

Primary Adenoid Cystic Carcinoma of the Mandibular Bone, an Extremely Rare Case Report

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Abstract

Adenoid cystic carcinoma (ACC) is a rare epithelial neoplasm, that may affect the salivary glands. It comprises 30% of epithelial minor salivary gland tumors. We present the case of a 56-year-old non-smoking woman with no significant medical history. The MRI finds a lesional process involving the right hemi mandible, the right buccal floor, which extends towards the region of the masticators and infiltrates the submandibular gland. A bone biopsy of the mandible under general anesthesia was performed, showing an adenoid cystic carcinoma. After announcing the diagnosis to the patient, she refused any treatment.

Keywords: Adenoid Cystic Carcinoma; Mandible; Magnetic Resonance Imaging

List of abbreviations: ACC: Adenoid Cystic Carcinoma; MRI: Magnetic Resonance Imaging

Introduction

Adenoid cystic carcinoma (ACC) is a rare epithelial neoplasm, that may affect the salivary glands. It comprises 30% of epithelial minor salivary gland tumors [1].

Primary central salivary gland carcinomas of the mandible are uncommon, and its origin is still controversial.

The present case report is to describe an unusual case of primary ACC of the right mandible without any medical history. The radiological findings shows a lesional process involving the right hemi mandible, the right buccal floor. The biopsy was performed under general anesthesia confirms an adenoid cystic carcinoma. The patient refused any treatment.

Case report

We present the case of a 56-year-old non-smoking woman with no significant medical history.

The onset of the disease dates back to a year by the appearance of right angular mandibular pain gradually increasing in intensity, and resistant to medical treatment, the patient's condition was complicated by a right juguale swelling with trismus initially treated as facial cellulitis without improvement.

On the radiological assessment, the dental panoramic X-ray finds an aspect of osteolysis of tooth 46 (Figure 1).

The CT scann shows an osteolysis and infiltration of the right soft tissues of the mandible with small collection of the right para pharyngeal region measuring 21 mm (Figure 2).

The MRI finds a lesional process involving the right hemi mandible, the right buccal floor, measuring 61.2 x 37 mm and which extends towards the region of the masticators and infiltrates the submandibular gland, as well as vertebral osteolytic lesions of the C4 -C5-C6 (Figure 3).

A bone biopsy of the mandible under general anesthesia was performed, showing an adenoid cystic carcinoma.

After announcing the diagnosis to the patient as well as the various therapeutic modalities, as surgery as well as radiotherapy, the patient refused treatment.



Figure 1: Panoramic X-ray showing osteolysis of the right mandible



Figure 2: CT scan showing an infiltration of the right mandible

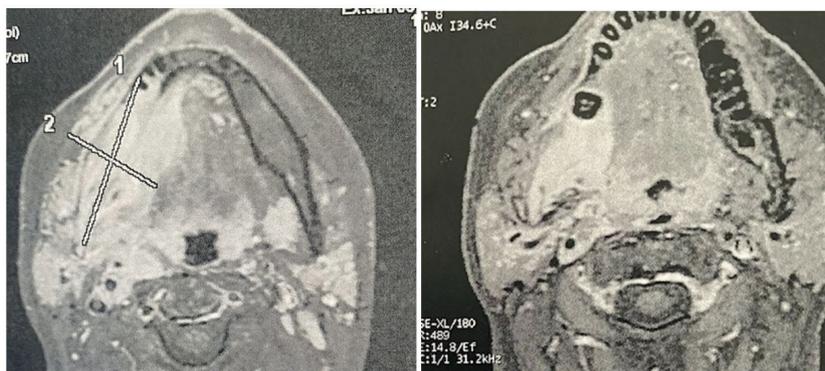


Figure 3: MRI aspect of the bone process of the right mandible

Discussion

ACC is a relatively uncommon tumor comprising about 1% of all head and neck malignancies and approximately 10% of all salivary gland tumors [2-4].

The palate is the most frequently affected site, followed by the tongue, buccal mucosa, lip, and floor of the mouth [5].

ACC is a slow-growing but highly invasive tumor, with high rates of recurrence and perineural spread [1,6,7].

Pathogenesis of central salivary gland tumors is unknown, but it is considered they originate from epithelial linings of cysts, particularly dentigerous cysts, or more probably from ectopic salivary gland tissue [8].

This lesion rarely affects the jaws [8].

ACC can occur in all age groups with high frequencies in the fifth and sixth decades of life, and a gender predilection was noted as an inconsistent feature in the literature [9].

ACC usually presents as a slowly growing, firm, unilobular mass, and pain is a common finding, occasionally occurring early in the course of the disease [6,10].

Clinically and radiographically they may mimic odontogenic cysts and tumors, and a final diagnosis can be obtained only after histological examination [11].

CT and MRI are important to delineate the extent, local invasion, and perineural spread of the tumor [12,13].

Strict diagnostic criteria have been established to confirm the central origin of salivary gland tumors involving the mandible:

1. Radiographic evidence of osteolysis;
2. Presence of intact cortical plates;
3. Absence of primary lesions within the salivary glands; and
4. Histological confirmation [11,14].

Conclusion

We have described a rare case of a primary bone ACC of the mandible, this case was extremely rare in terms of the site of origin and invasion pattern.

CT and MRI are important to delineate the extent, local invasion, and perineural spread of the tumor.

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Conflicts of Interests

The authors declare no conflicts of interests.

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