

CASE REPORT

Synchronous Bilateral Warthin's Tumors of the Parotid: A Case Report

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Abstract

Warthin's tumors are the second most frequent benign tumors of the parotid gland representing 6-10% of all tumors of the salivary glands. Multicentric Warthin's tumors are more common than any other salivary gland tumor. Most of the multifocal Warthin's tumors are unilateral, whereas bilateral Warthin's tumors are much more uncommon; bilateral Warthin's tumors are metachronous with few synchronous cases having been described in the literature.

Key Words

Parotid gland, Warthin's tumor, Bilateral tumors, Multifocal tumors

Introduction

Salivary gland tumors represent approximately 3% of all head and neck tumors. About 70-80% of these neoplasms occur in the major salivary glands, with the parotid being the most often affected site (1,2,3). Warthin's tumor (papillary cystadenoma lymphomatosum or adenolymphoma of the parotid gland) is the second most frequent benign tumor of the parotid gland representing 6 to 10% of all tumors of the salivary glands (4). These tumors present as asymptomatic, slowly growing masses, affecting predominantly men (10:1) in the 5th and 6th decade (5,6). Although pathogenesis is unknown, there is an association with cigarette smoking (6,7). Warthin's tumor is often multicentric and is bilateral in 10% to 15% of cases. It comprises 70% of all bilateral salivary gland neoplasms (8). Bilateral Warthin's tumors are metachronous with few synchronous cases having been described in the literature (9,10,11).

We hereby, present a rare, interesting case of a simultaneously occurring bilateral Warthin's tumor growing in the parotid glands.

Case Report

A 65 year old male presented with bilateral asymptomatic masses over the mandibular angles of both sides that had been enlarging slowly over many years (*Fig 1*). The patient was a chronic smoker. Clinical examination revealed bilateral mobile, non-tender, firm swellings measuring approximately 4 x 3.5 cm (right) and 4.5 x 2.5 cm (left) with normal intraoral examination. Fine needle aspiration biopsy carried out on both nodules yielded mucoid, murky fluid. Air dried and alcohol fixed smears were prepared from the aspirated material and stained with May-Grunwald Giemsa (MGG) and Papanicolaou (PAP) stains, respectively. Cytological examination of the smears demonstrated cohesive

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Fig 1. Clinical Photographs Showing Bilateral Parotid Masses

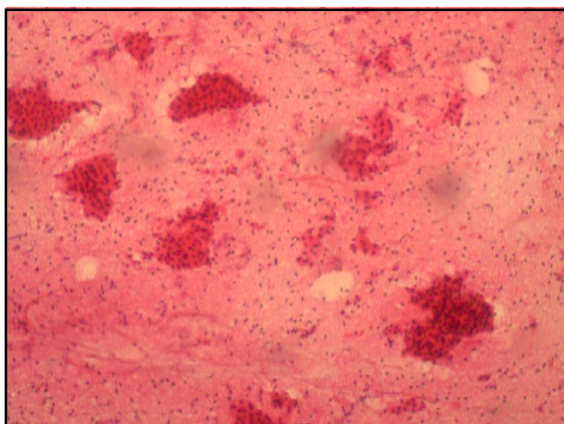


Fig 3. Papanicolaou Stained Smear Showing Sheets of Oncocytic Cells with Pinkish orange Cytoplasm. In the back Ground are Seen lymphocytes and Amorphous Granular debris (Pap x 100).

monolayered sheets of bland oncocytic cells having abundant dense cytoplasm (blue in MGG and pink to orange in Pap), small round nuclei with bland chromatin and inconspicuous nucleoli. Many mature lymphocytes along with amorphous and granular debris were seen in the background (Fig 2 & 3). These cytological features were characteristic of Warthin's tumor.

Discussion

The first description of adenolymphoma was given by Hildebrand in 1895 (12) and in 1929 Warthin published a series of so called papillary cystadenoma lymphomatosum leading to the well known term "Warthin's

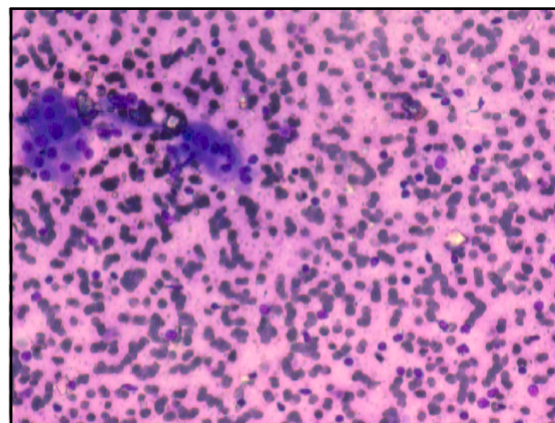


Fig 2. Photomicrograph Showing a Flat Sheet of Oncocytes Depicting Abundant Blue Cytoplasm and Uniform Round nuclei with lymphocytes and Debris in the background (MGG x 400).

tumor" (13). Cystadenolymphomas are the second most frequent benign tumors of the parotid gland representing 6-10% of all tumors of the salivary glands (4). However, it is difficult to estimate the real incidence of Warthin's tumor since there are regional, national and racial differences (5,6). Smoking seems to be a significant risk factor for development of Warthin's tumor. Some authors reported that the risk of bilateral Warthin's tumor correlated significantly with the amount of nicotine intake (7). Salivary gland neoplasms are usually solitary while multiple tumors occurring simultaneously in an individual are uncommon, albeit, there is evidence that multicentric Warthin's tumors are more common than any other salivary gland tumor; in fact, about 12% of patients develop multiple tumors (14). Most of the multifocal Warthin's tumors are unilateral, whereas bilateral Warthin's tumors are much more uncommon; bilateral Warthin's tumors are metachronous with few synchronous cases having been described in the literature (9,10,11).

