ABSTRACT
Warthin tumor was first described in the American literature, by Aldred Warthin, in 1929, the pathologist who named this tumor papillary cystadenoma lymphomatosum, but since then it was also known as adenolymphoma, cystadenolymphoma, and Warthin tumor. Because of its microscopically appearance and unknown origin, this tumor entity is still fascinating head and neck surgeons and pathologist. The etiology of Warthin’s tumors is controversial and whether they are true neoplasms or developmental malformations continues to be debated. Although this is the second most common benign tumor of the parotid, neither we nor our colleagues were familiar with it. Furthermore, we could find no mention of it in the family medicine literature. The following case illustrates a 50 year old male patient reported to our department with chief complaint of huge swelling on right side of face.

KEYWORDS: Warthins Tumor, Pleomorphic adenoma, oncocytic layer.

INTRODUCTION
The term “Warthin tumor”, named after Aldred Warthin, the pathologist who published the first two reports in the American literature in 1929, is the most commonly used term in order
to avoid confusion with malignant lymphomas, though the tumor is also known as adenolymphoma or papillary cystadenoma lymphomatosum.[1]

Warthin’s tumor is a controversial entity of benign salivary gland tumors because of its histopathological appearance and unknown origin. Histologically, the tumor is composed of bilayered oncocytic and basaloid epithelium forming cystic structures, papillae and glands that are accompanied by a dense lymphoid stroma. The lymphoid stroma becomes an issue when Warthin tumor’s origin is been discussed. Some authors believe that this tumor develops from the epithelial cells remnant inside the intraparotid lymph nodes within heterotopic salivary gland; other thinks that is an adenoma with lymphocytic infiltration. The most accepted hypothesis about the origin of Warthin’s tumor is that it develops from salivary ducts inclusions in the lymph nodes, after the embryonic development of the parotid gland, the stromal component is the lymph node.

This hypothesis is further supported by the frequent detection of salivary gland tissue in the peri- and intraparotid lymph nodes (Warthin’s tumor is almost exclusively located in the parotid region.

In the parotid region, lymph nodes were noted oncocytic and papillary changes, and the tumors presenting epithelial differentiations similar to those observed in Warthin tumors develops outside lymph nodes and has no lymphoid stromal component. With regards to luminal cells of tumor lining, the lymphoid stroma and cells reveal a similar aspect to the striated ducts of the normal salivary glands and have numerous mitochondria.[2]

These cells, called oxifile or oncocytic cells are swollen epithelial cells, with abundant eosinophilic granular cytoplasm, rich in mitochondria and enzymes. An increased number of oncocytic cells are also observed in the normal salivary glands once the person is getting older. The diffuse proliferation of the oncocytes without other changes has no pathologic significance and is called oncocytosis or oncocytic metaplasia.

**CASE REPORT**

A biopsy specimen from a private clinic was received, and the dentist was consulted for the history. On consultation the history was as a 52 years old male patient with the chief complaint of swelling in lower right side of face since 1 year. Patient was apparently alright 1 year back, and then he experienced swelling in mandibular right side. The swelling was pea
nut in size which gradually increased to the present size. Swelling was not associated with pain or discharge.

No contributory medical & dental history was relevant. He had habit of occasional tobacco chewing since past 15 years. Patient used to keep tobacco (powdered form) in the buccal vestibule for around 15-20 minutes prior to chewing. On extra-oral examination a circumscribed nodular swelling was noticed below pinna of ear, extending superio-inferiorly from base of the pinna till angle of mandible, mesially 1cm laterally from corner of the mouth, distally till angle of mandible, measuring about 4x4x3 cm in its greatest diameter, oval in shape with well defined borders, surface texture was smooth, firm in consistency, and free to underlying structure. Based on the clinical findings, a provisional diagnosis of pleomorphic adenoma was made. Excisional biopsy for the same was done.

On macroscopic examination, the specimen was brownish white in colour, roughly oval in shape, measuring about 5x3.5x4 cm in diameter and firm in consistency. The cut surface shows glandular area, lumen and capsule. Figure 1.

The histopathologic findings reveals lumen lined by epithelium and fibrous connective tissue stroma with numerous lymphoid follicle. Epithelium is lined by double layer of cells. Luminal cells were columnar showing reversal of polarity and other was oncocytic layer containing oncocytes. At places goblet cells were also appreciated. Figure 2,3,4,5. Histopathological diagnosis was made as Warthin’s tumor.

**Legends for figure**

Figure 1: On macroscopic appearance of excisional biopsy specimen was brownish white in colour measuring about 5x3.5x4 cm in diameter, firm in consistency. The cut surface of gross shows glandular area, lumen and capsule.

