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SQUAMOUS ODONTOGENIC TUMOR OF ANTERIOR MAXILLA: A RARE CASE REPORT

[1]*Dr. Arpita Kabiraj, MDS, [2]Dr. Tanya Khaitan, MDS, [3]Dr. Anil Singh, MDS, [4]Dr. Jagriti Gupta, MDS

[1]Senior Lecturer, Department of Oral Pathology & Microbiology, Index Institute of Dental Sciences, Indore, Madhya Pradesh, India - 452016.

[2]Tutor, Department of Oral Medicine and Radiology, Dental Institute, Rajendra Institute of Medical Sciences Ranchi – 834009, India.

[3]Prof. & HOD, Department of Oral Pathology & Microbiology, Saraswati Dental College, Lucknow, Uttar Pradesh, India - 227105.

[4]Reader, Department of Oral Pathology & Microbiology, Vyas Dental College & Hospital, Jodhpur, Rajasthan - 342001, India.

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***Corresponding Author**
Dr. Arpita Kabiraj
Senior Lecturer, Department of Oral Pathology & Microbiology, Index Institute of Dental Sciences, Indore, Madhya Pradesh, India - 452016.

ABSTRACT

Squamous odontogenic tumour (SOT) is a rare, benign but locally infiltrative epithelial odontogenic tumor arising from remnants cell rests of Serres, Malassez or gingival epithelium. It exhibits very little tendency for aggressive clinical behavior and practically no recurrences have been reported following curettage or conservative surgical removal. Clinically, SOTs presents as a slow growing, intrabony lesion with very few clinical signs and symptoms. Radiographically, they show a well-demarcated unilocular, triangular radiolucency between the roots of adjacent teeth with hypodense areas and hot spot in the focal lesional area in computed tomography and scintigraphy respectively. Histopathologically, they usually present as islands of benign squamous epithelium in mature connective tissue stroma without the evidence of peripheral columnar cells, palisading nuclei, or stellate reticulum and is characterized by squamous metaplasia of the epithelial cells. Here, we report a rare case of SOT on the left side of the maxilla in a 45 year old male with distinctive computerized tomography and radionuclide imaging features.

KEYWORDS: Metaplasia; Odontogenic; Radionuclide Imaging.

INTRODUCTION

Squamous odontogenic tumor (SOT) primarily described by Pullon et al in 1975 is thought to be an uncommon benign odontogenic neoplasm assumed to originate in the periodontium.[1] SOT is a benign but locally infiltrative epithelial tumor that arises from remnants of the dental lamina, or the Malassez or gingival epithelium.[2] To date, fewer than 50 cases have been reported in the literature. Forty-seven cases of SOT have so far been reported in the English literature (PubMed Database).The most common site of occurrence of the lesion in the mandible is the bicuspid‑molar region and in the maxilla, incisor‑cuspid area.[3] Here we report a rare case of SOT on the left side of the maxilla in a 45 year old male with distinctive computerized tomography and radionuclide imaging features.

CASE REPORT

A 45 year old male patient reported to the department with the complaint of swelling in the left side of face since 8 years. Initially, the swelling was apparently smaller in size which gradually increased to attain the present size. There was mild intermittent pain and discomfort associated with the swelling but no history of any blood/ purulent discharge and paresthesia in that region.

On extraoral examination, a diffuse swelling was seen in the left middle third of the face measuring approximately 3 x 4 cm in size, extending from the philtrum region to 2 cm parallel to cantho-meatal line obliterating the nasolabial fold. No secondary changes were associated with the swelling. On palpation, no local rise of temperature was noted; the swelling was non-tender, firm in consistency, non-compressible and non-reducible [Figure 1].

