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Unusual features of oral sarcomatoid carcinoma: a case report

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ABSTRACT

Oral sarcomatoid carcinoma (OSC) is considered a rare variant of oral squamous cell carcinoma (OSCC). It is extremely aggressive, affecting mainly older men with risk habits. We report an atypical case of OSC, in a 40-year-old black female that presented an invasive ulceration covering the left hard palate to the ipsilateral buccal mucosa with tissue loss and buccosinusal communication, measuring about 10 x 8 cm. The patient didn't refer any risk habits. After incisional biopsy, histopathological examination was suggestive for a spindle cell malignant neoplasm. Immunohistochemistry (IHC) revealed positivity for keratins (CK) 7,8/18 and p63 and negativity for calponin, S-100, α-SMA and SOX-10. The patient underwent 9 cycles of chemoterapy, but unfortunately, she died 3 months later. This is an atypical case of OSC, since it usually occurs in older male patients with risk habits. This tumor may lead pathologists to misdiagnosis and IHC is crucial for definitive diagnosis.

Keywords: *Sarcomatoid carcinoma; Head and neck cancer; Spindle cell carcinoma; Case report.*

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INTRODUCTION

Oral sarcomatoid carcinoma (OSC) is a rare and aggressive variant of oral squamous cell carcinoma (OSCC), most commonly affecting the tongue and floor of the mouth. Clinically, it presents as a rapidly growing exophytic or ulcerated mass, often associated with local invasion and a poor prognosis[1,2]. Due to its rarity and clinical complexity, OSC poses diagnostic and therapeutic challenges for clinicians[1,2]. Additionally, most cases reported in the literature involve patients with known risk factors such as tobacco and alcohol use, making cases without these factors uncommon and worthy of attention[1-4]. Reporting such atypical presentations contributes to a better understanding of OSC's clinical spectrum and diagnostic criteria.

OSC has a similar histology with sarcomas[1,2]. Sarcomatoid carcinomas are extremely aggressive, highly recurrent, and metastatic neoplasms, affecting mainly older men[3,4]. The etiology of OSC is unknown; however, some risk factors such as exposure to radiation, tobacco and alcoholic beverages may be associated with the appearance of the lesion[5]. The diagnosis of OSC is challenging and it is based on clinical, histopathological and immunohistochemical features[2,6-8]. Wide surgical excision, with or without neck dissection seems to be the most indicated treatment and the therapeutic modality with better results[1,3,9].

The aim of this paper is to report a case of oral sarcomatoid carcinoma (OSC) presenting with an unusual clinical profile, characterized by palatal localization in a female patient with no identifiable environmental risk factors. The diagnosis was confirmed through histopathological evaluation and immunohistochemical analysis, revealing features closely resembling a sarcoma.

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CASE REPORT

A 40-year-old black female patient was referred to a hospital in south Brazil with a complaint of pain and edema on the left side of the face lasting around three months. In the anamnesis, the patient reported that she was seen at a dental private clinic two months ago. At that time, she was diagnosed with infection of dental origin and tooth extraction was performed. Also, biopsy of the lesion was obtained with inconclusive histopathological diagnosis. At the hospital, a facial asymmetry with swelling on the left side of the face was observed at extraoral examination (**Figure 1**). It was also noticed that she had mouth opening limitation. Intraorally, it was observed an invasive ulceration covering the left hard palate to the ipsilateral buccal mucosa with tissue loss and buccosinusal communication (**Figure 2**). The patient reported no systemic disease and no smoking or drinking habit. Social and family history was not contributory. A cone bean computed tomography (CBCT) with contrast was performed, showing a massive infiltrative lesion in the left hemiface with heterogeneous appearance and predominantly peripheral to the contrast. The lesion determined extensive osteolysis of the maxillary sinus walls and the left hard palate with communication between the maxillary sinus and the nasal and oral cavities. The lesion was also invading the chewing space with infiltration of the masseter muscle and contact with parotid gland, measuring about 10.0 x 8.0 cm. (**Figure 3**).

