Case report

Value of imaging and aspiration cytology in the diagnosis of oncocytic carcinoma

Valore della radiologia e della citologia aspirativa nella diagnosi del carcinoma oncocitario

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SUMMARY

Oncocytic carcinoma of the parotid gland is a rare neoplasm. To date 70 cases have been described in 55 reports. To the best of our knowledge the simultaneous occurrence of oncocytic carcinoma and second malignancy in another site (outside the parotid gland) has not been reported. An oncocytic carcinoma of the parotid gland is described in a 56-year-old male with simultaneous breast cancer, emphasising the value of aspiration cytology and imaging procedures in the diagnosis of parotid neoplasms.

KEY WORDS: Parotid gland • Malignant tumours • Oncocytic carcinoma • Fine-needle aspiration cytology • Male breast cancer

RIASSUNTO

Il carcinoma oncocitario della parotide è raro. Sino ad oggi sono stati riportati 70 casi in 55 lavori. La contemporanea insorgenza del carcinoma oncocitario e di una seconda neoplasia maligna in altra sede non è stata mai descritta. Descriviamo il caso di un carcinoma oncocitario della parotide in un uomo di 56 anni con simultaneo sviluppo di carcinoma mammario sottolineando il valore della citologia aspirativa e delle tecniche radiologiche nella diagnosi della neoplasia parotidea.

PAROLE CHIAVE: Parotide • Tumori maligni • Carcinoma oncocitico • Esame citologico • Carcinoma mammella maschile

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Introduction

Physical examination does not always succeed in a pre-operative differential diagnosis between benign and malignant parotid tumours. Thus, diagnostic modalities, such as fine-needle aspiration cytology (FNAC) and imaging, play an important role in the work-up of patients with parotid mass lesions. Oncocytic carcinoma (OC) of the parotid gland is a rare neoplasm. Bauer and Bauer 1 reported the first case in 1953 and, to date, 70 cases have been described in 55 reports. The simultaneous occurrence of OC and a second malignancy in another site has not been reported up to now. Herein, the case is described of an OC of the parotid gland, in a 56-year-old male, with simultaneous breast cancer, stressing the value of aspiration cytology and imaging procedures in the diagnosis of parotid malignancy.

Case presentation

In June 2006, a 56-year-old male was referred to the Otorhinolaryngology Division for a painless right preauricular mass which had been gradually increasing in size for a year. Computed tomography (CT) showed an enhanced signal of the right parotid gland with a homogeneous hyperdense mass (3.5 cm in diameter) with regular contours; on the medial site, the mass was indistinguishable from the adjacent anatomic structures (Fig. 1). The facial nerve was functionally normal. FNAC revealed oncocytes with mild atypia. Total parotidectomy with facial nerve preservation was performed. The mass was firm, multi-nodular, unencapsulated, grey-brown and measured 3.5 x 3 cm. The frozen section showed a malignant infiltrative growth pattern.

Histology showed the typical features of oncocytic carcinoma (Fig. 2). The tumour cells were positive for immu-
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no-histochemical stain with anti-mitochondria antibody (Mitochondria antibody SPM198, Abcam).

Post-operatively, the patient received radiotherapy (6300 rad) to the surgical site. In January 2007, the patient was referred to the Surgical Division for a left mammary nodule. In the aspiration cytology, malignant cells were found. Total mastectomy with axillary lymphadenectomy was performed. The histological examination of the surgical specimen showed invasive ductal carcinoma, grade III (Nottingham Histological Score), with metastasis in 12 axillary lymph nodes (tumour deposits > 2 mm) (pT1c pN3a M0 G3). Immuno-histochemistry analysis exhibited 90% positivity for oestrogen receptor (Estrogen Receptor α clone SP1, Dako, Glostrup, Denmark), 80% for progesterone receptor (Progesteron Receptor clone SP2, Dako, Glostrup, Denmark), 40% for Ki-67 (Ki-67 clone SP6, Dako, Glostrup, Denmark). Her-2 evaluation showed a 10% of neoplastic positive cells, score 2 (HERCEP TEST, Dako, Glostrup, Denmark). FISH analysis showed Her-2 amplification. The treatment administered for breast carcinoma was Tamoxifen 20 mg/day and 17 cycles of Herceptin. For the parotid carcinoma, ultrasound follow-up was decided with no additional therapy. At present, the patient is free from disease.

