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CASE REPORT

A Rare Tumor of the Anterior Maxilla Masquerading as a Periapical Cyst: Case Report of Squamous Odontogenic Tumor

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ABSTRACT

Introduction: The squamous odontogenic tumor (SOT) is the most uncommon of all benign odontogenic neoplasms. It occurs from the first to eighth decades with a male predominance of 1.8:1. It may be solitary, multifocal or multicentric with the involvement of both jaws.

Case report: Here we are reporting a case of squamous odontogenic tumor in a boy of 12 years. The lesion was accidentally discovered in a radiograph taken after the patient had a fall injury while he was playing which revealed a radiolucent, unilocular, roughly triangular image localized in the periapical region of the right central incisor. On microscopic examination, the lesion was composed entirely of islands and variably sized nests and cords of squamous epithelium scattered in a fibrous stroma.

Management and prognosis: After complete surgical excision of the tumor and curettage the patient did not present any postoperative complications and no evidence of recurrence was noted for the past 6 months.

Conclusion: As the tumor can be easily misdiagnosed as other lesions like ameloblastoma, ameloblastic fibroma or squamous cell carcinoma that require more radical and mutilating treatments meticulous care should be taken while examining these lesions. Underdiagnosis of pseudoepitheliomatous hyperplasia or odontogenic proliferations can be harmful to the patient as SOTs may undergo a malignant transformation as observed in a few cases.

Keywords: Benign, Maxilla, Squamous odontogenic tumor.

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INTRODUCTION

The squamous odontogenic tumor is a benign slow-growing tumor that gradually invades the trabecular bone, destroys the cortical bone and infiltrates the adjacent structures.[1] It was first described by Pullon et al. in 1975 in a series of six cases.[2] Fewer than fifty cases have been recorded in literature until now. In the WHO classification, it was included in the family of benign odontogenic tumors of epithelial origi.[3] The central variant is the most common. Majority of unifocal squamous odontogenic tumors are asymptomatic and demonstrate indolent growth. Occasionally, associated pain, gingival swelling, mild erythema or mobility of adjacent teeth may be present. It usually presents as a semicircular or triangular radiolucency of the alveolar ridge between the roots of adjacent permanent teeth. Because of their benign character, these lesions are usually treated using a conservative surgical approach with curettage and surgical enucleation.[4] The purpose of this case report is to describe yet another case of a squamous odontogenic tumor in the anterior maxilla of a 12-year-old boy and review the literature.

CASE REPORT

A 12-year-old boy had a trauma as a result of a fall and was referred to the Department of Conservative Dentistry for the restoration of the fractured teeth. The extraoral examination did not show any swelling or asymmetry. Intraoral examination revealed normal mucosa and a mild degree of expansion of the labial surface in relation to 11 which was hard on palpation. There was no drainage or purulence from the area. A radiograph of the maxillary anterior teeth revealed a well defined, not very well circumscribed radiolucency in the periapical region of 11 measuring around 1 × 1.3 cm (Fig. 1). The initial assumption was that of a radicular cyst because of the history of trauma. A surgical flap was raised and the cortical bone was removed revealing a grayish rubbery mass which could be shelled out and submitted for histopathologic examination (Figs 2A and B). On microscopic examination, the lesion was composed entirely of islands and variably sized nests and cords of squamous epithelium scattered in a fibrous stroma (Fig. 3). The squamous cells were very uniform and exhibited no pleomorphism, nuclear hyperchromatism or mitotic activity. Occasionally, individual cell keratinization was present but no keratin pearls (Fig. 4). Intercellular bridges were

