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Myoepithelial dilemma of the hard palate: A case report and literature update.

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Abstract: *Salivary gland tumours constitute about less than 4% of all head and neck tumours and are commonly seen in adults[3].Myoepithelioma is a rare benign neoplasm of salivary glands, which accounts for less than 1% of all salivary gland neoplasms. It is more common in parotid glands followed by small salivary glands.Myoepithelioma once was considered as extreme variant of pleomorphic adenoma, but now many authors consider the myoepithelioma as distinct pathological entity as it is composed entirely of myoepithelial cells and behaves more aggressive than pleomorphic adenoma (PA). Even though these tumours are painless and slow growing ones, it is important to identify these cases rather early and extirpate them totally[16]. This case report discusses a case of myoepitheliomain male patient aged about 53 years old on the hard palate with a review of corresponding world literature and surgical treatment.*

Keywords: *Myoepithelioma, Minor salivary gland, palate.*

I. Introduction

Myoepithelioma is believed to be rare entity of all kinds of tumours of salivary glands. It was first described by Sheldon way back in 1943, when it was considered as variant of Pleomorphic adenoma[16].

Myoepithelial cells are contractile cells derived from ectoderm and are seen in major and minor salivary glands, lacrimal glands, breast, sweat glands and prostate [1] . These cells are thought to be responsible for expulsion and propagation of secretions from the acini and through the ductal network of these tissues.

These cells may assume several distinct forms- as a spindled, plasmacytoid, clear, stellate or a basket-like cell. The tumours arising exclusively from these cells are rare and account for less than 1% of all salivary gland tumours. Most of these tumours have been reported in the parotid gland and minor salivary glands on the hard palate. Although, clear cell myoepitheliomas have been reported at other sites, most of the cases of palatal myoepitheliomas have been plasmacytoid or spindle cell type. We did not find any report of a clear cell myoepithelioma of the hard palate in the review of medical literature[17]. The extreme rarity of such an occurrence prompted us to report this case.

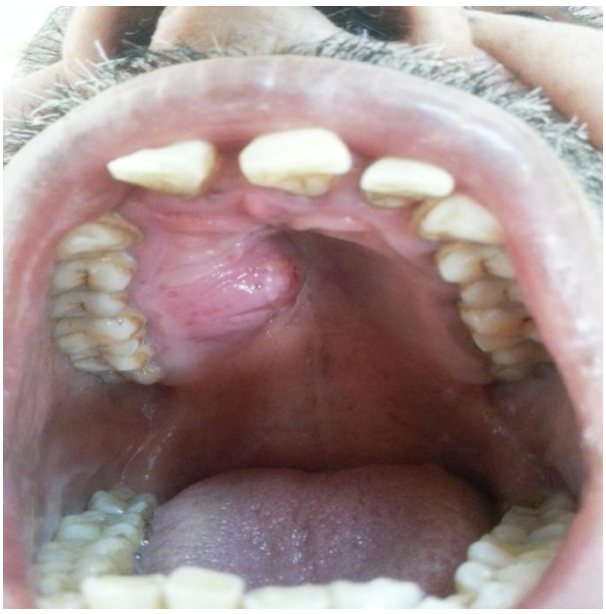
II. Case Report:-

A 53 year old male patient reported to the Department of Oral Medicine and Radiology with the chief complaint of an asymptomatic swelling over the right palatal region noticed six months back. The swelling was not interfering with the mastication or any other function. No associated constitutional symptoms were notedthe Past medical, dental, family and social history were not relevant. The patient gave a history of tobacco chewing habit since twenty years with frequency of six times a day and used to place it in his right and left buccal vestibule for 10-15 minutes and then spited out. On general examination, it was found that the patient was of moderate built and height. His vital signs were within normal range and review of systems revealed no abnormality.

