

# Squamos Odontogenic Tumor: A case report

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## Abstract

**A 40-year-old male patient presented with diffuse radiolucent lesions on the upper maxilla at the apex of the incisors, canines, premolars, and molars. Surgical excision of the lesions was performed, and the biopsy specimens were submitted for histological analysis: the lesions of the first quadrant were found to be a squamous odontogenic tumor, a rare benign neoplasm of odontogenic origin. The purpose of this article is to describe the clinical, radiographic and histological aspects of odontogenic squamous tumor.**

**Keywords:** Squamos Odontogenic Tumor, Radiolucent lesions, Maxilla.

## Introduction

Squamous odontogenic tumor (SOT) is a rare benign neoplasm that originates from the odontogenic epithelium. It was first described by Pullon in 1975 and was recognized by World Health Organization (WHO) in 2005 and, more recently, in 2017 (1,2)

The recent WHO classification of odontogenic and maxillofacial bone tumors defines SOT as "a benign epithelial odontogenic tumor in which the tumor cells show terminal squamous differentiation." SOT can manifest in different age ranges; however, it is mainly diagnosed in the fourth decade of life. The sites mainly involved are the anterior part of the upper jaw and the posterior part of the mandible (3).

The process of tissue formation and differentiation in squamous odontogenic tumor is multifactorial; Malassez remnants may underlie epithelial proliferation in lesions associated with the alveolar process while the dental lamina may be the cause of the occurrence of lesions associated with unerupted or impacted teeth (4,5).

Although SOT is more frequently solitary intraosseous lesion (central SOT), peripheral SOT and SOT-like proliferation arising in odontogenic cyst variants have also been described.

Peripheral SOT is rare and affects soft tissues producing 'saucerization' of underlying bone. (6)

Histologically they are typically described as mural outgrowths inside the walls of odontogenic cysts. In fact, SOT from a microscopic point of view shows large islands of benign squamous epithelium separated by a stroma of collagenous material with an outer layer of squamous cells. Some cases show calcifications both within the stroma and within the squamous islands. These islands may undergo cystic degeneration over time. (7, 8, 9)

The usual treatment is enucleation of the lesion with appropriate curettage of the affected site. Only one case of malignant transformation of intraosseous SOT at the mandibular level has been described in the literature, and cases of recurrence are also quite rare.

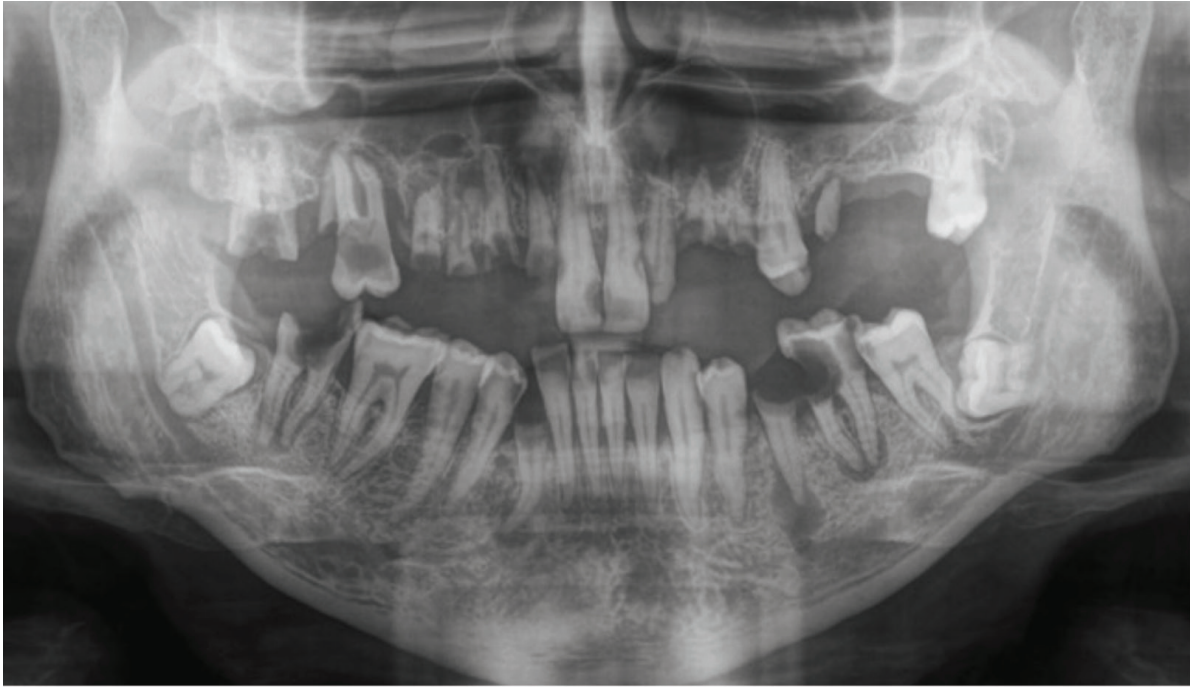
Clinical signs typically reported are asymptomatic swelling associated with tooth mobility and bone resorption. Diagnosis is made primarily during routine radiographic examinations in which predominantly unilocular radiolucency is evident although cases of SOT with a multilocular appearance have been described in the literature. Radiographically appearing with a triangular shape between the roots of the teeth, the treatment of election is surgical excision with curettage of the surgical site.

