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Squamous Odontogenic Tumour – A Paradoxical Pathology

Sam John Koshy¹, Dr Senthilnathan² and Madhulaxmi M³

¹Department of Oral & Maxillofacial Surgery Saveetha Dental College and Hospitals, Saveetha Institute of Medical And Technical Science, Saveetha University, Chennai- 600077, India ²Department of Oral & Maxillofacial Surgery Saveetha Dental College and Hospitals, Saveetha Institute of Medical And Technical Science, Saveetha University, Chennai- 600077, India ³Professor and Head Of Department, of Oral & Maxillofacial Surgery Saveetha Dental College and Hospitals, Saveetha Institute of Medical And Technical Science, Saveetha University, Chennai- 600077, India

ABSTRACT

Squamous odontogenic tumors (SOTs) are neoplasms being benign, local in origin and infiltrative in capacity that localize to the periodontium. In total, only less than 50 cases have been reported since the first description of SOTs in 1975. The most common site of occurrence of the lesion in the maxilla, incisor area and in the mandible, the bicuspid-molar region. Squamous odontogenic tumors show a characteristic triangular-shaped, unilocular radiolucency of the alveolar bone, with the wide base of the radiolucency localized between the diverging apices of the adjacent roots, radiographically. Here, we report an unusual presentation of the tumour in the periapical region of the mandibular incisor region mimicking radicular cyst clinically but with a major histological variation.

KEY WORDS: SQUAMOUS ODONTOGENIC TUMOR, ORAL AND MAXILLOFACIAL TUMOR, ODONTOGENIC TUMORS.

INTRODUCTION

Paradoxical refers to something that seems selfcontradictory. It often depicts an identity so impossible or difficult to understand because of two opposite facts or characteristics it contains. (PARADOXICAL | meaning in the Cambridge English Dictionary, no date) This case report is in fact of a paradoxical pathology because of how clinically contradictory it was when compared to all the literature present to refer to. Its contradiction has made this stand a notch different from all other cases judging by its clinical significance and presentation.

ARTICLE INFORMATION

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NAAS Journal Score 2020 (4.31) SJIF: 2020 (7.728) A Society of Science and Nature Publication, Bhopal India 2020. All rights reserved. Online Contents Available at: http://www.bbrc.in/ Doi: http://dx.doi.org/10.21786/bbrc/13.8/128 World Health Organization (WHO) tumor classification published in 1971(Philipsen and Reichart, 2006) has shed an understanding on the presence of a lesion called Squamous Odontogenic Tumor. A novel and completely revised WHO classification from 2005, encompassed the histopathological and genetic criteria of SOTs, described this pathology as a group of epithelial odontogenic tumors comprising within the ameloblastoma family, consisting of solid/multicystic, extraosseous/peripheral, desmoplastic and unicystic ameloblastoma, and squamous, adenomatoid, calcifying and keratocystic odontogenic tumors(World Health Organization and International Agency for Research on Cancer, 2005).

Squamous odontogenic tumors (SOTs) are rare benign tumors of the periodontium that possess an unknown etiology and were first described in 1975 by Pullon et al (Pullon et al., 1975; Perdigão et al., 2004). In the literature, a little over fifty cases have been reported worldwide (Reichart and Philipsen, 1990; Saxby, Rippin and Sheron,



1993). Squamous odontogenic tumors are slow-growing, locally infiltrating tumors with only a few clinical signs and symptoms which vary from patient to patient. Mobility of the teeth, increased periodontal pocket depth, sensitivity, swelling of the alveolar process, swelling and erythema of adjacent gingiva and moderate pain are the most widely accepted indicators for the underlying tumor. (Reichart and Philipsen, 1990; Saxby, Rippin and Sheron, 1993; Philipsen and Reichart, 1996). SOTs have also been reported to occur in various age groups, yet mainly affect adults in the third decade of life (Kim, Mintz and Stevens, 2007). Literature has shown that the male to female gender predilection is 1.4:1 (Reichart and Philipsen, 1990; Ruhin et al., 2007). The jaw most commonly affected is the mandible more often than the maxilla, with an increased preference of occurrence in the posterior premolar and molar area. Maxillary SOTs are described to be primarily present in the anterior area, and appear to be more aggressive in nature when compared with SOTs in the mandibular area(Reichart and Philipsen, 1990)(Reichart and Philipsen, 1990; Ruhin et al., 2007)(Reichart and Philipsen, 1990).

Squamous odontogenic tumors derive from the epithelial cell rests of Malassez where tumor appear on the lateral root surface. The typical radiographic presentation is a triangular radiolucent defect involving the lateral root surface of erupted and vital teeth(Cillo, Ellis and Kessler, 2005). The wide base of the radiolucency is localized between the diverging apices of the adjacent roots (Haghighat, Kalmar and Mariotti, 2002). The prognosis of squamous odontogenic tumor therapy is good if surgical enucleation of the whole lesion is done with curettage of the affected area with extraction of the affected teeth, if present. Recurrence appears to be rare, and may occur due to incomplete tumor removal.

