COEXISTENCE OF TUBERCULOSIS WITH WARTHIN TUMOR OF SUBMANDIBULAR SALIVARY GLAND: A CASE REPORT

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ABSTRACT

The concomitant occurrence of tuberculosis infection within a Warthin tumor of submandibular salivary gland is extremely rare as only one case has been previously reported to best of our knowledge. Warthin tumor is an exclusive tumor of the parotid gland and involvement of submandibular gland is very rare. Tuberculous infection of the salivary gland is also very rare. We report a case of a 50 yr old male who presented with a gradually increasing swelling of left upper neck. Surgical excision was done. Histopathology revealed warthin tumor with multiple epitheloid granulomas with caseous necrosis and the patient was treated with antituberculous drugs.

Key Words: Warthin tumor, Tuberculosis, Submandibular gland

INTRODUCTION

Primary salivary gland tumors account for less than 2% of all neoplasms in human.¹ Approximately 10% of all salivary gland tumors are localized in the submandibular gland of which 40% are malignant tumors.² Warthin's tumor forms 15% of epithelial salivary gland neoplasms and is almost exclusively a tumor of the parotid gland.¹ Involvement of submandibular salivary gland is rare.

Tuberculosis is the biggest health issue that lies around India. To prevent spread of tuberculosis, it is important to get treatment quickly and to follow it through to completion. Extrapulmonary tuberculosis is not at all uncommon in India and account for approximately 20% of overall active tuberculosis.³ Tuberculous lymphadenitis is the most common extrathoracic form. The cervical lymph nodes, including lymph nodes in and around the salivary glands are the ones most frequently involved.³ Salivary glands appear to be rarely infected.³

CASE REPORT

A 50-year-old male presented with a gradually increasing left neck swelling (level II) since last 2 months. He gave history of night sweats and weight loss but no history of any pulmonary symptoms. On Local examination there was a swelling in the left submandibular region of size 5 x 3 cm, firm, mobile, non tender. Routine laboratory test and X-ray chest findings were normal. The mass was excised and sent for histopathology.

Gross: Received a single globular tissue mass measuring 4.5 x 3 x 2 cm with a smooth external surface. Cut surface was solid grey white with few cleft like spaces & with no obvious calcification. Tiny areas of caseous necrosis were noted.

Microscopy: Revealed a tumor composed of cystic spaces separating the lobules of neoplastic epithelium consisting of double layer of eosinophilic epithelial cells based on a prominent lymphoid stroma, some with germinal centre. At places within the stroma areas of caseous necrosis were noted surrounded by epitheloid cells, multinucleated langhans type giant cells and lymphocytes.

DISCUSSION

In 1929, Aldred Warthin first described Warthin tumor in the American literature, he named this tumor papil-
lary cystadenoma lymphomatous, but since then it was also known as adenolymphoma, cystadenolymphoma, and Warthin tumor. Warthin tumor is commonly present in the sixth or seventh decade of life and has a definite male predominance. Warthins tumor which is exclusively found in parotid gland also has certain other sites of occurrence like lateral neck, palate, upper lip, nasopharynx, submandibular gland, sinuses, lacrimal gland.

Some think that this tumor develops from the epithelial cell rests within the intraparotid lymph nodes on heterotopic salivary gland, while some consider that it is an adenoma with lymphocytic infiltration. Recent molecular studies have shown that the epithelial component is polyclonal and does not exhibit clonal allelic losses, suggesting that this tumor is not a true neoplasm.

Warthin tumor has an epithelial component and a lymphoid stroma. The epithelial cells, the oncocytes, are disposed on two layers, a luminal layer of oncogenic columnar cells, supported by a layer of oncogenic basal cells. The nuclei of the luminal cells appear uniform and display palisading towards the free surface. The basal cells posses small, centrally located, round to oval nuclei with conspicuous nucleoli. The oncocytes cytoplasm is granular and eosinophilic due to accumulation of mitochondria. The lumen of the cysts contains thick proteinaceous secretions, cellular debris, cholesterol crystals, and sometimes-laminated bodies that resemble corpora amylacea. The cellular composition of the stroma is largely that of small lymphocytes with only few medium to large lymphocytes. The ratio of B & T cells is 0.8:1.

Von Stubenrauch in 1894 first described tuberculosis of salivary gland. More than 50% of extrapulmonary tuberculosis cases occur with no pulmonary disease. There is increased incidence of isolated head and neck tuberculosis. Salivary glands are relatively immune to tuberculosis because of thiocynates ions and proteolytic enzymes like lysozymes, which impart antibacterial property. Continuous flow of saliva which prevents lodging and growth of mycobacteria is also an important inhibitory factor. In majority of the cases of tuberculosis involving the salivary glands, the parotid is the commonest gland to be affected. Involvement of salivary gland is by first, a focus of mycobacterial infection in the oral cavity that liberates the mycobacterium which ascend into the salivary gland via its duct or pass to its associated lymph nodes via lymphatic drainage. The second pathway involves hematogeneous or lymphatic spread from a distant primary lung focus.

Primary tuberculosis of salivary gland may occur in two forms as an acute inflammatory lesion (mimicking acute suppurative sialadenitis) or as a chronic mass (tumor) lesion that may be asymptomatic for many years. The typical histology of tuberculosis presents as granulomas with caseous necrosis, epitheloid cells and Langerhans cells. Due to low concentrations of organisms in the tissue, acid-fast bacillus may not be demonstrable.

In 1959, the first case of tuberculosis associated with Warthin tumor was reported. The tumor presents as a slow growing nodular, painless mass, firm or fluctuant at palpation.

Coexistence of warthins tumor with tuberculosis has been reported but mostly of the parotid gland as in Sieferts’ series and in Sugoul’s series one case each. Wen & Chen have reported a single case of tuberculosis with Warthin tumor in parotid gland in an 81 yr old male. Watanabe has reported two cases of tuberculosis with warthin tumor one in parotid gland in a 75yr old female and other in submandibular gland in a 78 yr old male. Ozcan has reported one case of tuberculosis co-existent with warthin tumor of parotid gland in a 53 yr old male. Ulusan M et al has reported a case in a 41 yr old male. The majority of cases of coexistence of tuberculosis with warthin tumor have been reported in parotid gland and only one case of submandibular gland involvement has been reported by Watanabe et al.

**CONCLUSION**

Warthin tumor considered to be exclusively involving parotid gland should be considered in the differential diagnosis of submandibular gland tumors and a coexistent disease in a tumor must be kept in mind during histopathological examination, especially in a developing country like India where the incidence of tuberculosis is very high. It is important to give treatment for tuberculosis quickly and to follow it through.

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**CONFLICT OF INTEREST**

There are no conflict of interests.
AUTHORS CONTRIBUTION:

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REFERENCES

Figure 1: Gross photograph showing grayish white cut surface with cleft like spaces and necrosis

Figure 2: Microscopy showing warthin tumor and granuloma with central caseous necrosis.