Case Report

Indian Journal of Pathology: Research and Practice
Volume 5 Number 2, May - August 2016
DOI: http://dx.doi.org/10.21088/ijprp.2278.148X.5216.28

Sialadenoma Papilliferum of Buccal Mucosa : A Rare and Distinct Entity

Sadhana H. Khaparde*, Sanjay D. Deshmukh, Baba B. Shinde**, Prajacta Rane***, Shaleen Lamba******

*Professor and Head, **Professor ***Junior Resident, ****Senior Resident, Department of Pathology, Padmashree Dr. Vithalrao Vikhe Patil Foundations Medical College and Hospital, Ahmednagar, Maharashtra, India. 414111.

Abstract

Sialadenoma papilliferum (SP) is an unusual, benign neoplasm of predominately minor salivary gland which presents as an exophytic papillary lesion on oral of the mucosa. This lesion is included in the group of the ductal papillomas and accouns for less than 1% of minor salivary glands tumors .The present case of SP was observed in 66 year old male patient on left buccal mucosa.

Keywords: Salivary Gland; Buccal Mucosa; Ductal Tumor.

Introduction

Sialadenoma Papilliferum (SP) is a rare, distinctive benign tumour of salivary gland classified under the Ductal Papillomas by WHO [1]. This lesion is encountered in adults with mean age of 59.2 years and shows male preponderance, commonly involving minor salivary glands with the size ranging from 0.3–2 cm. In Indian literature this lesion is seldom documented [2]. We encountered a case of SP on buccal mucosa with distinct histomorphological features, which has prompted us to report this case.

Case Report

A 66 year old male, patient presented with complaint of swelling on the left buccal mucosa along the line of occlusion of second mandibular molar tooth. The patient had noticed small nodular mass 4 months back which was painless. However, it became painful since last 3 days. On clinical examination there was intraoral sessile nodular mass measuring 1.5 x 0.8 cm. The surface was rough and the swelling was tender on palpation. The lesion was provisionally diagnosed as benign minor salivary gland tumor and wide local excision under local anesthesia was done and submitted for histopathological examination.

On gross examination, partly nodular and partly cystic grayish-white lesion with papillary surface was noted, measuring 2 x 1.5 cm.

Histopathology

Multiple sections studied showed an unencapsulated lesion with biphasic pattern composed of squamous epithelial component and juxtaposed glandular component [Figure1]. The glandular component showed, surface papillary structures and deeper down variably dilated ducts. Surface stratified squamous epithelium revealed acanthosis and mild parakeratosis. Also seen were papillary structures overlined by double epithelial lining i.e. basal layer of cuboidal cells and surface lining of low columnar cells [Figure 2]. Beneath this ectactic ducts with variation in size and shape and lined by double layer with surrounded by mucinous minor salivary gland were noted [Figure 3]. In few of the glands there was marked basal cell hyperplasia with multilayering [Figure 4]. The stroma showed diffuse and dense infiltration by eosinophils, polymorphs, mucinous macrophages and few lymphoplasmacytic cells [Figure 5]. Based on these findings the diagnosis of Sialadenoma Papilliferum was arrived .

Corresponding Author: Sanjay D. Deshmukh, Professor, Department of Pathology, Padmashree Dr. Vithalrao Vikhe Patil Foundations Medical College and Hospital, Ahmednagar, Maharashtra, India. 414111.
E-mail: drsanjay123in@yahoo.co.in

Follow up

Patient was followed every month for 6 months. He was asymptomatic without any recurrence.