With a rich case bank established over 3 decades we have been able to publish extensively in our domain (Abdul Wahab et al., 2017; Eapen, Baig and Avinash, 2017; Patil et al., 2017; Jain and Nazar, 2018; J et al., 2018; Marimuthu et al., 2018; Wahab et al., 2018; Abhinav et al., 2019; Ramadorai, Ravi and Narayanan, 2019; Senthil Kumar et al., 2019; Sweta, Abhinav and Ramesh, 2019). Here, we report an unusual presentation of the tumour in the periapical region of the mandibular incisor region mimicking radicular cyst clinically but with a major histological variation.

Case Report: A 26 year old male patient reported to the Department of oral and maxillofacial surgery complaining of pain and swelling on the right side of his face for a duration of 3 years. Pain was intermittent and slowly increased in intensity over 3 years as the patient was negligent to the initial signs and symptoms. Swelling subsequently increased with accompanying pain and discomfort while mastication and speech. Patient had a history of bruxism. There were no other relevant medical, surgical, family or personal history. On clinical examination, a definitive swelling of the face on the right side was evident with a size of 3 X 3 centimeters extending from the right corner of the mouth, 3 centimeters medial to the angle along the lower border of the mandible. Skin discolorations were evident on the right side The skin over the swelling appeared shiny with a firm consistency, with absence of tenderness on the affected area. The borders were well defined with absence of warmth over the swelling, ulcerations and pulsations.

On intraoral examination, the lower anterior teeth of the left side (31) showed discolorations suggestive of non vitality. The remaining anterior teeth were tender on vertical percussion. A swelling of the right side vestibule extending from the region of the right mandibular canine to the left lateral incisors were observed with absence of pain on palpation of the vestibule. [Figure 1] On radiographic analysis, orthopantomography of the patient revealed well defined, circumscribed, definitive bordered periapical radiolucency, diffused triangular in shape extending from the peri apical region of the right mandibular canine measuring approximately around 3 X 5 millimeter in size periapically of each tooth. [Figure 2]

Figure 1: Intra oral image of the left, center and right side of the patient while in occlusion revealing discoloration of the lower anterior teeth of the left side (31) and swelling of the right side vestibule extending from the region of the right mandibular canine to the left lateral incisors.



Figure 2: Pre-operative orthopantamography revealing well defined, circumscribed, definitive bordered periapical radiolucency, diffused triangular in shape extending from the peri apical region of the right mandibular canines to the left mandibular canine measuring approximately around 3 X 5 millimeter in size periapically of each tooth



Patient history, clinical examination and radiological investigation suggested a provisional differential diagnosis of a benign pathology - radicular cyst in relation to the non vital teeth or a benign tumour. As patient was reluctant for local anaethesia procedure, treatment plan was made for an excisional biopsy under General anesthesia. Surgical procedure followed

a crevicular incision followed by releasing incisions extending from the region of the right mandibular canine to the left mandibular canine and the lesion was exposed. Enucleation of the lesion and post- enucleation prophylactic saucerization of the bone adjacent to the lesion was done. Simultaneous apicectomy with retrograde mineral trioxide aggregate filling and osseous reconstruction using Concentrated Growth Factors and xenograft Bone grafts (Geistlich Bio-Oss) was done as the lesion resembled a cyst on surgical exloration. [Figure 3].

Figure 3: Exposed pathology intra operatively.



Figure 4: (a) Enucleation of the lesion and post- enucleation prophylactic saucerized bone. (b) Concentrated growth factors along with the osseous reconstruction xenograft(Bio Oss) placed for bone regeneration.



Figure 5: Enucleated pathological specimen



Concentrated growth factors were extracted from the patients blood along with the osseous reconstruction xenograft(Bio Oss) acting as both a membrane and a growth potentiating component to enhance the development of neovascularization and osseous reformation. [Figure 4]. The specimen on histopathological analysis showed odontogenic epithelial lining with non keratinized stratified squamous epithelium of variable sizes. The connective tissue wall was dense and fibrous composed of several foci of intense chronic inflammatory cells. It also showed proliferating odontogenic epithelial rest cells with squamous metaplastic changes along with vacuolization of cells. Moderate vascularization and evidence of hemorrhage was also evident. Areas of peripheral resorbing bone were seen with evidence of an area of overlying stratified squamous epithelium. [Figure 5]

Figure 6: Histopathological microscopic examination of the pathological specimen.



The histopathological report stated it as suggestive of inflammatory odontogenic cyst with squamous odontogenic tumor-like projections. [Figure 6]. The patient underwent regular follow-ups every week for the first month post operatively and 6 months post operatively, with no clinical symptoms nor complaint about any pressure or sensitivity disturbances in the affected area. [Figure 7 & 8].



Figure 8: Image of One week follow up postoperatively.



