IOSR Journal of Dental and Medical Sciences (IOSR-JDMS)
e-ISSN: 2279-0853, p-ISSN: 2279-0861.Volume 14, Issue 5 Ver. III (May. 2015), PP 63-65
www.iosrjournals.org

Sclerosing Polycystic Adenosis of Submandibular Gland- A Rare Case Report

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Abstract*: Sclerosing polycystic adenosis (SPA) is a rare neoplasm of salivary glands, with a striking resemblance with benign fibrocystic disease of breast. Most common site of SPA is parotid gland; however cases in submandibular gland, minor salivary gland, and buccal mucosa have been reported. Generally SPA does not show any gender predilection and occurs over wide range of age (9-84 yrs). SPA is mostly unifocal, but cases with multifocality have been reported. Here we describe SPA of submandibular gland in a 55 years old woman. The exact nature of the tumor is unknown but until recently it has been believed to be a pseudoneoplastic reactive and inflammatory sclerosing process. Treatment is effected by localized surgical excision. Recurrence rate has been reported in 19% cases.*

Key Words- *Sclerosing polycystic adenosis, submandibular gland*.

I. Introduction

Smith et al in the year 1996 first described about sclerosing polycystic adenosis in a series of 9 cases.[1]SPA is a rare tumorous condition of salivary gland that has histopathological similarities with fibrocystic disease / sclerosing adenosis of mammary gland. Since then until today approximately over 50 cases have been published.[2] These tumor is a slow growing tumor of the salivary gland, mostly unencapsulated showing tubuloacinar adenosis with dilated ducts, apocrine metaplasia, epithelial hyperplasia and cystic changes associated with fibrosis.[3-6]

II. Case Report

Clinical findings

A 55 year old Indian female referred to Department of Surgery, Kamineni Institute of Medical Sciences with chief complains of painless swelling over right submandibular gland for last 4 months. Her past medical history was unremarkable, vitals were stable, no history of addiction. On examination a soft to firm mass noted over right submandibular gland, which is well defined, non tender, immobile, with no ulceration of surface mucosa or overlying skin. Radiological examination reveals benign cystic lesion of salivary gland. Surgical excision was done and the specimen sent to the Pathology department of Kamineni Institute of Medical Sciences.

III. Pathological Findings

Gross pathology:

We received a soft to firm well circumscribed cystic mass 5.5 × 4 × 3.5 cms. Outer surface is smooth, grey white in colour. Cut surface of the lesion show a cyst (Fig-1) measuring 2.8 × ×1.5 cms filled with clear fluid which is drained out, wall thickness of the cyst wall -3mms, along with solid grey white area measuring 4.2 × 2.8 cms. (Fig-2)

Histopathology:

Histopathology shows well circumscribed partially encapsulated lesion with adjacent normal salivary gland (Fig-3). Within the lesion there are islands of epithelial cells arranged in lobular pattern along with sclerotic stroma and cystically dilated glands which are more prominent in the centre of lesion (Fig-4). Within the lobule there are multiple ducts, glands, acinar structure and some solid aggregates of epithelial cells with foci of clear cell changes. Cystically dilated glands are lined by flattened to cuboidal epithelium with foci of stratification and foamy changes are observed. Sclerosing stroma shows lymphohistiocytic collections. There is no histological feature of malignancy noted in the studied sections.

