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Case Report

Warthin's tumour in a young female

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ABSTRACT

Warthin's tumor is the most common monomorphic adenoma of the parotid gland. It can be coexistent with other salivary gland tumors or can be metachronous, multifocal, unilateral, bilateral which perplexes the diagnosis hence delaying or over/under treating the ailment. Rare cases of Warthin's tumor are described in young population especially young non-smoker females. We present a case of Warthin's tumor arising in the left parotid gland of 20-year-old female.

1. Introduction

Warthin's tumour (WT) / Papillary cystadenoma lymphomatosum (PCL) is the second most common benign salivary tumor after pleomorphic adenomas.[1] It is the most common monomorphic adenoma, accounting for 3% to 17% of all parotid gland tumors.[2] It was first delineated by pathologist Aldred Scott Warthin in 1929.[3]

WT occurs almost exclusively within the parotid glands, mainly in superficial lobe, occasionally in the deeper lobe (10%) and rarely in the submandibular gland or cervical lymph nodes, minor salivary glands of the buccal mucosa, hard palate, lip, and oropharynx.[4–7]

It presents as a painless, soft, and smooth mass. It can be occasionally multicentric (12%–20%), and bilateral in 5%–14% of cases.[8] Malignant transformation of WT is extremely rare and accounts for 0.3% of the cases.[9]

Etiologic factors of WT have been aforementioned to cover tobacco, Epstein Barr virus infection, autoimmune disease, ionizing radiation, and chronic inflammation.[10–12]

It almost never occurs in young women, peak incidence in females being in the 6th decade, whereas it is in the 7th decade in men.[13] There is an apparent male predilection for the occurrence of WT.[14]

Histologically WT shows multiple cysts that have numerous papillations covered by bilayered columnar and basaloid oncocytic epithelium. The connective tissue portion shows proliferation of follicle- containing lymphoid tissue which necessitates careful distinction for diagnosis.[14]

Being a common tumor it is still considered distinctive because of its histological appearance and unknown origin and pathogenesis.

2. Case Report

A 20- year- old female with no history of substance abuse, presented with soft, painless swelling on left infra-auricular region whose size altered on chewing food. On examination a swelling was palpable in the left parotid region measuring 2.5 x 2 centimetre. It was soft, had restricted mobility and non-tender. Overlying skin and temperature were normal. Neck nodes were not palpable. Based on the history and

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clinical examination, a provisional diagnosis of benign tumour, of salivary gland origin, was made. The patient didn't give consent for fine needle aspiration cytology of the lesion hence the otolaryngologist proceeded with imaging. With regular margins of the lesion and no enlarged neck nodes patient was put up to surgical removal of the parotid gland.

Gross specimen comprised of a partially cystic to solid greyish white to brown encapsulated mass measuring 3.5 x 3.0 centimetre.(Figure 1)

