary gland, parotid

INTRODUCTION

Oncocytomas of the salivary gland is uncommon, accounting for only 2% of all salivary gland neoplasms and typically affecting individuals in their sixth to eight decades of life.[1,2] The malignant transformation of oncocytoma or oncocytic carcinoma is extremely rare. The definite criteria for malignancy are based on capsular, vascular or neural invasion with or without evidence of metastasis. The pre-operative assessment by fine-needle aspiration cytology may aid in the diagnosis, however, this alone may have limitation in deciding the extent of surgery.

CASE REPORT

A 70-year-old man presented with a painless left parotid swelling for 10 years which slowly increased in size. In the last 6 months, he complained of left facial weakness associated with constitutional symptoms. Physical examination revealed a firm, mobile and painless left parotid mass measuring 3 x 3 cm and grade 4 left facial nerve palsy. Magnetic resonance imaging (MRI) confirmed a mass within the deep lobe of the left parotid gland which extended into stylomastoid foramen (Fig. 1A). Pre-operative fine needle aspirations (FNA) were performed. The smears were sparsely cellular, composed of scattered cohesive, monolayered sheets of uniform oncocytic cells suggestive of an oncocytic neoplasm (Fig. 1B). There is no nuclear atypia seen. Clinically, a malignant lesion was considered based on the radiological findings and tumour presentation which was increasing in size and associated with facial paralysis. A left total parotidectomy and selective neck dissection were performed. During operation, the left facial nerve was found to adhere closely to the tumour and thus was sacrificed.

On macroscopic examination, there was a grossly circumscribed salivary gland tumour measuring 23x15x14 mm. The tumour exhibited a homogenous, brownish cut surface and was surrounded by a thin fibrous capsule, the latter was extensively sampled to look for evidence of tumour permeation. Microscopically, the tumour was mostly composed of uniform neoplastic

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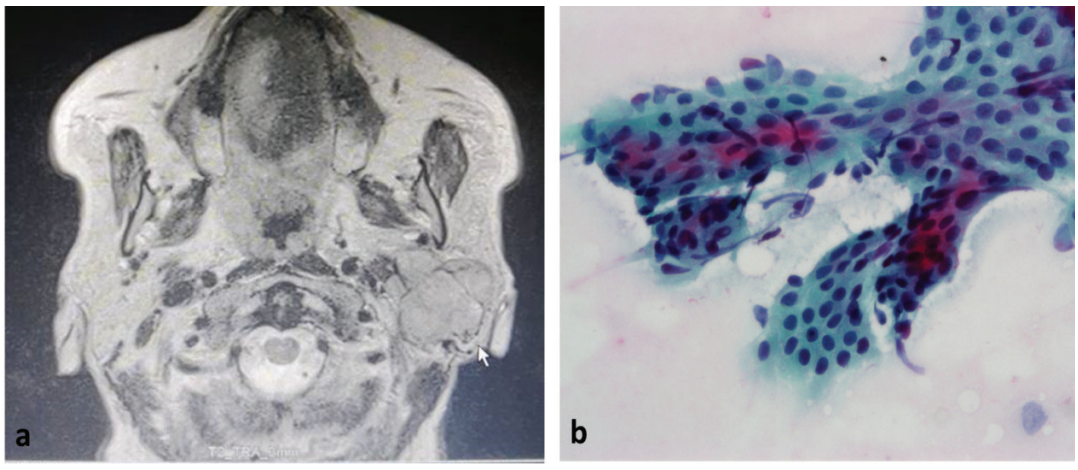

FIG. 1: (A) MRI revealed a left parotid mass (arrow) involving the deep lobe. (B) FNA of the mass showed monolayered clusters of uniform oncocytes exhibiting round nuclei within granular cytoplasm, low nuclear: cytoplasmic ratio and no significant atypia (cervical smear, x600).

oncocytes arranged in solid sheets and small cystic spaces, typical of benign oncocytoma. Nuclear atypia was focal and very minimal. Nerve entrapment was seen within the tumour. Focal capsular and perivascular permeation were also identified (Fig. 2A to 2D).