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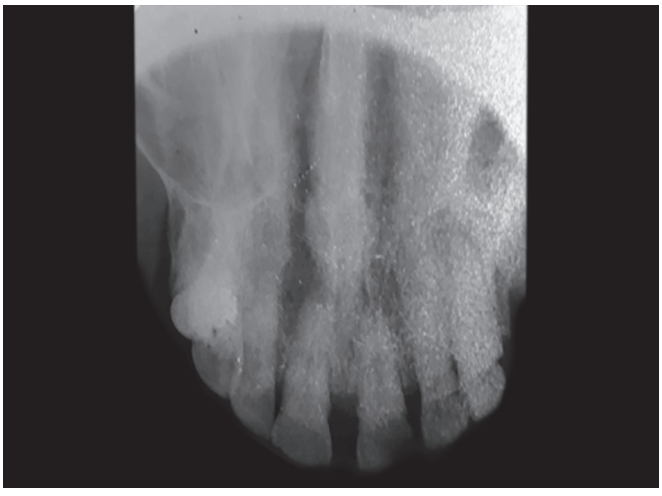


Fig. 1: Radiograph revealing a well defined, not very well circumscribed radiolucency in the periapical region of 11

present In some of these islands, the cells in the center were undergoing cystic degeneration (Fig. 5). Large and small lamellated calcified bodies were seen in some areas (Figs 6A and B). The peripheral layer was flattened, almost cuboidal. The fibrous stroma surrounding the epithelial islands was composed of mature bundles of collagen fibers. Immunohistochemistry with cytokeratin 13/16 showed heavy staining of the epithelial islands indicating proliferative activity of odontogenic epithelium (Figs 7A and B). Based on all these findings, a definitive diagnosis of the squamous odontogenic tumor was given.

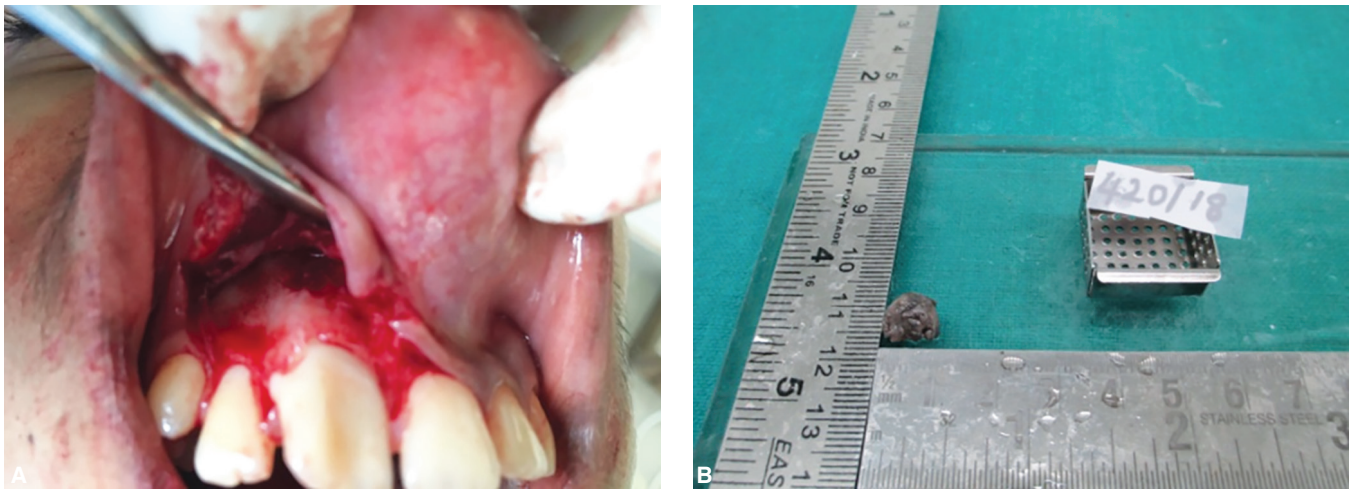
DISCUSSION

The squamous odontogenic tumor was a lesion without an established name until 1975 when Pullon et al. assembled, studied and published a series of six cases.[2] Before that pathologists gave this lesion such names as "benign epithelial odontogenic tumor," "acanthomatous ameloblastoma," "epithelial odontogenic tumor," and "acanthomatous ameloblastic fibroma." because the islands of squamous epithelium have a morphologic appearance similar to that seen in the follicular pattern of simple ameloblastoma.[5] However similar to ameloblastoma it may appear, the lesion has a distinct histopathologic appearance justifying the name "squamous odontogenic tumor."

