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Bulletin of the National Research Centre (2023) 47:41
https://doi.org/10.1186/s42269-023-01015-y

Bulletin of the National Research Centre

CASE REPORT

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Surgical and laser treatment for juvenile psammomatoid ossifying fibroma in an oncological patient: a rare case report

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Abstract

Background Juvenile psammomatoid ossifying fibroma is a rare variant of conventional ossifying fibroma that affects the maxillofacial complex in children and adolescents. It is a benign fibro-osseous neoplasm with aggressive clinical behavior and high recurrence rates, confirming the importance of early diagnosis and intervention to minimize undesirable aesthetic and functional possibilities resulting from surgical management, and added to the use of laser therapy, it can bring a better postoperative prognosis.

Aim To present the surgical removal of a juvenile psammomatoid ossifying fibroma, with the associated use of low-intensity laser and photodynamic therapy in a oncological patient.

Case presentation This case describes a senile cancer patient diagnosed with rapidly progressive and painful juvenile ossifying fibroma involving the inter-radicular region of the right maxilla. The treatment was excision with total tumor resection, photodynamic therapy and low-intensity laser follow-up of 48 months.

Conclusions Juvenile psammomatoid ossifying fibroma is a lesion with a high rate of recurrence and aggressive growth. Thus, early detection and treatment are essential. In this case, the use of photodynamic therapy after surgery and long-term follow-up with low-intensity laser brought a favorable prognosis in an oncology patient.

Keywords Ossifying fibroma, Oral neoplasms, Oral surgery, Low level light therapy, Photochemotherapy, Case report

Background

Juvenile psammomatoid ossifying fibroma (JPOF) is a rare neoplasm belonging to a group of fibro-osseous lesions characterized by the replacement of normal bone tissue by fibrous tissue, containing foci of mineralization of varying shapes and amounts (Chrcanovic and Gomez 2020). Due to its specificity, it is classified as an osteogenic neoplasm with clinical and histological characteristics different from juvenile trabecular ossifying fibroma, predilection for age and specific locations (Ranganath et al. 2014; Chrcanovic and Gomez 2020). They most frequently affect the bones of the maxillofacial complex of children and adolescents, involving the paranasal sinuses, orbit, frontoethmoidal complex and the mandible (Chandini et al. 2022; Sarode et al. 2018). According to the

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World Health Organization, 85% of cases are diagnosed before 30 years (Chandini et al. 2022).

Histological examination is characterized by fragments of fibro-osseous lesion composed of numerous small immature calcified bone structures of sarcomatoid appearance, randomly arranged in a hypercellular stroma composed of fusiform and stellate cells with hyperchromatic nuclei (Tenório et al. 2022).

The treatment protocol involves a multidisciplinary study, so that the surgical management removes the biological evolution of the lesion, reducing the possibility of recurrence, which makes the partial or total resection of the tumor an effective surgical procedure (Chrcanovic and Gomez 2020).

Moreover, complementary therapies with scientific evidence have emerged in this context, such as Low Level Laser Therapy (LLLT), which has proven to be an effective therapeutic tool due to its ability to improve tissue repair, reducing inflammation and edema and promoting analgesia (Pacheco et al. 2022a, b). The use of LLLT in biological tissue associated with a photosensitive substance (methylene blue) is called antimicrobial photodynamic therapy (aPDT). The aPDT consists of administering a photosensitizing dye, followed by irradiation of the lesion with visible light (Yang et al. 2020), forming reactive species in the presence of oxygen, such as singlet oxygen and free radicals (Pacheco et al. 2022a, b; Kwiatkowski et al. 2018). These chemically reactive species are cytotoxic and destroy microorganisms such as viruses, fungi and bacteria; thus, the laser stimulates antioxidant activity and protects the cell against oxidative damage during the healing process (Silveira et al. 2009). LLLT act as biostimulators and exhibit anti-inflammatory effects. Its application seems to accelerate healing through the stimulation of natural biological processes and has shown good results in controlling postoperative pain and edema (da Silva et al. 2018). However, there are no case reports in the literature that portray the use of LLLT and aPDT in cancer patients for the treatment of JPOF.