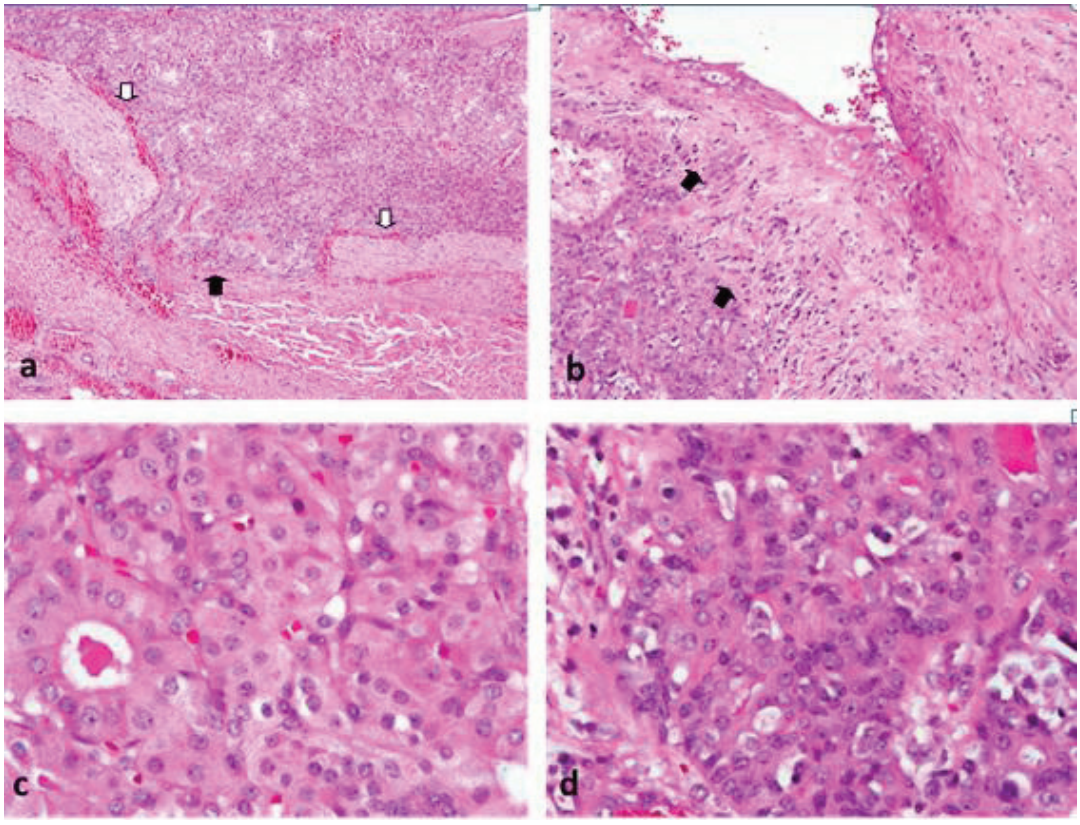

FIG. 2: On histology, the tumour showed (A) focal capsular invasion (black arrow) with nerve entrapment (white arrow) and (B) it permeated vascular wall (black arrow) (H&E x100, x200); (C) Most of the oncocytes had uniform round nuclei and abundant granular cytoplasm (H&E x600); (D) Nuclear atypia was only minimal and focal (H&E x600).

The lymph nodes were all negative for malignancy. A final diagnosis of oncocytic carcinoma was established. Subsequently, the patient received adjuvant radiotherapy with a total dose of 66 Gy given within six weeks. At 4 months follow up, there was no sign of local or regional disease recurrence.

DISCUSSION

Oncocytic cells are characterised by their dark, centrally located, round nuclei and abundant granular cytoplasm rich in mitochondria. Oncocytic cells can be seen in a range of salivary gland lesions, including reactive metaplasia or oncocytosis, or within the background of another parent tumour such as pleomorphic adenoma or mucoepidermoid carcinoma. Oncocytoma is a neoplasm composed solely of oncocytic cells where there are no features of other neoplasms. The oncocytes are arranged in sheets, duct-like and trabecular structures, surrounded by a thin fibrous capsule. Having no infiltrative quality, oncocytomas are considered benign and have extremely low malignant potential.

