



BILATERAL PRESENTATION OF WARTHIN TUMOUR-A CASE REPORT

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ABSTRACT

Salivary gland tumours are uncommon, representing less than 6% of head and neck neoplasm. Pleomorphic Adenoma is the most common benign epithelial salivary gland neoplasm, comprising 50%-74% of all parotid tumours. It is followed by warthin's tumour (4-14%). It is more common in males between 60 and 70 years of age and is closely related to smoking. The growth of this tumour is slow. Preoperative fine needle aspiration cytology (FNAC) of a parotid mass plays a pivotal role in differentiating neoplastic from inflammatory lesions as well as management of patients. We report a case of a 70 year old male chronic smoker who presented with bilateral painless swelling in the parotid region.

Key words:-Salivary Gland Tumours, Benign Tumour, Warthin's Tumor, Rare Tumour, Fine Needle Aspiration, Smoking, Bilateral.

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INTRODUCTION

Warthin's Tumour is a benign neoplasm. The Term "Warthin Tumor", was named after Aldred Warthin, the pathologist who published the first two reports in the american literature in 1929. Warthin's tumour is also known as Adenolymphoma or Papillary Lymphomatous Cystadenoma. Warthin's tumor is the second most common benign neoplasm of the parotid gland[2]. only rare cases demonstrate malignant transformation, constituting 0.3% of tumors. It occurs predominantly in the parotid gland and represents approximately 15% of parotid tumours. Now a days both sexes show an equal ratio[4]. The incidence rate is higher than that of salivary gland cancer but is lower than that of benign mixed tumor [1]. about 70% of all salivary gland tumours arise

in the parotid gland and the great number of this are benign tumours, with an average prevalence of 75% -85% of all parotid lesions[2,3]. parotid glands can give rise to a wide variety of benign and malignant neoplasm because of their mixed array of cells and tissues[6]. Tumours account for approximately 60-80% of parotid neoplasms[7]. Here, we are presenting a case of a 70 year old male chronic smoker who presented with bilateral painless swelling in the parotid region.

CASE REPORT: A 70 year old male patient reported to the department of oral medicine and radiology, Meenakshi Ammal Dental College and Hospital, Chennai, India with the chief complaint of swelling in his right and left side of the face for past one year. Patient gave a history of presence of swelling in the right and left preauricular region for past one year. He also gave a history of mild discomfort while chewing occasionally and there was no pain during rest. The swelling was slow

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in onset and gradually increased to attain the present size in one year. There were no other swellings present in the body. And there was No History of fever, palsy or paresthesia was elicited. Patient was hypertensive and diabetic for past 15 years and under regular medication for the same. His surgical and family histories were found to be insignificant. General examination revealed that the patient was moderately nourished and no other significant details were evident. Review of systems was not contributory. Extra orally on inspection bilateral diffuse Swellings were present in the right and left. The colour of the skin was normal to that of the adjacent skin. There was no sinus tract opening or pus discharge. Lifting of earlobe was absent. On palpation the inspeactory finding regarding site, size, extent was confirmed. The swellings was firm in consistency, tender on palpation, fluctuant, non compressible, non reducible. There was no fixidity to the underlying structures. There was no local rise in temperature. The skin over the swelling was pinchable. On Intra oral examination Oral mucosa appeared to be moist and hydrated with no obvious xerostomic signs. Stensen’s duct orifice and parotid papilla appear normal with no signs of inflammation. Milking of the right and left parotid gland showed normal salivary flow. No pathologic discharge was seen. Associated tenderness elicited during milking of the gland. Based on the history and clinical examination Bilateral chronic sialadenitis of right and left parotid gland was considered. Differential diagnosis of sialadenosis and pleomorphic adenoma was considered. Patient was alcoholic, diabetic and was under antihypertensive drugs which supports the feature of

sialadenosis but the age of onset and recurrence rate are the two features which differentiates it from sialadenosis. Pleomorphic adenoma was considered because of the site of occurrence and it was slow growing lesion. Female predominance and fourth decade of occurrence differentiates this lesion from pleomorphic adenoma.