The aim of this case report was to present the surgical removal of a juvenile ossifying fibroma in the interradicular region of the right maxilla, with the associated use of low-intensity laser and photodynamic therapy in an oncological patient, emphasizing the importance of early diagnosis and therapeutic planning for a good prognosis.

Case presentation

This manuscript was according to CAse REports (CARE) guidelines and was approved by the Research Ethics Committee of the Ribeirão Preto Cancer Hospital, Brazil in November 15, 2018 (Approval Number: 02018.11.15.0 00000000000000023).

A 63-year-old black female patient sought the Dentistry Service of the Hospital do Câncer de Ribeirão Preto, São Paulo, Brazil, reporting acute pain in the right maxillary region between dental element 13 and 14, for approximately 45 days. In the anamnesis, a positive medical history was observed for breast cancer with the use of Anastrozole and a year ago she underwent chemotherapy with the drugs Anthracycline, Cyclophosphamide and Taxanes. The progression of the lesion did not cause phlogistic signs of inflammation or submandibular lymphadenopathy. Clinically, the lesion was firm on palpation, with slightly violaceous mucosa, incipient edema and the patient presented pain on palpation. The tomographic study showed a unilocular and radiolucent lesion containing amorphous calcifications and cystic-necrotic areas in the inter-radicular space of teeth 13 and 14 (Fig. 1A). A sclerotic border and mild root resorption of the involved teeth delimited the lesion. Furthermore, it promoted its displacement, in addition to the thinning and discontinuity of the cortices (Fig. 1B).

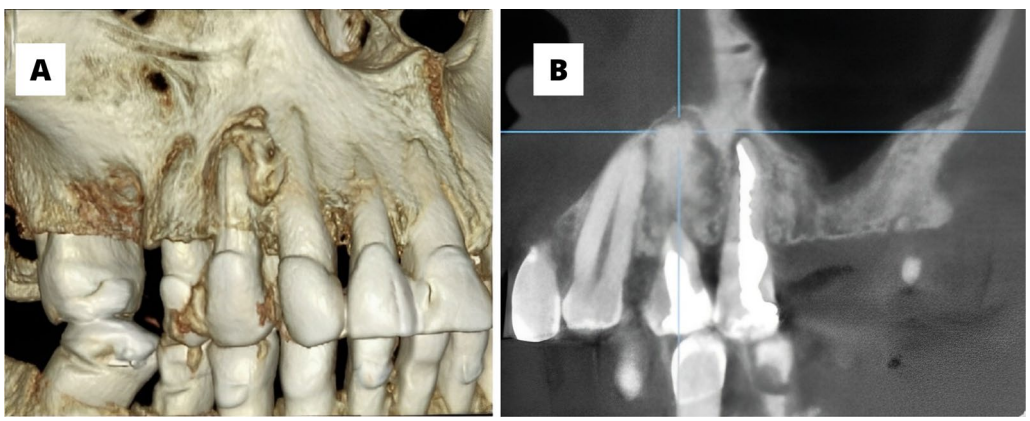

Fig. 1 Tomography. **A** Amorphous calcifications and cystic-necrotic areas in the interradicular space of teeth 13 and 14. **B** Areas of radiopaque and radiolucent displacement, with thinning and discontinuity of the cortices

An excisional biopsy of the lesion was performed (Fig. 2), followed by an anatomopathological-immunohistochemical examination, which confirmed the diagnosis of JPOF (Fig. 3). In the immediate postoperative period, antibiotic therapy was performed with Azithromycin 500 mg, 1 tablet per day for 4 days, Nimesulide 100 mg, 1 tablet 12/12 h for 2 days and also analgesia with Sodium Dipyrone 1 g, 1 tablet 12/12 h for 2 days. After ten days, the suture was removed. aPDT was started with chimiolux, methylene blue gel (0.01%) applied at the site between elements 13/14 (Fig. 4A), and after waiting 5 min (pre-irradiation time), the patient underwent to the application of the red wavelength laser, using the following parameters: EC therapy device, DMC equipment (Fig. 4B), red wavelength—660 nm; Power of 100 mW—Energy of 6 Joules, 1 point in the interradicular region, just one session of 60 s (Fig. 4B). And for three months, only LLLT (without methylene blue) was performed in the intra-oral area over the surgical site and extra-oral over the right nasolabial fold (Fig. 4C) to reduce edema, bone regeneration and analgesia, using the same device laser mentioned above, in the red/infrared wavelength, 3 Joules, 1 time per month lasting until the present day (48 months after surgery). The LLLT parameters are measured in wavelength (nm), energy (joules) and fluence (J/cm2) according to the parameters stipulated by the manufacturer (Table 1). Patient follow-up has been carried out for 48 months, with clinical and radiographic examination without recurrence (Fig. 5).

