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SQUAMOUS ODONTOGENIC TUMOR LIKE PROLIFERATIONS IN DENTIGEROUS CYST- A CHANCE FINDING.

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ABSTRACT

A female patient, aged 47 years came to the hospital with a dental complaint. During the examination, presence of intraoral swelling distal to the lower right second molar. The patient provided verbal consent to remove the tooth, and subsequent histopathological examination confirmed a diagnosis of squamous odontogenic tumor-like proliferations in dentigerous cyst . This entity is rare and has benign behaviour. Typically they are solitary and unilocular. Herein, we report a classic case of squamous odontogenic tumor-like proliferations in walls of dentigerous cyst .

Keywords : squamous odontogenic tumor-like proliferations , squamous odontogenic tumor, dentigerous cyst.

INTRODUCTION

The Squamous odontogenic tumor (SOT) is an infrequent, benign, locally invasive odontogenic neoplasm of epithelial origin which comprises variably sized islands of well-differentiated squamous epithelium supported in a fibrous stroma. The suggested determinants of SOTs include epithelial rests of Malassez, gingival surface epithelium, and remnants of the dental lamina.[12]

It is a slow growing lesion with tooth mobility, swelling of alveolar process and slight pain are possible indicators of the underlying lesion. Radiography of the central variant of SOT shows triangular unilocular radiolucency involving roots of adjacent teeth. Whereas, ‘saucerization’ of underlying bone is a sequel of a rare peripheral variant of SOT.[3] Till date, approximately 110 of SOTs have been reported in literature.[4]In contrast to odontogenic tumors, rarely SOTs have been

associated with impacted teeth. SOT like islands or SOT-like proliferations (SOT-LP) in walls of cysts is regarded as reactive hyperplasia of the cystic epithelium[3]. This article presents a case of SOT-LP in the walls of dentigerous cyst.

CASE PRESENTATION

A 47-year- old female reported to our institute with a chief complaint of pain and swelling in the lower right back region of the jaw since 1 month. Patient gave a positive medical history of hypertension and was under medications for the same. On intraoral examination presence of intraoral swelling distal to 47. (Fig 1) A panoramic radiograph revealed a well defined pericoronal radiolucency involving horizontally impacted tooth 48. (Fig 2) Based on clinical and radiographic findings a provisional diagnosis of a dentigerous cyst was made. The patient's consent was obtained to excise the lesion. Routine investigations were normal, and an excisional biopsy was performed under local anaesthesia.

Gross examination of the biopsy specimen showed a soft tissue bit measuring 2.5x2 cm in size, grayish white in color and firm in consistency.

On routine microscopic examination, hematoxylin and eosin stained soft tissue showed cystic epithelium resembling reduced enamel epithelium with areas of proliferation. Connective tissue capsule demonstrated presence of variable sized and shaped islands composed of benign squamous epithelial cells with occasional areas of vacuolization, keratinization and microcyst formation. Based on these characteristic histopathological findings, a confirmatory diagnosis of a dentigerous cyst with SOT-LP was made. (Fig 3,4)

Fig 1: intraoral swelling distal to 47.

Fig 2: OPG demonstrating well defined pericoronal radiolucency involving horizontally impacted tooth 48.

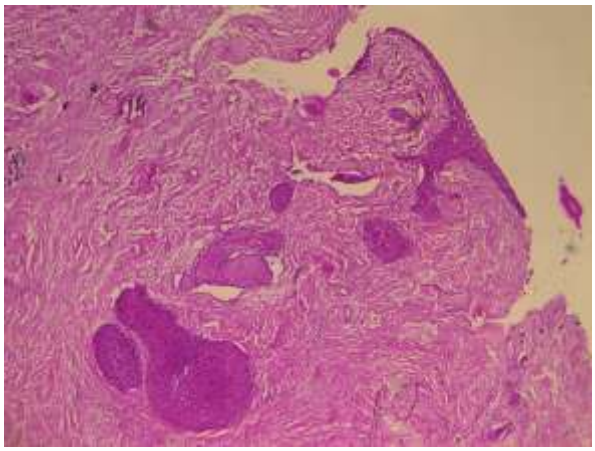

Fig 3: Hematoxylin and eosin stained soft tissue showing cystic epithelium resembling reduced enamel epithelium with areas of proliferation under 10x magnification.

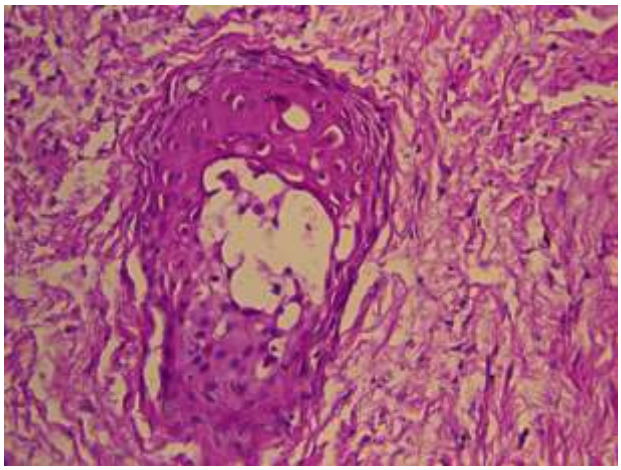

Fig 4: Hematoxylin and eosin stained soft tissue showing solitary island with benign squamous epithelial cells with occasional areas of vacuolization, keratinization and microcyst formation under 40x magnification.