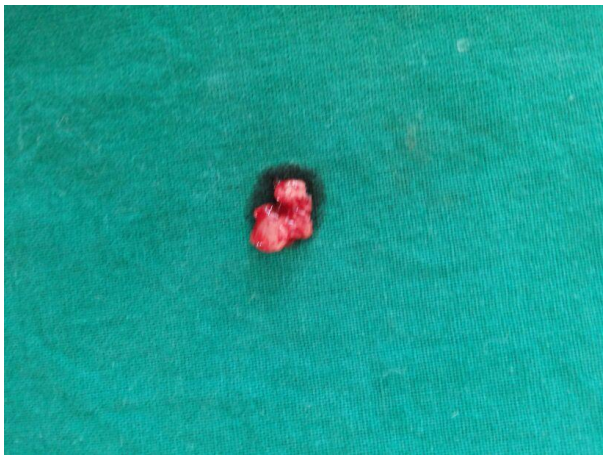
On extra oral examination, there was no facial asymmetry seen and lymph nodes examination showed no abnormality, intra-oral examination revealed deep periodontal pocket in relation to the mid palatal aspect of 16 with tendor on horizontal percussion associated with grade 1 mobility and a single, well demarcated, dome shaped swelling which approximately measured 1×1.5 cm and extendinganterio posteriorly from the imaginary line drawn from the mesial aspect of 16 and posteriorly extending up to an imaginary line drawn at the distal aspect of 17 and mesio laterally extending approximately 6-7mm from the marginal gingival in relation to the right first molar and second molar region, which did not cross the midline. The overlying mucosa was not ulcerated and was mobile over the swelling. On palpation, the swelling was found to be uniformly soft in consistency, non-tender and did not show any pus discharge. Hard tissue examination revealed no displacement or mobility of teeth however. On the basis of the history and the clinical examination, a provisional diagnosis of benign tumour of the minor salivary gland was made and a differential diagnosis of periodontalabscess, periapical abscess, infected radicular cyst, soft tissue tumours, malignant tumour of the minor salivary gland was considered.

The cross sectional maxillary occlusal projection radiograph was taken which did not show any bony invasion. A incision biopsy was done of the mass under local anaesthesia and the histopathological report of the biopsy specimen confirmed the diagnosis of pleomorphic adenoma.

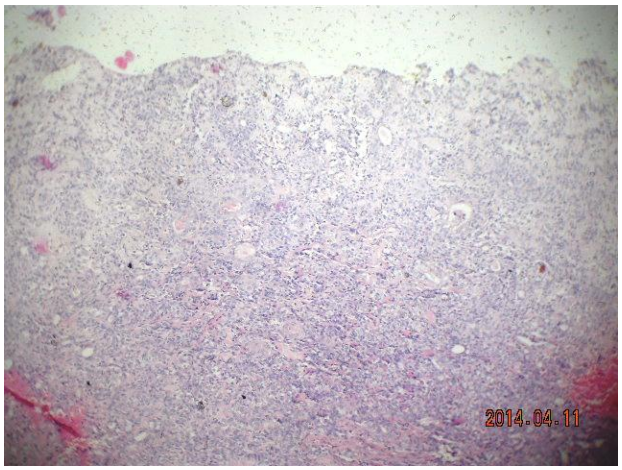
The patient was operated, under general anaesthesia, Wide excision of tumour with safe margin and drilling of under surface bone was done to avoid recurrence. There was no complication postoperatively, the palatal defect after the wide excision of the tumour mass was covered with a palatal obturator, for re confirmation, the whole excised lesion was again subjected to histopathological examination and diagnosed as myoepithelioma of a minor salivary gland in the hard palate, on regular follow up we did not find recurrence till date and this caused a diagnostic dilemma



A single dome shaped swelling approximately measuring 1 x 1 cm, extending 6 mm from marginal gingival in relation to right first molar to right second molar region, not crossing midline.[picture no 1]



Incisional biopsy done.
[Picture no 2]



Histopathological view. [Picture no 3]

III. Discussion:

Myoepitheliomas account for less than 1.5% of all salivary gland tumours. A review of the literature through 1993 yielded approximately 100 cases, this tumour appears to be rare, and this contrast with the active role of myoepithelial cells in the histogenesis of several type of salivary gland tumours.

The most common location of myoepithelioma of the head and neck are the parotid gland (40%) and the palate (21%). The age and sex distribution of myoepitheliomas is similar to that of mixed tumours. There are no distinctive clinical features and is similar to most other salivary gland tumors, myoepitheliomas present as asymptomatic, slowly growing masses[24].

Myoepithelioma is rare benign neoplasm of salivary glands. Among its four sub-types i.e. i)Spindle cell ii)plasmacytoidcell iii) polygonal epitheloid cell and iv)clear cell types spindle cell type is more common (seen in 70% cases) where as plasmacytoid cell type seen in only 20% cases[16].

Tumours arising from minor salivary glands of the palate may exhibit an overlap of clinical and biologic features which produce diagnostic and therapeutic dilemmas. Morphologic diversity is the hallmark of salivary gland adenoma, and considerable morphologic variation is often present within a tumor[9].Myoepithelioma which is involving minor salivary glands in the palate are mostly painless and also slow growing ones, and sometimes the growth rate could be fast. Rapid increase in size of the mass should lead to suspicion of intra lesion bleed / malignant transformation. The term myoepitheliomais used to indicate the histological presence of both epithelial and mesenchymal tissues.

