

## Bilateral Synchronous Salivary Gland Tumor: A Rare Presentation

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### Abstract

Warthin's tumor (WT) is the most prevalent monomorphic adenoma of the major salivary glands. It accounts for 2–15% of all parotid tumors. Bilateral occurrences of multifocal WT are considerably less frequent than unilateral occurrences. Most of these bilateral tumors occur metachronously, with only a few synchronous occurrences reported in literature. A 61-year-old male presented with bilateral parotid swelling, with intermittent pain and difficulty opening his mouth. MRI revealed solid lesions in the superficial lobes of both parotid glands, with the largest measuring 3.7x2.3x3.8 cm on the left. FNAC indicated a Category IVA neoplasm, favoring WT. Bilateral parotidectomy and level IB neck dissection were performed. Histology showed multifocal, well-defined tumors with bilayered oncocytic epithelium, papillary and cystic structures, and lymphoid stroma. Reactive hyperplasia in lymph nodes was observed. These findings were consistent with bilateral multifocal WT. This case highlights a rare instance of synchronous bilateral WT in a 61-year-old patient. It emphasizes the need for careful diagnostic evaluation and treatment, as WT is typically metachronous. The report adds valuable insights to the literature, stressing the importance of considering synchronous presentations in salivary gland tumors.

**Keywords:** Adenolymphoma, Head and Neck Neoplasm, Papillary Cystadenoma Lymphomatosum, Synchronous, Warthin's Tumor.

### Introduction

Warthin's tumor (WT) also known as papillary cystadenoma lymphomatosum (PCL) is the second most frequent benign tumor of parotid gland comprising 5–12% of all tumors of salivary glands. While exact pathogenesis remains unclear, there is a significant association with smoking [1]. The risk appears to be dose-dependent, with long-term smokers exhibiting a significantly higher incidence of both unilateral and bilateral WT. It is hypothesized that field cancerization or multifocal oncocytic transformation of salivary gland tissue, possibly triggered by chronic exposure to tobacco carcinogens, may play a role [2].

The World Health Organization has classified WT as one of the benign epithelial tumors of the salivary glands, along with sclerosing oncocytoma, myoepithelioma, canalicular adenoma, cystadenoma, ductal papilloma, sialadenoma papilliferum, lymphadenoma, sebaceous adenoma, intercalated duct adenoma, striated duct adenoma, basal cell adenoma, polycystic adenoma, pleomorphic adenoma, or keratocystadenoma [3].

Patients frequently report a painless, slowly expanding nodule near the angle of mandible [4]. This tumor is far prevalent in men and affects people in their 50-60 years [5]. WT is unilateral or bilateral and may be synchronous

or metachronous in occurrence. Bilateral tumors are extremely rare, occurring in 7–10% of cases; multifocal tumors and recurrences occur in approximately 2% of cases; and malignancy is rare [6]. The majority of bilateral tumors are metachronous, while there are few synchronous bilateral occurrences reported in literature.

This case report describes a rare occurrence of synchronous bilateral WT in a 61-year-old patient.

### Case Presentation

A 61-year-old gentleman presented with swelling in the right parotid region (Figure 1A)



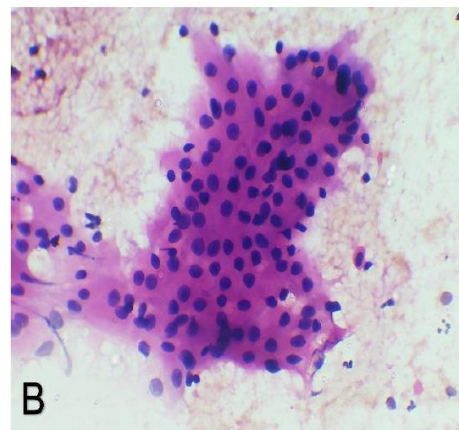
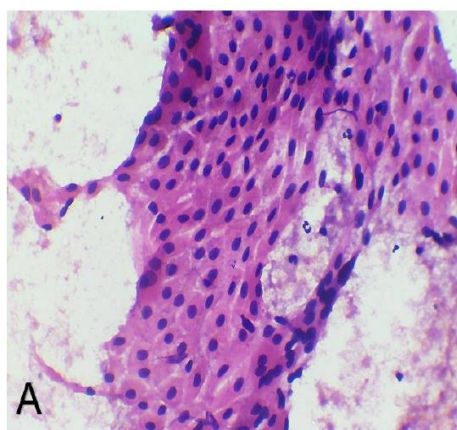
**Figure 1.** Clinical Photograph Showing Right Side Parotid Swelling(A) and Left Side Parotid Swelling(B)