DISCUSSION

WHO in 2005 categorized Squamous Odontogenic Tumor (SOT) as a benign odontogenic tumor of epithelial origin. Central variant is more common in contrast to the rare peripheral variant. Origin of SOT is debatable, some believe that it originates from remnants of dental lamina, gingival epithelium. Majority of researchers have theorized that epithelial rests of malassez in the periodontal ligament contribute to the origin due to occurrence of triangular radiolucency on the lateral surface of roots. Its histologic alike counterpart is designated as squamous odontogenic tumor (SOT) like proliferations, in which these proliferations are isolated in walls of odontogenic cysts.[5] The pericoronal follicle of impacted is another potential site of genesis for these proliferations.[5] This possible origin supports our case as it has also been identified at pericoronal site. SOT and SOT-LP exhibit significant histological similarity, characterized by clusters of well-differentiated, non-keratinized squamous odontogenic epithelial cells encircled by fibrous connective tissue. In our case the histological study of the lesion revealed the presence of variable sized and shaped islands isolated in the cystic wall, comprising of proliferating squamous odontogenic cells, and no signs of cellular atypia. Wright et al[6] in 1979 first reported five cases of SOT-LP in the walls of odontogenic cysts. There are documented cases of SOT-LP in walls of different cysts namely dentigerous cyst, radicular cyst, residual cyst, odontogenic keratocyst and glandular odontogenic cyst. To date, there have been no reports of recurrences or malignant alterations in the literature involving odontogenic cysts and SOT-LP. Nonetheless, periodic clinical and radiological examinations are required to identify potential neoplastic alterations and lesion recurrences.

Till date, 60 cases of SOT-LP have been reported, with the majority of cases in walls of radicular cysts (n=51), only seven cases involved a dentigerous cyst.[7] [8] [9] [10] [4] A summary of clinico-histopathological features of SOT-LP arising from dentigerous cysts is shown in Table 1.

AUTHOR	**AGE (In years)**	**SEX**	**SITE**	**INVOLVED TOOTH**	**BONE EXPANSION**	**HISTOLOGICAL FEATURES**	**DIAGNOSIS**
Wright et al[6]	45	Female	Mandible	47	No	Cyst composed of flattened stratified squamous epithelium of variable thickness. Cystic capsule demonstrated mural proliferations in the form of islands of variable size and morphology with squamous differentiation with no instance of peripheral palisading. Few islands showed microcystic degeneration. Few areas of the capsule were densely infiltrated by mixed inflammatory cells.	Squamous odontogenic tumor and dentigerous cyst, chronically inflamed.
Wright et al[6]	53	Male	Mandible	48	No	Cyst composed of stratified squamous epithelial lining. Cystic capsule was minimally inflamed and demonstrated islands of squamous odontogenic	Dentigerous cyst with mural epithelial proliferation.

						epithelium without peripheral palisading. A few of the islands were associated with cholesterol granuloma.	
Wright et al[6]	36	Male	Mandible	48	No	Cystic lesion lined with stratified squamous epithelium. The connective tissue wall showed a focal thickening containing irregular islands of squamous epithelium without peripheral palisading. A few of the islands showed cystic degeneration.	Dentigerous cyst with mural squamous odontogenic tumor.
Wright et al[6]	65	Female	Mandible	38	No	Cyst composed of fragments of edematous stratified squamous epithelium on the luminal surface. A focus of large. irregular islands of squamous epithelium were noted within the cystic capsule. No peripheral palisading was evident. Capsule was focally infiltrated by dense collections of chronic inflammatory cells.	Dentigerous cyst, chronically inflamed. (Because of the small quantity of epithelial proliferation.)
Fay et al [11]	60	Male	Mandible	37	Unknown	Unknown	

Leventon et al[12]	17	Female	Unknown	Unknown	No	Prominent islands of keratinizing squamous epithelium in the capsule of nonkeratinizing dentigerous cyst.	Dentigerous cyst with islands of keratinizing squamous epithelium.
Barbeiro et al[13]	42	Male	Maxilla	18	No	Cystic lesion lined by nonkeratinized, stratified squamous epithelium, containing several islands of benign-appearing squamous epithelium in the cystic capsule	Dentigerous cyst showing SOT-LP features.
Our case	47	Female	Mandible	48	No	Cystic lesion lined by nonkeratinized, stratified squamous epithelium. Cystic capsule shows presence of variably sized islands of benign squamous epithelial cells with occasional keratinization, areas of vacuolization and microcyst formation. No cellular atypia was noted.	Dentigerous cyst with SOT-LP.

Table 1. Review of reported cases of SOT-LP.

CONCLUSION

In light of the aforementioned, we believe it is imperative to perform a comprehensive histological analysis of every lesion extracted from the oral tissues. It is critical to avoid misdiagnosing SOT or other disorders such ameloblastoma or primary intrabony squamous cell carcinoma when a SOT-LP is present.

CONFLICT OF INTEREST

The authors declare NO conflicts of interests

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