DISCUSSION

Squamous odontogenic tumors are rare, benign, locally infiltrative neoplasms that many authors have elicited about. Many authors have indiscriminately and extensively studied and described cases of squamous odontogenic tumor in the past. (Pullon et al., 1975; Doyle et al., 1977; Hopper, Sadeghi and Pricco, 1980; McNeill, Price and Stoker, 1980; van der Waal, de Rijcke and van der Kwast, 1980; Carr, Carlton and Marks, 1981; Goldblatt, Brannon and Ellis, 1982; Cataldo, Less and Giunta, 1983; Norris et al., 1984; Kristensen, Andersen and Jacobsen, 1985; Monteil and Terestri, 1985; Warnock et al., 1985; Mills et al., 1986; Leider, Jonker and Cook, 1989; Baden et al., 1993; Kusama et al., 1998; Perdigão et al., 2004; Ruhin et al., 2007; Siar et al., 2010; Malathi et al., 2012; Bansal and Joshi, 2013; Virapara, Rajput and Shah, 2018)

On comparing with cases of SOT diagnosed world wide, the age at the time of initial diagnosis ranged between 8 and 67 years, with a mean age of 36.3 years. Patients aged 20–29 show an increased rise in the visibility of SOT's. The gender ratio of female to male predilection from as reported in the literature ranges from 1:1.2 to 1:1.4. The mandible was involved in 57.1% of all cases and the maxilla in only 38.8% with remaining having variations in the site of occurrence.

According to the present literature, the incidence of SOT is low with just over 50 cases reported worldwide. The tumor usually grows slowly and demonstrates a lack of symptoms for a long standing period of time. The radiographic and clinical characteristic features of SOT are neither unique nor sufficient for the final diagnosis and because of which, chances are that SOT may be confused with a number of other pathologies(Perdigão et al., 2004). Therefore, distinctive clinical, radiological and histological aspects are necessary for avoiding a misdiagnosis that may result in serious negative implications for the patient (Ide et al., 1999). There has been an increased predominance in the third decade for SOT, however studies have proved it to appear in any age. The youngest patient reported in the literature was a 9-year-old boy with maxillary SOT that was treated with local surgical tumorectomy.(Ide et al., 1999; Ruhin et al., 2007)(10). The maxilla appears to be involved more often in the region of the incisors, whereas the premolar and molar areas appear to be more involved in the mandible.

SOT presents as a locally infiltrative neoplasm and has been known to infiltrate into adjacent tissues, with resorption of the alveolar bone and invasion of the overlying gingival and oral mucosa. (Robson, 2001) The etiology of SOT is yet to be interpreted for correct and rightful diagnosis with timely effect. However, immunohistochemical evaluation performed in studies described in literature revealed positive reactivity of varying intensity in the neoplastic epithelial cells for the Notch1, Notch2 and Notch3 transmembrane receptors and their ligands. These findings suggest that

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these receptors play a role in the cytodifferentiation of squamous odontogenic tumors.(Siar et al., 2010).

Although squamous odontogenic tumors are considered to be benign neoplasms, the local expansiveness and clinical behaviour of the tumor indicates the possibility of carcinomatous transformation. In 1999, Ide et al (Ide et al., 1999) reported a rare occurrence of intraosseous squamous cell carcinoma arising in association with squamous odontogenic tumors. The enucleated specimen of the tumor revealed a characteristic pattern of SOT. However, within 2 months, aggressive bone destruction exhibiting the typical findings of intraosseous squamous carcinoma was described. Recent studies have revealed that an odontogenic tumor, which is difficult to access, is prone to the development of recurrence. This may be due to the inability of the surgical treatment to fully remove all tumor cells due to lack of accessibility or due to its normal like morphology..(Ide et al., 1999).

Treatment of SOT consists of the surgical enucleation of the whole lesion, curettage of the affected area with extraction of the affected teeth, if present. More extensive lesions with infiltrating neighboring structures require more radical interventions such as en bloc resection. (Jones et al., 2011; Badni, Nagaraja and Kamath, 2012; Tarsitano, Agosti and Marchetti, 2012; Bansal and Joshi, 2013): (Baden et al., 1993) CGF, Concentrated Growth Factors are described as a miracle in regenerative dentistry. CGF is a new regeneration platelet aggregate which is used widely in oral surgeries. It contains various growth factors which enhances its action and promotes wound healing and growth potentials. CGF is currently used best with autologous bone particles to help induce bone regeneration and connective tissue attachment. CGF is a fibrin tissue adhesive with properties of haemostasis and tissue sealant properties making it effective on placement. It accelerates osteogenesis and promotes wound healing. CGF improves the stability of the wound that is required for the attachment of a new connective tissue to the root surface. It promotes epithelial, endothelial and epidermal regeneration and decreases scarring.

It has antimicrobial properties due to high concentration of leukocytes. It acts as an anti-antigenic agent on chronic non healing wounds.(Y, 2018) Its most important factor is the induction of bone regeneration when added to xenografts like Bio Oss which potentiates and plays a vital role in the regeneration of lost or cauterized bone in the course of enucleation of a pathology. Many factors have contraindicated with literature in this described case making it indeed paradoxical pathology. The most common site of occurrence as per literature in mandible was the posterior region, (Perdigão et al., 2004) whereas in this case, SOT was present in the mandibular anterior region extending for the right mandibular canine to the left mandibular canine region. However in sync with the literature, the tooth was non vital and radiographically exhibited a triangular radiolucency of the peri apical region of the involved teeth.

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