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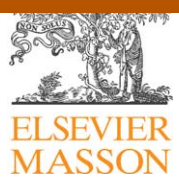

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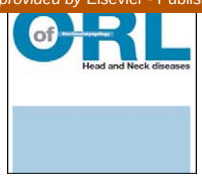

CLINICAL COMMENTARY

Mandibular osteosclerotic lesion of a parotid salivary duct carcinoma: Demonstration of the neural tropism of these tumors

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Summary
Introduction: Salivary duct carcinoma (SDC) is an uncommon entity of salivary gland cancers with a poor prognosis due to local aggressiveness or distant recurrences involving lymph nodes, lung, and long bones, in which secondary lesions are usually osteolytic. The authors report the first case of mandibular SDC, atypical due to its osteosclerotic presentation and its site, attributed to aggressive neural spread of the tumor along the trigeminal nerve.
Case study: This asymptomatic osteosclerotic bone involvement was diagnosed based on pathological enhancement of the trigeminal nerve demonstrated on MRI and was accompanied by facial nerve involvement up to its third intracranial portion. Radical surgery ensured disease control with continued good quality of life at the 4-year follow-up visit.
Conclusion: Nerve enhancement on MRI and determination of specific tumor markers (HER-2/*neu* and p53) should be taken into account to evaluate the prognosis of SDC and to propose appropriate surgical treatment.

Introduction

Salivary duct carcinoma (SDC) is an uncommon entity of salivary gland cancers (1—3% of salivary gland cancers [1] and 6—16% of parotid cancers [2,3]). Histologically, it is very close to mammary duct carcinomas, also expressing androgenic receptors [3]. Treatment is first surgical, even radical with lymph node evidement, followed by radiotherapy [2].

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Its negative prognosis stems from local recurrences or secondary remote lesions, occurring in more than half of the cases [1]. Metastases are to the lymph nodes, lungs, or long bones, in which lesions are usually osteolytic [2].

We report herein a case of SDC, atypical in its nerve invasion aggressiveness along the facial nerve up to its third intracranial portion and along the trigeminal nerve, leading to osteosclerotic mandibular extension.

Case study

A 53-year-old female patient consulted in our unit for the sudden and initial appearance of total left-sided facial paralysis, grade VI on the House and Brackman classification [4]. No parotid mass was palpable and there was no notable difference in size compared to the contralateral gland. Two ipsilateral adenopathies were noted. The otoscopy as well as audiometry were normal. The acoustic reflex was preserved, indicating facial nerve involvement situated after the emergence of the chorda tympani. Examination of the cranial nerve pairs demonstrated hypoesthesia of the left side of the chin and teeth 31—34, corresponding to the trigeminal innervation areas. No pain, lingual hypoesthesia, dental mobility, or oral mucosa lesions were observed. The diagnostic hypothesis at this time was an intraparotid malignant tumor invading the facial nerve and the trigeminal nerve via parapharyngeal lymph gland extension in contact with the foramen ovale.

MRI with gadolinium injection demonstrated homogenously increased volume of the left parotid gland, with intense contrast product uptake (Fig. 1) and pathological ipsilateral adenopathies. The radiologist also noted an abnormal signal within the left mandibular horizontal branch. The extracranial portion of the trigeminal nerve

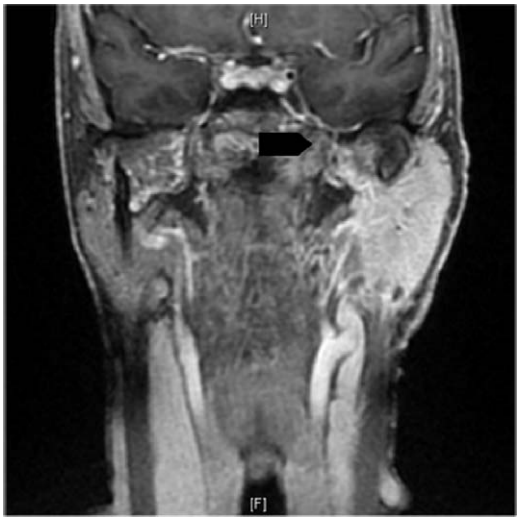

Figure 1 MRI (frontal plane) with gadolinium injection showing homogenous enhancement of the entire left parotid gland, bypassing the mandibular condyle and extending medially to the foramen ovale (black arrow) and laterally to the tympanic bone (not shown). The left parotid gland measures 36.3 mm in transverse diameter and 47.6 mm in anteroposterior diameter versus 28.3 mm and 45.1 mm for the right gland, respectively.

