Dental Research Journal

Case Report

Mucinous cystadenoma: A rare entity

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Received: May 2012
Accepted: May 2013

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ABSTRACT

Cystadenoma is a rare benign salivary gland tumor that chiefly originates in the minor salivary glands as a cystic growth with papillary projections into the cystic lumen without the lymphoid element. It is further classified into two histopathological variants that have been recognized by World Health Organization as the papillary and the mucinous forms of cystadenoma. Clinically, it is difficult to differentiate from other benign minor salivary tumors and mucous retention phenomenon. Diagnosis is chiefly based on characteristics histological features. It is believed that the salivary gland tumors are difficult to diagnose and interpret because there are varied patterns of presentation. The study of salivary gland disorders has increased in leaps and bounds. The authors report a case of mucinous cystadenoma of the minor salivary gland on the hard palate, which is even rarest of the rarely reported cystadenomas of the minor salivary glands.

Key Words: Benign neoplasm, hard palate, minor salivary gland tumor, mucinous cystadenoma

INTRODUCTION

Tumors of salivary glands constitute a heterogeneous group of lesions of great morphologic variation. Annual incidence of salivary gland tumors around the world is 1-6.5 cases per 100,000 people. They are more common on hard palate than soft palate, probably because there are greater number of gland aggregate on hard palate than on soft palate.[1]

Cystadenomas of salivary glands are benign neoplasms, in which the epithelium demonstrates adenomatous proliferation that is characterized by formation of multiple cystic structures. Several morphological variants of cyst adenoma have been described of which papillary and mucinous cystadenoma are important. World Health Organization (WHO) described papillary cystadenoma which closely resembles Wartins tumor, but without the lymphoid elements. If mucous cells predominate in the cell population of the lining epithelial cells, the tumor is termed as mucinous cystadenoma.

Cystadenoma is a rare, slowly and painlessly enlarging asymptomatic salivary gland tumor that rarely exceeds 1.5 cm in diameter and most commonly occurs in the minor salivary glands.[1] It comprises about 0.8-6.3% of all benign minor salivary gland tumors. Common sites of occurrence are palate and buccal mucosa, lip and the tonsillar area, but cyst adenomas have also been reported to occur on the tongue.[1,2] Older patients are most frequently affected with a female predilection.[1]

Cystadenoma is widely distributed among major and minor salivary glands. No large series of cyst adenoma with follow-up information has been reported. The likelihood of recurrence is low. A conservative surgical procedure ensures complete removal.[1] Recurrence may result from incomplete excision of the slow-growing cyst adenoma or when a cystadenocarcinoma is mis diagnosed. The use of a carbon dioxide laser has become common in oral and maxillofacial surgery for lesions of the oral mucosa. Advantages reported for the carbon dioxide laser in oral soft-tissue surgery include, minimal damage to adjacent tissue, good

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hemostasis, minimal inflammatory reaction, scar formation and precise cutting.

CASE REPORT

A 60-years-old female reported with an asymptomatic swelling in the right posterior palatal area in the region of missing 15,16 and 17 [Figure 1]. The swelling was present since several months and gradually increased to the presenting size. On examination, a well-defined, round mass, approximately 2 cm in diameter was revealed, which was soft to firm in consistency, non-movable, slightly compressible and painless. No carious tooth was found to be associated and there was no sign of tooth displacement. The mucosa over the swelling appeared normal.

A provisional clinical diagnosis of benign minor salivary tumor was given For differential diagnosis, pleomorphic adenoma, a low- grade minor salivary gland malignancy acinar cell adenocarcinoma (AcAC), adenoidcystic carcinoma, mucoepidermoid carcinoma), residual cyst, retention phenomenon, lipoma, neurofibroma and palatal odontogenic keratocyst were considered.

Patient was investigated with a panoramic and a maxillary occlusal radiograph, which showed no bony alteration in the area of the swelling. An intra-oral periapical radiograph was performed to rule out possible odontogenic origin. Fine needle aspiration cytology of the swelling yield a hemorrhagic aspirate of insignificant volume. H and E stained smear of the aspirate showed presence of mostly *red blood cells* and some chronic inflammatory cells. Routine hematological investigations were advised before the patient was referred to oral surgery for excision of the palatal mass.

