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# Use of photobiomodulation in the treatment of tissue complications after resection of leiomyosarcoma of the maxilla

*Uso de la fotobiomodulación en el tratamiento de las complicaciones tisulares tras la resección de un leiomyosarcoma del maxilar*

Bianca de Fátima Borim Pulino,<sup>\*</sup> Daniel Natri de Luca,<sup>‡</sup> André Luiz Luchini Predin,<sup>§</sup> Henry García Guevara,<sup>¶</sup> Eduardo Hochuli Vieira,<sup>||</sup> Robert Alexander Sader,<sup>\*\*</sup> Raphael Capelli Guerra<sup>††</sup>

## Keywords:

Leiomyosarcoma, sarcoma, soft tissue tumors, head and neck cancer.

## Palabras clave:

Leiomyosarcoma, sarcoma, tumores de tejidos blandos, cáncer de cabeza y cuello.

## ABSTRACT

Leiomyosarcoma is a type of cancer that originates from smooth muscle cells in the uterus, gastrointestinal tract, and other soft tissues. The rare occurrence of leiomyosarcoma in the oral cavity has been correlated with the scarcity of smooth muscle structures in this location. It is a rare and aggressive soft tissue tumor that accounts for approximately 5-10% of all soft tissue sarcomas, with only 3% of these tumors occurring in the head and neck region. Differential diagnosis can be problematic. The physical appearance can be confused with non-malignant conditions. Clinical differential diagnosis of the lesion includes glandular tumors (pleomorphic adenoma, mucoepidermoid carcinoma and adenoid cystic carcinoma) and mesenchymal tumors. We report a case of maxillary leiomyosarcoma in an 86-year-old patient who underwent tumor resection and had almost complete necrosis of the surgical flap as a complication. Through laser therapy, we achieved complete healing of the flap and implant and dental rehabilitation with immediate loading.

## RESUMEN

El leiomyosarcoma es un tipo de cáncer que se origina en las células musculares lisas del útero, el tracto gastrointestinal y otros tejidos blandos. La rara aparición del leiomyosarcoma en la cavidad oral se ha correlacionado con la escasez de estructuras musculares lisas en esta localización. Se trata de un tumor de tejidos blandos raro y agresivo que representa aproximadamente 5-10% de todos los sarcomas de tejidos blandos, y sólo 3% de estos tumores se producen en la región de la cabeza y el cuello. El diagnóstico diferencial puede ser problemático. El aspecto físico puede confundirse con afecciones no malignas. El diagnóstico diferencial clínico de la lesión incluye los tumores glandulares (adenoma pleomórfico, carcinoma mucoepidermoide y carcinoma adenoide quístico) y los tumores mesenquimales. Relatamos un caso de leiomyosarcoma maxilar en un paciente de 86 años que fue sometido a una resección tumoral y tuvo como complicación la necrosis casi completa del retalto quirúrgico. Gracias a la terapia con láser, logramos la curación completa del colgajo y el implante y la rehabilitación dental con carga inmediata.

## INTRODUCTION

Leiomyosarcoma is a malignant tumor of smooth muscle origin. It is a rare tumor that was first described by Bartkowiak.<sup>1</sup> Most leiomyosarcomas arise in the gastrointestinal tract, uterus, and skin. Few cases occur in the oral cavity, and leiomyosarcoma of the jaw are extremely rare.<sup>2</sup>

Soft tissue sarcomas of the head and neck are rare, approximating less than 10% of all soft tissue sarcomas and less than 1% of all neoplasms in this region. They present as malignant tumors of the smooth muscle that account for only 4% of head and neck sarcomas and only 41 cases of leiomyosarcoma of the head and neck structures have been reported in the English literature to date.<sup>3</sup>

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\* PhD Student-Oral and Maxillofacial Surgery. Department of Oral and Maxillofacial Surgery, School of Dentistry, Sao Bernardo do Campo, Methodist University of Sao Paulo; Department of Oral and Maxillofacial Surgery, Dasa, Grupo Leforte; Department of Diagnostics and Surgery, School of Dentistry, Aracatuba, Sao Paulo State University-Unesp, Brazil. ORCID: 0000-0003-0519-3246.