Discussion

The role of imaging in the assessment of a salivary gland tumour is to define the intra-glandular vs extra-glandular location, to detect malignant features, to assess local extension and invasion and to detect nodal metastases and systemic involvement.

For parotid lesions, ultrasound (US) is a sensitive and efficient procedure for relatively superficial structures accessible by high resolution US which provides excellent resolution and tissue characterization without a radiation hazard. Cervical node involvement can also be assessed. US has a limited visualization of the deep lobe of the parotid gland.

A series of reports demonstrated the superiority of magnetic resonance imaging (MRI) over CT in delineating parotid mass lesions. CT evaluates the cortical involvement and the presence of calculus disease in sialoadenitis (which may mimic a tumour) in a more precise way; but MRI is superior in defining tumour characteristics and extension, particularly an eventual perineural spread.

Among MRI features, only poorly defined margin and infiltration on non-enhanced non-fat-suppressed T1-weighted images yielded a significant difference between benign and malignant parotid lesions.

In the recent literature, it was reported that the sensitivity of parotid FNAC ranges from 54% to 95%, specificity from 86% to 100% and accuracy from 84% to 97%.

In the study of Inohara et al., of 81 patients with a parotid mass lesion (60 benign and 21 malignant), the sensitivity/specificity/accuracy of FNAC and MRI were 80%/95%/94% and 81%/92%/89%, respectively. FNAC or MRI served equally to predict the malignant nature of parotid mass lesions.

The role of nuclear medicine and PET scan, in the imaging of parotid masses, has not yet been established.

In conclusion, MRI is the procedure of choice in the identification of parotid malignancy, and the combination of FNAC and MRI yielded no diagnostic advantage over either modality alone.
When malignancy is suspected on the basis of clinical or US data, MRI should be performed for all the patients candidate to surgery. In the articles regarding FNAC and imaging of the parotid lesions, OC is not reported. Only a scrupulous review of single cases or small series of OC would permit clinical features, to be established, as well as outcome and value of FNAC and imaging in the management of this neoplasm. Regarding the 70 reported cases, age and sex were given in 61 patients. The age ranged from 30 to 93 years, with a mean and a median age of 62.7 years. Clinically, they usually presented as parotid masses. The size of the tumour ranged from 0.4 to 8 cm in maximum diameter (mean 4.01).

The function of the facial nerve was studied in 54 out of 70 patients and facial nerve paralysis was found in 16 cases.

Initial treatment was specified in 60 out of 70 patients (one patient refused the treatment) and the follow-up time was available in 44 cases, ranging from 3 to 150 months (mean 32 months).

Initial conservative surgery (tumorectomy, superficial parotidectomy) was performed in 13 cases for OC, in 3 cases for oncocyotma, in 1 case for pleomorphic adenoma, and in 1 case for undifferentiated malignant carcinoma, out of the 59 patients with clinical records of the treatment. Radiation therapy was performed in 26 cases: in 1 case as the only treatment, in 10 cases associated with conservative surgery and in 15 cases associated with total parotidectomy.

Metastases to the cervical lymph nodes were present in 30 out of 70 patients. Other metastatic sites were lungs, liver, bone, parapharyngeal region and skin. A total of 11 out of the 44 patients with available follow-up died of the disease and 3 from unrelated causes (pneumonia, cardio-respiratory failure and cerebro-vascular accident in the absence of local or metastatic disease).

FNAC and imaging procedures were reported in a small number of cases (Tables I-II) 10-30.