Intraorally, solitary well-defined swelling was present in the left maxillary region measuring approximately 4 x 5 cm in size, extending from midline to the mesial aspect of 25 mediolaterally and from rugae area to labial vestibular region anteroposteriorly obliterating the vestibule. Swelling was smooth surfaced, lobulated and same mucosal color with mild discrete erythematous and white keratinized areas due to trauma from opposing teeth. On palpation, it was non-tender, soft and fluctuant in the centre between two lobes and firm in consistency towards the periphery [Figure 2]. Based on the history and clinical features, a provisional diagnosis of odontogenic keratocyst and ameloblastoma was well thought of.

The patient was subjected to radiological investigations. Intraoral periapical radiograph and anterior maxillary occlusal radiograph revealed multilocular radiolucency extending from the midline to mesial aspect of 25 [Figure 3]. Lateral Cephalogram showed a large well-defined multilocular radiolucency surrounded by a thin corticated border in the left anterior maxillary region. Paranasal sinus view revealed a similar multilocular radiolucency involving left maxillary sinus obliterating maxillary sinus and left nasal fossa [Figure 4]. Computerized tomographic scan showed hypodense area in the left anterior maxilla [Figure 5]. Radionuclide imaging was also planned for the patient with Positron Emission Tomography (PET). With the patient fasting for 6 hrs, (18) F-fluorodeoxyglucose (FDG) was injected intravenously and PET-CT was performed. Physiological concentration was observed in the heart, gut, brain, kidneys, and bladder. Increased concentration of FDG was seen in the focal lesion in left anterior maxillary region (coronal, sagittal and axial aspect) measuring approximately 4x5 cm in size [Figure 6].

Considering a favourable prognosis, the entire lesion was excised under local anaesthesia and sent for histopathological examination. Microscopically, the haematoxylin and eosin stained section revealed a connective tissue stroma consisting of mature collagenous fibres. Individual epithelial islands comprising of peripheral layer of flat epithelial cells were also seen. Individual cell keratinisation depicting squamous metaplasia was also evident within the epithelial islands [Figure 7].

Therefore, based on the clinical, radiological and histopathological findings, a final diagnosis of Squamous odontogenic tumor was confirmed. The patient was kept under periodic recall after 15 days, 1, 3 and 6 months and no recurrence noted till date.

FIGURES

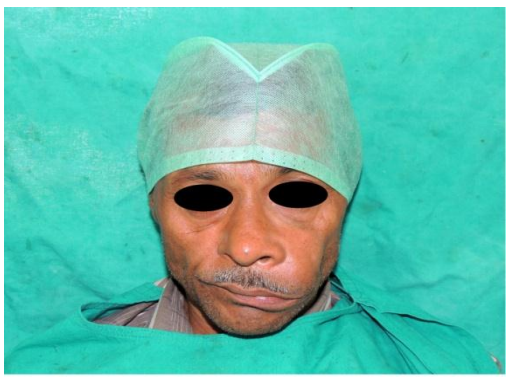

Fig. 1: Clinical photograph of the patient showing diffuse swelling in the left middle third of face.