The aim of this case report is to describe the diagnostic course and surgical and squamous cell tumor treatment in a patient treated at our department.

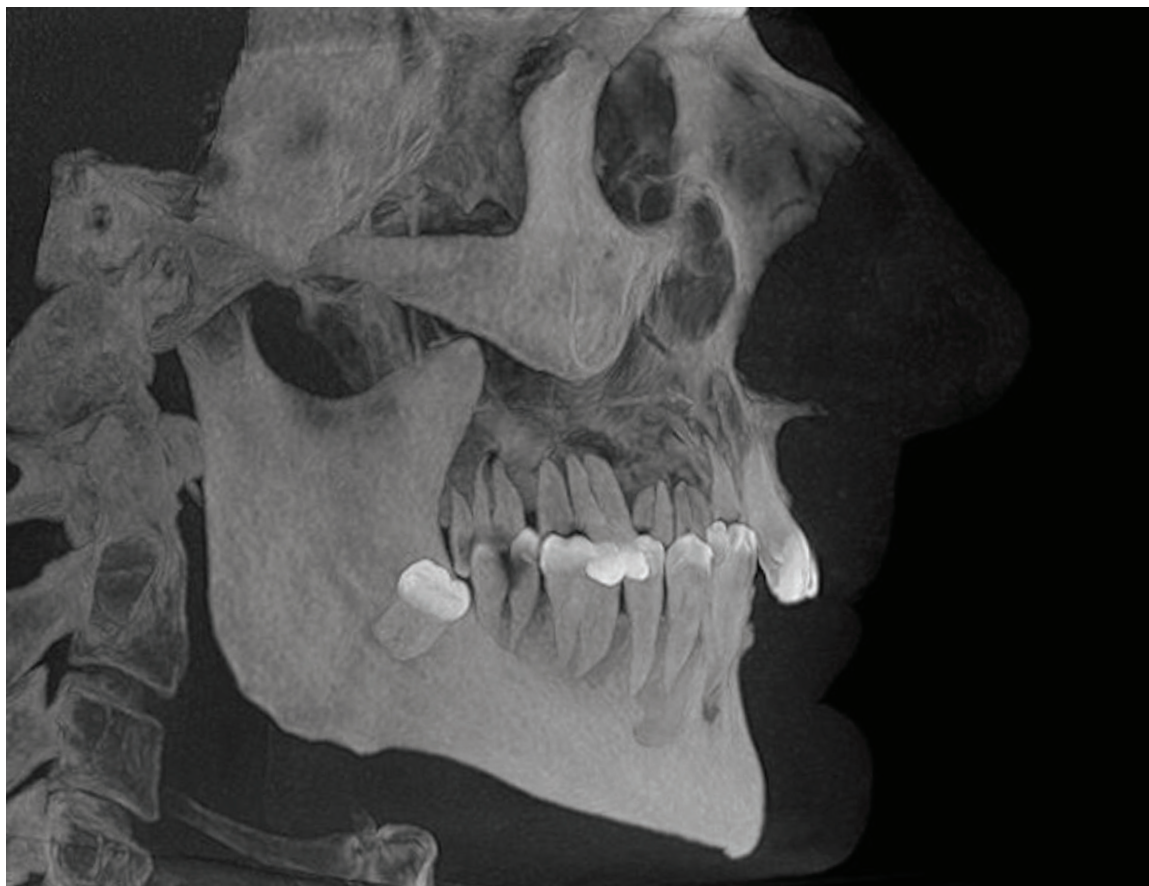
## Case report

A 40-year-old male patient came on clinical examination with multiple cavities and root remnants of lateral incisors, canine, premolar and second molar in the first quadrant; canine, first premolar, first and second molar in the second one.