On routine radiographic investigation, OPG Revealed No of teethpresent: 25, Missing: 24, 26, 37, 32, 47, 48. Root stumps: 18, 36, 45, 46, Dental caries with apical periodontitis: 17, 35, 34, 33, 38. Generalized attrition. Chronic generalized periodontitis (Fig.2).

Patient was subjected to Ultrasono Graphy, which revealed bilateral enlarged parotid gland with heterogenous parenchyma with increased vascularity suggestive of bilateral mixed parotid tumour. Following which, Magnetic Resonance Imaging was taken, Evidence of Fairly Well Defined Lobulated Heterogenous Solid Mass Lesion Involving The Central And Posterior Aspect Of Superficial And Deep Lobe Of Parotid Gland Measuring 3.4*3*4.1cm With Eccentric T2 Hypointensity With No Extraparotid Extension Or Bony Invasion Or Calcific Foci Within The Possibility Of Pleomorphic Adenoma.(fig.3)

FNAC: Smear Shows Papillary Clusters Cohesive Groups And Singly Dispersed Epithelial Cells With Oncocytic Features In A Background Of Numerous Lymphocytes, Cyst Macrophages And Proteinaceous Eosinophilic Material suggestive of warthin’s tumour in right and left parotid gland. Based on the FNAC report and clinical finding superficial parotidectomy of both the right and left parotid lobes were done.(fig.4)

Fig. 1. Profile Picture



Fig. 2. OPG

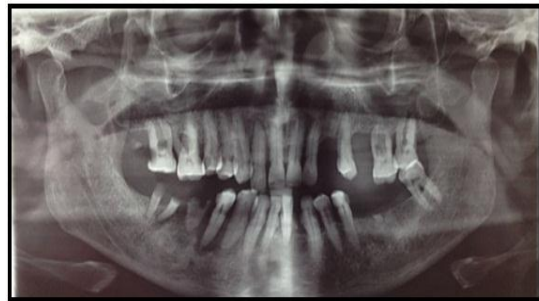


Fig. 3. MRI

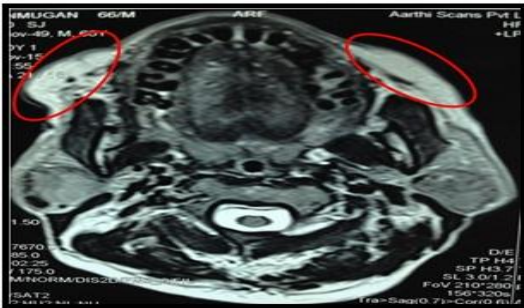
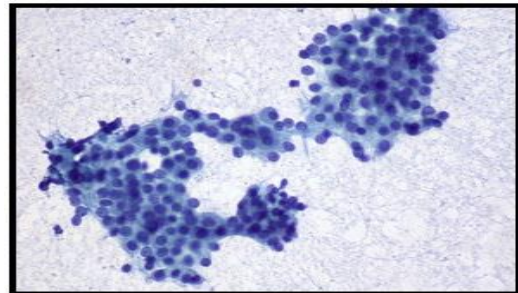


Fig. 4. FNAC



DISCUSSION

Papillary cystadenoma lymphomatosum was first described by Hildebrand in 1895 as a form of congenital cyst of the neck. It is most commonly found in parotid gland,[8]but cases have also been reported in sub maxillary gland. Warthin's tumor is multicentric in 12-20% of patients and it is bilateral. Unlike other benign neoplasm of the salivary glands, this tumour has a tendency towards bilateral involvement and approximately 90% of lesions occur in the superficial lobe of the parotid gland[2]. Warthin tumor is a controversial entity of benign salivary gland tumors because of its histopathological appearance and unknown origin [1]. The most accepted hypothesis about the origin of Warthin tumor is that it develops from salivary ducts inclusions in the lymph nodes, after the embryonic development of the parotid gland, the stromal component is the lymph node. This hypothesis is further supported by the frequent detection of salivary gland tissue in the peri- and intraparotid lymph nodes[1]. Warthin tumor generally has been regarded as the second most common benign tumor of the parotid gland after pleomorphic adenoma. It occurs largely in middle-aged and older men and usually in the parotid gland or periparotid region, mostly involving the inferior pole of the gland. Multicentric occurrence is seen more often with Warthin tumor than any other salivary gland tumor. Warthin tumors usually are spherical to ovoid, smooth surfaced, measures 2 to 4 cm in diameter. Smoking is an important risk factor for the development of Warthin tumor. Some authors report that the risk is closely related to the amount smoked and that smokers are eight times more likely to develop the tumor than nonsmokers.[9,10]