An informed consent form was obtained, signed by the patient, and properly archived. A new incisional biopsy was performed on palate under local anesthesia, and microscopic examination revealed spindle cell proliferation invading skeletal muscle with areas of coagulative necrosis and mild atypia. Individually, the tumor cells were pale and plump with eosinophilic cytoplasm, vesicular nuclei, prominent nucleoli, pleomorphism, and frequent mitotic figures. Based on these features, our first hypothesis was mesenchymal malignancy, such as sarcoma (**Figure 4**). Immunohistochemistry (IHC) (**Figure 5**) was performed for definitive diagnosis. Immunostaining was positive for cytokeratin 7 (Roche Diagnostics, Switzerland), cytokeratin 8/18 (Cell Marque Corporation, California, USA) and p63 (Biocare Medical, California, USA), and negative for calponin (Cell Marque®), alpha actin smooth muscle (Cell Marque®), S-100 (Roche Diagnostics®) and SOX-10 (Bio SB Inc., Santa Barbara, California, USA). Based on the microscopic and IHC features, the diagnosis was OSC. After the diagnosis, the patient was referred to the service of Oncology of the hospital. The treatment suggested for the case was chemotherapy with gemcitabine combined with docetaxel and radiotherapy. Three months later, the patient concluded 9 cycles of chemotherapy and died before starting radiotherapy.

Figure 1: Extra-oral presentation showing facial asymmetry with edema and hematoma on the left side of the face.

Figure 2: Intra-oral presentation revealing an invasive ulceration covering the left side of the hard palate and extending to the buccal mucosa.

Figure 3: Imaging findings. CBCT showing an extensive and expansile lesion on the left hemiface, involving both osseous and soft tissues (white arrows). Presence of heterogeneous osteolytic areas with marked bone destruction, consistent with necrotic processes (yellow arrows).

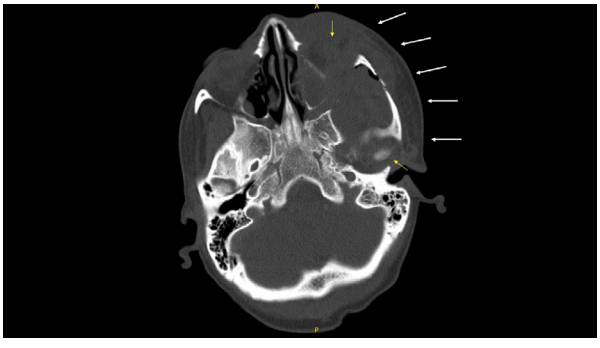


Figure 4: Microscopic Findings. Microscopic examination revealed fusocellular proliferation invading skeletal muscle with areas of coagulative necrosis and mild atypias. Individually, the tumor cells were pale and plump with eosinophilic cytoplasm, vesicular nuclei, prominent nucleoli, pleomorphism, and frequent mitotic figures. (amplifications: **A** and **B**: 40x; **C** and **D**: 100x; **E**:200x and **F**:400x)