Discussion

Fibro-osseous lesions are considered a group of lesions that affect the maxillary and craniofacial bones and are characterized by the replacement of normal bone tissue

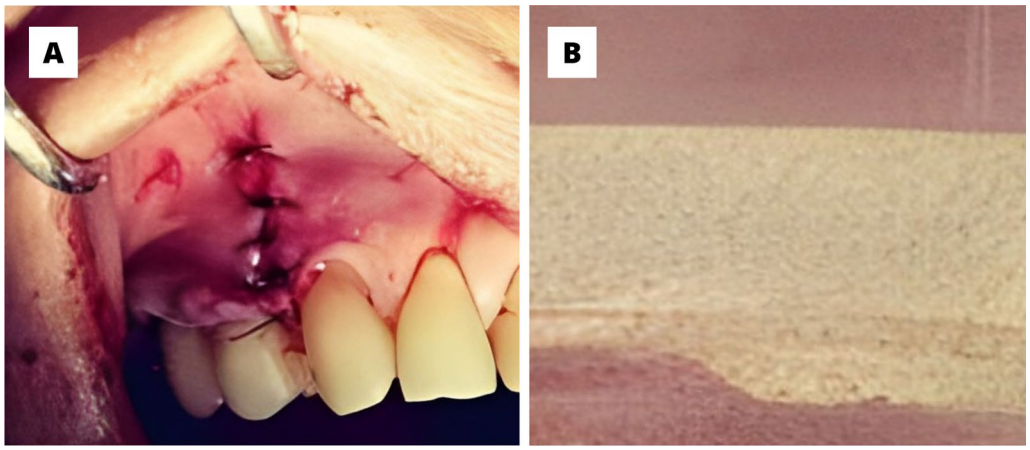

Fig. 2 Post-surgery. **A** Surgical site suturing and **B** Excisional biopsy fragment

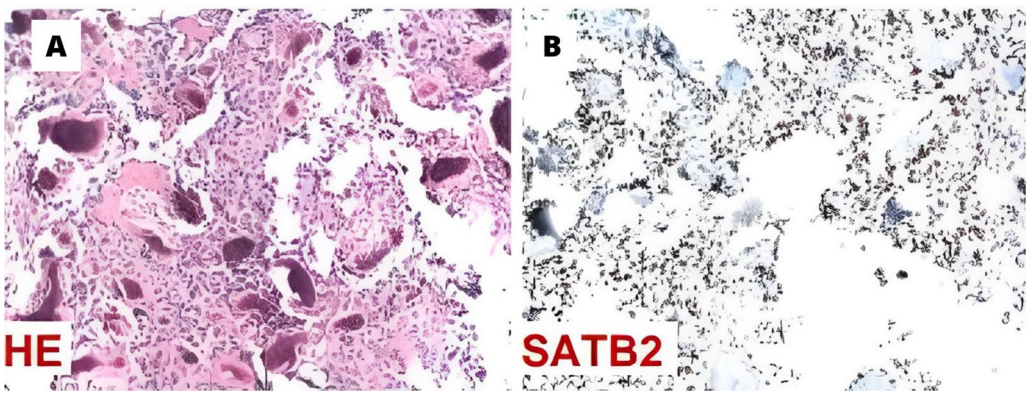

Fig. 3 Immunohistopathological examination. **A** Fragments of fibro-osseous lesions with calcified immature bone structures, with a "psamomatoid" appearance, randomly arranged in a hypercellular stroma composed of fusiform and stellate cells with hyperchromatic nuclei. **B** SATB2 positivity characteristic of hypercellular fibro-osseous lesion, favoring the diagnosis of JOPF

