Case Report

Squamous odontogenic tumour-like proliferation in a maxillary dentigerous cyst – An unusual finding

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Abstract

Squamous odontogenic tumour-like proliferations (SOTLPs) in the wall of odontogenic cysts are rare occurrences. Due to the histopathological similarity of these proliferations to neoplasms, such as squamous odontogenic tumour, intraosseous well-differentiated squamous cell carcinoma, and acanthomatous ameloblastoma, their correct elucidation is of paramount importance to avoid unnecessary and unwanted treatment. SOTLPs are uncommon in dentigerous cysts and rare in those that occur in the maxilla particularly the anterior region. This paper presents a case of maxillary dentigerous cyst involving 33 and a mesiodens in a 32 year old male which on histopathological examination showed SOTLPs in a dentigerous cyst.

Keywords: Dentigerous cyst, epithelial island, odontogenic cyst, squamous odontogenic tumour like proliferation

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INTRODUCTION

Squamous odontogenic tumour (SOT) is a rare benign epithelial odontogenic tumour that can invade into trabecular bone, destroy the cortical bone and infiltrate into its adjacent tissues.[1] It was described by Pullon *et al*.[2] (1975) for the first time in a series of six cases. Most of the SOTs develop in the periodontal ligament of permanent teeth. SOT can occur in three forms. The most common is the intraosseous/intrabony or central type. Apart from the SOTs that occur intraosseously, SOTs have been found to occur extraosseously (peripheral variant) and also in the form of squamous odontogenic tumour-like proliferations (SOTLPs) in the connective tissue wall of odontogenic cysts.[3,4] Calcifying epithelial odontogenic tumours has also been found in association with SOTLPs.[5] SOTs localized in edentulous areas, and multicentric ones have also been described.[6]

Histopathologically, SOT is characterized by islands and nests consisting of well-differentiated squamous epithelial cells located in a fibrous stroma.[2,7] It is important that SOTLPs are differentiated from SOT because they are non-neoplastic and may represent reactive inflammatory hyperplasia of the cyst epithelium.[6] The presence of SOTLPs in the walls of odontogenic cysts is a rare phenomenon.[3,8]

In this paper, we report a case of SOTLP in a maxillary dentigerous cyst, an uncommon finding.

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CASE SUMMARY

A 32-year-old male patient reported a chief complaint of painless swelling in the inner aspect of his upper front teeth region for a week. Intraoral examination revealed vestibular obliteration and palatal swelling in relation to 21 and 22. The swelling was approximately 1×1×0.5 cm^3 in measurement, roughly oval in shape and showed a smooth surface, as shown in Figure 1. The swelling was firm in consistency. There was a missing tooth 23. Intraoral periapical radiograph revealed unilocular radiolucency around impacted 23, and an inverted mesiodens was also noticed, which was confirmed by computed tomography [Figure 2a and 2b].

Based on the clinical findings, radiographic features and computed tomography (CT) findings, a provisional diagnosis of dentigerous cyst was given.

Surgical excision was performed to remove the cyst along with impacted 23 and mesiodens. The specimen was sent for histopathological examination. Histopathological examination revealed a cystic lesion with a two- to three-cell thick epithelial lining and connective tissue capsule. Epithelium resembled reduced enamel epithelium [Figure 3a]. In some areas, goblet cells and ciliated epithelium was evident [Figure 3b]. In addition, few islands showed squamous epithelium in the cystic capsule with flat and few cuboidal cells in the periphery of the islands [Figure 4a and 4b]. Dyskeratotic cells and keratin pearl formation were not seen in the islands [Figure 4b]. The connective tissue capsule was fibrocellular and showed moderate vascularity with mild inflammatory cell infiltrate. Few calcifications were also evident. Based on these features, a diagnosis of dentigerous cyst with SOTLP was given.

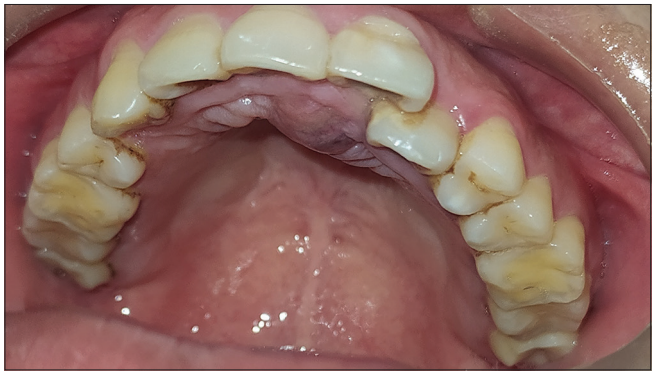

Figure 1: Clinical image showing a swelling on the palatal aspect of 21, 22 region

