

Warthin's Tumor in Submandibular Salivary Gland: An Uncommon Extra Parotid Location

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Abstract

Introduction: Salivary gland tumors are complex to diagnose both on histopathology and cytology. Majority of these tumours arise in the parotid glands, followed by submandibular glands, and rarely in minor salivary glands. Warthin's tumor (WT), also known as papillary cystadenoma lymphomatosum, is characterized by a dense lymphoid stroma and a double layer of oncocytic epithelium with papillary and cystic architecture. Only 6-10% cases have been reported in the extra-parotid locations i.e. cervical lymph nodes, submandibular gland and larynx.

Case Report: Our patient presented with painless bilateral submandibular swellings for 1.5 months with history of chronic smoking of two bundles of biddis per day for 32 years.

Conclusion: Hereby, this case was unique in its presentation as it was located in an extra parotid region with bilateral origin, as these locations carry clinical significance in diagnosing and ruling out conditions like lymphoma.

Keywords: Cytology; Fine Needle Aspiration Cytology; Submandibular Gland; Warthin's Tumor.

Key Messages: Although Warthin's tumor is usually located in parotid salivary gland, a possibility of them arising from extra parotid sites like submandibular gland, lymph nodes etc should also be kept in mind as they carry important clinical manifestations.

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INTRODUCTION

Salivary gland shows wide variety of both benign and malignant tumors which are complex to diagnose on histology as well as cytology. Majority of these tumours arise in the parotid glands, followed by submandibular glands and rarely in minor salivary glands.¹ There are two most common benign tumors in the parotid gland, pleomorphic adenoma and Warthin's tumour

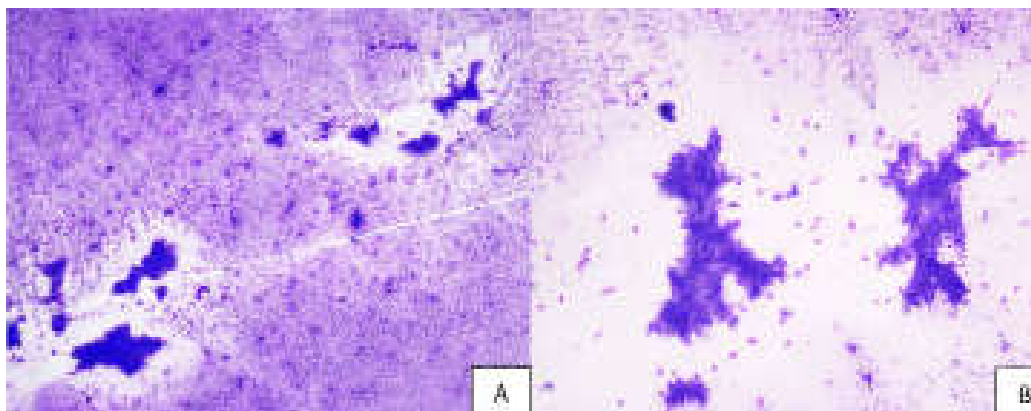
(WT).² Because of its histological features, which include a dense lymphoid stroma and a double layer of oncocytic epithelium with a papillary and cystic architecture, WT is also known as papillary cystadenoma lymphomatosum.³ Most of these tumours are located in the parotid gland's lower pole. These locations have clinical significance to diagnose and rule out other causes.² We hereby report a case of bilateral WT of submandibular salivary gland diagnosed on fine needle aspiration cytology (FNAC).

CASE REPORT

A 42-year-old male patient presented to us with painless bilateral submandibular swellings for 1.5 months with history of chronic smoking of two bundles of biddis per day for 32 years. On examination of both the swellings in submandibular region, right side measured 3x2 cm and left side measured 2x2 cm in size. The two swellings were oval in shape, had smooth surfaces, and were soft to firm in consistency. The swellings were nontender, non-pulsatile, were seen originating from submandibular glands during bimanual palpation and had no calculus. The ultrasound findings suggested ovoid lesions in right side of submandibular gland measuring 3.5x1.5x 0.5 cm with well-defined margins and sponge-like anechoic areas. Another lesion was seen in left submandibular region measuring 2x1.5x1 cm with regular margins and anechoic areas. Both the lesions showed cystic contents with septations. Our patient was advised FNAC of bilateral salivary gland swellings, as it is a straightforward and noninvasive method for preoperative diagnosis of salivary gland tumours. The whole FNAC