### FNAC Findings

Unguided FNAC from both sides exhibited several oncocyte clusters (Figure 2A&B) surrounded by lymphocytes, lymphoid follicular centre cells, and granular debris.

for 2 years and a swelling in the left parotid region (Figure 1B) for 1.5 years, with intermittent pain and difficulties opening his mouth. On examination a firm swelling of size 2\*2cm noted in right parotid region and swelling of size 3\*3cm noted in left parotid region. MRI revealed solid lesions dispersed across superficial lobes of parotid glands. Largest lesion appeared 3.7\*2.3\*3.8cm located in left parotid gland. MRI in addition shows a few symmetrically enlarged level IB lymph nodes in upper cervical region inferior to parotid glands.

Findings were as per category IVA Neoplasm favouring WT as reported by MILAN system of reporting salivary gland cytology [7]. Following which patient underwent Bilateral parotidectomy with bilateral level IB neck dissection.



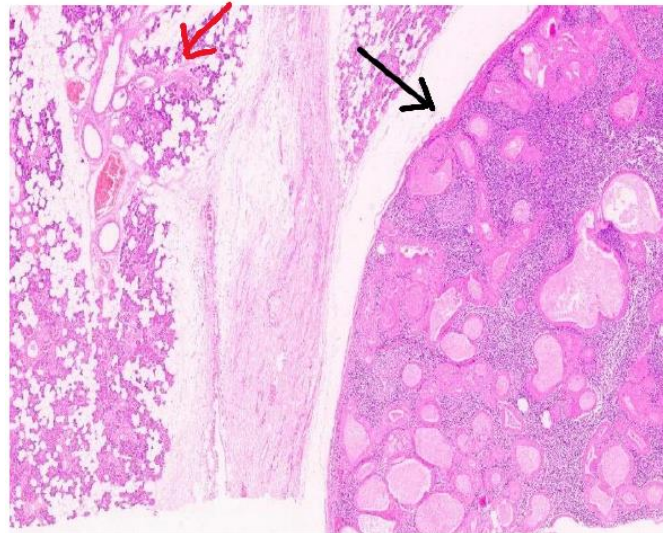
**Figure 2.** (H&E 40x magnification): [A]FNAC from right parotid swelling [B] FNAC from left parotid swelling

Both A and B shows an oncocytic cluster of cells with eosinophilic granular cytoplasm, well defined cytoplasmic margins and rounded nuclei.

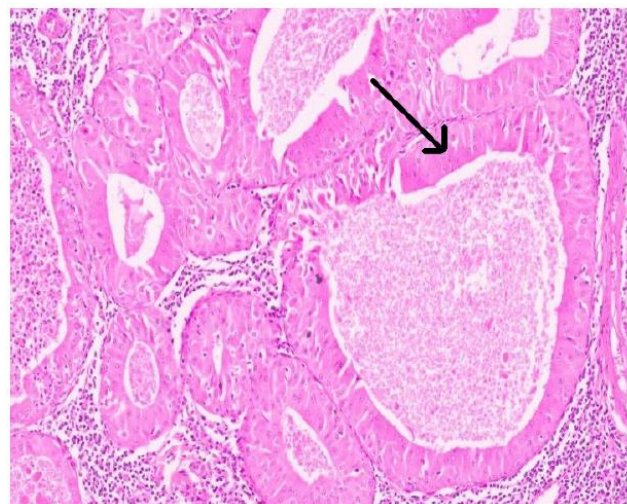
### Histopathology Findings

Haematoxylin and Eosin (H&E) sections from both sides show a multifocal, well-defined

tumors lined by bilayered oncocytic epithelium composed of mixed proportions of papillary and cystic structures (Figure 3). Oncocytic columnar cells contribute to inner luminal layer of epithelium, while oncocytic cuboidal cells form discontinuous outer layer. (Figure 4).