Palpation of the affected area did not result in discharge of purulent material, and the area appeared slightly compressible. Radiographic examinations (Orthopantomogram and Cone Beam) showed well-circumscribed radiotranslucent lesions of spherical shape nearby all root remnants. In the premolar region of the first quadrant such lesions appeared overlapped. (Fig. 1-2; 3-4)



**Figure 1.** Ortopantomography.



**Figure 2.** Lateral Telecranium.

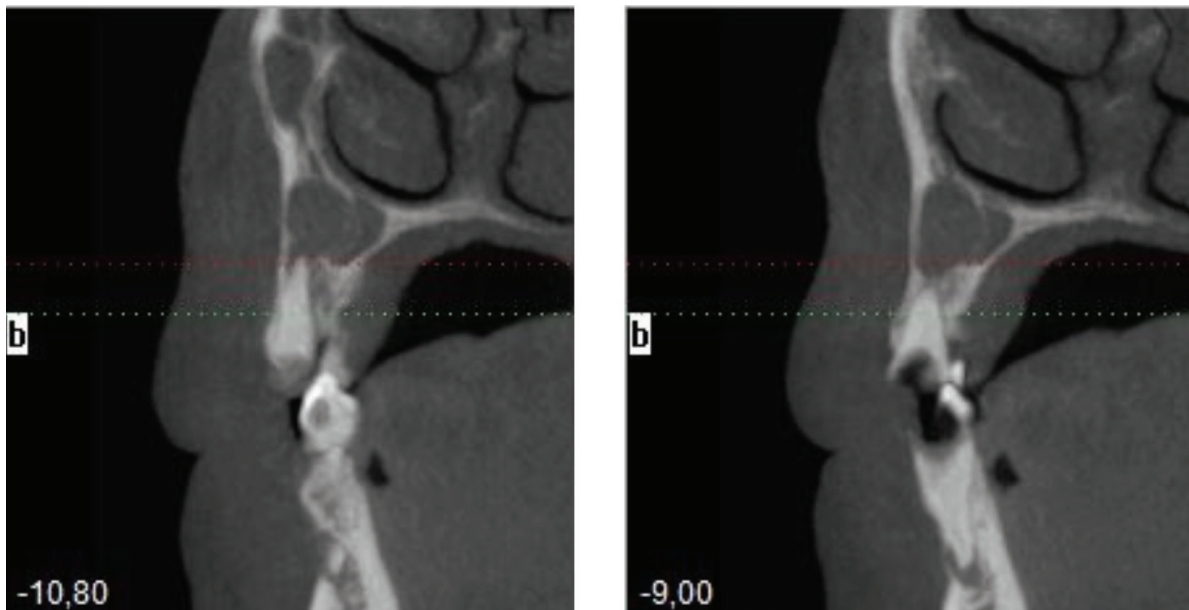


Figure 3. Element 1.2 CBCT frames

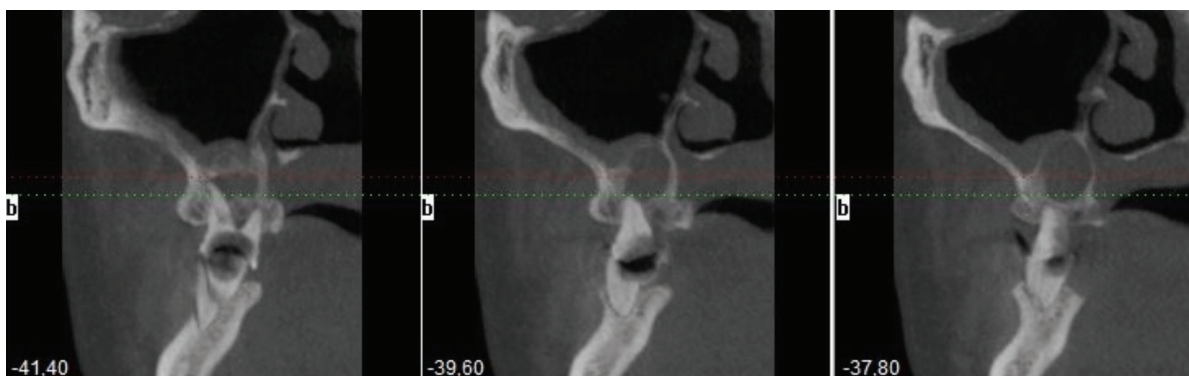


Figure 4. Element 1.6 CBCT frames.

