

Research Article**UNUSUAL PRESENTATION OF BILATERAL WARTHINS TUMOR IN SUBMANDIBULAR GLANDS WITH SQUAMOUS CELL CARCINOMA OF LIP****Khare M¹, Sharma U B², Airun A³ and Mohanty S⁴**^{1,2,3}Department of Pathology Jnuimsr Jaipur⁴Department of Plastic Surgery Jnuimsr JaipurDOI: <http://dx.doi.org/10.24327/ijrsr.2017.0806.0366>**ARTICLE INFO****Article History:**Received 16th March, 2017
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Published online 28th June, 2017**ABSTRACT**

Cystadenolymphoma is the second most frequent benign tumor of the parotid gland representing 6 to 10% of all tumors of the salivary glands. Multicentricity is more common in warthims tumor, however bilateral warthims tumors are much less common. Warthims tumor is very rare in the submandibular glands. Here we present a rare case of bilateral warthims tumor in submandibular glands associated with the squamous cell carcinoma of the lip

Key Words:

Warthin's tumor, bilateral submandibular gland, squamous cell carcinoma

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INTRODUCTION

Cystadenolymphoma is the second most frequent benign tumor of the parotid gland representing 6 to 10% of all tumors of the salivary glands.¹ Multicentricity is more common in warthims tumor compared to other salivary gland tumors.² Multicentric warthims tumors can show same or different morphological features, may occur unilaterally or bilaterally.³ However bilateral warthims tumors are much less common than unilateral tumors.^{3,4,5} Warthims tumor is very rare in the submandibular glands. Since Honda *et al.* showed that the epithelial tumor components are polyclonal cell populations these lesions cannot be classified as a true neoplasia anymore and are classified as tumor-like-lesions.⁶ In this case report we are presenting a rare presentation of the patient with squamous cell carcinoma with simultaneous bilateral warthims tumor in submandibular glands.

Case Report

A 50 years male presented with the growth on the lower lip since 6 months. The growth was looking malignant and a biopsy was performed from the lip, which on histological examination came out to be well differentiated squamous cell carcinoma. A CT scan head and neck of the patient was performed which showed a growth on the lip which was

infiltrating the mandible along with bilateral level I, II and III cervical lymphadenopathy.



Fig 1 Picture Showing Surgical Procedure with Complete Resection of Lower Lip and Complete Neck Resection

Surgery of the patient of lip resection along with partial mandibulectomy and complete neck dissection was performed.

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(fig 2) The specimens were send to the laboratory for the histopathological examination.



Fig 2 Picture Showing Specimen of Submandibular Gland with Tiny Grey White Areas

On gross examination, a irregular grey white growth was seen on the lip which was involving mandibular bone. Multiple lymphnode were identified in neck dissection specimens along with part of submandibular glands in level II lymphnode dissection specimen.(fig 2)

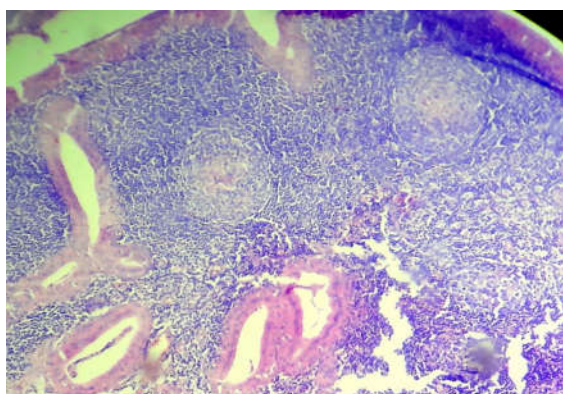


Fig 3 Photomicrograph Showing Low Power View of Warthins Tumor

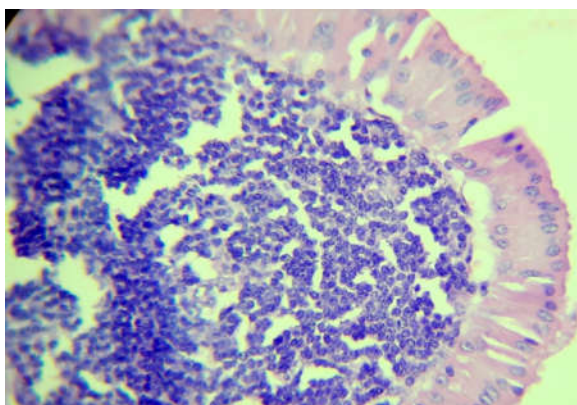


Fig 4 Photomicrograpp Showing Warthins Tumor with Bilayered Oncocytic Cell Linning and Lymphoid Tissue

Multiple sections from the tumor proper confirmed the diagosis of well differentiated squamous cell carcinoma. Bilateral level I and III neck dissection lymphnodes showed features of reactive lymphoid hyperplasia. Section from bilateral level II lymphnode dissecton showed similar features. Multiple

sections from the bilateral submnadibular saliavary glands showed features of warthims tumor along with normal saliavary gland parenchyma. (fig.3). Sections showed papillary structures lined by bilayered oncocytic cells and in between lying was the lymphoid tissue with many germinal centre. (fig 4)

DISCUSSION

Thus Warthin's tumors have their origin from the epithelial inclusions.⁷ This hypothesis puts Warthin's tumors in vicinity of lateral cervical cysts, which are thought to origin from proliferating heterotopic inclusions of tonsillar tissue in cervical lymph nodes. The hypothesis of heterotopia may explain the origin of salivary tissue in parotideal lymph nodes, in the periparotideal region and the upper neck, but cannot explain the origin of heterotopic salivary tissue in the lower neck lymph nodes. A relationship between a defective closure of the sinus of the his of the branchial apparatus with heterotopic salivary tissue was suggested by Youngs and Scofield as a possible reason for heterotopic salivary tissue of the lower neck.⁸ Other authors propose that aberrant salivary tissue of the pharyngeal pouch may migrate downwards with the thyroid and parathyroid gland.⁹

Most of these tumours occur in patients over the age of 60. The average size of the tumor at presentation varies from a few millimeters to centimeters, averaging 2 to 4 cm.^{10,11,12} A sudden increase of the tumor can indicate secondary infection in the location or the need to add another diagnostic hypothesis. Smoking seems to be a significant risk factor for development of Warthin tumours. Some Authors have correlated the amount of cigarette smoking with the bilaterality of the tumor.¹³ Some Authors have pointed out that the incidence of multifocal Warthin's tumour is probably underestimated since multiple Warthin's tumour with small extensions may escape histopathological evaluation if sampling is not adequate.^{2,3} More than 100 cases are reported for bilateral parotid gland warthims tumor but bilateral submandibular gland warthims tumor is very very rare.³

A detailed clinical history and physical examination can help in reaching the diagnostic hypothesis toward Warthin tumor. Additional tests such as ultrasonography may be useful in elucidating the content of the lesion. Computed tomography and magnetic resonance imaging may be requested in special situations, such as multiple tumors, suspicion of other neoplasms, and cases of relapse.¹¹ The treatment for bilateral tumours is the surgical approach, similar to that indicated for solitary tumours, i.e., partial gland excision. An alternative for partial excision is the extra-capsular tumour dissection (ECD).¹⁴ This surgical modality is based on meticulous dissection immediately outside the tumour capsule.

To conclude as per my knowledge, this is the first case presentation of well differentiated squamous cell carcinoma lip along with bilateral warthims tumor in the saliavary gland. Recent studies were able to proof that these tumors are no true neoplasia and consequently were classified as tumor-like-lesions.

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