‡ Specialist in Oral and Maxillofacial Surgery, Orofacial Harmonization, TMD and Orofacial Pain. Collaborating Professor at the Center for Research and Teaching of Phototherapy in Health Sciences, Institute NUPEN, Sao Carlos, Brazil. Owner of Ecodonto Dental Clinic, Sao Carlos, SP, Brazil. ORCID: 0000-0002-8005-5719.

§ Oncological surgery in private clinic. ORCID: 0000-0002-7879-6382.



<sup>†</sup> Oral and Maxillofacial Surgery, Department of Oral and Maxillofacial Surgery, Children's Orthopedic Hospital of Caracas, Chief of the Department of oral surgery, Santa Maria University-Venezuela. ORCID: 0000-0002-1840-7568.

<sup>‡</sup> PhD, Professor-Oral and Maxillofacial Surgery, Department of Oral and Maxillofacial Surgery, Group Leforte, DASA; Department of Diagnosis and Surgery, School of Dentistry, Araraquara, Sao Paulo State University-Unesp, Brazil. ORCID: 0000-0003-4040-9313.

<sup>\*\*</sup> PhD, Cranio-Maxillofacial and Facial Plastic Surgery, Head of Department for Oral, Cranio-Maxillofacial and Facial Plastic Surgery Hospital of the Wolfgang Goethe University Frankfurt. ORCID: 0000-0002-0265-0301.

<sup>††</sup> PhD, Oral and Maxillofacial Surgery, Department of Oral and Maxillofacial Surgery, School of Dentistry, Sao Bernardo do Campo, Methodist University of Sao Paulo; Department of Oral and Maxillofacial Surgery, Dasa, Grupo Leforte, Brazil. ORCID: 0000-0002-9071-7827.

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This tumor occurs in regions with scarce or absent smooth muscle structures, such as the jawbone and oral tissues, possibly arising between the blood vessels, myoepithelial cells, and/or mesenchymal cells.<sup>4</sup>

Leiomyosarcomas usually have a distinctive morphology and architectural features with the following characteristics: they are composed of cells with elongated nuclei and eosinophilic cytoplasm, sometimes with longitudinal fibrils. More rarely, these tumors are formed by rounded epithelial cells with eosinophilic or transparent cytoplasm (epithelioid leiomyosarcoma). The presence of frequent and atypical mitotic figures and necrotic foci indicate an aggressive behavior.<sup>5</sup>

We report a case of leiomyosarcoma of the maxilla in an 86-year-old patient who underwent tumor resection and presented with almost complete necrosis of the surgical flap. Through laser therapy, it was possible to achieve complete healing of the flap, and dental implantation and rehabilitation with immediate loading.

## CLINICAL CASE

Patient MM, aged 86 years, was referred by an otorhinolaryngologist due to lesions and pain in the maxilla for six months. He had a maladjusted upper total prosthesis, which induced hyperplasia. He changed his total prosthesis two months ago, but he reported worsening, though there was no local secretion. He reported hypertension, continued use of antihypertensive medications, and denied having any allergies.

Intraoral clinical examination revealed a painless clinical lesion, measuring 15 × 20 mm, swollen and with a smooth surface, compression an increase in volume in the alveolar ridge by the palate adjacent to the upper right second premolar and upper right first molar. No fistula or secretion was present, but there was hardened tissue with a slightly purplish color (*Figure 1*). In the computed tomography scan of the face, there were no signs of bone involvement (*Figure 2*).