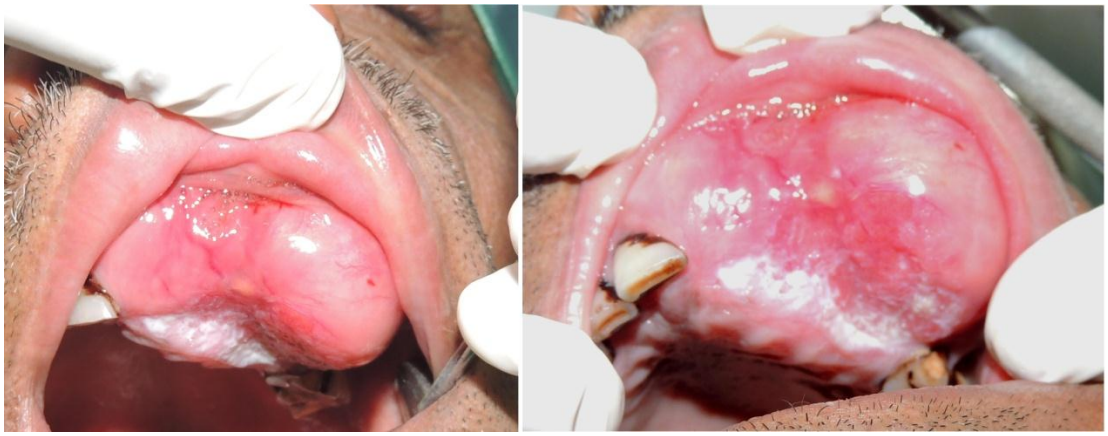

Fig. 2: Intraorally, solitary well-defined swelling seen in the left maxillary region extending from midline to the mesial aspect of 25 mediolaterally and from rugae area to labial vestibular region anteroposteriorly.

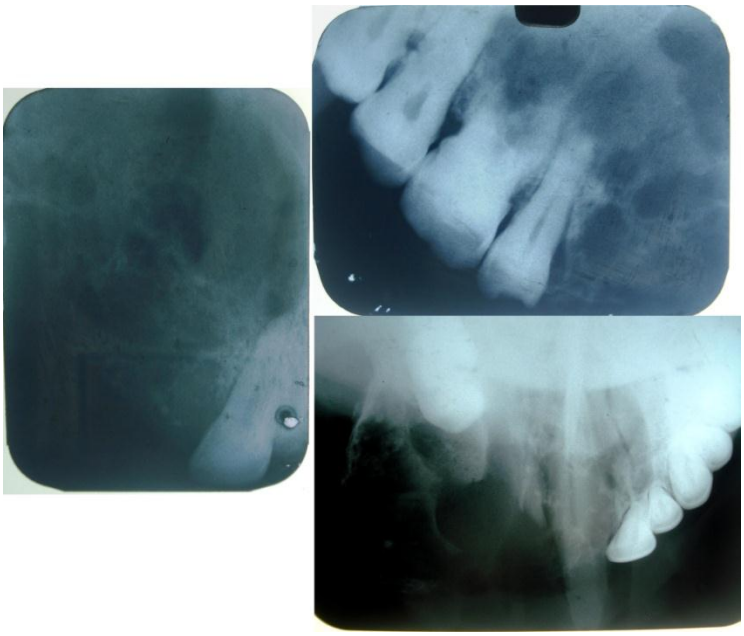

Fig. 3: Intraoral periapical radiograph and anterior maxillary occlusal radiograph showing multilocular radiolucency extending from the midline to mesial aspect of 25.