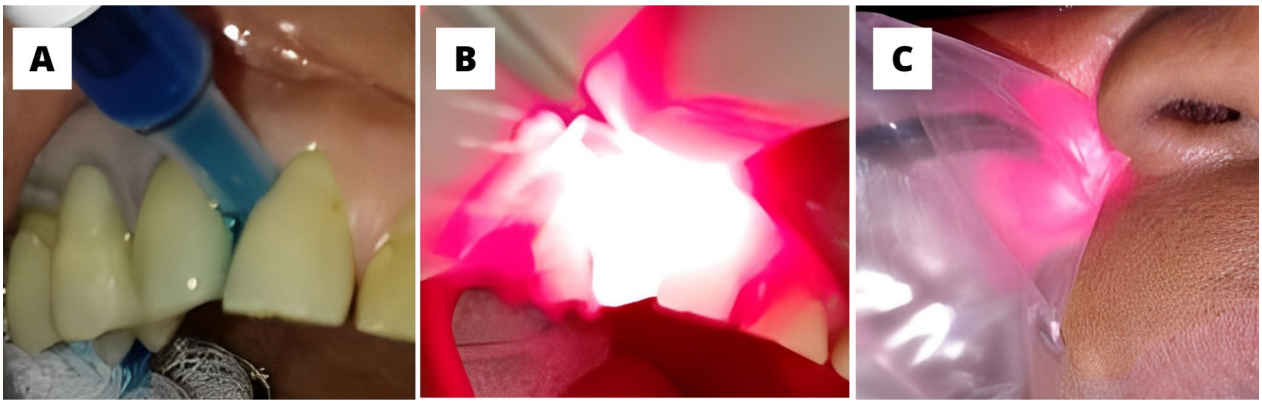

Fig. 4 Laser therapy. **A** aPDT technique with application of methylene blue on the surgical site and **B** application of laser with red wavelength—660 nm; 100 mW of power, intraoral and **C** extraoral in the right nasolabial region, with LLLT therapy, without methylene blue

Table 1 Irradiation parameters

Irradiation parameters	Unit of measurement	Description
Wavelength	nm	It is the laser emission characteristic defined by different colours of the visible (400–700 nm) and invisible spectrum
Fluence	J/cm^2	Fluency or dose is a description of the energy flow divided by the area of the laser emitter tip
Energy	Joule	Energy is represented by the power of the equipment multiplied by the treatment time

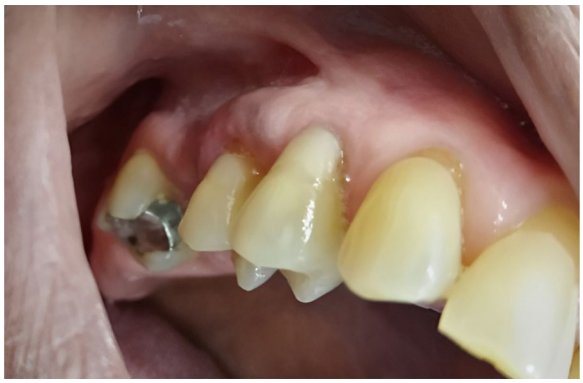

Fig. 5 Intraoral image after 48 months. The patient's follow-up has been carried out for 48 months, without signs of recurrence and within normal limits

by cellular fibrous tissue, containing various forms of ossification (Chrcanovic and Gomez 2020). Its manifestation occurs in more than 70% of the head and neck region (Macdonald-Jankowski 2004). Juvenile ossifying fibroma originates from the differentiation of precursor cells, demonstrating non-random chromosomal breakpoints in patients affected by the sarcomatoid variant (Nikitakis et al. 2022).

This subtype usually affects children and young adults (Rai et al. 2012), unlike the case presented where the patient was elderly aged 63 years. In the literature, a few cases report affecting older ages (Diniz et al. 2020; Chandini et al. 2022).

The patient presented pain on palpation. For some authors, painful symptoms may be the result of expansive growth (Barth et al. 2004; Han et al. 2016; Diniz et al. 2020). Due to its rapid bone progression, JPOF can promote undesirable functional and aesthetic effects, compromising adjacent structures, trismus, facial asymmetry and malocclusion (Chrcanovic and Gomez 2020; Leimola-Virtanen 2001), as occurred with the patient.