procedure was explained to the patient and consent was taken. Five clean slides along with a 20 ml disposal syringe and 22-gauge black needle were used. The slides were labeled with diamond pencil as A for right side and similarly B for left side. Then under aseptic precautions, first pass was taken from right side according to Zajdela's modified technique. As soon as the material aspirated in hub of the black needle, it was put on to the slides with the help of syringe and smears were made with the help of spreader. Similar procedure was done for left side. Two air dried smears were stained by May Grünwald Giemsa (MGG) and two alcohol fixed smears were stained by Papanicolaou (PAP) stain and hematoxylin and eosin (H&E) respectively from both the swellings. The cytomorphological findings of both sides showed oncocytic epithelial cells arranged in cohesive clusters and sheets with abundant granular cytoplasm, prominent nucleoli in few cells. No cytological atypia was noted. These oncocytic cells were admixed with abundant lymphocytes and few cystic macrophages in a murky background. (Fig. 1).

Later, both bilateral salivary glands were sent for histopathological examination in same container. On gross inspection, two greyish brown globular tissue pieces were received, larger measuring 4.5x2x2 cm and smaller measuring 3x2.5x2 cm. Both the pieces were well encapsulated and their cut section revealed partially solid and cystic areas along with areas of hemorrhage. Under light microscopy, the two glands displayed similar histomorphological features: a well encapsulated lesion with different sized papillary cystic structures surrounded by a lymphoid stroma with germinal centres and lined by bilayered oncocytic epithelial cells. (Fig. 2) Thus histopathological findings were in concordance with cytology.



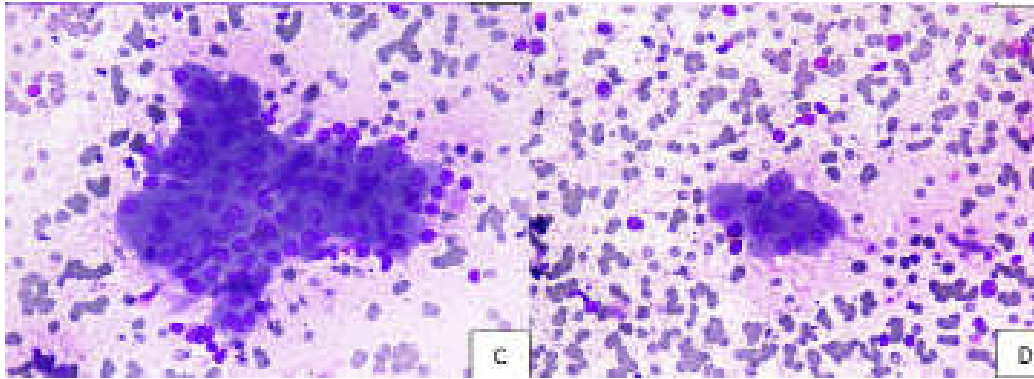


Fig. 1: (A) Cytomorphological features showing murky background (L&G, x40), (B) oncocytic epithelial cells arranged in cohesive clusters (L&G, x100), (C) & (D) oncocytic cells with abundant granular cytoplasm and lymphocytes in background (L&G, x400).