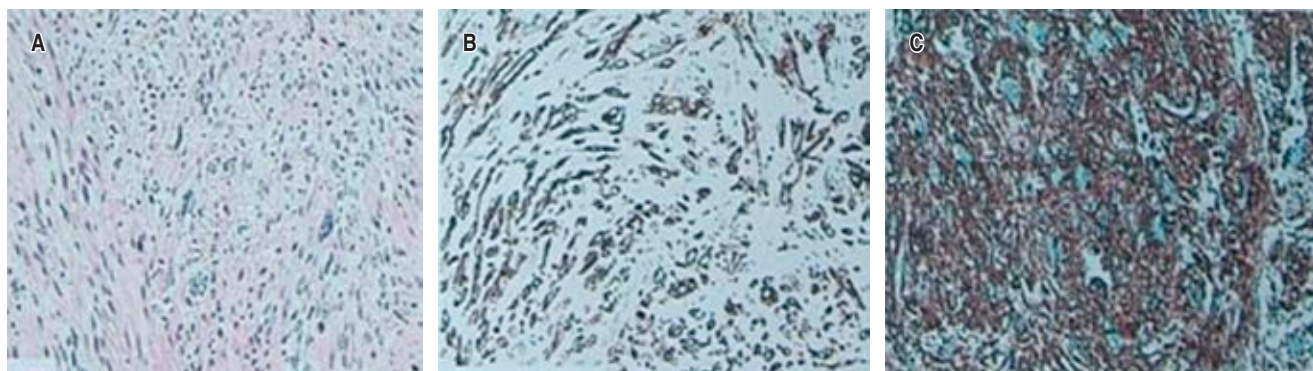
**Figure 1:**

*The intraoral clinical examination reveals an increase in volume in the alveolar ridge by the palate in the region of the upper second right premolar and upper first right molar.*



**Figure 2:**

*The computed tomography scan of the face does not show changes in bone structures and soft tissues in the maxilla.*



**Figure 3:** **A)** The tumor is shown to be positive for the antibody desmin (intermediate filament of the muscle cell) in the immunohistochemical technique. **B)** The tumor is shown to be positive for the smooth muscle actin antibody in the immunohistochemical technique. **C)** The tumor is shown to be positive for the epithelial membrane antigen antibody in the immunohistochemical technique.

An excisional biopsy was performed under local anesthesia with complete enucleation of the lesion, which was easily removed with simple peeling and curettage using a Molt's peeler. After removal, we performed superficial vaporization of the cavity and hemostasis in the region of the palatine artery with a 2-W-pulsed surgical diode laser (980 nm; Thera Lase, DMC Sao Carlos/SP). Continuous suturing was performed, chlorhexidine gel was placed, and the patient was instructed to wear the upper total prosthesis normally.

Pathological examination revealed high-grade leiomyosarcoma 2 and 3. To define the diagnosis, we utilized an immunohistochemical technique using the antibodies desmin (intermediate filament of the muscle cell) (Figure 3A), smooth muscle actin (Figure 3B), and epithelial membrane antigen (EMA) (Figure 3C).

Because the lesion did not show a good prognosis with chemotherapy and radiotherapy treatment, resection of the lesion with a soft tissue safety margin was scheduled together with the oncological head and neck surgeon.

Under general anesthesia, the patient underwent surgery, during which an extensive area of soft tissue was removed from the right posterior palatal region. After removal, slight changes in the coloration and texture of the palatal alveolar bone were observed. A chisel and hammer were used to perform a partial posterior osteotomy of the right maxilla. To close the surgical wound, a pedicled flap of the right jugal mucosa was rotated and sutured using Monocryl 4.0. A nasoenteral tube was used for feeding, and the patient was discharged from the hospital 24 h after the procedure; the patient was stable at this time. The patient was re-evaluated by the oncologic surgeon to remove the suture, and almost complete necrosis of the flap was observed (Figure 4), evolving to extensive buccosinusal communication. The patient was treated with clindamycin, ketoprofen, and dipyrrone, and the possibility of starting laser therapy by an oral and maxillofacial surgeon was discussed.



**Figure 4:** A necrotic area on the palate can be observed after the excisional biopsy due to a deficiency in local vascularization.

When evaluating the patient, we proposed treatment with decontamination through photodynamic therapy with methylene blue at 0.01% (Chimiolux - DMC, Sao Carlos/SP) and photobiomodulation using a low-power red laser at 660 nm and 100 mW in the bleeding parts and infrared at 808 nm and 100 mW (Therapy EC - DMC, Sao Carlos/SP) around the lesion. In addition, the patient's mold was taken, which was used to make an acetate plate to be applied in the jaw with chlorhexidine gel (PerioKIN; Laboratorios KIN, San Salvador, El Salvador) to protect the wound and prevent the entry of liquids and contact with the tongue (Figure 5). Six sessions were performed within 30 days, with healing and closure in almost 90% of the affected area.

The patient underwent implant installation and placement of a fixed prosthesis in the maxilla with immediate loading 90 days postoperatively (Figure 6). A palatal obturator prosthesis with retention at the edges of the fixed dental prosthesis was also made.

Upon the patient's return for the six-month control, the buccosinusal communication was partially closed, and

he reported that he was no longer wearing the obturator prosthesis (Figure 7). Upon the patient's return for the nine-month control, there was no more visible buccosinus communication, although there was a hole that could be penetrated with an exploratory probe.

