Ultrasound and CT imaging features in a patient with salivary duct carcinoma of the parotid gland: a case report with literature review.

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Abstract

The aim of this paper was to present the ultrasound (US) and computed tomography (CT) appearance of a patient with salivary duct carcinoma of the parotid gland. US showed a voluminous mass of the parotid gland, with multiple calcifications. Furthermore, it revealed regional multiple lymph nodes with malignant characters. Sonoelastography of the lesion and lymph nodes detected increased rigidity. Contrast enhanced CT scan of the neck completed the data description regarding the mass expansion and invasion of surrounding tissues. US and CT imaging features played a key role in establishing the malignant character of the mass and lymph nodes.

Keywords: parotid gland, carcinoma, salivary duct, lymph nodes

Introduction

Salivary duct carcinoma (SDC) is an uncommon high grade primary neoplasm of the major salivary gland (1-3 % of all malignant salivary gland tumors) [1]. It was first described by Kleinsasser et al in 1968 and included in the recent World Health Organization Classification [2,3].

Given the aggressive behavior of the tumor (with local recurrence, early distant metastasis and high mortality) early diagnosis is mandatory. The majority of cases occur in the parotid gland with a predilection for elderly men in the sixth to seventh decades of life [4-9]. Pathologically, SDC has a significant resemblance to ductal carcinoma of the breast especially due to intraductal and ductal patterns [10].

Besides the clinical and biological aspects, imaging investigations have a significant contribution in the diagnosis of SDC, US being the first step. In this case report, we present the gray scale, color Doppler, sonoelastography and computer tomography (CT) imaging features and pathological correlation of a case with salivary duct carcinoma and lymph node metastasis.

Case report

A 65 year old man, diagnosed 20 years prior presented with a 2 cm tumor in the right parotid region (without pain or functional disorders during this period) was admitted to the Maxillo-Facial Surgery Unit due to the tumor growth over the last six months, associated with the advent of pain, facial asymmetry, anxiety, and eating disorders. US showed a tumoral mass, deeply located in the right parotid gland with multiple calcifications both clustered and dispersed. Cranially the tumor extended to the skull base, abutting to the mastoid. Medially the tumor extended to the parapharyngeal space and inferiorly it was confined to the parotid parenchyma (fig 1a). The parotid mass showed vascularization with increased resistance indices (fig 1b) and was highly stiff (fig 1c) at elastography.

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Fig 1. SDC of the right parotid gland in a 65 year patient. a) Transverse and coronal grey-scale US scan shows an ill defined hypoechoic tumor with clustered and dispersed calcifications; b) Doppler ultrasound of the tumor reveals high impedance vascular signal inside the tumor; c) Sonoelastogram showing high stiffness of the tumor, in the presence of calcifications.

Fig 2. US appearance of a metastatic cervical lymph node: a) grey scale US of the lymph node shows rounded, hypoechoic, heterogeneous aspect; b) color Doppler US showing irregular distribution of the vessels and multiple pedicles; c) sonoelastogram of the lymph node showing high stiffness.

US also showed multiple hypoechoic oval nodular images, within the gland parenchyma and also in the inframastoid region, behind the gland. Laterocervical lymphadenopathy with features suggestive for malignancy affected the internal jugular chain up to the lower third of the cervical region. Some nodules showed the tendency of matting, while others were heterogeneous, with an appearance suggestive of necrosis (fig 2a). The nodes were

Fig 3. Parenchymal phase enhanced CT scan of the neck a) transverse and b) sagittal planes revealed a large, ill-defined, heterogeneous soft tissue mass, with calcifications, located in the deep portion of the right parotid gland, with slight peripheral enhancement; extraparotid infiltration and the presence of lymph nodes, extending along the internal jugular chain down to the lower third of the cervical region is also noticed.

Fig 4. Histopathology of the salivary gland: a) HEx4 shows a fragment with atypical epithelial cells with fibrotic stroma and some tubular and cystic structures; b) CK(x10) positivity in the metastatic lymph nodes.

hypervascularized with multiple pedicles and chaotic distribution of the vessels (fig 2b). Elastography of the lymph nodes with malignant appearance detected increased stiffness compared to the neighboring muscles (fig 2c).

CT scan of the neck showed a mass isodense with the parotid parenchyma, located in the deep portion of the right parotid area. The tumor presented ill-defined margins, extraparotid infiltration and was heterogeneous due to the presence of multiple calcifications. After contrast administration, the lesion revealed slight peripheral enhancement. The CT scan also showed multiple enlarged laterocervical lymph nodes (fig 3).