In this case report, there was an immediate intervention after symptoms were reported by the patient, which allowed all interventional measures to be planned and executed quickly. The recurrence of JPOF is mainly due to incomplete removal of the tumor, which can reach up to 90% of cases and usually appears after 06 months and can reach up to 19 years, in some cases (Goulart-Filho, et al. 2018).

Thus, as it is a neoplasm with a high rate of recurrence, the follow-up of this patient has been carried out once a month for 48 months, without clinical and radiographic alterations. In addition to carrying out an appropriate approach, with the aim of preserving vital structures, prolonged proservation is importance to avoid possible recurrences (Ollfa, et al. 2017).

To the best of our knowledge, this was the first reported case of an oncology patient with JPOF where laser was used as an auxiliary therapy after surgery. Although rare, metastasis to the oral cavity can occur from distant tumors such as breast cancer (D'Silva et al. 2006; Vetri et al. 2022), emphasizing the importance of early diagnosis and intervention.

Photodynamic therapy was used in the tumor region with the photosensitizer methylene blue, which has high affinity with the cell mitochondrial membrane, with phototoxic activation. aPDT is able to generate cytotoxic effects in the generation of reactive oxygen species that consequently increase the expression of inflammatory mediators promoting inflammation and ultimately inducing cell death by necrosis (Kwiatkowski et al. 2018). A fact that we can verify the possibility of having a tumoricidal immune response due to the presence of neutrophils, which causes long-term immunological protection in the treatment of cancer (Petrellis 2019). Also, the anti-inflammatory effect of the LLLT is based on reducing the concentration of prostaglandin, altering the arachidonic acid pathway and reducing the effect of tumor necrosis factor and with promising results in pain control (Park et al. 2022; Aimbire et al. 2006; Romão et al. 2015). Antunes et al. (2017) and Bensadoun et al. (2002) suggest that the use of LLLT may decrease the recurrence of lesions in cancer patients.

A case report of a patient with JPOF was presented; however, more studies should be carried out that include a larger sample and with paired and randomized controls. The importance of this manuscript is due to the stabilization of the condition in a cancer patient through a well-executed surgery and the periodic use of LLLT, which maintains action in cell maturation, increased metabolism and cell proliferation, showing acceleration of the healing process bone formation, without altering the physiological cellular pattern (Romão et al. 2015).

From the patient's perspective, the injury happened quickly and unexpectedly, impaired chewing and even brushing on the left side, which caused great discomfort and interfered with the patient's daily routine. However, after the treatment, there was no more pain and discomfort and there was the resumption of masticatory functions and regular hygiene.

Conclusions

Juvenile psammomatoid ossifying fibroma is a lesion with a high rate of recurrence and aggressive growth. Early detection, clinical, radiological and histopathological characteristics, adequate treatment and long-term follow-up are essential for good planning. The insertion of initial photodynamic therapy and long-term follow-up with low-intensity laser brought a favorable prognosis in the last 48 months. The patient will continue to be followed up at this hospital unit indefinitely due to the high rates of recurrence in the world literature.

Abbreviations

JPOF	Juvenile psammomatoid ossifying fibroma
LLLT	Low level laser therapy
aPDT	Antimicrobial photodynamic therapy
CARE	Case reports

Acknowledgements

The authors are grateful for the contributions of the Ribeirão Preto Cancer Hospital teams and the support of University of São Paulo.

Author contributions

JAP reviewed the literature and authored the case and surgical history. JAP, BT and HP contribute to laser treatment. KFM and JAP contributed to the final report. EVV supervised the project. All corresponding authors have read and approved the final version of this manuscript.

Funding

Not applicable.

Availability of data and materials

Not appropriate.

Declarations

Ethics approval and consent to participate

This study was approved by the Research Ethics Committee of the Ribeirão Preto Cancer Hospital, Brazil.

Consent for publication

Written consent from patient obtained.

Competing interests

The authors declare there are no competing interests.

Received: 3 January 2023 Accepted: 2 March 2023
Published online: 13 March 2023

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