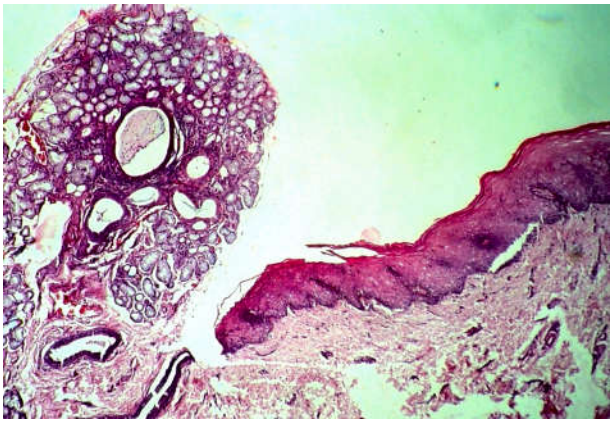

Fig. 1: Photomicrograph low power view showing biphasic pattern of squamous epithelium and salivary glandular component. (H&E stain, x40)

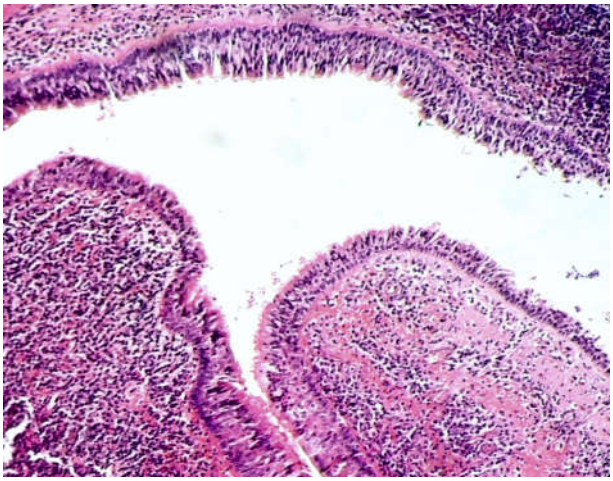

Fig. 2: Photomicrograph showing Papillary folds lined by basal layer of cuboidal cells and luminal lining of Columnar cells. (H&E stain, x100)

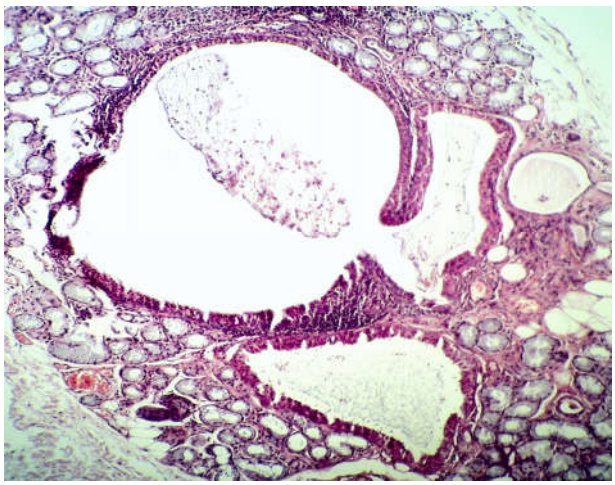

Fig. 3: Photomicrograph showing minor salivary gland with ectactic ducts lined by double layer and containing mucinous material. (H&E stain, x40)

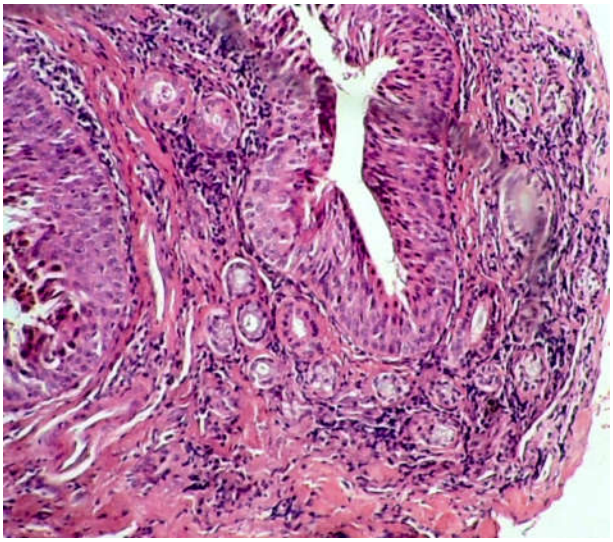

Fig. 4: Photomicrograph showing high power view of basal cell hyperplasia.(H&E stain,x400)