In the diagnosis of OC, FNAC was first used by Eneroth 10. Then, other reports described the use of this procedure in the diagnosis of this kind of neoplasm, with controversial opinions (Table I). In accordance with Harrison et al., we retain that OC aspiration cytology may

### Table I. Aspiration cytology in the diagnosis of malignant oncocytoma. Review of the literature.

<table>
<thead>
<tr>
<th>Authors</th>
<th>Findings</th>
<th>Diagnosis</th>
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<tbody>
<tr>
<td>Eneroth 1965</td>
<td>Usual oncocyes mixed with oncocytic cells with marked atypia of the nucleus as well as morphologically typical carcinoma cells</td>
<td>Malignant Oncocytoma</td>
</tr>
<tr>
<td>Austin 1987</td>
<td>Single cells with copious, dense, granular cytoplasm and eccentric nuclei</td>
<td>Not performed</td>
</tr>
<tr>
<td>Haberman 1990</td>
<td>NOS</td>
<td>Consistent with oncocytoma</td>
</tr>
<tr>
<td>Abdul-Karim 1991</td>
<td>Cytologically “benign” oncocyes</td>
<td>Oncocytoma</td>
</tr>
<tr>
<td>Scher 1991</td>
<td>NOS</td>
<td>Malignancy with oncocytic features</td>
</tr>
<tr>
<td>NOS</td>
<td>Malignant epithelial neoplasm with oncocytic features</td>
<td></td>
</tr>
<tr>
<td>NOS</td>
<td>Carcinoma with oncocytic features</td>
<td></td>
</tr>
<tr>
<td>Ramakrishna 1992</td>
<td>NOS</td>
<td>NOS</td>
</tr>
<tr>
<td>Lafora 1994</td>
<td>Cellular polymorphism, nuclear atypia, hyperchromatism, with nuclear membrane irregularities and prominent nucleoli</td>
<td>Consistent with a malignant tumour, confidently of oncocytic nature</td>
</tr>
<tr>
<td>Rajan 1994</td>
<td>Cells were predominantly dissociated, showed nuclear enlargement and pleomorphism, prominent nucleoli, multinucleation and contained abundant homogeneous cytoplasm</td>
<td>Malignant oncocytoma</td>
</tr>
<tr>
<td>Kandiloros 1995</td>
<td>NOS</td>
<td>Warthin’s tumour</td>
</tr>
<tr>
<td>Harrison 1995</td>
<td>Partly cohesive cluster of oncocyes showing abundant, finely granular cytoplasm, mild nuclear pleomorphism and some nuclear overlapping. Cohesive sheets of polygonal oncocytic cells with narrow intercellular spaces conferring a “squamoid” appearance</td>
<td>Primary salivary gland neoplasm of an unusual type, with characteristics suggestive of oncocytic or possibly squamous differentiation</td>
</tr>
<tr>
<td>Ardekian 1999</td>
<td>NOS</td>
<td>Adenocarcinoma</td>
</tr>
<tr>
<td>Cinar 2003</td>
<td>First aspiration: NOS</td>
<td>Warthin’s tumour</td>
</tr>
<tr>
<td>Second aspiration: oncocyes and atypical cells</td>
<td>Malignant epithelial tumour</td>
<td></td>
</tr>
<tr>
<td>Guclu 2005</td>
<td>Oncocytes and atypical cells</td>
<td>Malignant epithelial tumour</td>
</tr>
<tr>
<td>Caloglu 2006</td>
<td>Atypical epithelial cells</td>
<td>NOS</td>
</tr>
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* Aspiration cytology was performed on metastases near left stylomastoid foramen.
display a spectrum of appearance from apparently benign to clearly malignant cells. MRI was used in 5 cases, but the radiological reports are inadequate because the features of malignant parotid lesions (poorly defined margin and infiltration on non-enhanced non-fat suppressed T1-weighted images) were not described.

In the present case, FNAC was inconclusive; CT had a limited value in determining the malignant nature of the neoplasm. We should have performed MRI but we did not manage to programme it before the surgical intervention and we based our diagnosis on the frozen section. The simultaneous presence of two malignancies gives rise to problems in the choice of the treatment. Consequently, the description of single cases of simultaneous malignant lesions could contribute to the comprehension of the biological behaviour.

Male breast cancer (MBC) is a rare tumour and accounts for less than 1% of breast cancers, but the incidence seems to be increasing. Some aspects of the aetiology of MBC are similar to those of the female counterpart. It has been estimated that there is a family history in about 5% of MBC patients.

In the review of the literature, O'C has never been reported before in association with other primary malignancies of other sites, while MBC has only been reported in association with hepatocellular carcinoma.

MRI is the procedure of choice in the O'C diagnosis when FNAC contains atypical oncocytic cells.

### References


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