

PAPILLARY CYSTADENOMA OF PALATE: A CASE REPORT

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ABSTRACT

A case report of the papillary cystadenoma from minor salivary gland in palate of a 52-year-old man is described.

INTRODUCTION

Papillary cystadenoma of salivary glands is an uncommon benign neoplasm. In two large reviews, it constituted only 2% and 4.7%, respectively, of all minor salivary gland neoplasms, and 4% and 8.1%, respectively, of all benign epithelial minor salivary gland neoplasms.^{1,2} Cystadenoma of salivary glands is an uncommon benign neoplasm that presents intraluminal papillary projections.

The most frequent locations for this tumor in the oral cavity are: hard palate, cheek, and posterior region of the tongue.⁴ This lesion has a tendency to recur if not appropriately excised⁵. There are case reports of papillary cystadenoma of the oral region^{3,5,6,7,8,9,10}, however, Papillary Cystadenoma arising from a minor salivary gland is rare.^{11,12}

CASE REPORT

A 52-year-old man presented with an asymptomatic mass of the hard palate that had been present for 2 years. Clinical examination revealed a 3-cm-diameter, well-circumscribed, solid, round mass that was not tender (Figure 1). A CT scan showed a soft tissue swelling of the palate with small extension into antrum but without erosion of antral floor or bony involvement (Fig 8, 9). The orthopantomogram and blood

tests were non contributory. A provisional clinical diagnosis of pleomorphic adenoma was made. Incisional biopsy was done under local anesthesia. The histopathological diagnosis was suggestive of pleomorphic adenoma.

The histological examination showed epithelial and mesenchymal components. The epithelial component was composed predominantly of proliferating ductal and myoepithelial cells. Some of the epithelial cells showed typical intercellular bridges. Few mucous cells and plasmacytoid cells were also seen. Numerous cystic spaces containing eosinophilic coagulum and mucoid material were present. The connective tissue stroma was fibrous in nature showing areas which are myxomatous in appearance and hyalinised areas of hemorrhage. The diagnosis was suggestive of Pleomorphic Adenoma.

It was planned to do total excision. Under general anaesthesia with nasotracheal intubation the procedure was performed uneventfully. A clear acrylic surgical obturator, fabricated on a cast made prior to surgery, was given. The post excision biopsy report came as papillary cystadenoma (Fig 2, 3, 4). The patient was followed up for ten months and there was no evidence of recurrence (Fig 5).

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The histologic examination showed the tumour mass to be encapsulated by thick fibrous connective tissue and arranged in the form of lobules, sheets and ductal pattern. The mass was also composed of numerous cystic spaces lined by epithelium which are thrown into papillary projection and in few areas they were surrounded by thickened basement membrane. The lining epithelium comprised of cuboidal ductal cells with vesicular nuclei and showed no atypia. Thin fibre connective tissue core admixed with few areas of haemorrhage and hyalinization were seen. The capsule was infiltrated in few areas by the ductal pattern of tumour mass. Final diagnosis was confirmative of papillary cyst adenoma of the minor salivary gland (Fig 8, 9).

DISCUSSION

The World Health Organization (WHO)¹³ described Papillary Cystadenoma as “a tumor that closely resembles Warthin tumor but without the lymphoid