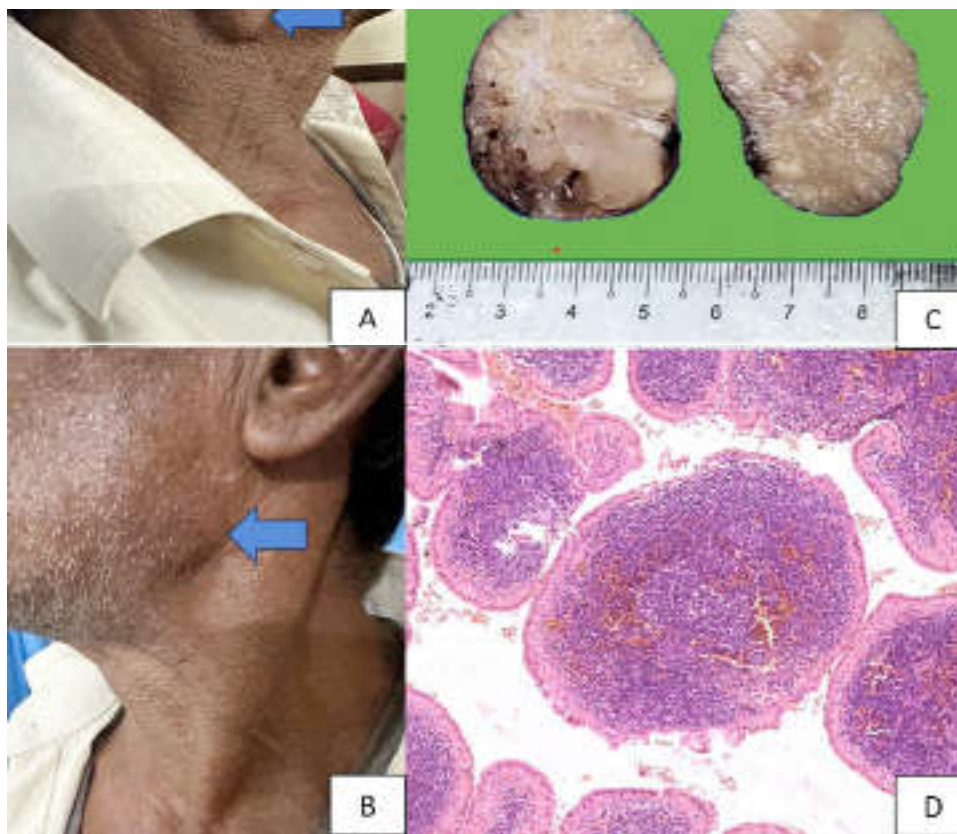


Fig. 2: (A) & (B) Bilateral swellings in the submandibular region, (C) Gross photograph showing a well circumscribed tumour with both solid and cystic areas of hemorrhage, (D) Histopathological examination showing bilayered oncocytic epithelium with lymphoid background (H&E, x400)

DISCUSSION

WT is the salivary gland's second most common benign tumour. Since the retrograde flow of chemicals from tobacco raises the risk of cancer, therefore these tumors are more common among male smokers.^{4,5} However, pathogenesis of WT is not clear, though many theories have been laid forward for its development. One such hypothesis includes 'hypothesis of heterotopias' in periparotid

and intraparotid lymph nodes. Other hypothesis postulates the theory that WT are adenomatous tumor with lymphocytic infiltration.⁶ Diagnostic accuracy with preoperative FNAC has been reported as 95% and 74%, by Flezar *et al.* and Syed *et al.* respectively.^{7,8} Viguer and Kim *et al.* stated that FNAC of WT showed features such as lymphocytes and amorphous granular debris with double rows of oncocytic cell layers.^{9,10} Many pitfalls are also associated with FNAC like less

cellular smears, scarcity of diagnostic features of tumour, presence of atypical or metaplastic cells or preponderance of only one component of tumour leading to misdiagnosis.^{6,8} For example, smears comprising of only oncocytes are typical for WT, but too much presence of these cells can lead to wrong interpretation of oncocytoma. Mukunyadzi *et al.* reported in one of their articles regarding oncocyte arrangements, i.e. when they are present in small and flat clusters, it is more in favour of WT, but when these group of cells form 3-dimensional clusters it is suggestive of oncocytoma and oncocytic carcinoma.^{5,11,12} Most Extra Parotid WT have been reported in periparotid lymph nodes, however English literature falls short of case reports of WT in bilateral submandibular gland locations.^{13,14}

CONCLUSION

This case was unique in its rarity as it was located in extra parotid region with a bilateral origin. Both cytological and histological morphology favoured diagnosis of WT. Such an extra parotid location has both academic and research importance, as a possibility of lymphoma and other pathologies always need to be worked up.

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Conflict of Interest: None

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