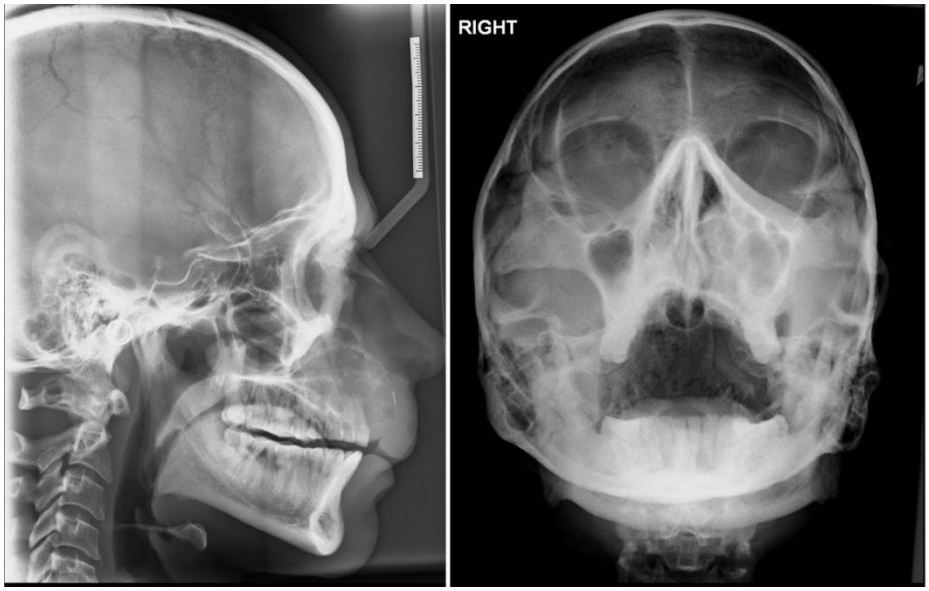

Fig. 4: Lateral Cephalogram and Paranasal sinus view showing a multilocular radiolucency involving left maxillary anterior region obliterating maxillary sinus and left nasal fossa.

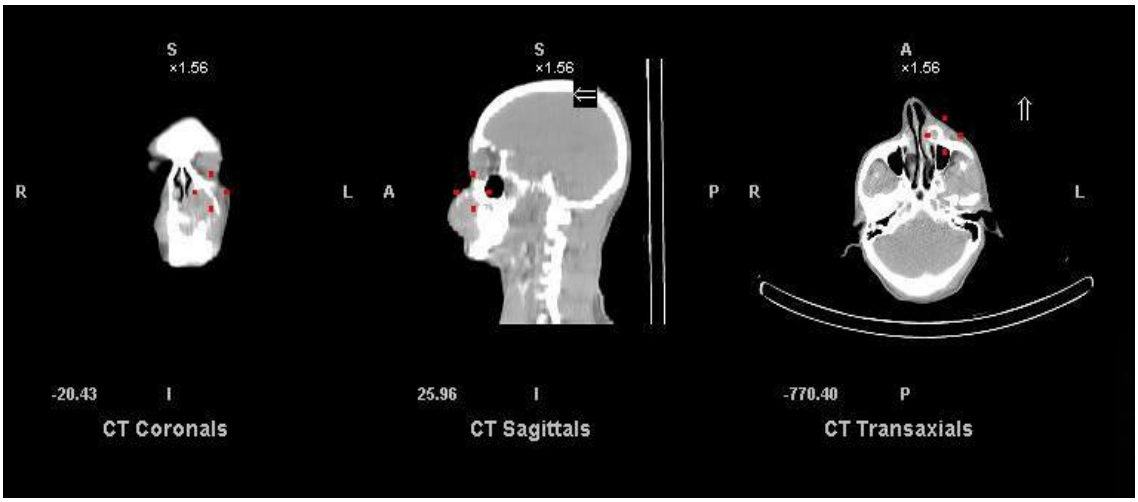

Fig. 5: Computerized tomographic scan showing hypodense area in left anterior maxilla (marked by red).