The lesion was excised completely along with the nerves and vessels under local anesthesia by raising a mucosal flap [Figure 2a] and a single soft-tissue bit of about 2 cm × 1.6 cm [Figure 3] was sent for histopathological examination. Primary closure of the overlying mucosa was performed [Figure 2b] and the wound healed by primary intention without any intra-or post-operative complications [Figure 2c]. Periodontally compromised adjacent maxillary molar was later extracted.

The histopathological examination revealed numerous large and small cystic spaces interspersed in a fibrous connective tissue stroma [Figure 4]. The cystic lumens were lined by 1-3 cell layer thick epitheliums, which varied from flattened to cuboidal to tall columnar cells

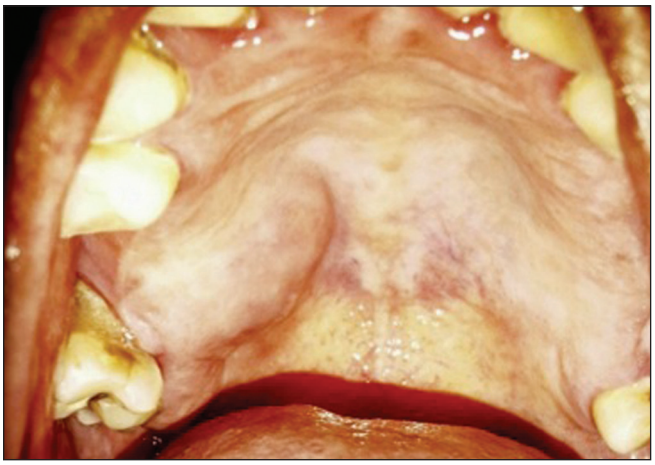

Figure 1: Swelling in the right posterior palatal region

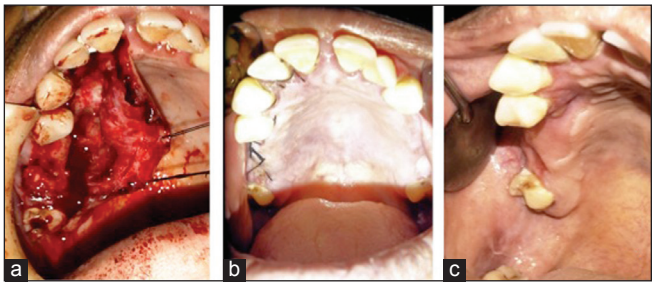

Figure 2: Surgical excision by raising a full thickness mucosal flap (a), Primary closure of the wound (b), Post-operative: Healing after 1 month (c)

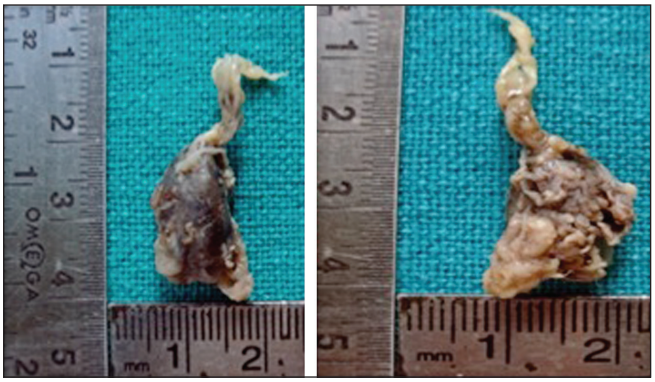

Figure 3: Soft-tissue bit sent for histopathology (approx. 1.6 cm × 2.0 cm)

[Figure 5]. The stroma exhibited multiple hemorrhagic areas and focal aggregates of chronic inflammatory cells along with few mucous cells. Normal appearing mucous acini could also be appreciated in the deeper sections [Figure 6]. Cellular atypia and mitosis were absent. A diagnosis of mucinous cyst adenoma was made. Patient is under follow-up for identification of any recurrence.

