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CASE REPORT

Alveolar soft-part sarcoma of the masseter and mandibular ramus: Report of a case and review of the literature

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Abstract Alveolar soft-part sarcomas are clinically and morphologically distinct soft-tissue sarcomas, with an unknown histogenesis. When the tumors affect the region of the head and neck, they are often located in the orbit and tongue. We report a case of an alveolar soft-part sarcoma in the left masseter of a 28-year-old female. The patient had chronic pain and paresthesia of her left lower lip. Panoramic radiography and computed tomography showed a well-delimited radiolucent mass in the left ramus. An incisional biopsy was performed, and the sample submitted for histopathological study. The tumor showed positive periodic acid-Schiff diastase-resistant granules. Immunohistochemically, the tumor cells were diffusely positive for myoglobin, and focally positive for actin and desmin.

Introduction

Alveolar soft-part sarcomas (ASPSs) are clinically and morphologically distinct soft-tissue sarcomas first defined and named by Christopherson et al in 1952.[1] ASPSs are rare, uniform malignant neoplasms with no benign counterpart that account for <1% of all soft-tissue sarcomas.[1–7] Tumors are usually associated with muscle, but the histogenesis

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remains uncertain.[2,3,8] Most of the tumors occur in adolescents and young adults (age 15—30 years), and females are more often affected.[5,8] There are two main locations of these tumors. In adults, they are seen predominantly in the lower extremities. In infants and children (27%), tumors are often located in the region of the head and neck, especially the orbit and tongue.[1,2,4–7,9] ASPSs usually present as slowly growing, painless masses that almost never cause functional impairment. Because of the relative lack of symptoms, they are easily overlooked. In a number of cases, metastasis to the lung or brain is the first manifestation of the disease.[1]

We present a case of a 28-year-old female, suffering an apparent toothache, who presented with ASPS of the masseter and left mandibular ramus, and review the literature of the past 10 years.

Case report

A 28-year-old female was referred to the Department of Oral Pathology and Surgery of Andres Bello University, for evaluation of chronic pain of her left mandible and paresthesia of her left lower lip, within a period of 2 months, in which her general practitioner performed a root canal on tooth 3.7. In the intraoral examination, no problems in the mouth opening or oral mucosa, and no tooth alterations were seen. Upon palpation, the patient presented an ill-defined 3-cm-long painful mass that arose from her left ramus, and was covered by the normal mucosa. A panoramic radiograph showed a well-delimited radiolucent mass in the left ramus with effacement of the mandibular channel (Fig. 1). Computed tomography (CT) showed that the lesion was destroying the entire ramus and infiltrating the masseter (Fig. 2). A malignant lesion, osteosarcoma like, was suggested by the Imagenology Department of the Naval Hospital. An incisional biopsy under local anesthesia was performed. During this procedure, abundant hemorrhage and pain refractory to a local anesthetic were observed.

Pathology

Several irregular and soft-tissue sections of the lesion were sent to the Oral Pathology laboratory of Andres Bello University, with a presumptive diagnosis of an oral sarcoma.

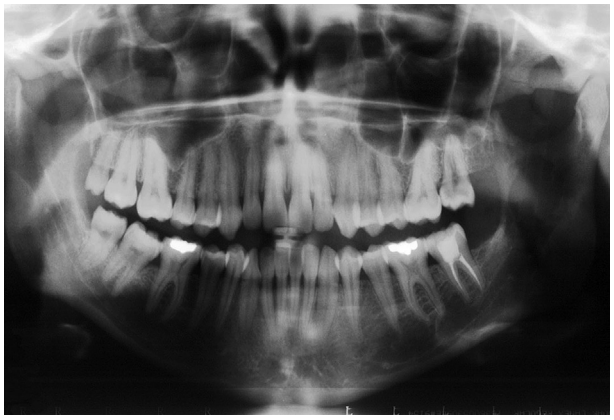

Figure 1 Panoramic radiograph showing a well-defined radiolucency in the left mandibular ramus.

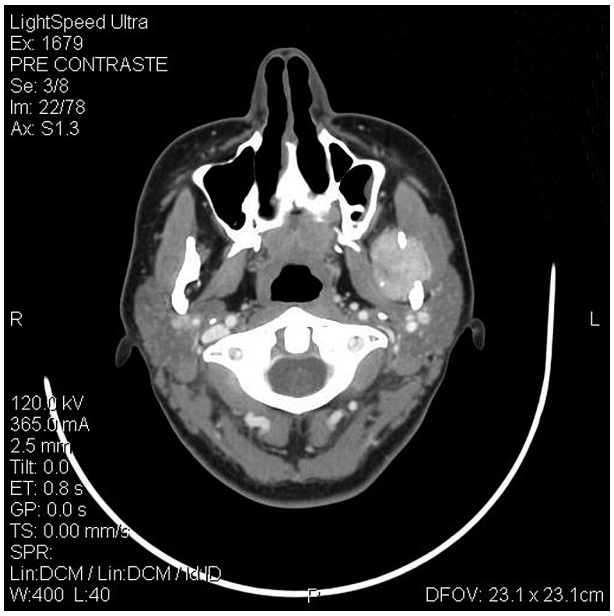

Figure 2 Axial computed tomography scan of the patient showing a well-circumscribed mass involving the left masseter and mandibular ramus.

A microscopic study using routine techniques with hematoxylin and eosin showed proliferation of large polygonal cells separated by thin fibrous septa and numerous vascular channels disposed in a lobular arrangement. The nuclei were round and disposed centrally, and few and normal mitoses were seen. Cells contained abundant granular eosinophilic cytoplasm (Fig. 3).

The patient was sent to the Renal Pathology Department to rule out the possibility of a clear cell metastatic carcinoma of the kidney. Echography of the kidney was taken, and no changes were observed.

The periodic acid-Schiff technique was used, and positive diastase-resistant granules were focally seen. On immunohistochemistry (IHC), tumor cells were diffusely positive for myoglobin, focally positive for actin and desmin (Fig. 4), and negative for cytokeratin, S-100, and chromogranin.

Samples were discussed with two general pathologists of the main hospitals of the region, and both agreed with our diagnosis of ASPS.

Treatment and follow-up

The patient was referred to the Oncology Department of a local hospital, where complete excision of the lesion and a partial hemimandibulectomy were performed. The histopathological findings were similar to those of the previous biopsy, and the diagnosis of ASPS was corroborated. One lymph node was examined, but no tumor-cell infiltration was observed. A complete CT scan was performed, and no metastases were seen. Additional radiotherapy was given, because there was a microscopic positive margin. The orientations of the edges were impossible to locate due to the

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