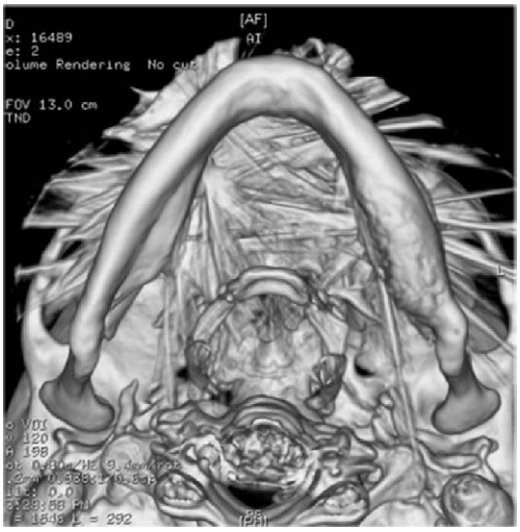

Figure 2 Bone window CT image in 3D reconstruction (axial plane) of the osteosclerotic lesion of the horizontal branch of the left mandible.

also took up the contrast product pathologically, particularly at its penetration into the mandible. Its intracranial portion was normal, however, CT, performed to better evaluate the left hemimandible, showed local hyperdense enlargement with periosteal appositions (Fig. 2). The workup was completed with (18)F-fluorodeoxyglucose positron emission tomography (PET), demonstrating substantial enhancement (standardized uptake values [SUV] >6) of the left parotid, the mandible, and adenopathies in zone IIA. There was no distant fixation. The parotid and mandible lesions appeared to be clearly distinct (Fig. 3).

In this atypical context of synchronous osteosclerotic mandible and parotid lesions, diagnostic biopsies were done, revealing SDC. The androgen receptors (monoclonal AR27 receptors; ZYMED, Cergy Pontoise, France) were expressed

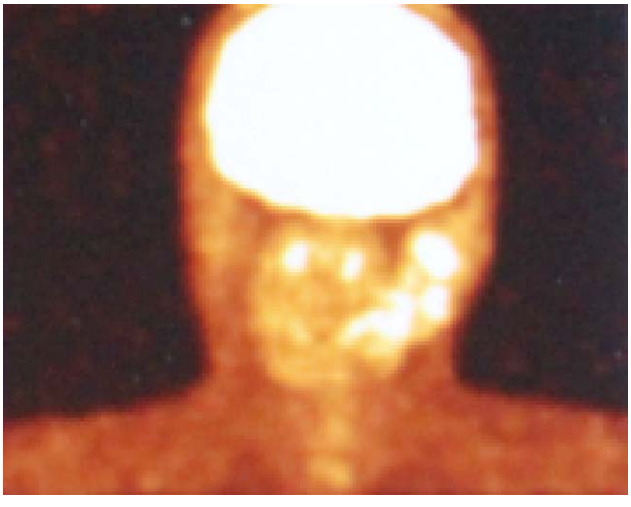

Figure 3 (18)F-Fluorodeoxyglucose positron emission tomography (frontal plane): pathological enhancement of the inferior pole of the left parotid, an adenopathy in zone IIA, and the horizontal mandibular branch. The fixations of these three lesions are distinct and intense.

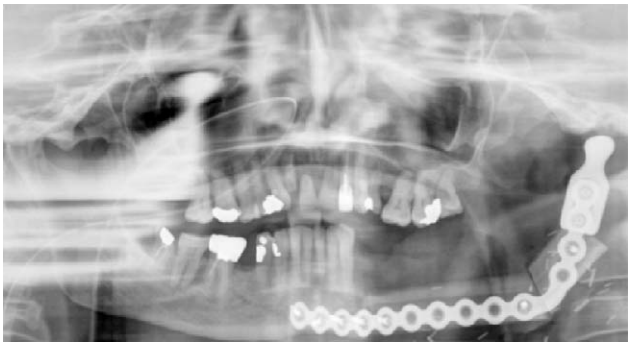

Figure 4 Dental panoramic X-ray of the horizontal branch of the left mandible reconstructed with an osteocutaneous fibular free flap.

by the tumor, but the expression of HER-2/*neu* and p53 (DAKO, Trappes, France) was low. Low HER-2/*neu* and p53 expression has been reported to be a factor of better prognosis [1], and therefore the multidisciplinary oncology meeting decided to propose radical surgery with ipsilateral lymph node evidement. The surgery associated left radical parotidectomy, petrosectomy, and hemimandibulectomy, reconstructed with a local flap from the temporal muscle and a microanastomosed osteocutaneous fibular free flap (Fig. 4).

Examination of the histological specimen confirmed parotid SDC, comprising an invasive carcinomatous proliferation made up of pseudonodular cavities filled with mucoid material. There was considerable perineural sheath tumoral invasion. The intramandibular lesion was histologically identical, surrounded by osteosclerosis of trabecular bone (Fig. 5). The facial nerve was invaded up to its intramastoid portion; the resection was entirely in healthy tissue. The trigeminal nerve was also involved but distant from its emergence from the foramen ovale (where it was resected). Lymphatic evidement included five metastatic lymph nodes with no capsular breakage. Surgery was therefore completed with chemoradiotherapy.