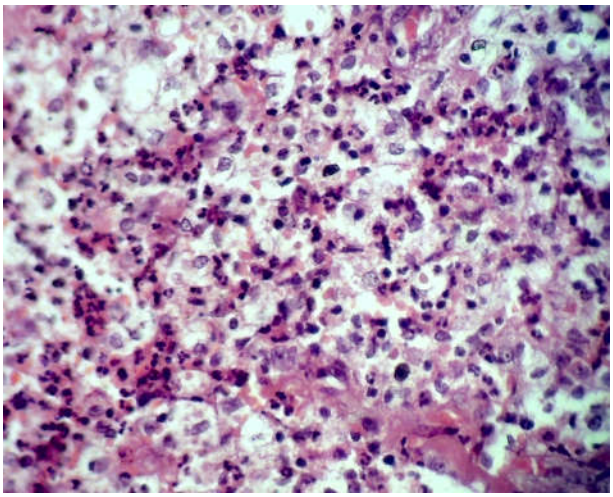

Fig. 5: Photomicrograph showing stromal infiltrate by eosinophils, polymorphs and mucinous macrophages(H&E stain, x400)

Discussion

Since the lesion of Sialadenoma Papilliferum was first described by Abrams and Finck in 1969, the histogenesis has been controversial [3]. However, WHO has given specific definition that Sialadenoma Papilliferum is an exophytic papillary and endophytic proliferation of mucosal surface and salivary duct epithelium and hence included in the lesions of ductal system of salivary glands [1]. Reviewing the literature, SP occurs in minor salivary glands of palate, upper lip, buccal mucosa and rarely in parotid gland [2,4]. In the present case the lesion was located on the left buccal mucosa along the occlusion line at level of second mandibuar molar tooth. We observed biphasic proliferation of squamous and ductal epithelium so

also, ectactic ducts lined by double layer of luminal columnar cells and basal cuboidal cells. Distinct proliferation and multilayering of cuboidal cells was noted. The deeper tissue showed dense and diffuse acute on chronic cell infiltration, this may be attributed to trauma during mastication, as the lesion was located at the closure line in our case.

By and large, SP behaves in benign fashion. However, lack of capsule coupled with proliferating ductal components may lead to mistaken diagnosis of malignancy. The differtial diagnosis includes lesions like: Inverted ductal papilloma; mucoepidermoid carcinoma; sqamous papilloma and intraductal papilloma [1]. Inverted ductal papilloma, shows well circumscribed and cystic lesion. The papillary structures extend into lumen; however they lack glandular complexity seen in SP. The distinctive squamous element as seen in SP is also absent. Mucoepidermoid carcinoma is characterized by squamous i.e epidermoid, mucous producing cells and cells of intermediate type. In addition clear, columnar and/or oncocytic cells may be present.

Our case on follw up did not show recurrence. It must be noted that rare cases of recurrence and even malignant transformation have been documented [5].

Our experience with case adds to cumulative knowledge of this rare lesion.

References

1. Barnes L, Eveson JW, Reichart P, Sidransky D.10th ed. Lyon:IARC Press; World Health Organization(WHO) Classification of Tumors, Pathology and genetics of head and neck tumors. 2005; P271-2.
2. Anuradha A, Ram Prasad VV, Kashyap B, Srinivas V.Sialadenoma papilliferum: clinical misdiagnosis with a histological decree.2012 Case Rep Dent. 2012; 2012: 356271. [PubMed].
3. N.Abrams AM, Finck FM: Sialadenoma papilliferum. A previously unreported salivary tumor.Cancer. 1969; 24: 1057–63[PubMed].
4. Kubota Y, Kubo C, Mori Y: Sialadenoma papilliferum of the hard palate: A case report. J Oral Maxillofac Surg. 2012; 70(7): 1609–12 [PubMed].
5. Liu W, Gnepp DR, Vries E et al: Mucoepidermoid carcinoma arising in a background of sialadenoma papilliferum: A case report. Head Neck Pathol. 2009; 3: 59–62 [PubMed].