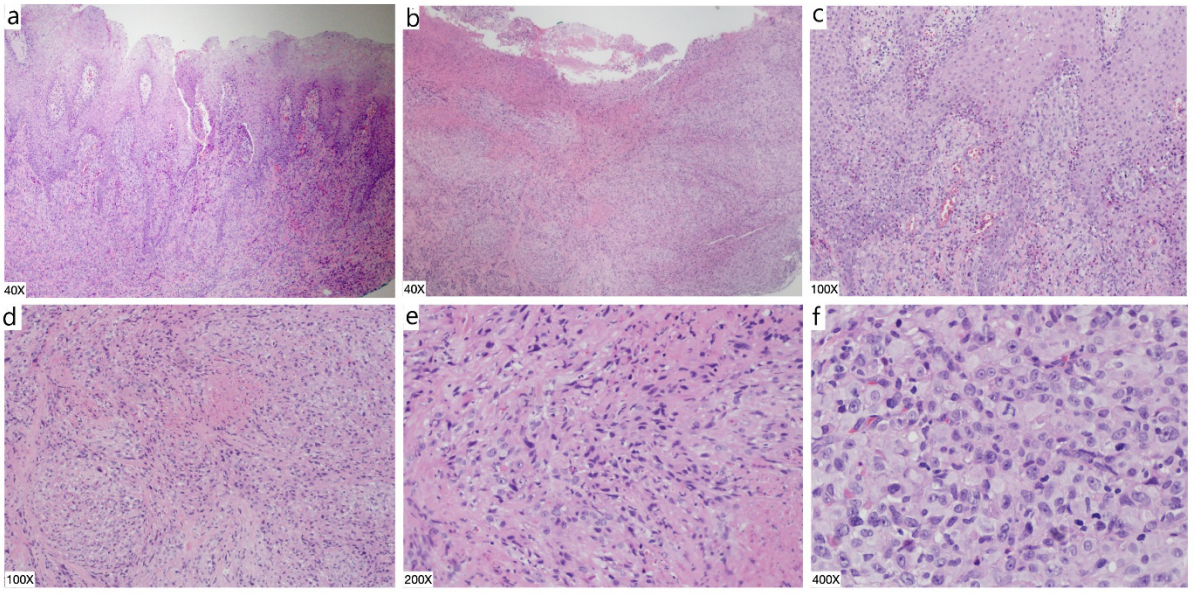
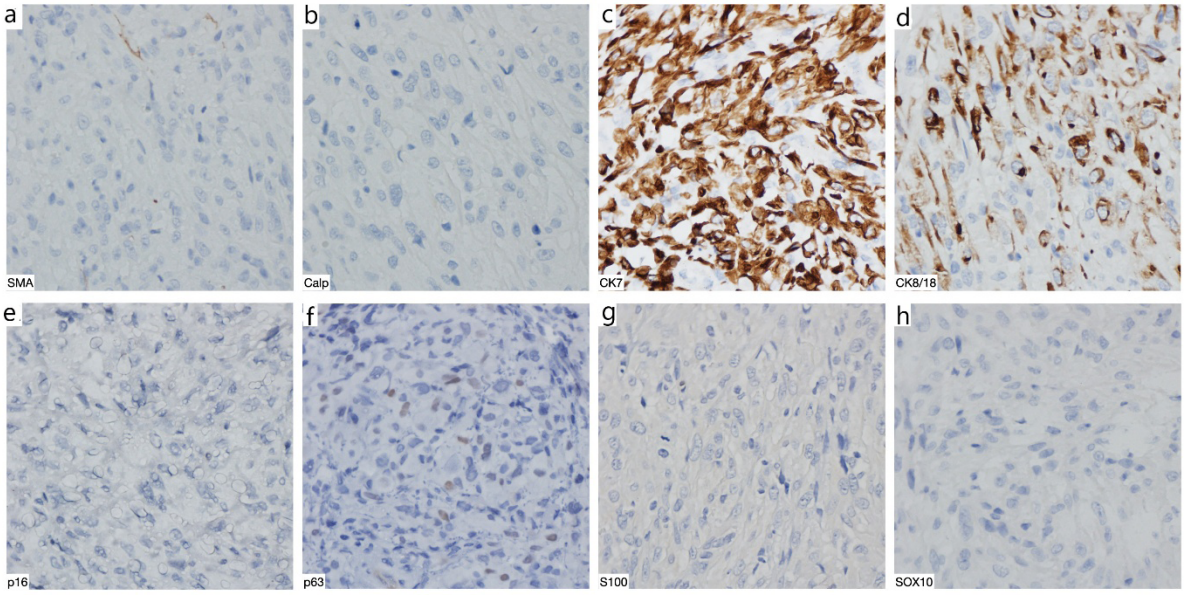


Figure 5: Immunohistochemical features. Negative immunostaining for smooth muscle actin (**A**), calponin (**B**), p16 (**E**), S-100 (**G**), and SOX-10 (**H**). Positive immunostaining for CK7 (**C**), CK8/18 (**D**) and p63 (**F**).