Multiple occurrence of Warthin's tumor seems to be related to its histologic origin. There is no intraglandular compartmentalization between the epithelial and lymphoid components in the early stages of embryogenesis of the parotid gland. Epithelial cells, the

precursors of the salivary duct-acinar system, may remain embedded within the lymphoid component. These epithelial inclusions may give rise to Warthin's tumor following an unknown tumourogenic event (9).

Though the position of fine needle aspiration cytology in the diagnosis of Warthin's tumors or lesions of the parotid gland in general has been mired in controversy, however, several studies have been able to prove a high sensitivity and specificity for the diagnosis of Warthin's tumor with FNAC (15,16). Cytology when coupled with clinical and image findings may also permit conservative tumor management (16).

The treatment for bilateral tumors is the surgical approach, similar to that indicated for solitary tumors, i.e., partial parotidectomy when it occurs in the superficial lobe, or total parotidectomy with the identification, dissection and preservation of facial nerve branches when located in a deep lobe.

In view of the possible association of Warthin's tumor with extra-salivary neoplasms, extensive work-up of the patients harbouring multiple Warthin's tumors is, therefore, indicated. A long-term follow up is mandatory, because of possible occurrence of metachronous salivary and extra-salivary tumors even after prolonged time intervals (17,18).

References

1. Ellis GL, Auclair PL, Gnepp DR. Surgical pathology of the salivary glands. Philadelphia: W.B. Saunders; 1991, .pp.165.
2. Loyola AM, de Araujo VC, de Souza SO, *et al.* Minor salivary gland tumors. A retrospective study of 164 cases in a Brazilian population. *Eur J Cancer B Oral Oncol* 1995; 31B: 197-201.
3. Hilton JM, Phillips JS, Hellquist HB, *et al.* Multifocal multi-site Warthin's tumour. *Eur Arch Otorhinolaryngol* 2008; 265: 1573-1575.
4. Chapnik JS. The controversy of Warthin's tumor. *Laryngoscope* 1983; 93: 695-716.
5. Teymoortash A, Werner JA. Tissue that has lost its track: Warthin's tumour. *Virchows Arch* 2005; 446: 585-588.
6. Teymoortash A, Krasnewicz Y, Werner JA. Clinical features of cystadenolymphoma (Warthin's tumor) of the parotid gland: a retrospective comparative study of 96 cases. *Oral Oncol* 2006; 42: 569-573.
7. Peter Klussmann J, Wittekindt C, Florian Preuss S, Al Attab A, Schroeder U, Guntinas-Lichius O. High risk for bilateral Warthin tumor in heavy smokers - review of 185 cases. *Acta Otolaryngol* 2006; 126: 1213-1217.
8. Turnbull AD, Frazell EL. Multiple tumors of the major salivary glands. *Am J Surg* 1969; 118: 787-789.
9. Maiorano E, Lo Muzio L, Favia G, *et al.* Warthin's tumour: a study of 78 cases with emphasis on bilaterality, multifocality and association with other malignancies. *Oral Oncol* 2002; 38: 35-40.
10. Yu GY, Ma DQ, Zhang Y, *et al.* Multiple primary tumours of the parotid gland. *Int J Oral Maxillofac Surg* 2004; 33: 531-534.
11. Ethunandan M, Pratt CA, Morrison A, *et al.* Multiple synchronous and metachronous neoplasms of the parotid gland. The Chichester experience. *Br J Oral Maxillofac Surg* 2006; 44: 397-401.
12. Hildebrandt O: Innate epithelial cysts and growths of the neck. *Arch Klin Chir* 1895; 49: 167-206.
13. Warthin A: Papillary cystadenoma lymphomatosum. *J Cancer Res* 1929; 13: 116-125.
14. Shah N, Tighe JV, Barrett AW, *et al.* Bilateral intraparotid and extraparotid Warthin's tumours. *Br J Oral Maxillofac Surg* 2007; 45: 238-239.
15. Veder LL, Kerrebijn JD, Smedts FM, den Bakker MA: Diagnostic accuracy of fine needle aspiration cytology in Warthin's tumors. *Head Neck* 2010; 32: 1635-1640.
16. Viguer JM, Vicandi B, Jimenez-Heffernan JA, Lopez-Ferrer P, Gonzalez-Peramato P, Castillo C. Role of fine needle aspiration cytology in the diagnosis and management of Warthin's tumor of the salivary glands. *Cytopathology* 2010; 21: 164-169.
17. van der Wal JE, Davids JJ, van der Waal I. Extraparotid Warthin's tumours - report of ten cases. *Br J Oral Maxillofac Surg* 1993; 31: 43-44.
18. Scasso CA, Papini M, Eligi C, *et al.* An unusual neck mass: the Warthin's tumor. *Acta Otorhinolaryngol Belg* 1998; 52: 55-7.