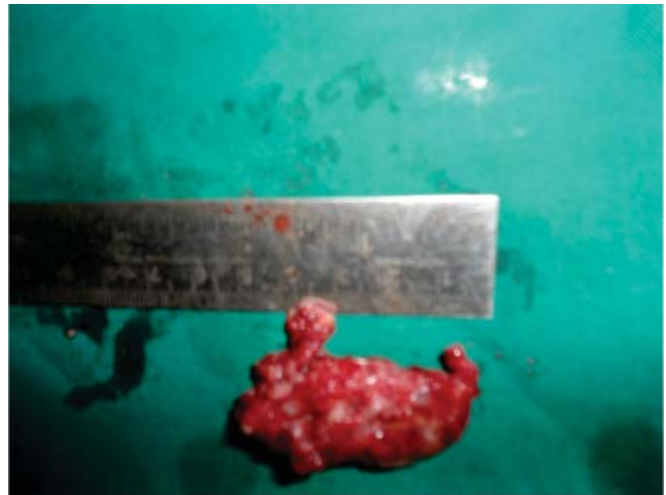


Fig. 3



Fig. 4



Fig. 1



Fig. 2

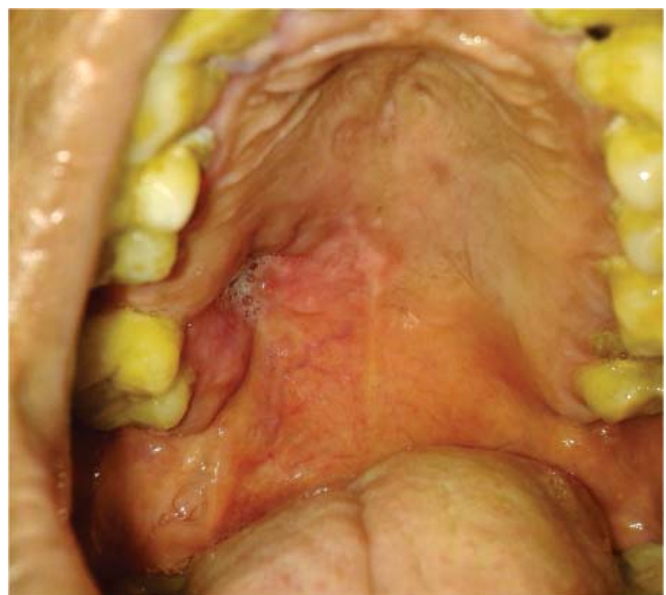


Fig. 5

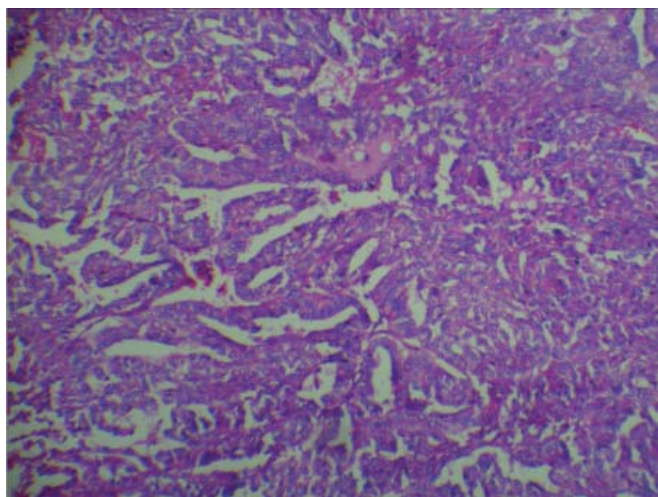


Fig. 6



Fig. 9

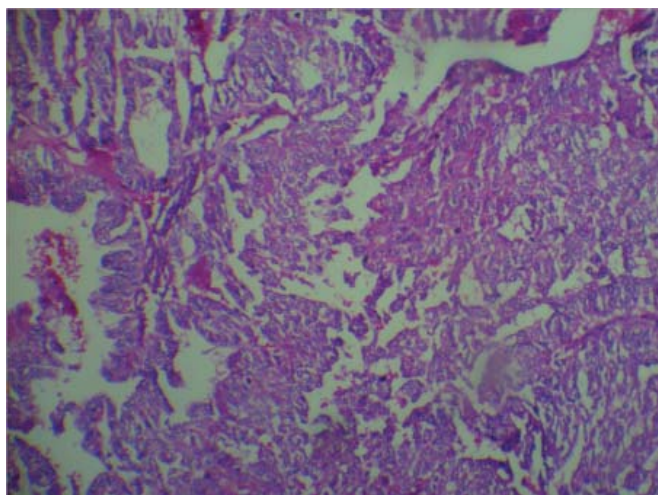


Fig. 7



Fig. 8

elements, constituting multiple papillary projections and a greater variety of epithelial lining cells.” It is believed that salivary glands tumors are difficult to diagnose or interpret because there are many possible patterns of presentation. In addition, papillary cystadenoma of the minor salivary gland is rare.^{11,12}

First review of the clinical, histologic, and biologic features of the papillary cystadenoma¹⁴ show that it appears to occur more frequently in women, most patients older than 50 years of age, with several in their seventies. The most common sites are the palate and buccal mucosa; however, tumors in the lip and tongue also have been described. The usual presentation is an asymptomatic mass.

In a case series of 834 salivary gland tumors reported from their hospital from 1980 to 2004, only two cases of cystadenoma of a minor salivary gland were found, in the upper lip. On microscopic examination, the neoplasm is usually well circumscribed and may be surrounded by a rim of fibrous tissue. There are solid areas (usually limited in extent) and cystic areas into which project papillae lined by cuboidal to columnar cells usually two layers thick. The cells usually have eosinophilic cytoplasm, and goblet cells may be present.^{16,12,11}

Perhaps the most important entity in the differential diagnosis of papillary cystadenoma is cystadenocarcinoma; sometimes the distinction may be difficult because the neoplasms have similar architecture, and also because cystadenocarcinoma often shows little atypia.¹⁶ Both neoplasms usually reveal papillary proliferation of the epithelial lining and are composed of

cells that possess bland cytomorphic features. Differentiation of tumour types depends largely on the identification of actual infiltration of salivary gland parenchyma or surrounding connective tissue by either cystic or solid epithelium in cystadenocarcinomas. Step sections of a borderline tumour may yield unequivocal evidence of invasion.¹⁵ Most recent report of a case involving cytological analysis and review of the literature, reported that in the authors' best knowledge, theirs was only the 12th case of this tumor seen in the palate.¹⁷

CONCLUSION

Conservative surgical removal is the treatment of choice for Papillary Cystadenoma of minor salivary gland and these lesions rarely recur.

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