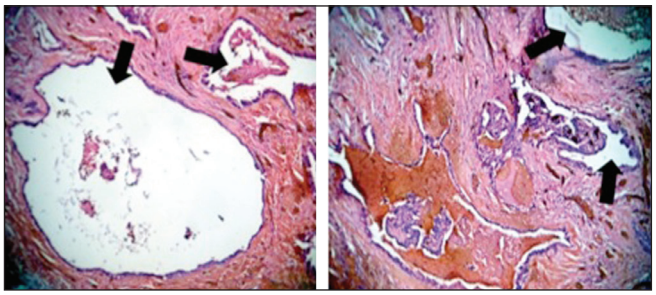

Figure 4: Photo micrograph depicting large and small cystic spaces (black arrows) in moderately fibrous connective tissue stroma along with numerous hemorrhagic areas (H and E, ×40)

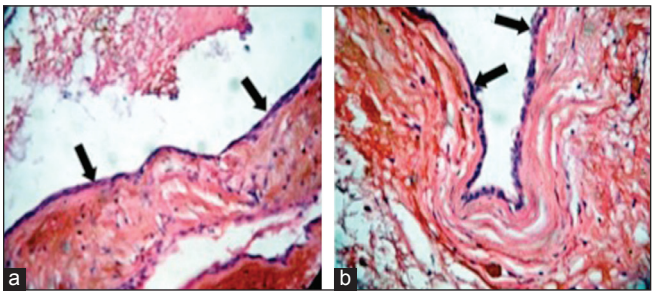

Figure 5: Photo micrograph depicting cystic lumen with flat (a) and cuboidal (b) epithelial lining (black arrows). (H and E, ×40)

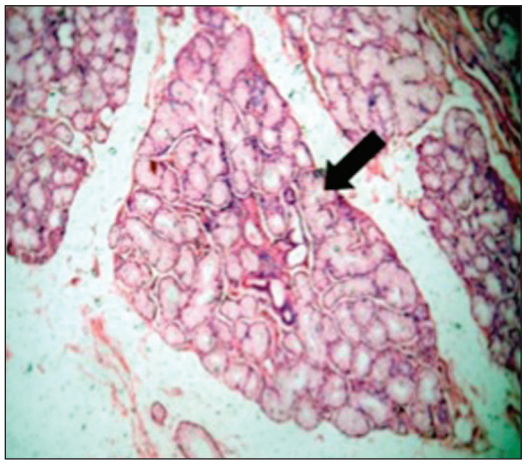

Figure 6a: Photo micrograph showing normal appearing mucous acini (black arrow) (H and E, ×100)

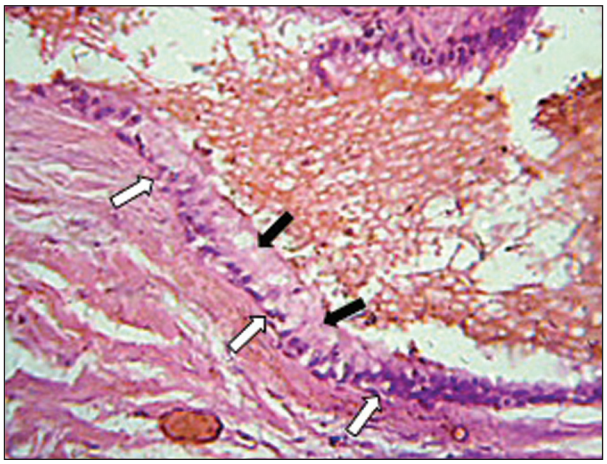

Figure 6b: Photo micrograph 6b showing layer of epithelial cells(white arrow) lining a cystic cavity with foamy mucous secretion on the surface (H and E, ×40)

DISCUSSION

Cyst adenoma is a rare benign neoplasm of epithelial origin found chiefly affecting the minor salivary glands. Although cases involving the major salivary glands have been reported.[3] WHO, in its revised classification in 1992, sub-classified cyst adenoma into two distinct histopathological varieties — mucinous and papillary.[4]