IV. Discussion

Sclerosing polycystic adenosis is a very rare case having similarity with fibrocystic disease of breast. It occurs most commonly in parotid gland (80.5%), submandibular gland (7.3%) and minor salivary glands (12.2%). Among the minor salivary glands commonly involved sites are hard palate, floor of mouth and buccal mucosa. Sclerosing polycystic adenosis in the major salivary glands usually present with slow growing, deep seated, rounded, palpable mass with or without pain and tenderness whereas in minor salivary gland it usually present with asymptomatic freely mobile nodule with white, creamy or yellow in colour.[7] Histologically sclerosing polycystic adenosis is a sharply circumscribed, poorly encapsulated with acinar and ductal components with more prominent fibrosis is noted. Other feature like apocrine like metaplasia, cribriform pattern, clear cell changes, squamous and oncocytic like changes are observed.[1,7] Histochemically sclerosing polycystic adenosis is PAS positive with resistance to diastase digestion. Immunohistochemically epithelial cells are positive for AE1/AE3, CAM 5.2, EMA, focal estrogen progesterone positivity and myoeithelial cells are positive for smooth muscle actin, S100.[3,7]

All reported cases of sclerosing polycystic adenosis are benign and cured by localized surgical excision however Gnepp et al reported recurrence rate of 19% in their study.[8] However recurrence is mostly due to inadequate surgical excision and multifocality of the tumor compared to true recurrence. Prognosis of sclerosing polycystic adenosis is favorable, with no reported death so far.

The pathogenesis of sclerosing polycystic adenosis is uncertain. Most of the histological feature such as sclerotic fibrosis, epithelial proliferation point towards reactive post inflammatory process. However mild to moderate cellular atypia and carcinoma in situ are observed by some authors.[5,9]

V. Conclusion-

Sclerosing polycystic adenosis is a benign tumor with positive outcome but high incidence of recurrence and dysplasia, carcinoma in situ suggests that possible low grade malignant potential of sclerosing polycystic adenosis is yet to be possible.

Reference

[1]. Smith BC, Ellis GL, Slater LJ, Foss RD. Sclerosing polycystic adenosis of major salivary glands. A clinicopathological analysis of nine cases. Am J Surg Pathol 1996;20;161-70.

[2]. Fredrick Peterson. Sclerosing Polycystic Adenosis of Salivary Glands: A Review with Some Emphasis on Intraductal Epithelial Proliferations. Head neck pathol.2013;Jul 7 (suppl 1): 97-106.

[3]. Bin Bin Li, Tie Jun Li. Sclerosing Polycystic Adenosis of the Sublingual Gland:a Case Report.The Chiness journal of Dental Research.2008;11(2):134-36.

[4]. Noonan VL, Kalmar JR, Allen CM, Gallagher GT, Kabani S. Sclerosing polycystic adenosis of minor salivary glands: report of three cases and review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2007;104:516-20.

[5]. Skálová A, Michal M, Simpson RH, Stárek I, Prádná J, Pfaltz M. Sclerosing polycystic adenosis of parotid gland with dysplasia and ductal carcinoma in situ. Report of three cases with immunohistochemical and ultrastructural examination. Virchows Arch 2002;440;29-35.

[6]. Bharadwaj G, Nawroz I, O'Regan B. Sclerosing polycystic adenosis of the parotid gland. Br J Oral Maxillofac Surg 2007;45;74-6.

[7]. Shabnum Meer, Mario Altini. Sclerosing Polycystic Adenosis of the Buccal Mucosa. Head Neck Pathol. 2008 Mar; 2(1): 31–35.

[8]. Gnepp DR. Sclerosing polycystic adenosis: a lesion that may be associated with dysplasia and carcinoma in situ. Adv Anat Pathol.2003(10):218-22.

[9]. Gnepp DR, Wang LJ, Brandwein-Gensler M, Slootweg P, Gill M, Hille J. Sclerosing polycystic adenosis of salivary gland: a report of 16 cases. Am J Surg Pathol 2006;30:154-64.

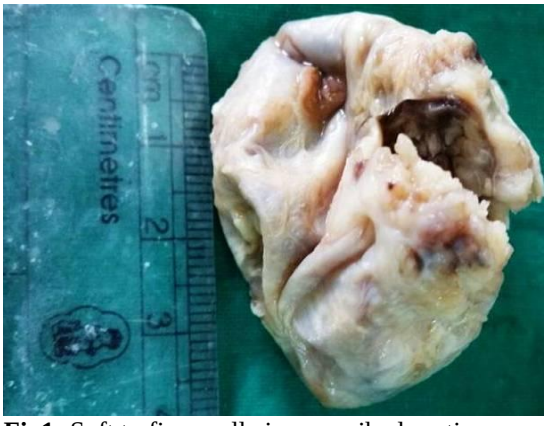

Fig1- Soft to firm well circumscribed cystic mass.