As the tissue healed and the edges epithelialized, vaporization of the hole was suggested to de-epithelialize the edges and promote closure. The procedure was performed only with topical anesthetic, and the 1-W surgical diode laser in pulsed mode (980 nm, Thera Lase - DMC, Sao Carlos/SP) was used. A 2-J punctual low-power res laser was then applied over the fistula (660 nm and 100 mW; Therapy EC DMC, Sao Carlos/SP). The procedure was repeated after eight days.

During the patient's return four years after the last procedure, it was found that he maintained complete healing and stability. Up until the time of writing, there have been no recurrences (Figure 8).

## DISCUSSION

Leiomyosarcoma is a type of cancer that originates from smooth muscle cells located in the uterus, gastrointestinal tract, and/or other soft tissues, such as the retroperitoneum.<sup>6</sup> It is a rare, aggressive soft tissue tumor that accounts for approximately 5-10% of all soft tissue sarcomas, with only 3% of these tumors occurring in the head and neck region.<sup>7</sup>

This tumor often presents as an enlarged, painless, and slow-growing mass, and a variety of symptoms may appear based on its location. In the head and neck region, especially when the maxillary or other paranasal sinuses are involved, it commonly presents with nasal obstruction, epistaxis, local pain, and/or facial edema.<sup>8</sup>

In the oral cavity, the maxillary sinus, mandible, and maxilla seem to be prone areas for leiomyosarcoma, but



**Figure 5:** An acetate plate with chlorhexidine gel was applied to the jaw to protect the wound and prevent the entry of liquids or contact with the tongue.



**Figure 6:** The radiographic examination reveals the installation of five dental implants in the maxilla for a dental prosthesis.

other intraoral locations reported are the cheek, tongue, hard and soft palates, lips, and gums. The rare occurrence of leiomyosarcoma in the oral cavity has been correlated with the scarcity of smooth muscle structures in this location compared to its abundance in other locations. It is a malignant neoplasm of smooth muscle, which can originate from the tunica media of the blood vessels, circumvallate papillae of the tongue, and/or pluripotent mesenchymal cells. Leiomyosarcomas are usually divided by anatomical location into three groups: 1) leiomyosarcomas of deep tissues, the most frequently occurring group, predominantly involving the uterus, gastrointestinal tract, and retroperitoneum; 2) leiomyosarcomas of vascular origin, from the tunica media; and 3) those that affect the skin and more superficial subcutaneous tissues and the extremities.<sup>9</sup> The tumor has no specific age or sex predilection.<sup>10</sup>

When they occur in the respiratory tract, the most common sites are the nasal cavity, the maxillary sinus, and the ethmoid sinus. Therefore, our patient exhibited a rare case of leiomyosarcoma of the right maxilla.

The differential diagnosis of this rare tumor can be problematic. The physical appearance can be confused with non-malignant conditions.<sup>11</sup> The clinical differential diagnoses for the lesion include benign and malignant salivary gland tumors (pleomorphic adenoma, mucoepidermoid carcinoma, adenoid cystic carcinoma, and polymorphic low-grade adenocarcinoma) and benign and malignant mesenchymal tumors.<sup>12</sup>

Owing to the lack of a distinct radiographic presentation, these tumors can mimic any expansive or destructive lesion of the maxilla and mandible. They may present as lytic lesions with ill-defined margins, periosteal elevation, or cortical destruction.<sup>13</sup>

Therefore, the diagnosis is supported by immunohistochemistry and ultrastructural investigations.

He histological features of leiomyosarcomas vary but usually show arrangements of spindle-shaped cells with

eosinophilic cytoplasm, blunt stumps, and cigar-shaped nuclei. In the epithelioid variant, which develops occasionally, the neoplasm is composed of round epithelioid cells with clear or eosinophilic cytoplasm. When the tumor presents with a well-differentiated pattern or low degree of malignancy, it is difficult to distinguish it from leiomyoma. Because of this difficulty, some clinical and histological criteria have been proposed as indicators of differentiation between leiomyosarcoma and leiomyoma: 1) rapid growth of the lesion is considered a clinically useful indicator of malignancy; 2) neoplasia in the gastrointestinal system sized > 30 mm should also be considered a possibly malignant entity; 3) the presence of neoplastic cells in the lymph nodes or of distant metastases is conclusive of malignancy; and 4) a high incidence of mitoses is the most important finding from a histological point of view in the definition of malignancy. As a parameter, five to ten mitoses should be observed in the high-magnification field. Other important findings include cellular pleomorphism, necrosis, hyperchromatism, and nuclear atypia.<sup>4</sup>

In our case, it was necessary to perform an immunohistochemical study to help elucidate the diagnosis using the antibodies desmin (intermediate filament of the muscle cell), smooth muscle actin, and EMA.