Figure 2: Low power of H & E stained section reveals lumen lined by epithelium and connective tissue stroma is fibrous with numerous lymphoid follicle.

Figure 3: Low power view of H & E stained section reveals lumen lined by epithelium surrounded by fibrous capsule with numerous lymphoid follicle.

Figure 4: Low power view of H & E stained section reveals lumen lined by epithelium surrounded by numerous lymphoid follicle.
Figure 5: At high power view H and E stained microphotograph showing epithelium is lined by double layer cells. One is columnar cell with reverse polarity and oncocyte layers containing oncocytes with few places goblet cells.
DISCUSSION

Warthins tumor, also known as papillary cystadenoma lymphomatosum, is a fairly common tumor. It makes up 14% to 30% of parotid tumors and is well known among otolaryngologists. The case described above is typical of Warthins tumor. The patient’s age, smoking history, lack of symptoms, and delay in presentation are all typical. Warthins occasionally occurs in young patients, most studies have found an average age at diagnosis in the early 60's. Warthin’s tumors most commonly present as an asymptomatic, slowly growing mass usually affecting men in the 5th and 6th decade. The male to female ratio ranges from 2.6:1 to 10:1.6.

Teymoortash et al. in 2005 reported the male to female ratio was 3.3:1. The comparatively significantly greater tumor incidence in men might indicate a hormone dependence of this disease. This is in accordance with our case in which the tumour was diagnosed in the male patient of 52 years.[4]

Warthin’s tumor is the most common bilateral and multifocal parotid neoplasm. In about 4–10% of Warthin’s tumors, there is bilateral tumor development, which is commonly metachronous. On contrary, the present case showed unilateral involvement of parotid gland. In about 4% of the cases multiple Warthin’s tumors may be observed in one parotid gland.

Warthin tumors are described as having a monotonous aspect, but Seifert G observed a variable quantitative rapport between the stromal and epithelial component. The relative proportions of epithelial and lymphoid components in Warthin tumors vary.

Seifert G recognizes four subtypes: Subtype 1 (classic Warthin tumor) in 50% epithelial (77% of all Warthin tumors); Subtype 2 (stoma poor) is 70–80% epithelial (14% cases); Subtype 3 (stroma-rich) is only 20–30% epithelial (2%); and Subtype 4 is characterized by extensive squamous metaplasia.

The present case correlates with the Subtype 1 having an epithelial component alongwith lymphoid stroma. Cystic spaces containing eosinophilic homogenous material were also noted in conjunction with orderly arranged bilayered, luminal columnar cells & basally cuboidal cells. Lymphocytic infiltrate is of greater proportion than the epithelium seen.

Numerous theories have been proposed for this tumor. Currently accepted theory is that tumor arises in salivary gland tissue entrapped within paraparotid or intraparotid lymphnodes.
during embryogenesis. Allegra et al (2008) suggested that Warthin’s tumor is more likely a delayed hypersensitivity disease, the lymphocytes being an immune response to salivary ducts which undergo oncocytic change. Hsu and coworkers recently studied the tumor immunohistochemically and have suggested that the lymphoid component of the tumor is an exaggerated secretory immune response.\[^5\]

The epithelial component can undergo metaplastic changes to squamous, mucous or even ciliated cells, especially in response to inflammation or infarction. Sometimes the tumor undergoes infarction, either spontaneously or following fine-needle aspiration, and the tumor cells can be obscured by necrosis, granulation tissue, inflammatory reaction, and fibrosis. Worse still, cellular atypia and a pseudoinfiltrative appearance of the metaplastic squamous epithelium in the residual tumor often invite an erroneous diagnosis of squamous cell or mucoepidermoid carcinoma.

With regard to the differential diagnosis, Warthin’s tumor, in general, has a highly distinctive morphology and poses no problem in diagnosis. It differs from oncocytoma in the presence of a prominent lymphoid component, papillae and glands rather than trabeculae and packets and conspicuous basal cells. Squamous metaplasia of Warthin tumor usually lacks keratinization, which is seen in most squamous cell carcinoma.

In contrast to low-grade mucoepidermoid carcinoma, there is no definite infiltrative growth and the tumor cells appear more frankly squamous. Lack of true infiltrative growth into the surrounding parenchyma and merging of the atypical squamous islands with oncocytic epithelium should point to the correct diagnosis.

**CONCLUSION**
Warthin’s tumor remains one of the most histologically recognizable salivary gland tumor and is a classical example of those tumor in which luminal and non luminal cells can differentiate. The columnar oncocytic cells, papillary and cystic growth pattern and lymphocytic component provide the neoplasm with distinctive histopathology.

**REFERENCES**