Even after four decades only around fifty cases of the intraosseous squamous odontogenic tumor were found in literature making it the rarest of all benign odontogenic tumors. The tumor often appears as a triangular-shaped unilocular expansile radiolucency in the alveolar process between the roots of teeth. This involvement of the alveolus is typical although large tumors may extend into the body of the mandible, or in the maxilla, they may involve the maxillary sinus.[6] Some of the larger lesions have appeared as multilocular radiolucencies and can be associated with impacted teeth.[7] Multiple sites have been involved in about one-fourth of cases.[8] The mandible was affected more frequently.[9] McNeil et al. reported a case of multiple SOTs in all four quadrants.[10] In the present case, the solitary lesion was noted in the maxilla in relation to a central incisor. Initially, the lesion was confused with a radicular cyst because of the periradicular location and history of trauma, but histopathological examination proved otherwise.

The histogenesis of SOT is controversial as many researchers have supported the theory of its origin from the cell rests of Malasezz although some believe that it arises from cell rests of Serres or reduced enamel epithelium. It is possible that it could arise from any residual odontogenic rests.[2,3,11]

Other than the central variant, SOT may present as a peripheral tumor in the gingiva.[12] Doyle et al. reported the first completely extraosseous case.[13,14] Histologically similar lesions have been reported as mural growths within the walls of odontogenic cysts called squamous odontogenic tumor-like proliferations (SOTLP).[15] Ide et al. reported a cystic SOT in a 46-year-old woman.[16]



Figs 2A and B: (A) Raised surgical flap; (B) The cortical bone was removed revealing a grayish rubbery mass

A possible hamartomatous nature of the SOT was also suggested because of multiple site involvement in some cases.[2] A familial pattern was reported with multiple lesions in three siblings.[17] Another case of bilateral maxillary SOT was found in association with a primary squamous cell carcinoma of the mandible.[18] A squamous odontogenic tumor following an orthodontic micro-screw was reported recently.[19]

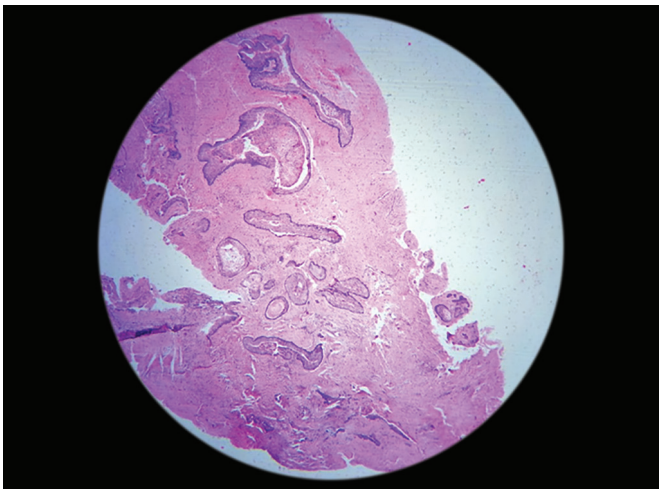


Fig. 3: The lesion composed entirely of islands and variably sized nests and cords of mature squamous epithelium (H&E- 4X)

Malignant transformation of the squamous odontogenic tumor to intraosseous squamous cell carcinoma has been described in the mandible.[4,16,20] Although rarely reported this consequence should always be taken into consideration.

This tumor is characterized histologically by epithelial islands of various size and shape. These islands are usually round in shape though they may appear irregular and cord like in some instances as in the present case. This pattern is very much similar to desmoplastic ameloblastoma. The peripheral layer is flattened and devoid of palisading, polarized cells as in ameloblastoma. Lamellated calcified bodies may be seen in some of the islands.[3,11] The epithelial cells at the center of the islands undergo individual cell keratinization and microcystic changes

