

SHORT COMMUNICATION

Rare presentation of radicular cyst with sebaceous differentiation

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Abstract

Sebaceous glands in the oral mucosa are said to be a normal variation, but the presence of the sebaceous gland in the jaw is extremely rare. Sebaceous gland differentiation in dentigerous cysts and in keratocystic odontogenic tumor (earlier odontogenic keratocysts) has been reported, but it has never been reported in any radicular cyst. We presented a case of a radicular cyst in an 18-year-old male with sebaceous gland differentiation.

Key words: Metaplasia, odontogenic cyst, radicular cyst, sebaceous differentiation, sebaceous gland

Introduction

Sebaceous glands and dermal adnexal structures present in the oral mucosa and are termed Fordyce granules. In rare instances, they are associated with odontogenic cysts. Hofrath, Gorlin and Spouge documented the occurrence of sebaceous gland in dentigerous cyst.^[1,2] Branon in his review of 312 odontogenic keratocysts (OKC), found three cases contains sebaceous gland.^[3] Recently, Shamim *et al.* have reported a sebaceous gland differentiation in an OKC.^[4] A review of the available English literature revealed no case of sebaceous gland differentiation with the association of radicular cyst.

Case Report

An 18-year-old male presented with a chief complaint of pain in the front, upper, and right region of the upper jaw. Intra-oral periapical revealed an ovoid radiolucency

covering the roots of 11 and 12 [Figure 1]. A provisional diagnosis of radicular cyst was given thereafter.

Apicoectomy of the tooth was performed [Figure 2] and some soft tissue attached along with a root piece was sent for histopathological evaluation [Figure 3]. After the surgical procedure, the mucoperiosteal flap was sutured properly [Figure 4].

Histopathological examination revealed a 5-8 cell layered thick cystic lining of parakeratinized stratified squamous epithelium, which was hyperplastic at a few places. Arcading pattern of epithelium was noted and connective tissue was seen enclosed within the arcades. Fibrous capsule was made up of bundles of collagen fibers, fibroblast, fibrocytes, and dilated blood vessels. Chronic inflammatory cell infiltrate was also seen, chiefly composed of lymphocytes [Figure 5]. In the deeper connective tissue, an unusual finding of sebaceous tissue was also noted, which was surrounded by chronic inflammatory cells [Figures 5 and 6].

Based on all these histopathological features with the correlation of clinical and radiological features, the final diagnosis of radicular cyst with sebaceous differentiation was given.

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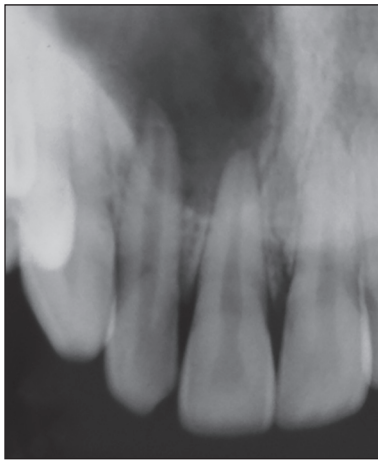


Figure 1: Radiolucent lesion associated with roots of 11 and 12



Figure 2: Surgical removal of the lesion

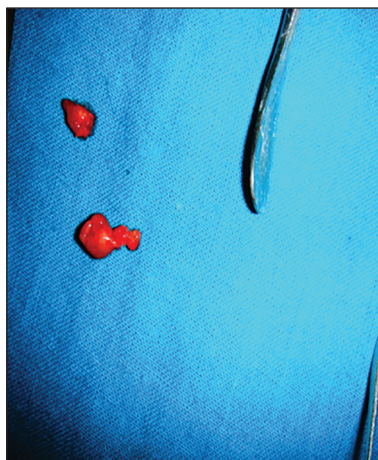


Figure 3: Gross specimen

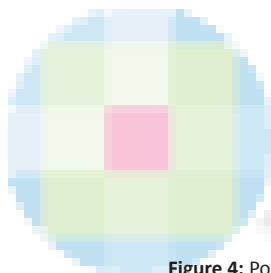


Figure 4: Postoperative picture after suturing

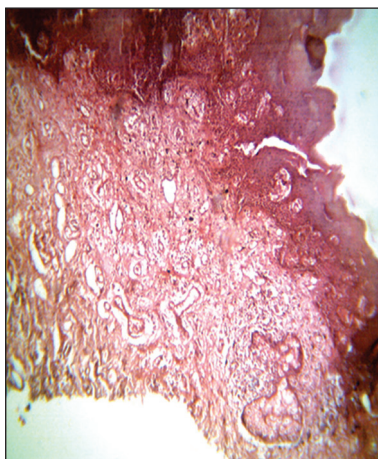


Figure 5: Hyperplastic epithelium shows arcading pattern and sebaceous gland like structure present in deeper connective tissue (H and E, 10x)

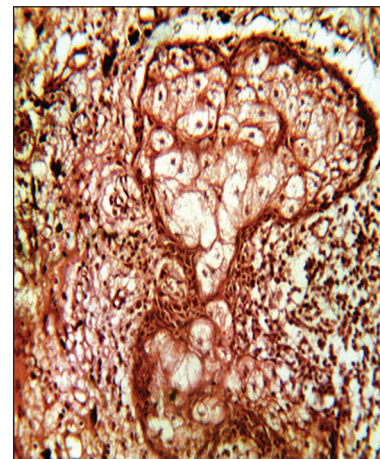


Figure 6: Sebaceous gland like structure (H and E, 40x)

Discussion

Fordyce granules are such a frequent intra-oral finding that they can be considered a normal anatomic variation rather than an ectopic phenomenon.^[4,5] In addition,

aberrant or ectopic sebaceous glands have been described in various locations, including the parotid gland, orbit, larynx, and esophagus. Intraosseous jaw cysts with sebaceous elements are rare, and various interpretations of such cysts have been given in the

literature. Some authors have described these lesions as orthokeratinized odontogenic cysts exhibiting sebaceous differentiation, whereas others have preferred to consider these lesions as intraosseous dermoid cysts or unusual variants of dentigerous cysts.^[5] Chi *et al.* have reported five cases of jaw cysts with sebaceous elements and reviewed the literature concerning these unusual lesions.^[6] The sebaceous glands located deeper within the cyst wall, as per our case, potentially could originate from metaplasia of sequestered epithelial rests.^[7] Many investigators have commented on the pluripotentiality of the odontogenic epithelium with the capacity to differentiate into sebaceous cells, mucous cells, respiratory epithelial cells, and other cell types.^[8]

In our opinion, as mucoepidermoid carcinoma is one of the potential complications of a dentigerous cyst with mucous metaplasia, likely sebaceous metaplasia of odontogenic epithelium of a cyst can also undergo malignant transformation if not treated promptly and appropriately.

Conclusion

The behaviors of these usual jaw cysts with unusual finding of adnexal structures are uncertain as per the paucity of the reported cases. Its recognition is important due to its capabilities for further metaplastic transformation to adnexal tumors. Hence, we conclude the present lesion to be a rare case of radicular cyst with

adnexal metaplasia, which may otherwise get under-diagnosed. Therefore, both the maxillofacial pathologists and maxillofacial surgeons should have a thorough and complete understanding of this unusual variant, in order to approach to an accurate treatment and follow-up plan. Moreover, we recommend that such types of cysts should be categorized as a separate entity.

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