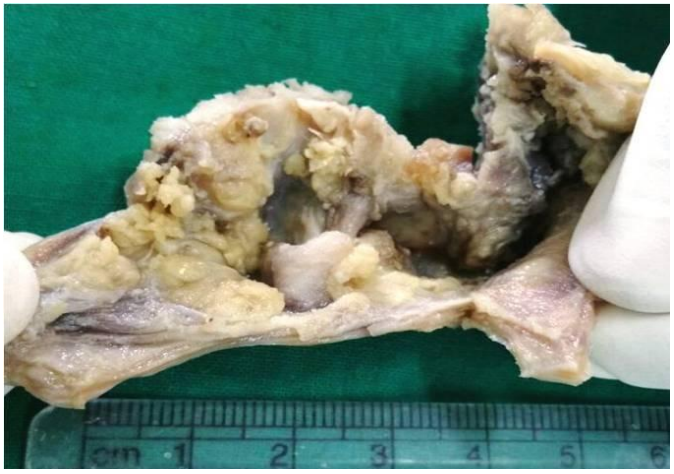

Fig2- Solid grey white mass with cyst wall.

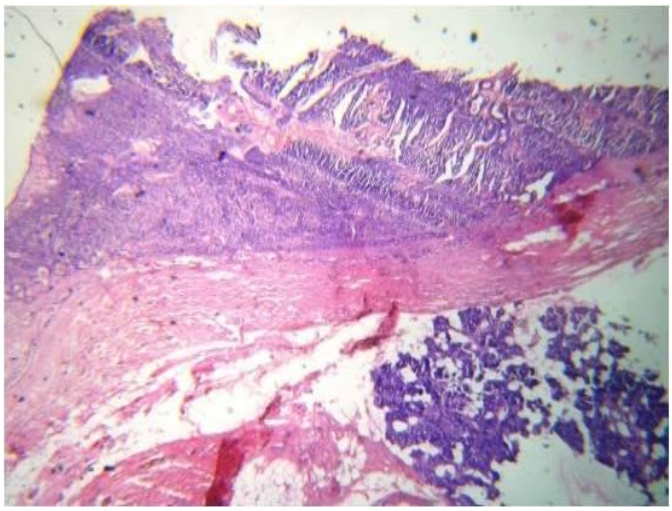

Fig3- Encapsulated tumor, normal salivary tissue with sclerotic stroma.

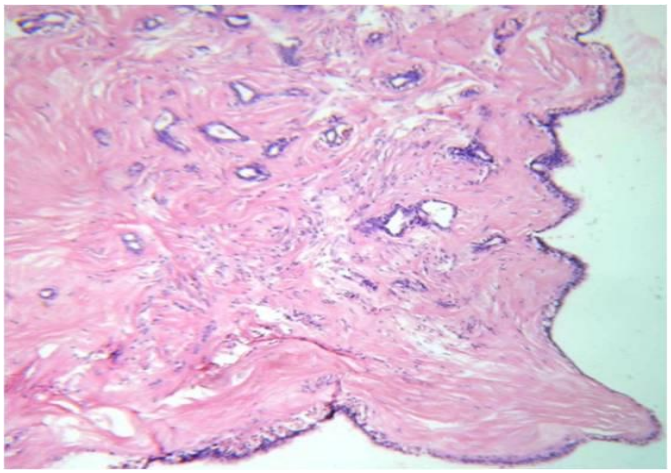

Fig4- Cyst wall , sclerotic stroma and many cystically dilated glands.