## Treatment

Treatment included extraction of root remnants and compromised dental elements, removal of lesions for histologic analysis, and adequate curettage of the post-extraction cavity and washings with physiological saline. The patient was treated under general anesthesia and since the lesion corresponding to elements 22 23 24 was the most extensive the surgery started from the second quadrant: after plexus anesthesia with adrenaline 1:100.000, a full thickness crestal flap with distal discharge was made from the second molar area up to the central incisor. A mucoperiosteal flap was detached until a good view of the areas affected by the cystic lesions was obtained.

An osteotomy was then performed until the right cleavage was obtained to mobilize the cystic epithelium until it was completely “shelled out”. The lesion excised from the surrounding bone consisted of fibrous and compressible tissue with a citrine yellow liquid content. Curettage of the cavity and abundant washing was then carried out.

Moreover, severely compromised elements 22 and 24 and root residues 23 24 and 26 were extracted.

In order to get primary intention healing, all the sharp corners of the crest were smoothed with a diamond bur and silk 4/0 sutures were used.

Then, the same crestal full thickness-flap was subsequently made in the first quadrant from the incisal area to the last molar. The dental elements and root residues were all extracted except for 11.

In this area there were multiple lesions of a cystic nature, the major one involving the lateral incisor and they appeared very similar to that of the second quadrant that we had previously enucleated. Once all the lesions had been excised, an accurate curettage was performed with abundant physiological washes. The severely compromised buccal cortex was smoothed and made as regular as possible before the suture. It was not possible to completely close the flap in correspondence of the cavity at the level of the buccal molar area where a dehiscence remained. All the lesions were collected for histological analysis.

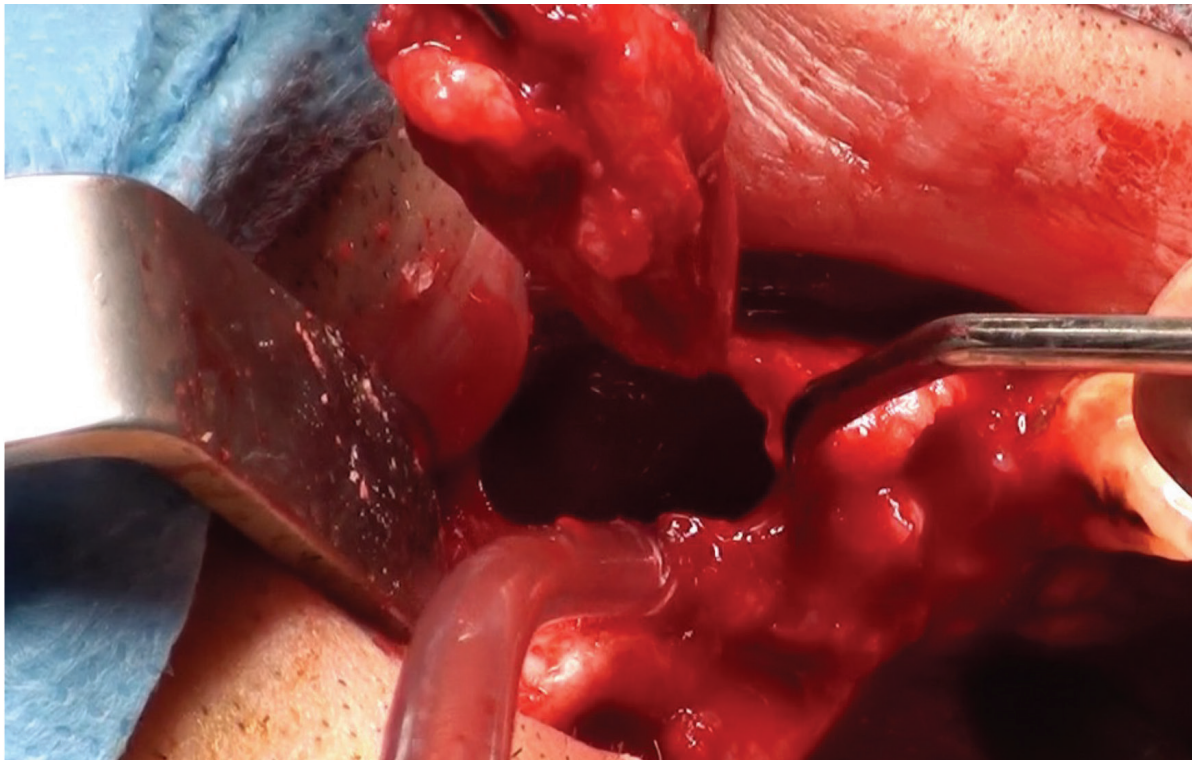
After the surgery, the patient was given zitromax (500 mg) for three days, ibuprofen (600 mg) for four days and mouth-rinses with clorexidine 0,12 % , twice a day, for a week. Sutures were removed after one week and a dehiscence was noticed on the first quadrant with bone exposure: a revision and curettage of the zone was performed and it was closed by primary intention.