DISCUSSION

SOTLPs in the connective tissue wall of odontogenic cysts are rare histopathological findings, the pathogenesis of which is not yet known.[3] SOTLPs in odontogenic cysts have been reported in the literature.[3,7-11] Since SOTLPs have histopathologic similarities with SOT, several investigations have been performed to know its clinical, histopathologic spectrum, histogenesis, prevalence and biologic behaviour. Though the literature available is mainly based on SOTLPs found in radicular cysts, central SOT and SOTLP differ in the mean age of the patients (older patients in case of SOTLPs), location of the lesion (SOTLPs more frequently observed in maxilla), mobility of tooth/teeth associated with the lesion (more frequently observed with central SOTs), cortical bone perforation (only observed in central SOTs) and locularity appearance in radiological examinations (all SOTLPs presented with a unilocular appearance).[10] Notable case series studies on SOTLPs have been conducted by Wright,[11] Chrcanovic *et al*.,[10] Parmar *et al*.[8] and Zargaran *et al*.[9] all of whom have provided substantial information on the occurrence of SOTLPs in odontogenic cysts. SOTLP was, for the first time, described by Doyle *et al*.[12] in the wall of a radicular cyst. The majority of the cases of SOTLP have been reported to be associated with radicular cysts,[13] whereas only six cases have been found to be reported in dentigerous cysts.[13,14] The radicular cysts showing SOTLPs were prevalent in the

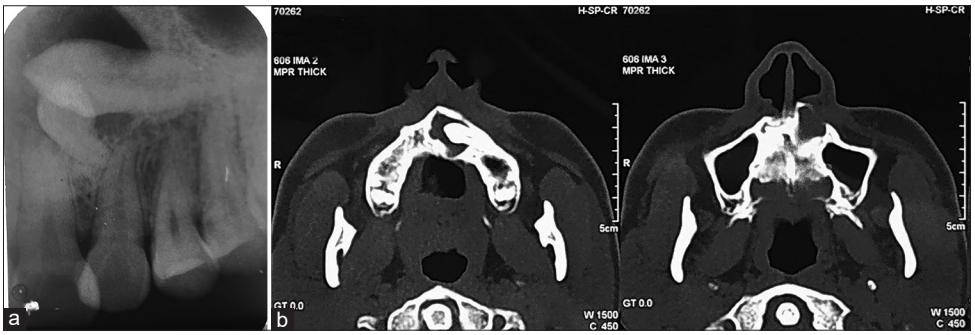

Figure 2: (a) Intraoral periapical radiograph showing unilocular radiolucency around 23 and inverted mesiodens. (b) Computed tomography scan (CT) showing impacted 23

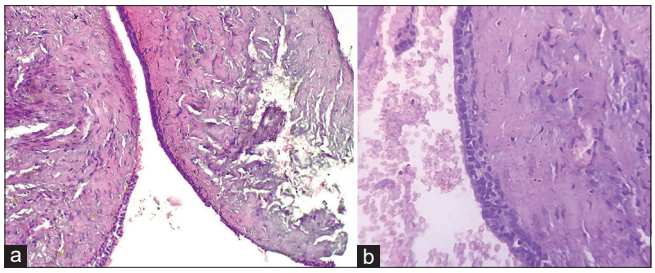

Figure 3: (a) Histopathological picture showing lining epithelium of dentigerous cyst (H&E, 10X). (b) Histopathological picture showing ciliated epithelium in focal area of dentigerous cyst lining (H&E, 40X)

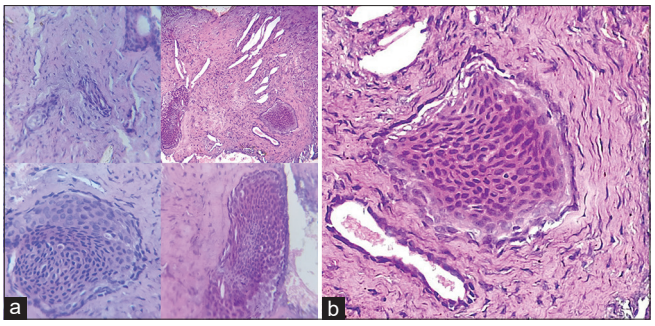

Figure 4: (a) Histopathological picture showing SOTLPs within the connective tissue wall of the dentigerous cyst (H&E, 10X). (b) Histopathological picture showing an island composed of well-differentiated squamous cells with bland appearance within the connective tissue wall of the dentigerous cyst (H&E, 40X)

maxilla in comparison to the mandible and in the anterior region in comparison to the posterior region. Of all the dentigerous cysts reported with SOTLPs, only a single case has been reported in the maxilla to date by Barbeiro *et al*.[13] whereas the rest have been reported in the mandible.[8,10] The single maxillary case was a male 42 years old, and the tooth involved was 18, in the posterior region of the upper jaw. The present case also occurred in a maxillary dentigerous cyst, an uncommon finding. Our patient was a 32-year-old male and the tooth involved was 23 with an inverted mesiodens, involving the anterior region of the upper jaw. SOTLPs have been found in ages ranging from second decade to ninth decade.[9,11]