Oncocytic carcinoma, on the other hand, is extremely rare with fewer than 100 cases reported.[3,4] It accounts for 11% of all oncocytic salivary gland neoplasms, 0.5% of all epithelial salivary gland malignancies and 0.18% of all epithelial salivary gland tumours.[5] Most oncocytic carcinomas occur in the parotid gland. They can arise de-novo or in up to 50%, in association with a benign oncocytoma.[6,7] Typically, oncocytic carcinoma present with a slow-growing mass, which can be even longer if it has arisen from an underlying oncocytoma. Malignancy is defined by local infiltrative nature (capsular, vascular or neural invasion) or metastasis either locally or at a distant site. Other features in support of malignancy include lack of encapsulation, frequent or atypical mitosis, and necrosis.[8] Most malignant oncocytoma exhibit a degree of nuclear pleomorphism, although in occasional cases, they may have deceptively bland cytologicy.[9]

In our case, the presentation of a very slow-growing, otherwise symptomless mass over 10 years duration may indicate a benign lesion, however, there is no previous tissue diagnosis available. The histopathological evaluation shows the majority of the tumour has benign and uniform morphology, with an absence of significance nuclear pleomorphism, mitosis and necrosis. However, the presence of capsular breach, vascular wall permeation and neural invasion give unequivocal evidence of malignant behaviour of the tumour.

Preoperative evaluation by FNA cytology plays an important role in the immediate assessment of salivary gland tumours. Unfortunately, the diagnostic accuracy of FNA for oncocytic salivary lesions is reportedly low, possibly partly due to rarity of the lesion and sampling error.[10] The diagnostic difficulty may arise on cytological smears as oncocytes can be present in oncocytosis, oncocytic neoplasm or as part of other parent tumours. Evaluation of accompanying cells and stromal tissue characteristic of the underlying condition would be helpful. These include a predominance of myoepithelial cells and fibrillar matrix in pleomorphic adenoma, presence of squamous, intermediate, and mucin-secreting cells in mucoepidermoid carcinoma and heavy lymphoid background in Warthin tumour. Other tumours which may also demonstrate oncocytic changes include myoepithelioma, basal cell adenoma and polymorphous low-grade adenocarcinoma. In disputed cases, histopathological examination is mandatory.[11]

Differentiating benign oncocytoma from oncocytic carcinoma on cytology may be relatively straightforward if the tumour displays clear malignant features such as nuclear pleomorphism, frequent and/or atypical mitosis, and coupled with a necrotic background.[7] However, benign oncocytoma may show a degree of nuclear atypia while oncocytic carcinoma, on the other hand, maybe deceivingly bland, just as demonstrated by our case. In such situations, the term 'oncocytic neoplasm' is preferable, with definite diagnosis only made possible in the resected specimen.[11,12]

The widely accepted treatment for oncocytic carcinoma is a combination of total surgical resection of the tumour with a safe margin and neck dissection. Adjuvant radiotherapy to improve local control is recommended by some authors.[5] In view of its rarity, the prognosis of oncocytic carcinoma is not well established. Currently, it is considered as a high-grade tumour based on the follow-up data in current series.[9]

CONCLUSION

A high suspicion is necessary for distinguishing between benign oncocytoma and oncocytic carcinoma, as both entities may appear clinically similar. Preoperative radiological imaging can be helpful in assessing the extent of such lesions.

Cytological smears must be interpreted with caution as oncocytic cells may be present in a wide range of condition. Benign nuclear features on cytology do not entirely exclude malignancy and vice versa. Definite evidence of malignancy is only possible by demonstrating evidence of local invasion or metastasis.

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Conflict of interest: The authors declare they have no conflict of interests.

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