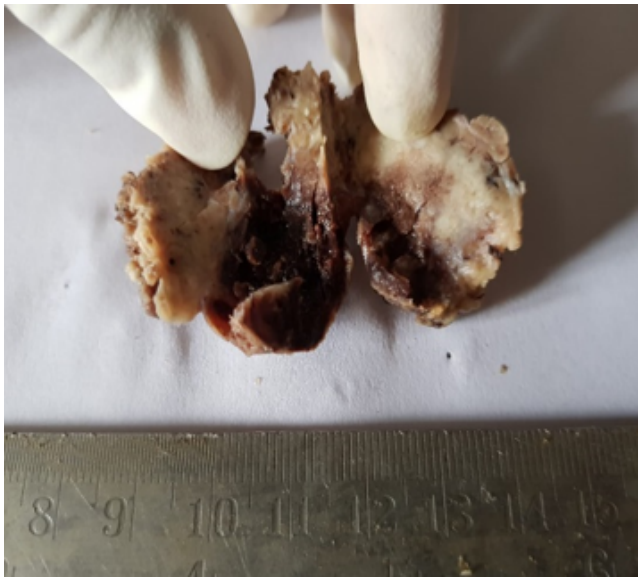

Fig. 1: Gross image

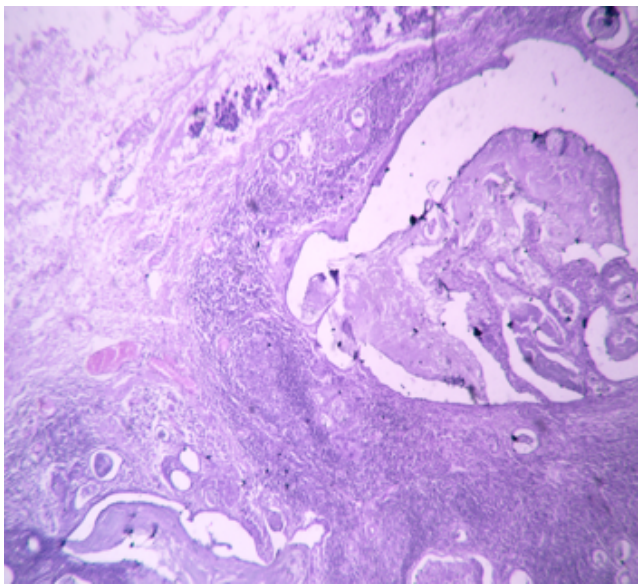

Fig. 2: Solid, Cystic Tumor With Lymphocytic Infiltrate. H& E 400X

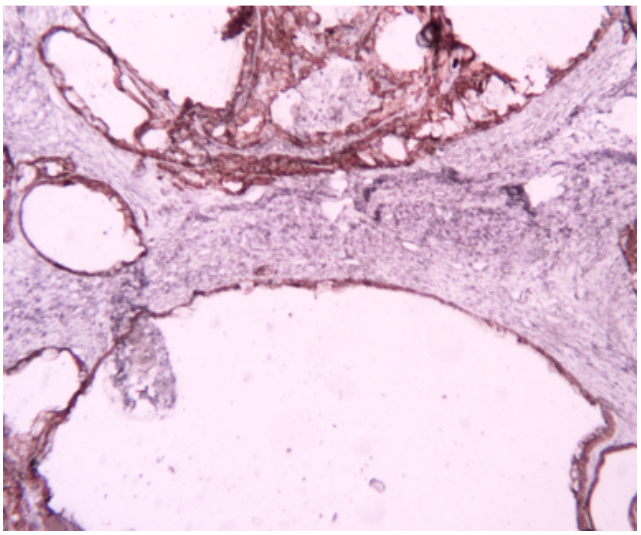

Fig. 3: IHC: p63 positive in myoepithelial cells

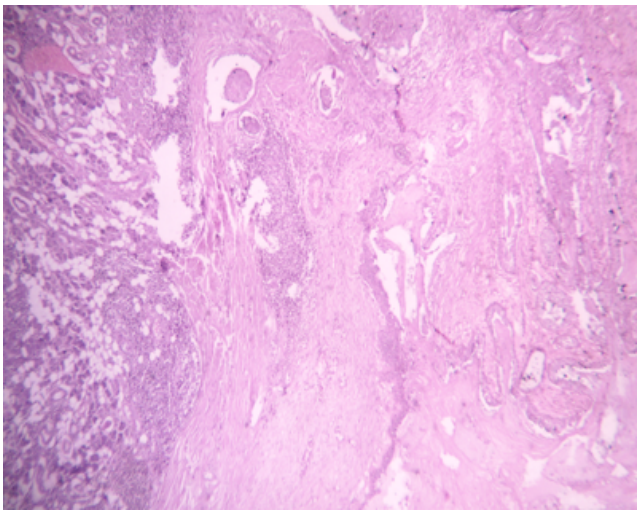

Fig. 4: H& E 400X (tumour arising from salivary gland)

Microscopically, a well encapsulated benign tumor comprising of cystic solid elements lined by double epithelial layer resting on lymphoid stroma with variable germinal centres was seen. Some oncocytic columnar cells palisading over basal layer and few papillary projections were noted.(Figures 2 and 4)

Immunohistochemistry showed p63 positive myoepithelial cells.(Figure 3) Diagnosis of Warthin's tumor was signed out.

3. Discussion

Salivary gland tumors are 2%–6.5% of all head and neck neoplasms occurring in both major and minor salivary glands, WT being the second most common arising most frequently in the parotid gland.[2–4]

WT usually presents after 40 years of age, with the mean age of diagnosis being 62 years. [2,4]

Most studies showed that WT is associated with cigarette smoking with a male predilection with male-to-female ratio up to 10:1 while aacording to later studies the difference has been on decline probably due to increased number of female smokers. [13,15,16]

Clinically, WT presents as a rounded /ovoid nodular painless, slow-growing, fluctuant to soft nodule. It can be unilateral, bilateral, or multicentric and is asymptomatic in 90% of cases.

On ultrasound, most tumors tend to be ovoid, with well-defined margins and multiple irregular, small, sponge-like anechoic areas. Tumors that are large (e.g., >5 cm) tend to have a higher proportion of cystic content than smaller lesions had and, in some cases, can be composed almost entirely of cystic material. [17]

Grossly WT is two to four centimetres on average, well-circumscribed spherical to oval mass. On cut section, there are solid areas and multiple cysts with papillary projections. [18] The cystic spaces often contain mucoid creamy brown or white fluid. [19] Aspiration cytology may suggest differentials of mucoepidermoid and adenoid cystic carcinoma, however both lack a prominent lymphocytic background.

Microscopically WT are composed of varying proportions of papillary- cystic structures lined by oncocytic epithelial cells and a lymphoid stroma with germinal centres. The epithelial component is formed of inner columnar and outer cuboidal cells.

Malignant transformation of Warthin's is suspected when there is

1. Transition from a benign oncocytic to a malignant epithelium.
2. An infiltrating growth in the surrounding lymphoid tissue.

The most frequent histological types of malignant transformation in a WT are mucoepidermoid carcinoma, squamous cell carcinoma, undifferentiated carcinoma, oncocytic adenocarcinoma, and adenocarcinoma.

The treatment for WT is primarily surgical, either with a superficial parotidectomy or enucleation of the tumor. [2,3]

Warthin's tumor has a favourable prognosis and with recurrence rate of 2%–5.5% in parotid WT, which is thought to be due to multifocality. [2]

4. Conclusion

This case presents WT at an unusual age and gender of presentation with no associated predisposing factors in the parotid. Clinicians ought to therefore include WT in their differential diagnosis of an infraauricular mass even in young females before surgical intervention as it is difficult to acquire the correct pre-operative diagnosis in unusual clinical scenarios of salivary gland tumors like these. Thus, surgery in cases like these with parotid gland neoplasm should be designed to remove the tumor completely with an adequate margin.

The definite diagnosis was achieved only after the histopathological examinations thus guiding further management of patient. Though the lesion is common, a greater number of incidences will help clinicians to understand the unusual presentations and pathology of Warthin's tumour in great more detail.

5. Source of Funding

None.

6. Conflicts of Interest

There is no conflict of interest.

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