**Figure 3.** (H&E Scanner 5x): Shows a normal salivary gland (red arrow) with well demarcated neoplasm (black arrow) composed of papillary and cystic structures.



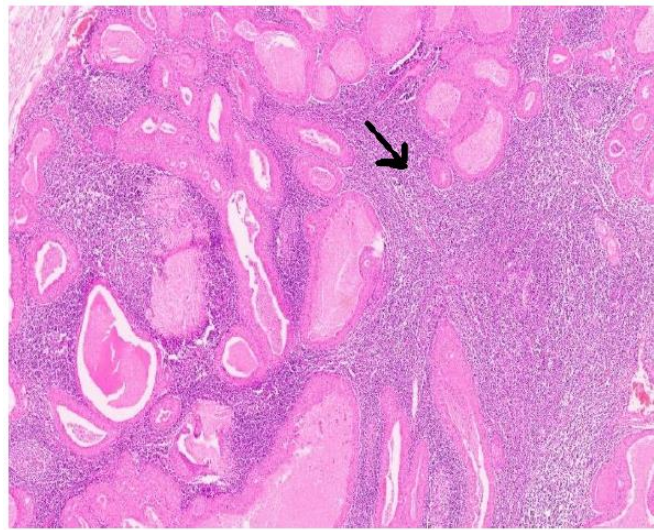
**Figure 4.** (H&E 40x): The characteristic bilayered oncocytic epithelium (black arrow) with inner columnar and outer cuboidal cells. The cyst lumen shown thick proteinaceous secretion.

Luminal cells have uniform nuclei that palisade toward free surface and contain plenty of eosinophilic cytoplasm. Round to oval nuclei and occasionally small, noticeable nucleoli appear in basal cells. Downward extension of epithelial cells forming loosely arranged to

closely packed tubular glands and nests were identified. Focal metaplastic changes of lining epithelium to squamous and mucous cells were noted. Thick proteinaceous secretions, cholesterol crystals, inflammatory cellular debris, and small number of eosinophilic

laminated bodies have been detected in cyst lumens. Small lymphocytes mixed with plasma cells and histocytes have been detected in surrounding dense lymphoid stroma (Figure

5). Germinal centres were also noted. Patchy areas of stromal fibrosis, neutrophilic abscess, and haemorrhage were identified.



**Figure 5.** (H&E 10x): Shows dense lymphoid stroma (black arrow) surrounding the papillary and cystic structures.

The features were compatible with bilateral multifocal WT. Right and left intraparotid and level Ib nodes showed reactive hyperplasia.

## Discussion

Adenolymphoma, PCL, and cystadenolymphoma are additional terms for WT. Hildebrand initially recognized it as a kind of congenital cervical neck cyst in 1895. Pathologist Alfred Scott Warthin of the University of Michigan reported two additional cases later in 1929 and termed them PCL. Martin and Ehrlich introduced term Warthin's tumour in 1944 upon reporting 22 cases of parotid gland PCL.

WT is the second most frequent benign salivary gland neoplasm [8]. It often occurs in the inferior pole of superficial lobe and periparotid lymph nodes. According to WHO it accounts for 5-20% of all salivary tumors with slight male predominance and mean age of occurrence being 62 (range 12-92). WT subtype is Infarcted/metaplastic WT [9].

Clinical presentation is a slow-growing painless, firm, or fluctuant swelling. Pain or facial nerve palsy may be present in

metaplastic/infarcted variants [9]. They can appear unilaterally or bilaterally and are multifocal in 12-20% of patients, either synchronously or metachronously. Few synchronous cases of bilateral WT have been reported in literature; most are metachronous. It is rare to find multiple synchronous bilateral WT [10].

One major risk factor that is directly associated with the development of WT is smoking. According to the authors' research results, there appeared to be a substantial correlation between the probability of bilateral incidence and nicotine consumption. However, researchers have demonstrated that tobacco smoke induces ductal epithelium to become irritated and that in turn promotes development of tumors [11]. Strongly linked to cigarette smoking if it occurs bilaterally. Its incidence has increased in recent decades, a trend possibly attributed to higher smoking rates among men in earlier generations [12].