DISCUSSION

The occurrence of OSC is extremely rare, with only a limited number of cases reported in the literature[2,3,6,9-12]. Epidemiologically, OSC accounts for less than 1% of all oral squamous cell carcinomas, highlighting its rarity and diagnostic challenge. A brief review[2,3,6,9-12] of the literature reveals scattered case reports and small case series documenting this lesion, reinforcing the value of each new case report in expanding the current understanding.

OSC is considered a variant of OSCC that tends to present in older male patients, usually as a distinctive polypoid growth[1]. Typically, OSC affects predominantly males over 60 years of age who have a history of tobacco and alcohol use, and the

lesion often presents as a rapidly growing, polypoid or exophytic mass, frequently with ulceration and a destructive pattern in the oral cavity. In contrast, our patient is fairly atypical to the clinical presentation of OSC, being a 40-year-old female non-smoker without any known environmental risk factors. This divergence from the common demographic and clinical profile underscores the uniqueness of this case and reinforces the importance of considering OSC even in patients who do not fit the classic risk factors or demographic characteristics.

OSC may present as a diagnostic dilemma especially to the pathologists signing out the biopsy case who may confuse it with mesenchymal malignancies. Thereby, in view of the location, destructive appearance, and histopathological features, it seems reasonable that our first hypothesis was sinonasal sarcoma (SS). SS is a rare spindle cell neoplasm in the sinonasal region[1,13,14]. This pathology seems to be more common in females, at a median age of 48 years, aggressive and recurrent[13]. Histopathological features frequently reveal spindle cell proliferation arranged in fascicles, plump cells with abundant eosinophilic cytoplasm and aberrant mitosis[13,14]. Some of these features are seen in this case. However, the SS immunohistochemical panel includes positivity for S-100 and smooth muscle actin in the spindle cells[13], and these markers were negative on the IHC panel performed in this case (see **Figure 5**).

OSC presents controversies regarding its possible origin, due to its histological characteristic since both carcinomatous epithelial cells and sarcomatous spindle cells can be found in it[1,6]. Immunohistochemical analysis is required for a more accurate diagnosis and exclusion of true spindle cell sarcomas[6,8]. In the absence of immunohistochemical analysis, if there is a predominance of sarcomatoid components, it can be easily confused with a sarcoma[2,8]. Thus, immunohistochemical analysis is essential for the diagnosis. The most sensitive markers for detecting the epithelial origin of the sarcomatoid component seem to be mainly cytokeratins (CK)[8]. In the present case, an immunohistochemical study was performed using CK7 and CK8/18, which showed positivity for both markers. Mesenchymal markers such as calponin and alpha actin smooth muscle, and neural markers such as S-100 and SOX-10 were not expressed in the sample of this study. These findings supported ruling out neoplasms of mesenchymal or neural origin.

Wide surgical excision seems to be the most indicated treatment and the therapeutic modality with better results[1,3,9]. Adjuvant therapy such as chemotherapy is indicated for patients with advanced and unresectable sarcomatoid carcinoma, as well as for those with compromised surgical margins or lymph node metastasis[3,9]. Radiotherapy can be indicated despite controversies in the literature[2,6]. In the present case, the tumor was already at a very advanced stage that surgical approach was not indicated. Therefore, chemotherapy seemed to be the most viable alternative, but without regression of the lesion. Unfortunately, the patient died three months after the diagnosis.

OSC is a rare lesion and its diagnosis is challenging. Microscopic and immunohistochemical characteristics are essential for the accurate diagnosis of this type of tumor. An agile diagnosis must be made for better prognosis and increased survival rates.

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