Histological features ofmyoepithelioma

1) The abundance of cytoplasmic myofibrils.
2) In addition, pinocytotic vesicles, cell junctions, remnants of basement membrane, microvilli, nuclear indentation, and widely distributed glycogen and endoplasmic reticulum are ultra-structural features previously shown to be characteristic of myoepithelial cells[27].

Histological features of pleomorphic adenoma:

1. Islands of spindle cells over myxoid background
2. Inner layer of epithelial cells
3. Outer layer of myoepithelial cells

An interesting aspect about myoepitheliomas is the scarce information we have about its biological behaviour, because of its low incidence rates. Some authors consider it to be more aggressive than pleomorphic adenomas, while another investigation noticed, through PCNA expression, that there were no differences as far as proliferative activity is concerned between myoepitheliomas and pleomorphic adenomas[25].

These tumours are encapsulated and hence complete removal ensures cure. Care should be taken to leave at least 1mm margins around the lesion. While removing the mass, rupture of the capsule is to be avoided to minimize recurrence[3].

Several investigators have shown that myoepithelial cells are a component of most mixed tumor of salivary glands and some suggest that the mesenchymal-appearing components of these lesion are attributable to the myoepithelial cells. In addition, there are reports of pure myoepithelial neoplasms, several of which have been described in minor salivary glands.

In an analysis done by Sciubbaand Brannonon 23 cases, there was no differences in sex distribution evident, men and women were equally affected. Age ranges were similar to more studies those previously reported, although this series noted an older average age **(62.9** years vs. 33.3 years). Overall anatomic distribution in the current series **is** similar to the previously reported cases although their literature states that the parotid gland was by far the most frequently affected site.

Recurrences are rare. According to Sciubba& Brannon who could follow 16 cases out of 23 over a period of 1 year found recurrence only in one case(3). Recurrences can be picked up by regular follow up[16], similarly our case also did not show any recurrence till date which is kept on regular follow up.

IV. Conclusion:

Myoepithelioma of palatal minor salivary gland is a rare entity. It is relatively more aggressive than other benign neoplasms of salivary glands,both the radiologist, oral physician and the surgeon need to be aware of its different presentation as it caninfluence both radiological diagnostic assessment and treatment. Imaging should be considered in ruling out bone erosions in these patients. Complete extirpation of the mass is curative,one word of caution is that the capsule should not be breached, when attempting to surgically excise the mass because the breach of capsule is associated with increasing recurrence rates.

References:

[1]. Pleomorphic adenoma of the hard palate; Gupta M, et al. BMJ Case Rep 2013. doi: 10.1136/bcr-2013-008969.
[2]. Pleomorphic Adenoma of the Palate; Amit Kumar Gothwal et al. Journal of Clinical and Diagnostic Research. 2012 August Vol-6(6): 1109-1111.
[3]. Pleomorphic adenoma hard palate a case report and literature review– Professor Balasubramanian Thiagarajan Balasubramanian Thiagarajan; ENT SCHOLAR;March 18, 2013; Rhinology.
[4]. Pleomorphic adenoma (benign mixed tumour) of the salivary glands: its diverse clinical, radiological, and histopathological presentation; R.K. Lingam et al. / British Journal of Oral and Maxillofacial Surgery 49 (2011) 14–20.
[5]. Myoepithelial carcinoma ex pleomorphic adenoma of salivary glands: A problematic diagnosisSaid and Campana; Oral Surg Oral Med Oral Pathol Oral RadiolEndod; 2005; 99:196-201.
[6]. Carcinoma ex pleomorphic adenoma: Immunoprofile of the cytoskeleton proteins.S. Sousa, A. Altemani, M. Martins, N. Arau´ jo, V. Arau´ jo, University of Sa˜ o Paulo, University of Campinas, and Study CenterSa˜o Leopoldo Mandi, Brazil; OOOOE Volume 98, Number 2.
[7]. Pleomorphic adenoma of the palate: a case report and review of the literature;MansurRahnama, UrszulaOrzędała-Koszel, ŁukaszCzupkałło,corresponding author and MichałŁobacz;ContempOncol (Pozn). 2013; 17(1): 103–106.
[8]. Metachronous bilateral pleomorphic adenoma of the parotid gland; Jose´ da Silva et al. Vol. 101 No. 3 March 2006.
[9]. Slow-Growing Palatal Mass: A Challenging Differential Diagnosis; Marques et al. Challenging Diagnosis of Maxilla Tumour. J Oral MaxillofacialSurge 68:1884-1889, 2010.
[10]. Ellis GL, Auclair PL, Gnepp DR: Surgical Pathology of the Salivary Glands (ed 1). Philadelphia, WB Saunders, 1991.
[11]. Jansisyanont P, Blanchaert RH Jr, Ord RA. Intraoral minor salivary gland neoplasm: a single institution experience of 80 cases. Int J Oral MaxillofacSurg 2002; 31:257–61.
[12]. Garcia Berrocal JR, Ramirez Camacho R, Trinidad A, Salas C. Mixed tumour (pleomorphic adenoma) of the head and neck. Typical and atypical patterns. An OtorrinolaringolIbero Am 2000; 27:333-40.
[13]. Toida M, Shimokawa K, Makita H, Kato K, Kobayashi A, Kusunoki Y, et al. Intra-oral minor salivary gland tumors: a clinicopathological study of 82 cases. Int J Oral MaxillofacSurg2005; 34:528-32.
[14]. Kondo T. A case of lipomatous pleomorphic adenoma in the parotid gland. DiagnPathol 2009; 4:16.
[15]. Barnes L, Eveson JW, Reichart P, Sindransky D: World Health Organization Classification of Tumours: Pathology and Genetics of Head and Neck Tumors (ed 1). Lyon, IARC Press, 2005, p 209
[16]. Dr.Janardhan.N, Dr.Ramanjaneyulu.J, Dr.Rajendra Rao, Dr.Murali Mohan;Myoepithelioma-Plasmacytoid variant of minor salivary gland: Case report with emphasis on histopathological findings;Medical Science;Vol :2 Issue: 1 Jan-June 2013.
[17]. A Agarwal, A Sethi, S Chopra, D Sareen;Clear Cell Myoepithelioma Of The Hard Palate;The Internet Journal of Head and Neck Surgery;2006 Volume 1 Number 2.
[18]. BinduJ.Nair,Velayudhannair,vivek,TrivandrumT.Sivakumar,AnnaP.Joseph,Babyamm Raghavanpillai Varun, and Vinod Mony;Clear Cell Myoepithelioma of Palate with Emphasis on Clinical and Histological Differential Diagnosis;ClinPract. Mar 27, 2014; 4(1): 628.
[19]. Zormpa MT, Sarigelou AS, Eleftheriou AN, Assimaki AS, Kolokotronis AE;Plasmacytoidmyoepithelioma of the palate: case report;Head Neck Pathol. 2011 Jun; 5(2):154-8.
[20]. Swarna YM, Ali IM, Annigeri RG (2014) A Rare Case Report of PlasmacytoidMyoepithelioma of Hard Palate. J Clin Case Rep 4:348. doi: 10.4172/2165-7920.1000348.
[21]. Lucas Gomes Patrocinio; Priscila Garcia Damasceno; José Antonio Patrocinio;Malignant myoepithelioma of the hard palate: 9-year follow-up;Braz. j. otorhinolaryngol. (Impr.) vol.75 no.4 São Paulo July/Aug. 2009.
[22]. Gore CR, Panicker N K, Chandanwale S S, and Singh BK. Myoepithelioma of minor salivary glands - A diagnostic challenge: Report of three cases with varied histomorphology. J Oral MaxillofacPathol 2013; 17:257-60.
[23]. E Ferri, I Pavon, E Armato, S Cavaleri, P Capuzzo, and F Ianniello;Myoepithelioma of a minor salivary gland of the cheek: case report;ActaOtorhinolaryngol Ital. Feb 2006; 26(1): 43–46.
[24]. M. Politia, C. Toroa, N. Zermanb, L. Mariuzzic, M. Robionya;Myoepithelioma of the parotid gland: Case report and review of literature;Oral Oncology ExtraVolume 41, Issue 6, July 2005, Pages 104–108.
[25]. Éricka Janine Dantas da Silveira; Antonio Luiz Amaral Pereira; Maria Carmen Fontora; Lélia Batista de Souza; Roseana de Almeida FreitasV;Myoepithelioma of minor salivary gland - An immunohistochemical analysis of four cases;Rev. Bras. Otorrinolaringol. vol.72 no.4 São Paulo July/Aug. 2006.
[26]. Sciubba and Brannon; Myoepithelioma of Salivary glands;Cancer 49:562-572, 1982.
[27]. John D. Crissmamnd, John A. Wirmanm, D, and AnthonyhArrism, D;Malignant Myoepithelioma of the ParotidGland;Cancer 40:3042-3049, 1977.