CASE STUDY

Salivary Duct Carcinoma: Diagnostic Clues, Histology and Treatment☆

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Received 24 May 2011; accepted 20 October 2011

KEYWORDS
Salivary duct carcinoma; Salivary neoplasm

Abstract Salivary duct carcinoma is a very rare, highly malignant epithelial tumour. We present a case of a 75-year-old man with a rapidly growing salivary duct carcinoma in the parotid gland, of one month’s evolution. Histopathologically, salivary duct carcinoma is characterised by its resemblance to ductal carcinoma of the breast. It usually develops aggressively with possibilities of early distant metastasis and local recurrence. The tumour is managed with total parotidectomy, ipsilateral neck dissection and adjunctive radiation. In advanced cases, we recommend treatment with anti-Her-2 monoclonal antibodies such as trastuzumab.

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PALABRAS CLAVE
Carcinoma ductal salival; Neoplasias salivales

Resumen El carcinoma ductal salival es un tumor epitelial de alto grado, muy poco frecuente. Presentamos el caso de un varón de 75 años con carcinoma ductal salival de rápido crecimiento, en la glándula parótida, de un mes de evolución. Histopatológicamente se caracteriza por su gran similitud con el carcinoma ductal de mama. Suele tener un comportamiento muy agresivo, con recurrencia local y metástasis a distancia tempranas, por lo que hay que realizar parotidectomía total con vaciamiento cervical ipsilateral y radioterapia adyuvante. En casos avanzados se recomienda tratamiento con anticuerpo monoclonal selectivo para el HER-2/neu como trastuzumab.

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Introduction

Salivary duct carcinoma (SDC) is a rare malignant tumour, first described in 1968 by Kleinsasser et al. In 2005, the World Health Organisation published its third classification of salivary gland tumours, divided by histological grades, and included SDC in the category of high level of malignancy, along with high-grade mucoepidermoid carcinoma and carcinoma ex pleomorphic adenoma. The most striking feature about this tumour is its great histological similarity with breast ductal carcinoma, requiring a differential diagnosis with possible metastases through immunohistochemistry analysis among patients with a previous history. Following a diagnosis of high-grade salivary carcinoma, distinguishing among histological subtypes is the most laborious part due to the similarity between their cytological characteristics.

Up to 85% of these tumours appear in the parotid gland. Most patients are males with a mean age of onset between the fifth and sixth decades of life. Clinically, they appear as a rapidly growing and painful mass, with facial nerve involvement in 25%–60% of cases. Most patients present early cervical lymphadenopathies and distant metastases in the lungs and bones, so the prognosis of this entity is highly unfavourable. In Spain, there is almost no literature on this type of carcinoma. Thus, we present the case of a patient with ductal carcinoma of the parotid gland and review the published literature.

Case Report

The patient was a 75-year-old male who presented a 2.5 cm mass in the tail of the left parotid gland with 1 month evolution. We performed fine needle aspiration, which was not diagnostic. Magnetic resonance imaging (MRI) and computed tomography (CT) scans showed a well-encapsulated lesion (Fig. 1). Following suprafacial parotidectomy, the anatomopathological study found cystic formations with a ductal-type, cribriform epithelial lining. The cells presented a pattern of oncocytic metaplasia (Fig. 2). Immunohistochemical techniques showed strong positivity for cytokeratin 7 (CK7) and Her-2/neu, and were also positive for ki67, cyclin D1, P53, S100, epidermal growth factor receptor, GCDFP-15, epithelial membrane antigen and carcinoembryonic antigen. The results were negative for CK20 and PSA.

The definitive diagnosis was parotid ductal carcinoma, so we conducted parotidectomy totalisation with preservation of the facial nerve and ipsilateral cervical lymph node dissection of levels I–III. The patient also had a perineural, vascular and lymphatic embolisation, with free surgical margins. The control PET-CT scan was negative.

Following the TNM classification by the American Joint Committee on Cancer, we recorded the case as a stage II (T2N0MO) and treated with radiotherapy at 60 Grey on the parotid site.

Discussion

Ductal carcinoma of the salivary glands is a rare adenocarcinoma stemming from cells of the glandular excretory duct. Its reported incidence varies between 3% and 9% of malignant salivary gland tumours. However, it is difficult to determine the true incidence of this tumour, since it has not been recognised in numerous series or has been confused with other tumours due to the scarce knowledge about it.

These entities generally arise de novo, although some cases develop from a pleomorphic adenoma and, less frequently, from low grade adenocarcinoma. There have been cases reported in young patients and in locations other than salivary glands, such as the larynx and paranasal sinuses. However, their most common occurrence is among men aged 50–60 years, with involvement of the parotid gland. The finding of tumoural calcifications on CT scans may be suggestive of SDC in 33% of cases. MRI scans help to evaluate signs which suggest malignancy, such as ill-defined margins and comedonecrosis. It is advisable to perform a PET-CT for the diagnosis and monitoring of possible distant metastases.

The entity is manifested as a painful mass, often associated to paresis/facial nerve paralysis. Perineural and

Figure 1  Magnetic resonance imaging and computed tomography scans showing a well-encapsulated lesion in the left parotid tail.
lymphatic invasion are common histological features. In a series of 15 patients, Sefik Hosal et al. published 83% of cases with perineural invasion and 73% with lymphatic invasion, of which 50% were N0. These data highlight the importance of lymph node dissection in patients with SDC.

Our patient was above the mean age. We noted the rapid growth of the tumour, as well as the benign characteristics found on the MRI scan.

Affected patients have a survival rate of 35%-65% at 5 years. The main causes of death are distant metastases in the lungs and bones, although some patients have been reported to survive for over 10 years, in which case we refer to healing criteria. There is controversy regarding the negative prognostic factors, which include tumour size and lymph node involvement, followed by distant metastases and expression of Her-2/neu.

Histologically, SDC is characterised by its great similarity to breast ductal carcinoma, with areas of invasion and areas of intraductal carcinoma. The intraductal pattern shows different growth types, such as cribriform, papillary and solid, generally associated with areas of central comedonecrosis. The invasive pattern presents a desmoplastic stroma. Immunohistochemically, it presents strong positivity for RA and cytokines. In our case, there was positivity in 10 of the 12 immunohistochemical determinations studied. The patient had good prognostic factors except for Her-2/neu expression, and is currently asymptomatic after completion of radiotherapy 2 months ago.

Regarding treatment, the recommendation is to start by total parotidectomy with ipsilateral lymph node dissection and adjuvant radiotherapy. In advanced cases with distant metastases, some studies have shown favourable results following treatment with trastuzumab, a monoclonal antibody selective for Her-2/neu, which is also expressed in breast ductal carcinoma.

Conflict of Interests

The authors have no conflict of interests to declare.

References