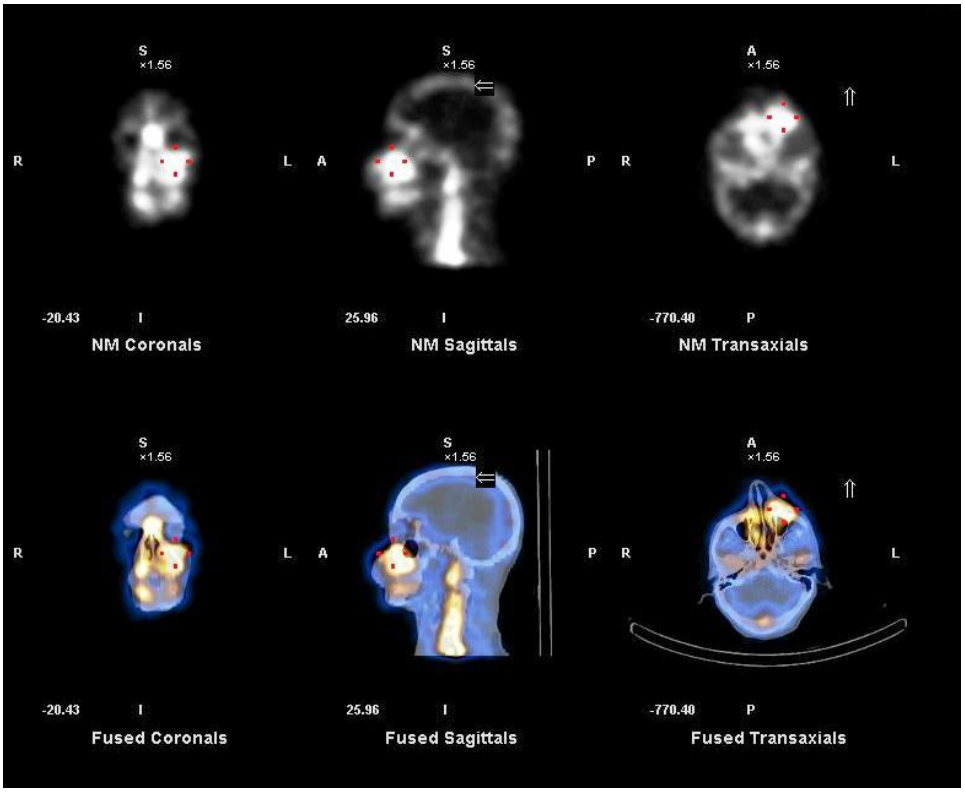

Fig. 6: PET-CT showing increased concentration of FDG in left anterior maxillary region.

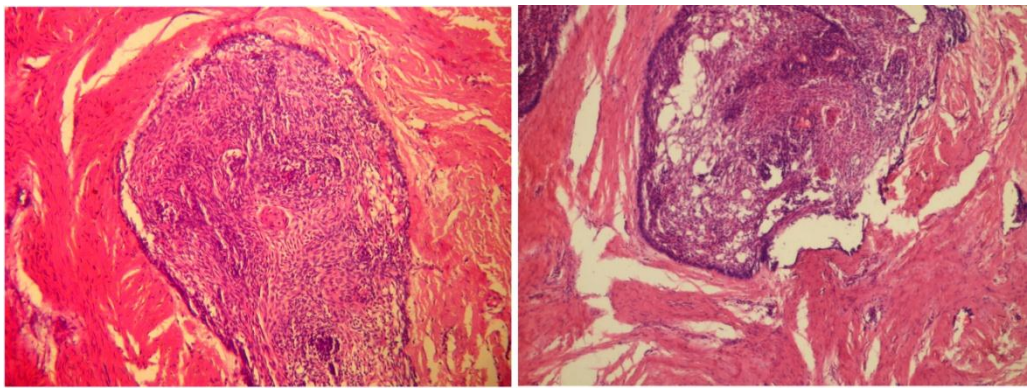

Fig. 7: H & E stained section revealed connective tissue stroma with mature collagenous fibres. Presence of epithelial islands with peripheral layer of flattened epithelial cells and individual cell keratinisation was evident. (40X).

DISCUSSION

World Health Organization (WHO) in 2005 has classified SOT as a benign epithelial odontogenic tumor with around 50 cases reported in the literature till date.[4] SOT is a very atypical, locally-infiltrative epithelial neoplasm with three possible sources of origin depending on its location. Intraosseous SOT presumably originates from the cell rests of

Malassez whereas its peripheral counterpart (extraosseous variant) arises from the dental lamina remnants (rests of Serres) or surface of the gingival epithelium. SOT must be distinguished from other intraosseous neoplasms notably ameloblastoma and primary intraosseous squamous cell carcinoma.[5] Even though there is an association with bone involvement and being benign in nature, this tumor has been seen to infiltrate adjacent soft tissues, resulting major esthetic and functional mutilation.[6]