One week later, the area completely healed and the next step will be the rehabilitation of the upper jaw with a removable prosthesis and a follow-up at 6 months.

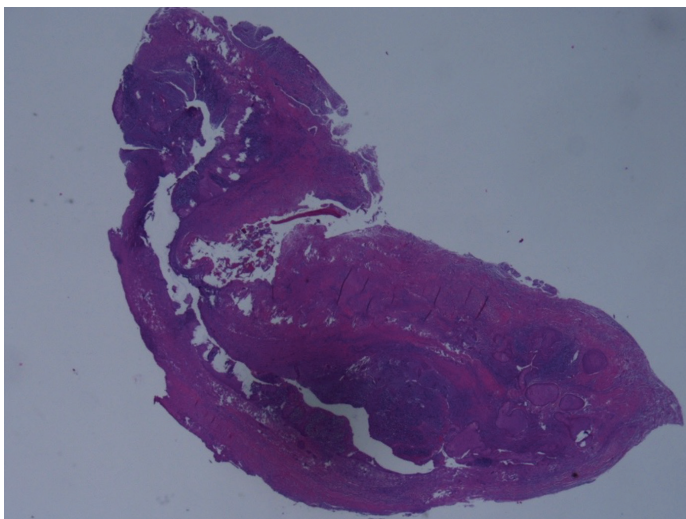
As regards the lower jaw, also multiple cavities, root remnants and radiolucent lesions are detectable: elements 48,47,43,35,36,38 will be extracted and all the related lesions will be enucleated and analyzed (Fig. 5).

## Diagnosis

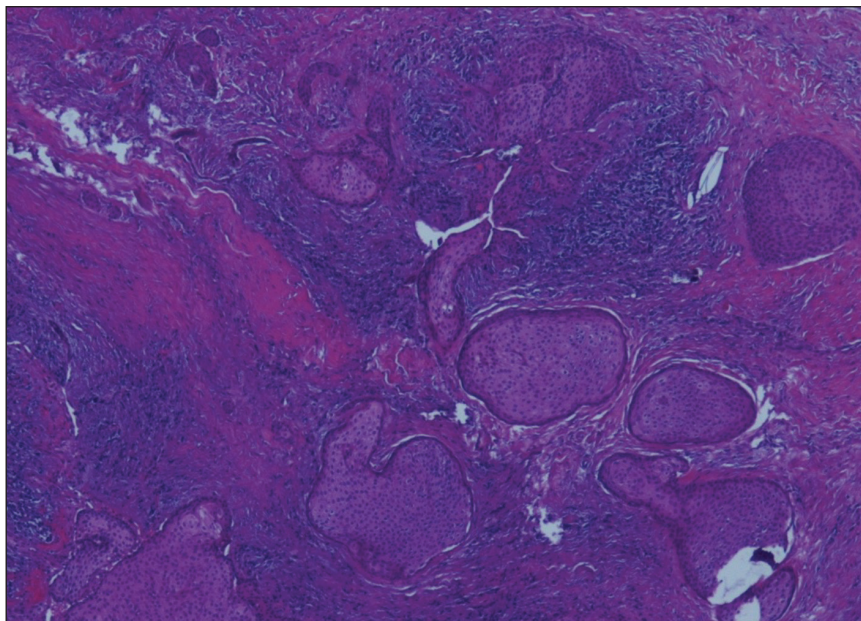
Histologic analysis of the lesion from the second quadrant revealed a cystic formation with multi-layered non-keratopoietic hyperplastic epithelial lining due to a chronic inflammatory process: it was compatible with radicular cyst. As regards the first quadrant, three samples were analysed and they revealed a multi-layered epithelial lining associated with surrounding nests of variably shaped and sized epithelial cells without atypia: the samples were compatible with the diagnosis of squamous odontogenic tumor (SOT). (Fig. 6-7)