It is believed that this relationship is justified by the retrograde flow of substances in cigarette smoke to the salivary ducts or the excretion by the cigarette of noxious substances in the ducts. [9,11]. The association with cigarettes is independent of sex and age. There are few case reports demonstrating a malignant tumor adjacent to a benign Warthin lesion. Most of these cases were associated with previous radiotherapy[3,12] ionizing radiation represents a decisive factor in the appearance of this tumour. This theory is confirmed by the high incidence of this tumour among the Japanese people who survived the atomic bomb explosions and among subjects irradiated at young age for treatment of ringworm[13,14]. Teymoortash et al. (2001) performed immunohistochemical analysis on tissue samples of cystadenolymphoma, pleomorphic adenoma and normal samples of the parotid gland, searching for oestrogen and progesterone receptors. Positivity for progesterone receptors was found in six out of nine samples tested. progesterone receptors in the salivary duct system of the normal parotid gland is expected to influence the regulation of water and electrolyte transport. The detection of sex hormone receptors in

cystadenolymphoma might give evidence of a hormone dependence of this tumour that could explain the higher proportion of men affected by the disease[13,15]. The association between Warthin's Tumour and autoimmune disease were first hypothesized by Allegra in 1971. On the basis of morphological analogies between the histological features of Warthin's tumour and those observed in several organ-specific immune disorders, particularly in Hashimoto's thyroiditis and autoimmune disease involving salivary and lacrimal glands (that is, Sjogren syndrome), Allegra postulated the involvement of cell-mediated immune mechanisms of the delayed hypersensitivity type in its pathogenesis[13]. Further studies are required to confirm these hypothesis and to investigate the role of the other etiological factors in the development or progression of the lesion.

Since Warthin's tumours can be multifocal, a pre-operative diagnosis by means of FNAB is mandatory and complete bilateral screening of the gland by MRI is needed to programme the surgery.

The warthin tumor has one of the most distinctive histopathologic patterns of any tumor in the body. Although the term papillary cystadenoma lymphomatosum is cumbersome, it accurately describes the salient microscopic features. The tumor is composed of a mixture of ductal epithelium and a lymphoid stroma and the epithelium is oncocytic in nature, forming uniform rows of cells surrounding cystic spaces. The cells have abundant, granular eosinophilic cytoplasm and are arranged in two layers. The inner luminal layer consists of tall columnar cells with centrally placed, palisaded and slightly hyperchromatic nuclei. Beneath this is a second layer of cuboidal or polygonal cells with more vesicular nuclei. The lining epithelium demonstrates multiple papillary in foldings that protrude into the cystic spaces. Focal areas of squamous metaplasia or mucous cell prosoplasia may be seen. The epithelium is supported by a lymphoid stroma that frequently shows germinal center formation [9,11, 16].

Surgical removal is the treatment of choice for patients with Warthin's tumor. The procedure usually is easily accomplished because of the superficial location of the tumor. Some surgeons prefer local resection with minimal surrounding tissue; others opt for superficial parotidectomy to avoid violating the tumor capsule and because a tentative diagnosis may not be known preoperatively. 6% to 12% recurrence rate has been reported. Many authors, however, believe that the tumor is frequently multicentric in nature; therefore, it is difficult to determine whether these are true recurrences or secondary tumor sites. Malignant Warthin tumors (Carcinoma Ex Papillary Cystadenoma Lymphomatosum) have been reported but are exceedingly rare [11].

CONCLUSION

Finally, even if a benign parotid tumour has been detected by both imaging and FNAC or biopsy, appropriate surgery is always recommended more than clinical observation. Clinical and radiological findings might result in some cases discordant with definitive

diagnosis due to the variable clinical presentation and the histological heterogeneity of parotid tumours. Only surgery can give histological certainty of benignity and definitively prevents long term malignant degeneration or lump infection or risk of size-dependent surgical complications.

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