Other risk factors are radiation exposure in atomic bomb survivors and autoimmune diseases especially thyroiditis. The Epstein-Barr virus has a strong association with WT

etiology, according to research conducted by Santucci et al [11].

As mentioned above WT occurrence is multiple. This is related to histological origin. Epithelial and lymphoid components don't intra-glandularly compartmentalize during early development. An unidentified tumorigenic process may result in WT development, as epithelial cells remain embedded within lymphoid components. Parotid gland's tail contains multiple lymph nodes, leading to probability that tumors may develop [13].

Ultrasound is widely regarded as the initial imaging modality for evaluating disorders among the main salivary glands. Ultrasound usually shows a well-defined, ovoid or spherical, hypoechoic mass in the case of WT, often exhibiting microcystic or anechoic components due to its cystic nature [14]. While ultrasound provides a useful initial assessment, MRI is the preferred imaging technique for in-depth evaluation of salivary gland lesions. It provides critical information about lesion size, internal architecture, tumor margins, and potential perineural spread. On MRI, WT usually presents as a well-demarcated lesion exhibiting strong T1-weighted images with low-to-intermediate signal intensity, while T2-weighted images have high signal intensity. Following contrast, WT typically demonstrates low or heterogeneous enhancement due to its cystic and lymphoid components [15].

The diagnostic technique known as FNAC (fine-needle aspiration cytology) helps assess abnormalities in the salivary glands, including WT. WT typically shows oncocytic epithelial cell cohesive sheets or papillary clusters surrounded by granular, frequently cystic debris and a large number of lymphoid cells. The existence of these oncocytes with centrally placed nuclei and abundant eosinophilic cytoplasm, alongside lymphocytes, is considered a hallmark of WT [16]. However, diagnostic challenges arise in variants such as metaplastic or infarcted WT.

WT exhibiting extensive squamous metaplasia are referred to as metaplastic or infarcted WT. In such cases, the presence of the characteristic bilayered oncocytic epithelium remains the most reliable morphological feature for accurate diagnosis. The absence of MAML2 gene rearrangement is a useful molecular diagnostic for metaplastic Warthin's tumors since these tumors routinely test negative for MAML2 gene fusions differentiating them from Warthin-like mucoepidermoid carcinoma [17].

On gross examination, most WT are usually well-circumscribed and spherical to ovoid in shape, except situations where secondary inflammation is present. Due to the potential for multicentricity, particularly within the parotid gland, parotidectomy specimens must be carefully assessed for the presence of additional, separate tumor foci. A thin capsule usually encapsulates the lesion, clearly delineating it from the surrounding salivary gland parenchyma [18].

Microscopically, they are characterized by a combination of solid and cystic constituents, comprising both lymphoid and epithelial components. The cystic areas are usually lined by a bilayered oncocytic epithelium and frequently contain eosinophilic fluid. This epithelium commonly forms papillary projections, supported by fibrovascular cores embedded within a dense lymphoid stroma, which frequently contains germinal centers [18].

Bilateral gland screening by MRI and FNAC pre-operative diagnostics is required. MRI scans are an excellent choice for diagnosing parotid gland tumors due to their exceptional ability to distinguish soft tissue [19]. Research have demonstrated that FNAC is more sensitive and specific for WT and for differentiating between benign and malignant neoplasm. Surgical resection is the treatment in case of multiple parotid gland tumors. Definitive diagnosis is only after histopathological report [20].

## Conclusion

A rare case of synchronous bilateral WT in a 61-year-old gentleman is described in this case report. The diagnosis was established through imaging and fine needle aspiration cytology, leading to successful surgical excision of the tumors. Given that WT is usually metachronous, this case emphasizes the importance of considering synchronous bilateral occurrences into account. The results emphasize the requirement for comprehensive

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diagnostic examination and suitable therapeutic methods for individuals with salivary gland tumors. The findings contribute valuable insights to the existing literature on this uncommon presentation.

## Conflict of Interest

The authors declare no conflict of interest.

## Acknowledgments

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