The therapeutic approach was tumor excision with preservation of the facial nerve and extemporaneous histopathological examination.

Histopathology of the tumor tissue showed hyaline masses with the presence of squamous tumor cells, arranged in tubular and cystic structures (fig 4a). Adjacent to the salivary gland, pathology revealed carcinoma lymph nodes metastasis with positive cytokeratin 7 (CK7) immunostaining (fig 4b). Following histopathological examination, functional right laterocervical lymph node dissection was performed.

The final diagnosis was lymph node metastasis in a patient with salivary duct carcinoma of the parotid gland.

The patient was discharged with antibiotic and symptomatic treatment and was referred to the oncological department.

Discussions

According to Motoori et al [11] SDC is a rare, aggressive malignant tumor. Although occasional submandibular, sublingual and minor salivary gland localisation of the SDC has been reported, this tumor shows a predilection for the parotid gland, representing 6-12% of all cases of parotid cancer. This type of tumor is more frequently encountered in elderly male patients [11]. Clinically, this type of tumor includes the presence of painless nodules, facial palsy or nodal metastasis, suggesting its aggressive behavior. Lymph node metastases have the highest incidence (57-73%) of all cancers of the parotid [6-8]. In our case, the patient had lymph node metastasis at the time of presentation and diagnosis. Pre-existing benign pleomorphic adenoma of the parotid gland may be a precondition for the occurrence of SDC [6-8,12,13]. From this point of view, our patient was known to have had a painless parotid mass for 20 years, but histological examination could not identify the benign components.

Surgery is the treatment of choice, followed by post-operative radiotherapy, but despite this aggressive attitude (local resection and prophylactic ipsilateral neck dissection), the survival time is reported to be no more than 2-3 years [11,12,14,15]. In addition, very frequent locoregional recurrence (16-55%) has an important role in poor prognosis of SDC [15]. Histologically, SDC is composed of atypical epithelioid tumor cells arranged in different patterns with fibrotic stroma being immunoreactive for low- and high- molecular- weight cytokeratin and other markers. [16-19]. Gray scale and color Doppler US have an important contribution in establishing the differential diagnosis between benign and malignant parotid tumors. It is, however, difficult to completely rule out malignancy [20,21].

According to Lee et al [20], high-resolution US of the salivary glands provides excellent tissue characterization, multi-planar information and vascular pattern with Doppler technique. A malignant salivary gland tumour typically shows ill-defined border with heterogeneous aspect (internal necrosis, cystic change and the presence of calcifications) [21]. Doppler shows hyper vascular tumour with increased resistance indices ($RI > 0.8$ and a $PI > 2$) [21]. In this regard, all of the above, highly suspicious for malignancy, were detected in our patient's tumor description, such as ill defined borders, hypervascularity with

RI of 1 and PI of 2.6 and a heterogeneous echostructure hereby calcifications. Local invasion may be seen, and malignant nodes are identified by their round shape, heterogeneity, loss of hilar architecture, abnormal, disorganized vascularity with or without internal necrosis and extra-capsular spread [20,22]. Thus, our patient showed local tumoral invasion and metastatic lymph nodes.

To the best of our knowledge, this is the first case report that describes the ultrasonographic appearance of SDC.

After Dumitru et al, the role of elastography in differentiating malignant from benign parotid gland tumor has been shown to be limited because of the stiffness of benign tumors (especially pleomorphic adenomas) [23]. From this point of view, our patient presented increased stiffness of the tumor (with a score of 10) more likely due to the presence of calcifications.

According to Weon et al, the presence of multiple intratumoral calcifications was also described on CT scans, as being suggestive for the possibility of SDC [11]. In this case report, the tumor showed an ill-defined margin, infiltration into the adjacent structures and intratumoral calcifications. As for the regional lymph nodes, the US and CT imaging features correlated well with the pathological findings.

The particularities of the case lie in the rarity of this type of tumor, insidious onset and tumor aggressiveness.

In conclusion, it is important to suspect malignant transformation in a patient known to have a benign parotid tumor which presents sudden tumor growth associated with the advent of pain. US is the first imaging modality used for diagnosis. The presence of calcifications (detected both at US and CT) and intraparotid and laterocervical lymph nodes may raise suspicion of a SDC with lymph node metastasis. Despite the non-specific character of US and CT imaging findings, they are useful methods for tumor staging of SDC.

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