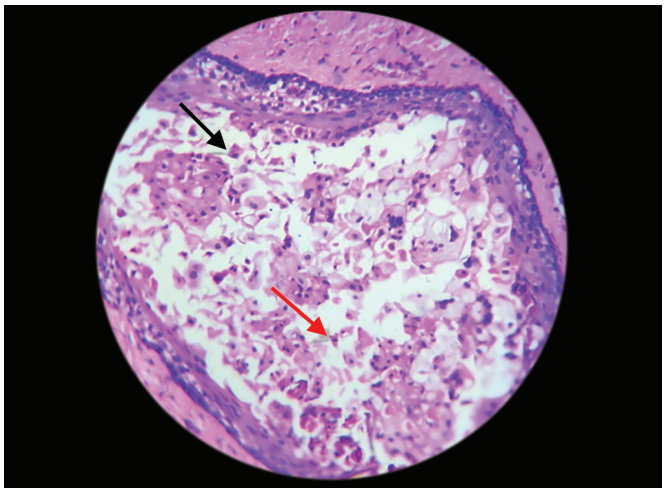


Fig. 4: Flattened peripheral layer. The epithelial cells at the centre of the islands undergoing individual cell keratinization (black arrow) and microcystic changes (red arrow) (H&E 40X)

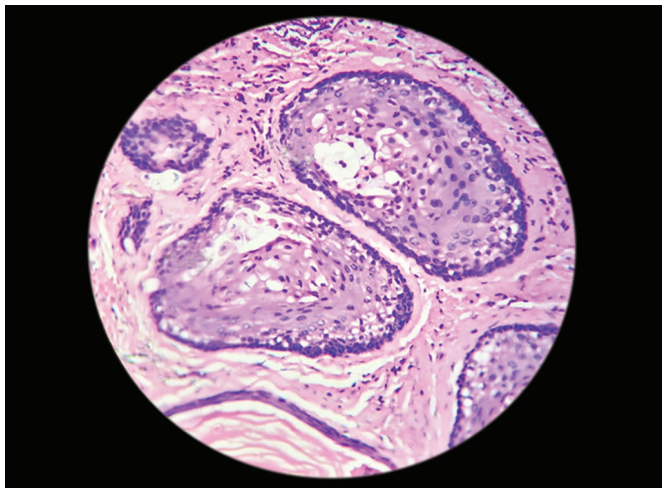
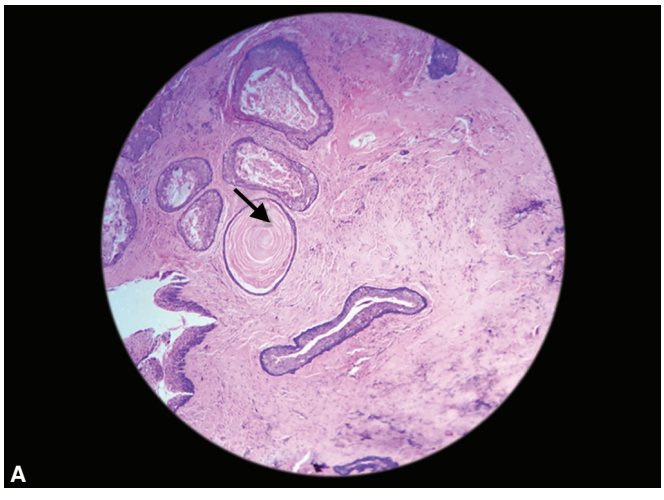
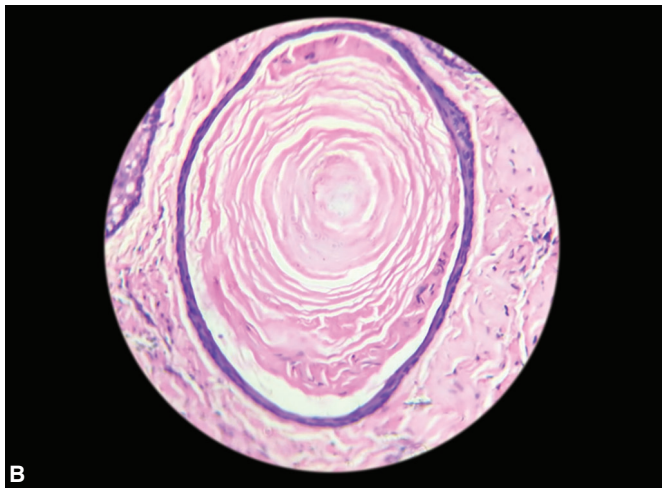
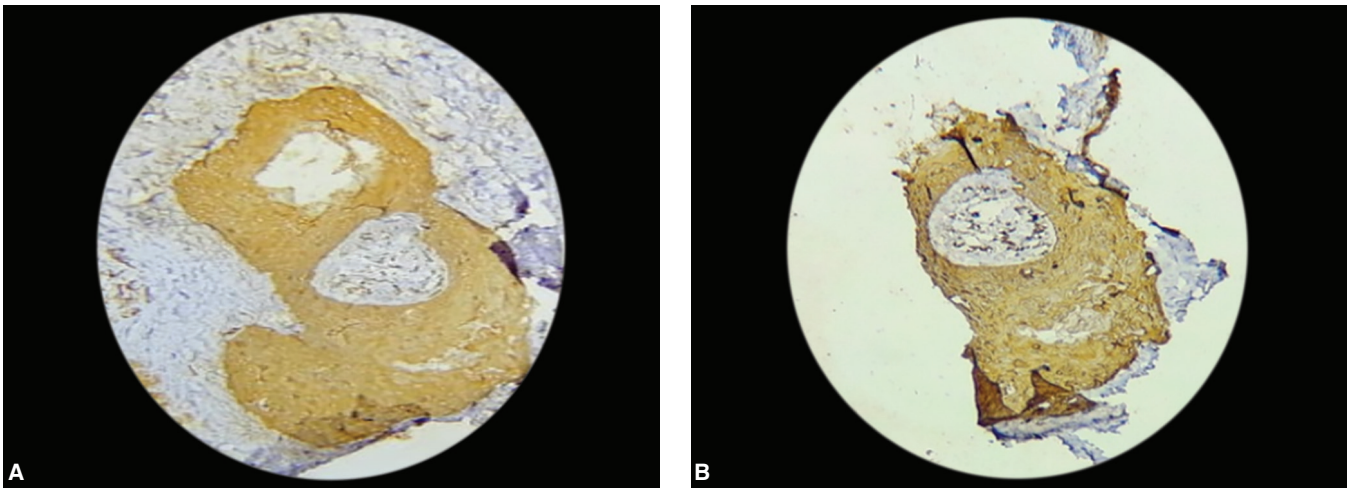