Radiographically, all the odontogenic cysts showing SOTLPs show no signs of cortical perforation or root resorption of adjacent teeth, and all have an unilocular appearance.[10] Multilocular lesions are rarely seen.[10] Before Doyle *et al*.,[12] all published cases of SOTLPs were considered as latent or incipient neoplasms. Contrary to this belief, Goldblatt *et al*.[1] were of the opinion that the absence of cellular atypia in SOTLP indicates that they are not true neoplasms. The aetiology of SOTLP is not known, although it is believed that they may develop from rests of Serres, or epithelial rests of Malassez or pericoronal follicle of unerupted or impacted teeth.[2,12] The most frequent location of SOTLP is in the wall of an odontogenic cyst, most commonly in radicular cyst, followed by dentigerous cyst and less commonly in residual cyst, lateral periodontal cyst, glandular odontogenic cyst and odontogenic keratocyst.[7,14-16] Our case was in a maxillary dentigerous cyst associated with an unerupted permanent canine and mesiodens. Variable-sized islands of bland appearing well-differentiated squamous cells in the connective tissue wall of odontogenic cysts characterize SOTLPs.[11] In radicular cysts, it is common to find the SOTLPs in close proximity to the lining epithelium. The cells at the periphery of the islands are flat or cuboidal basaloid. The squamous cells do not exhibit features such as dyskeratosis, keratin pearl formation, microcyst formation and mitosis.[8] Evidence of inflammation may be seen mostly in cases of radicular cysts, which may vary from mild to moderate to severe, with the cells chiefly consisting of lymphocytes and plasma cells.[7] Budlike extensions and the presence of rests of Malassez may also be seen. Opinions vary on the origin of the SOTLP epithelium in odontogenic cysts. The cell rests of Malassez could be an important source of epithelial proliferation, considering the proliferative ability of these rests, their ability to undergo squamous differentiation and the close proximity of the cysts with SOTLPs to the roots of the teeth.[7] Philipsen and Reichart[6] are of the opinion that SOTLPs are a result of reactive, inflammatory hyperplasia of the epithelial cyst lining. Another theory of the origin of the SOTLPs is a budding type of hyperplasia of the lining epithelium of radicular cysts, which can be attributed to a response to subsiding inflammation because it usually occurs in areas without inflammation.[9,10] Parmar *et al*.[8] believe that inflammation might not be the primary stimulus for SOTLP in radicular cysts, as SOTLPs are found in inflammation-free areas in their study of 42 SOTLP cases. Oliveira *et al*.[3] observed similar findings regarding the lack of inflammation as the proliferative stimulus. Further, SOTLPs are not confined to inflammatory cysts as they are also seen in developmental cysts such as dentigerous cysts, glandular odontogenic cysts and odontogenic keratocysts. The fact that SOTLPs occur in developmental cysts suggests that factors other than inflammation may be responsible for the phenomenon.[8]

Histopathologically, SOTLPs are morphologically similar to epithelial islands that form SOT, although they do not have any tendency to develop into a solid tumour.[7,8,11] Wright *et al*.[11] and Zargaran *et al*.[9] found that all the SOTLP cases were radiographically diagnosed provisionally as a

cyst and were also cystic in terms of gross description as was in our case.

Sometimes, SOTLPs may histopathologically overlap with neoplasms such as acanthomatous ameloblastoma, primary intraosseous squamous cell carcinoma (PISCC) and squamous cell carcinoma (SCC) arising from cystic lining. Ameloblastoma typically has islands with peripheral columnar ameloblast-like cells which have a clear cytoplasm and nucleus with reverse polarity.[8] The squamous islands in our case showed peripheral cells, which were flat with cuboidal cells in some areas. Though budding from the epithelial lining was not seen in our case, when the islands of squamous cells are seen adjacent to the cyst lining, it can create confusion with an SCC arising in an odontogenic cyst. The islands in PISCC and SCC would exhibit cytologic atypia including altered nuclear cytoplasmic ratio, nuclear hyperchromatism and mitotic activity.[8] The epithelial islands, in our case, did not exhibit any cellular atypia or other features suggestive of a malignant process. Neoplastic transformation of SOTLPs to a solid SOT does not occur, and their clinical behaviour is no more aggressive than the cysts in which they are found.[8] Enucleation is the treatment modality most often described in the literature.[8,10] SOT is an odontogenic neoplasm that shows more aggressive biologic behaviour than SOTLPs, and both lesions have distinct clinicopathologic features.[8,10]

CONCLUSION

SOTLPs in the lining epithelium of odontogenic cysts are rare, and the correct diagnosis of such finding is of paramount importance. As SOTLPs may at times bear close resemblances to SOT or acanthomatous ameloblastoma or an intraosseous carcinoma, histopathological misinterpretation may lead to unwanted and unnecessary treatment. To date, malignant transformation of SOTLPs in odontogenic cysts has not been reported.

Key messages

SOTLPs may be misinterpreted as neoplasms such as squamous odontogenic tumour, acanthomatous ameloblastoma, desmoplastic ameloblastoma or an intraosseous carcinoma because of its histopathologic features that overlap with these neoplasms which may lead to unwanted and unnecessary treatment.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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