Alexis and Dembrow reported the incidence of cyst adenoma to be 2% of all minor salivary gland tumors and 4.7% of benign epithelial salivary gland tumors.[5] Waldron *et al*. reported a higher rate of occurrence of 4% and 8.1% of all the minor salivary gland tumors and of all benign epithelial salivary gland tumors respectively.[6] Chaudhry *et al*. reported 2% cases of cyst adenoma out of 800 intra oral minor salivary gland tumors.[7]

Bauer and Bauer suggested that the cyst adenoma arises principally from the undifferentiated epithelium of the intercalated ducts of the salivary gland.[8] They are characterized by multi cystic growth within a fibrous connective tissue stroma. The papillary variety of cyst adenoma exhibits papillary proliferations that project into the cystic lumens and may closely resemble the Warthin's tumor with the absence of the lymphoid element.[1] The less frequently seen mucinous variety predominantly shows mucous cells in the epithelial lining of the cystic lumens, with absence of distinctly visible papillary projections.

Cyst adenoma has been reported to occur in patients between 17 years and 86 years of age with a mean age of about 61years.[9] It frequently occurs in the 6th decade of life more frequently in females, with a female: Male ratio being 3:1.[1,2] The age of occurrence in our case is in terms with what has been reported in the literature. The most common site of occurrence is the palate, as described in our case, followed by the buccal mucosa and the tongue.[1,9-11] It has also been reported to occur on the upper and lower lip and floor of the mouth. [6,9,10,12] Mucinous cyst adenoma of minor salivary gland of the nasal fossa has been reported by Cano Cuenca *et al*.[13]

As such, cystadenoma presents no distinct clinical features and may be impossible to differentiate clinically from other benign salivary gland tumors,

mucous retention or extra vasations phenomena and low-grade minor salivary gland malignancies such as papillary cystic variant (PCV) of ACC, adenoid cystic carcinoma and muco epidermoid carcinoma.

Cyst adenoma must be carefully differentiated from ACC-PCV. Acinic cell carcinoma although is considered a low grade malignancy with a 5 year survival of 90%, but its papillary variety (ACC-PCV) has been reported to be universally fatal in 10 years.[14] ACC-PCV is usually encapsulated and frequently shows capsular invasion. Papillary folds are seen interspersed with cystic spaces, which may be small or large with well-defined papillary growths and glandular epithelial masses stranded by cords of fibro vascular tissue.[1,14] Multiple vacuolated and acinar cells may also be seen along with hob nailing or tomb stoning of luminal cells, which is characteristic of ACC-PCV.[14] However, the reported case is of the mucinous variety of cyst adenoma typically showing absence of papillary projections within the cystic lumen. Further, ACC-PCV is reported to occur in younger patients (16-40years),[15] contrary to our case of a 60-year-old female.

When cystadenoma occurs on the palate, such as in our case, it must be differentiated from radicular/residual cyst and palatal space abscess and a possible odontogenic origin must be ruled out by necessary radiographic examination.

Diagnosis of cystadenoma is strictly histopathological. The histopathological features described in our case were characteristic and in general agreement with what has been reported in the literature for cystadenoma.[6,16] 25% cases of cystadenoma have been reported to show a distinct fibrous capsule,[6] which was not present in our case. Since salivary gland tumors show great histomorphological diversity, differential diagnosis of cystadenoma must include intra ductal papilloma, cystadeno carcinoma, low-grade mucoepidermoid carcinoma and Warthin's Tumor.[2]

CONCLUSION

Most cases of cystadenomas, including our case, are treated by simple surgical excision.[10,12,16] Regular follow-up of the patient is necessary for early identification of recurrence, which may occur due to incomplete excision. To the best of authors 'knowledge, very few cases have been reported of mucinous variety of cystadenoma and that too at extra oral sites. The aim of this case report is to document the rarely reported mucinous cystadenoma and may be the first reported mucinous cystadenoma in the oral cavity among the Indian population.

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How to cite this article: Rai S, Rana AS, Gupta V, Jain G, Prabhat M. Mucinous cystadenoma: A rare entity. Dent Res J 2013;10:685-8.

Source of Support: Nil, **Conflict of Interest:** None declared.