Electron microscopy revealed intracytoplasmic microfilaments with numerous dense cytoplasmic bodies, micropinocytotic vesicles, and a partially discontinuous lamina basale.<sup>9</sup>

The treatment of this type of lesion is difficult because of the high recurrence and metastatic rates and poor prognosis.<sup>14</sup>

Complete surgical resection is generally recommended. Margins must be removed to control local recurrence, and adjuvant radiotherapy or chemotherapy is considered;



**Figure 7:** The clinical examination reveals that the buccosinusal communication is partially closed.



**Figure 8:** No alteration of tissues can be observed in the maxilla 6 months after the installation of the dental prosthesis.

however, they have little beneficial effect in decreasing leiomyosarcoma recurrence or increasing survival time. However, at some specific anatomical sites, such as the vicinity of the infratemporal fossa, maxillary sinus, pterygoid plates, and mandibular condyle, it may be less technically feasible to achieve tumor-free margins due to difficulty, possibly resulting in residual microscopic disease, which would lead to local tumor recurrence and a worse prognosis.<sup>4</sup>

In our case, it was possible to resect the tumor with safety margins, and, during the four years follow-up, there was no recurrence of the lesion.

Some cases of metastatic leiomyosarcoma in the oral cavity have been described. Distant intraoral leiomyosarcoma metastases occur in up to 39% of these cases. With oral leiomyosarcomas, metastasis to the regional lymph nodes is relatively rare, and the most common site of metastasis is the lungs.<sup>9</sup>

The process of tissue repair is complex and comprises vascular and cellular alterations, epithelial and fibroblast proliferation, collagen synthesis and deposition, elastin and proteoglycan production, revascularization, and wound contraction. Trophic-regenerative, anti-inflammatory, and analgesic effects also stand out.<sup>15</sup>

It was also affirmed that low-power laser therapy can generate increased mitochondrial activity, with a consequent increase in adenosine triphosphate (ATP), vasodilation, protein synthesis, a decrease in prostaglandin levels, presence of cell mitosis, migration and proliferation of keratinocytes, and occurrence of the neoangiogenesis phenomenon. The increase in collagen production occurs through photostimulation mechanisms, on which certain frequencies or doses may act, thus modulating cell proliferation and increasing the amount of fibroblast growth factors. Another possible explanation for this is that this type of energy can be better absorbed by the mitochondria, leading to a higher production of ATP and nucleic acids, the result of which is the increase in collagen

production, acceleration of epithelial repair, and facilitation of granulation tissue growth.<sup>11</sup>

The excitatory doses (up to 8 J/cm<sup>2</sup>) are indicated when the objective of the intervention includes potentiation of the sodium/potassium pump, stimulation of ATP production, restoration of the membrane potential, and increased metabolism and cell proliferation.

Despite the aggressiveness of the tumor, the case reported in this article presented a good prognosis after resection, with local healing using laser therapy resources to correct complications during treatment.

Dental implants can be installed, and rehabilitation can be performed through immediate loading, thereby restoring the patient's chewing and aesthetic function. However, it is necessary to monitor patients for intervention if any tissue or imaging alterations occur at the site.

## CONCLUSION

Leiomyosarcoma is a relatively rare tumor in the oral and maxillofacial regions and has a poor prognosis because of its high recurrence rate. A thorough morphological analysis and careful interpretation of immunohistochemical markers are necessary to reach a correct diagnosis. An accurate diagnosis, a multidisciplinary approach and treatment are essential for favorable results. Photobiomodulation has been shown to be effective for tissue repair in the treatment of tissue complications after tumor resection.

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### Correspondence:

**Bianca de Fátima Borim Pulino - PhD Student**

Department of Diagnosis and Surgery, School of Dentistry, Aracatuba, Sao Paulo State University - Unesp, 1193 José Bonifácio st, Sao Paulo, Brazil. Postal Code: 16015-050. Phone: (+55) 18 3636 3237

**E-mail:** biancapulino@icloud.com