**Figure 5.** Enucleation of the lesion.



**Figure 6.** SOT 1x section stained with Hematoxylin-Eosin.



**Figure 7.** SOT 10x section stained with Hematoxylin-Eosin.

## Discussion

Squamous Odontogenic Tumor (SOT) is a benign lesion with slow proliferation rate that is thought to origin from dental lamina remnants. However, it is locally infiltrative and it may cause tooth displacement or un-eruption and root divergency, irregular swelling and bony expansion due to the perforation of the cortical bone; other cases of mild discomfort, soreness or a feeling of pressure when the nerves are involved have been reported; sometimes, it is totally asymptomatic and discovered with routine exams. In our case the patient did not have any kind of symptom related to the growth of the lesions and was referred to the oral surgery department to treat the multiple cavities and dental remnants. As reported in literature, the most common location of the SOT is the premolar-canine in maxilla or the molar region of the mandible. Our patient showed multiple radiolucent lesions in all quadrants and we attempted to analyze the ones affecting the first and the second ones. (10)

The histological analysis revealed the presence of both SOT and a radicular cyst, which come from proliferation of Malassez remnants after inflammatory stimulus. Also SOT is frequently associated with necrotic or unerupted teeth which may have cause the proliferation of dental lamina, thus suggesting a mild correlation between the tumor and the cyst.

A review by Chrnovic and Gomez (2018) analyzed cases of proliferation of SOT and squamous odontogenic tumor-like proliferation in odontogenic cysts (SOT-LPOC) thus concluding that they are different clinicopathological conditions since SOT showed a more aggressive growth pattern: the former had unilocular or multilocular aspect and was associated with cortical bone perforation in 62 % of central lesions and tooth displacement while the latter showed no signs of bone erosion or root resorption and were most associated with radicular and dentigerous cysts, as it appeared most of the times as an unilocular lesion. (11)

Also Barbeiro et al. (2021) described a case of maxillary dentigerous cyst involving tooth 18 which showed squamous odontogenic tumor like proliferation: their group managed to treat it by enucleation of the lesion and involved tooth and obtained complete healing and no recurrence after 3 year follow-up. Considering all clinical differences between the two kind of lesions it is not possible to assert that SOT is the natural progression of SOT-LPOC. (12)

The election treatment modality is conservative enucleation with curettage; other cases reported in literature received a more aggressive treatment consisting on en bloc resection, hemimaxillectomy or radical alveolectomy. Moreover, it is suggested to treat multilocular, multifocal and/or recurrent lesions involving maxilla, which is known to be more porous than mandible, with an extensive intervention. (13)

Cases of recurrence of SOT are reported in literature especially when enucleation was performed but were not influenced by the age of the patient, location of the lesion, expansion and perforation of cortical bone and locularity in radiological exams. Since there are no guidelines about the gold standard of treatment of SOT, it is suggested that the surgeon would evaluate the suitable surgical treatment for the case.

In our case, the enucleation was completed with accurate curettage and physiological washes due to the wide extension of the lesions: we preferred not to extend the resection beyond the cavities and preserve the residual bone crest to a future rehabilitation.

## Conclusion

Our study is an additional case of intraosseous multifocal SOT, which has been described in literature as a rare but locally aggressive odontogenic tumor with low recurrence rate: our approach was conservative since the patient had multiple lesions and we attempted to preserve the bone volume. The limitation of the study

is the short follow-up, which will be updated, and the absence of a surgical protocol treatment, since in literature there are still not data and guidelines to treat SOT with predictable results.

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