SOT can manifest as a single tumor or affect multiple sites at the same anatomic location (multicentric).[6] The site of occurrence of the neoplasm has an equal predilection for the posterior maxilla and mandible. However, tumors located in the anterior region of the mandible are quiet infrequent whereas, in maxilla, it mostly occurs in cuspid‑first premolar region.[3] Out of the several reported cases, 20 have been located in posterior region of mandible. The present case discussed here was an unusual one since the site of occurrence was the anterior maxilla. These neoplasms are locally infiltrative, nonetheless, tumors of the maxilla tend to perform more aggressively due to the anatomical organization and the dispute it creates for the surgeon.[7] SOT is thought to arise in a extensive age range (1st to 8th decade of life) with mean age of occurrence being 38.2 years. The female to male ratio is 1:1.8 among the many cases reported, thus showing slightly more male predilection.[8] This was found to be in concordance to our case reported here.

Radiographically, central variant of SOT shows a well-defined unilocular, triangular radiolucency between the roots of adjacent teeth. The peripheral variant may show saucerization of underlying bone which probably owes to a pressure phenomenon rather instead of tumor infiltration.[8] When bony evaluation is required, CT is considered to be superior to MRI scans. Recently, studies have demonstrated that the PET-CT can accurately evaluate the extent of tumor and its stages when compared with CT alone with the advantage being that it can image the whole body at one time and detect unsuspected lesions. It provides more accurate evaluation of the primary site, possible recurrence and distant metastases, which frequently results in changing patient management.[9] The present case emphasizes on the radionuclide features along with other imaging modalities.

SOT by and large fabricates a readily decipherable pattern on histopathologic examination. The tumor is characterised to be composed of several round to oval shaped islands of proliferative squamous epithelium uniformly scattered in a connective tissue matrix. These

squamous epithelial islands are easily recognized with swirling or "whirlpool" pattern of central squamous cells and are distinctly distinguished and contoured from the surrounding stroma by a flattened layer of cells at their periphery. Presence of cystic change centrally within the epithelial islands is frequently evident and individual cell keratinization of the central squamous cells may be present.[7] Intraepithelial microcystic degeneration and circular areas of fibrous condensation around few epithelial islands could be seen, possibly depicting a reactive phenomenon of the connective tissue stroma to epithelial proliferation. These features were quiet evident in the case reported here. The undamaging manifestation of the epithelium exemplify the lesion and differentiates it from other such lesions with histologic features like ameloblastic nuclear polarization and a palisade-like arrangement of peripheral columnar cells that are absent in SOTs. Furthermore, the epithelial proliferations seen in the walls of inflammatory odontogenic cysts are not considered a manifestation of SOT. Therefore, the histopathological differential diagnoses for SOT include the acanthomatous and desmoplastic ameloblastoma and gingival squamous cell carcinoma which is extremely different from SOT.[6]

The treatment of SOT comprises of enucleation, complete curettage or conservative surgical removal. However, more hostile intervention might be essential in the case of maxillary tumors owing to the aggressive probability of the lesion. Recurrence of this odontogenic neoplasm is hardly ever reported in literature and is accredited to partial removal of the initial tumor with no reported malignant changes till date. According to Ide et al. (1999), SOT could transform into a malignant form like intraosseous squamous cell carcinoma.[10] However, only one case of recurrence was observed among the six cases evaluated by Pullon et al.[6] King Kim et al. in 2007 proposed that the neoplasm might decorticate the dense mandibular bone and may affect the therapeutic and diagnostic modalities of these reputed innocent lesion.[11]

CONCLUSION

SOT being a benign odontogenic neoplasm of distinctive histological features has imposed significant dilemma in the establishment of the lesion as a separate entity. The erratic reports in the literature have enhanced the knowledge of the propensity of the lesion to occur in the jaws. However, the need for a comprehensive diagnostic evaluation considering all clinical, radiological and especially histopathological features is mandatory in order to guarantee the success of treatment indicated in each case, endorsing quality of life for patients with SOT.

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