After 4 years of follow-up, the patient has presented no local recurrence or secondary lesions, either clinically or on complementary exams (MRI, PET).

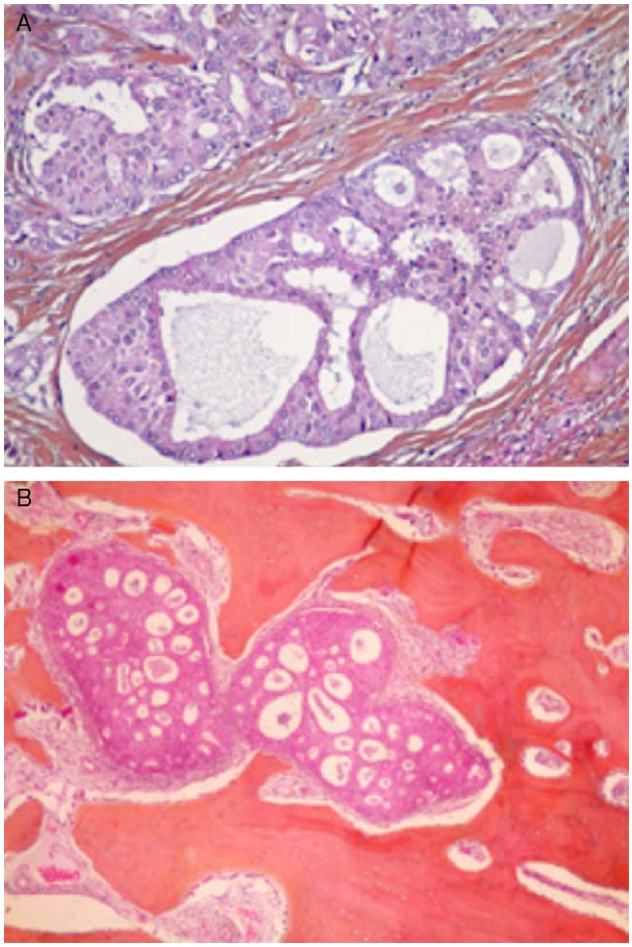

Figure 5 Infiltrating SDC (hematoxylin-eosin-saffron [HES staining], moderate magnification). A. Within the parotid gland: note the cribriform ductal component and the necrosis, the large cells with a pink cytoplasm and pleomorphic nuclei. B. Within the mandible: carcinoma infiltrating bone and pronounced osteosclerosis.

Discussion

It seems that this case is the first reported case of an osteosclerotic SDC lesion in a short bone. SDCs usually affect men older than 60 years of age [1,2] presenting a painful parotid mass (33% of cases [2]) and facial paresis in one zone (42—62% of cases [2,5,6]). Metastases, when they occur in long bones, appear after a medial time of 2 years [1,2]. The atypical clinical presentation reported herein, i.e., a painless development without a distinctive mass and delayed sudden total facial paralysis, resulted in the patient consulting late. Moreover, the mandibular lesion was diagnosed by the radiological exam given that the bone invasion had no clinical expression and the chin and tooth hypoesthesia was initially attributed to trigeminal nerve invasion by tumor extension close to the foramen ovale (after its division into its two terminal branches). Mandibular bone metastases are usually osteolytic and are suspected with hypersalivation, pain, paresthesia, abnormal palpation, tooth mobility, and/or trismus at presentation [7]. In the present case, the different diagnoses for this mandibular lesion were metastasis, invasion from the adjacent tissues, or a secondary lesion with diffusion along the inferior alveolar nerve. However, the presence of this sclerosing lesion simultaneously with a parotid tumor were not highly suggestive of metastasis in this context [1,2,7]. In addition, the absence of an oral mucosa lesion associated with visualization of two clearly distinct lesions on PET, with no uptake between the two, also argued against the hypothesis of invasion from adjacent tissues. The mandibular lesion was therefore related to the tumor dissemination along the trigeminal nerve given the pathological enhancement on MRI and the association with extensive facial invasion. Even though perineural invasion of SDC is well known (80% of cases) [2,6], only a single study has reported the case of symptomatic intracranial extension along the trigeminal nerve [8].

The prognosis of SDC is negative, with overall survival of 56 months [1]. However, quantification of HER-2/*neu*

and p53 tumoral markers can aid in management by providing additional prognostic indices [1]. An alternative to surgery when this is impossible could be antiandrogenic hormone treatment, which has been successfully attempted in one case [3]. This therapy requires more detailed assessment.

Conclusion

This first case of an osteosclerotic location of SDC illustrates the strong neural tropism of this tumor. With an attentive search for pathological enhancement on MRI and quantification of survival markers, treatment adapted to the prognosis can be proposed.

Conflict of interest statement

None.

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