Turgut Özal Tıp Merkezi Dergisi **6(3):1999**

WARTHIN TUMOR (PAPILLARY CYSTADENOMA LYMPHOMATOSUM) OF THE SUBMANDIBULAR GLAND

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Papillary cystadenoma lymphomatosum , adenolymphoma or with its more frequently used name, Warthin tumor is an unusual type of salivary gland tumor which occurs almost exclusively in the parotid gland. The most widely accepted histogenesis is from salivary gland tissue entrapped within paraparotid or intraparotid lymph nodes during embryogenesis. We present a case of this peculiar tumor diagnosed in a 20 year old woman in the submandibular gland region.

Key words: *salivary gland tumor, Warthin tumor, adenolymphoma, submandibular gland.*

Submandibular Gland Warthin Tümörü (Papiller Kistadenoma Lenfomatozum)

Papiller kistadenoma lenfomatozum, adenolenfoma veya daha sık kullanılan ismiyle Warthin tümörü büyük çoğunlukla parotisde oluşan olağandışı bir tükürük bezi tümörüdür. En sık kabul edilen histogenez embriyonik hayatta parotis çevresi veya içindeki lenf düğümlerinde kalmış olan tükürük bezi dokusundan geliştiği yönündedir. Bu değişik tümörün bir örneğini 20 yaşında bir kadında submandibular gland yerleşimli olarak sunmaktayız.

Anahtar kelimeler: *Tükürük bezi tümörü, Warthin tümörü, adenolenfoma, submandibular bez*

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Tumors of the salivary glands are relatively uncommon and constitute a heterogeneous group of lesions with great morphologic variations.[1-3] Papillary cystadenoma lymphomatosum , adenolymphoma or with its much frequently used name Warthin tumor is almost exclusively seen in the parotid gland and constitutes about 4 - 15 % of the tumors of major salivary glands with occasional bilateral or multifocal tumor locations. The minor salivary glands are even more rarely involved with few cases being

reported in the literature.

The abundant lymphoid tissue which accompanies the epithelial component has resulted in a widely accepted hypothesis of histogenesis from paraparotid or intraparotid lymph nodes. In this report we describe a peculiar case of Warthin tumor located in the submandibular gland region in a 20 year old woman

CASE

A previously healthy non-smoker woman noticed a slowly growing mass located in the right submandibular region during the last six months. She had no additional complaint or symptom other than this mass. Physical examination revealed a freely movable smooth contoured solid mass with dimensions of 5x2 cm measured at ultrasound. Additionally the ultrasound showed heterogeneous cystic and necrotic areas within the mass. The laboratory results were unremarkable. An aspiration was attempted and cytology of the very small amount of cyst fluid did not reveal any diagnostic cells. The mass was resected with an uneventful clinical outcome; the patient was discharged in a few days. The resected specimen consisted of a pink, tan nodular mass of 4x3x2 cm with smooth surface. There were multiple cystic spaces of varying size filled with semiviscous, mucoid fluid upon sectioning. The microscopic sections revealed an abundant lymphoid tissue with prominent follicles and active germinal centers. High columnar eosinophilic epithelial cells with a low nuclear cytoplasmic ratio (oncocytic cells) were also noticed in close juxtaposition to the lymphoid tissue, these cells were lining the cystic spaces with occasional papillary projections (Figure 1). In addition, a second layer of reserve cells cuboidal and centrally nucleated with a tendency to focal squamous differentiation were also noticed below the eosinophilic columnar epithelial cells forming two rows of cells lining the cystic spaces (Figure 2). The lymphoid component did not reveal any atypical proliferation. The capsular area was free of any infiltration and nearby

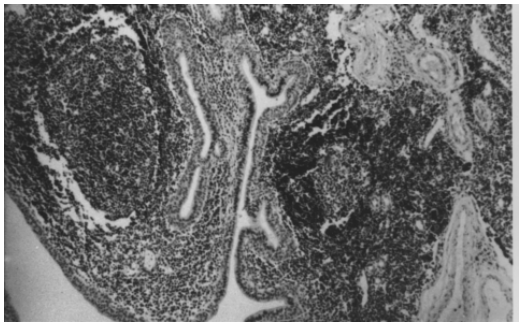

Figure 1. Low power view of the tumor showing prominent lymphoid tissue and the cystic spaces (H. E. 100 X)

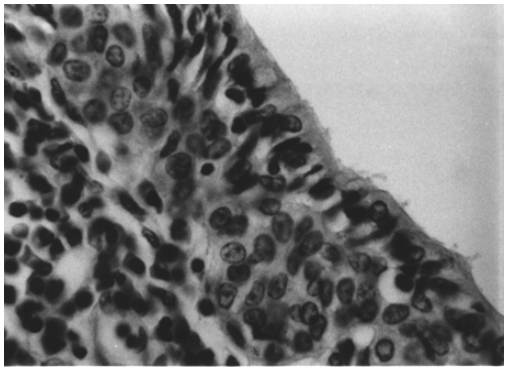

Figure 2. High magnification revealing the two rows of epithelial cells close to the lymphoid tissue. (H. E. X 400)

submandibular gland tissue was also noticed in the pericapsular area.

DISCUSSION

The uncommon Warthin tumor exhibits a definite predilection for men with an average age of presentation being after the fifth decade.[1-4] Recent studies have documented a high incidence among people of Asian origin excluding dark skinned groups again with a male predominance.[5] There is also a clear cut high risk for smokers.[6] The tumor is almost exclusively located in the parotid gland in a superficial location and rarely attains a size exceeding 3 to 4 cm in diameter. This tumor has two histologic components: lymphoid tissue and epithelial. The epithelial proliferation consists of columnar or cuboidal cells usually

aranged in two rows, the inner layer sometimes being several layers thick. These cells are eosinophilic with cytoplasmic mitochondrial hyperplasia and are referred as oncocytes. Warthin tumor may be divided into three groups in regard to the epithelial and lymphoid tissue components i.e. epithelial, so called classic and lymphoid types respectively.[7] Our case was heavily dominated by the lymphoid tissue component and could be classified as predominantly lymphoid. Morphometrical analyses of Warthin tumor suggested an initial adenomatous proliferation and secondary lymphoid infiltration.[7] This view, against a developmental defect hypothesis that we had mentioned above, it is not surprising to see this tumor in extra parotid locations. The treatment of choice is local surgical excision, since the lesion is encapsulated and recurrences are rare.[8] Malignant transformation is exceedingly rare in either the lymphoid or epithelial component. Our case is unique in the rare submandibular location and presenting in a young female. Pathological and clinical differential diagnosis in this case include branchial cysts, caseating tuberculous lymphadenitis, metastatic carcinoma to lymph nodes, lymphomas, carotid body paraganglioma and lastly salivary duct cysts.[1-3] The above mentioned entities were discarded by the histopathologic examination which revealed microscopic uniformity and orderly organization of the epithelial eosinophilic cells (oncocytic cells) with the lymphoid tissue typically described for Warthin tumor. An additional differential diagnosis in HIV positive patients nowadays must also include lymphoepithelial cysts.[1] Our patient did not have HIV positivity and the microscopic sections did not show the clear cut squamous component of these cysts.

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