Fig. 5: Centre of the epithelial islands showing microcystic changes. (H&E 40X)





Figs 6A and B: Lamellate calcified bodies in some of the epithelial islands.(H&E 10X, 40X)



Figs 7A and B: Immunohistochemistry with (A) Cytokeratin 13; (B) Cytokeratin 16 showing heavy staining of the epithelial islands indicating proliferative activity of odontogenic epithelium (10X)

and may resemble acanthomatous ameloblastoma.[3] If secondarily infected these lesions may show inflammatory cell infiltration. So there are chances of misdiagnosing SOT as ameloblastomas or intraosseous squamous cell carcinoma. In such cases, clinical behavior of the lesion has to be correlated with the histopathological features. The present case shows histological features typical of the squamous odontogenic tumor and the patient belonged to a younger age group. After complete surgical excision of the tumor and curettage, the patient did not present any postoperative complications and no evidence of recurrence was noted for the past 6 months.

CONCLUSION

As the tumor can be easily misdiagnosed as other lesions like ameloblastoma, ameloblastic fibroma or squamous cell carcinoma that require more radical and mutilating treatments meticulous care should be taken while examining these lesions. Underdiagnosis of pseudoepitheliomatous hyperplasia or odontogenic proliferations can be harmful to the patient as SOTs may undergo a malignant transformation as observed in a few cases. SOT may mimic various other odontogenic and nonodontogenic lesions of the alveolus where clinical and radiographic findings may not be very much characteristic of the tumor as in the present case. Accuracy of histopathologic diagnosis is the key to successful treatment of the lesion in such circumstances. Though rare squamous odontogenic tumor should always be considered as a differential diagnosis while examining most lesions of the alveolus.

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