EUROMEDITERRANEAN BIOMEDICAL JOURNAL

for young doctors

Case Report

FAMILIAL OCCURRENCE OF WARTHIN TUMOUR: OUR EXPERIENCE AND REVIEW OF THE LITERATURE

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Summary

The tumour of Warthin (WT) is the second parotid tumour for frequency after the pleomorphic adenoma, it represents about 2% to 15% of all parotid tumors. The pathogenesis of familial Warthin Tumour is unknown, even if cytogenetic abnormalities have been identified in Warthin Tumour. Among the inducing factors there is the smoke which has an important role. Familial occurrence of Warthin Tumour is rare, only five reports in the literature. Two of them were reported in brothers, two in a mother and son and one in monozygotic twins. We quote a case of familiar Warthin Tumour arisen in two brothers and we present a review of the Literature.

Introduction

The Warthin tumour, called also adenolymphoma, cystoadenolymphoma, oncocitoma owes its name to Warthin who in 1929 (1) checking the parotid tumours and the branchial cysts of the neck, found two cases and called them teratomas of the salivary glands. The elective seat is the parotid gland, in fact only in 2 – 5% of the cases it's the submandibular gland and in the 0,5% also extrasalivary structures. It is a benign cystic tumor of the salivary glands and its etiology is unknown, but there is a strong association with cigarette smoking.

In the latest two years, we have examined a case of Warthin tumour (WT) in a family, that consists of two brothers.

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Received: *18th June, 2016 — **Revised:** 22th July, 2016 — **Accepted:** 10th September, 2016*

ISSN 2279-7165 - Euromediterranean Biomedical Journal [online]

Case reports

Case 1

The younger brother is admitted to our hospital in April 2010. He smokes 20 cigarettes a day and he has no past medical history of note. About eight months ago he refers the appearance of a sidecervical tumefaction which gradually increased. Treatment consists of subtotal parotidectomy with preservation of the facial nerve in general anesthesia. No postoperative complications occurred. The hystological diagnosis is of cystoadenolymphoma (Figure 1).

Case 2

The older brother is admitted to our structure in March 2011 for a bilateral tumefaction at the parotid higher on the left side. The anamnesis revealed that he is a smoker too (20 cigarettes a day). The surgical procedure is a left superficial parotidectomy, defined by the resection of tumours located in the superficial portion of the gland (above the nerve). Contralateral parotidectomy is performed at different times, beginning on the side where the tumour is larger.

The post-operative course is regular. The hystological examination makes a diagnosis of WT (Figure 2).

Whereby, one year later because the right neck tumefaction became higher, a superficial parotidectomy was performed. The hystological exam makes again the diagnosis of WT (Figure 3).

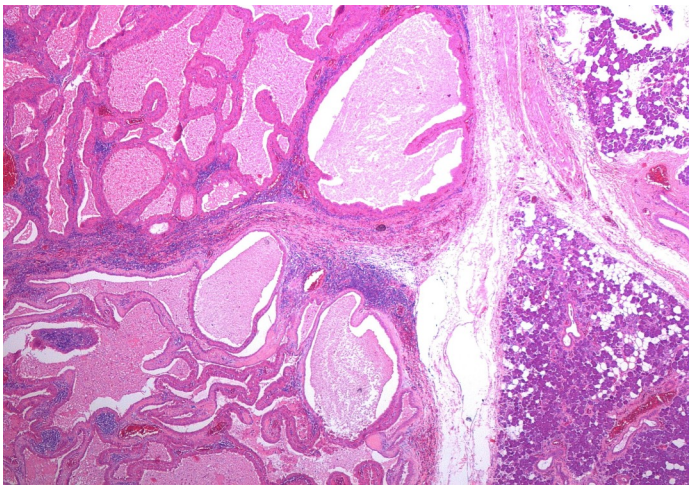

Figure 1: Hystological examination of case 1: cystoadenolymphoma

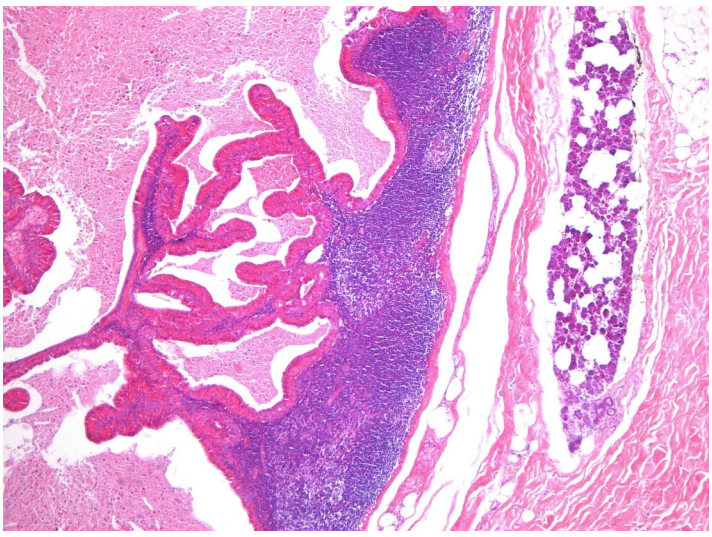

Figure 2: Hystological examination of case 2: whartin tumour

Discussion

The WT is the second parotid tumour for frequency after the pleomorphic adenoma, it represents about 2% to 15% of all parotid tumors (2, 3). Sometimes it is multiple, sometimes it is bilateral (whereas bilateral WTs are much more uncommon) (4, 5), more frequent in males than in females, the most affected age is between 50 and 70 years (6).

Among the inducing factors the smoke plays an important role (7).

The WT appears as an ovalar mass of hard elastic substance, mobile. The growth is slow, but can grow quickly for the breaking of the tumoral capsule which causes a leakage of serous or mucoid fluid being in the cystic spaces with the consequent infective reactions of the surrounding parenchyma. It can compress the facial nerve. Hystologically the tumour is formed by epithelial cells. The stroma contains a variable quantity of limphoid tissue. The mitosis are rare. The metaplasia would be caused by the carcinogenic substances being in the smoke or by nutrional or hormonal factors.

The pathogenesis of familial WT is unknown. Cytogenetic abnormalities have been identified in WT. A complex translocation involving t(11;19)(q21;p13) was detected in rare cases (6, 8), although when the fusion gene is present in this tumor type, it seems to be restricted to special cases with indeterminate morphology and may constitute a definitive subgroup. In contrast, cases showing reciprocal translocations involving the 6p region were also described, suggesting that the short arm of chromosome 6 contains a region consistently involved in the origin of WT (6).

Current reviews and conclusions

Familial occurrence of WT is rare, only 5 are reported in the literature. Two of them were reported in brothers (9, 10), two in a mother and son (10, 11) and in one case it is a monozygotic twins (6).

We report a case of familial WT arisen in two brothers, the older one has bilateral synchronous tumours. This feature joins our case with the one described by Skerlaway (10) in 1976 and strengthens the hypothesis that in the development of this neoplasia there are genetic factors. Besides the age of our brothers , between 45 and 55 years is almost similar to the one of the three familial cases quoted by Talmi (9). Both our patients are smokers , similarly to the cases quoted by Russo (8) confirming a role of the cigarette smoking as the literature data reports. Up to now the modest number of cases of familiar WT makes diffi-

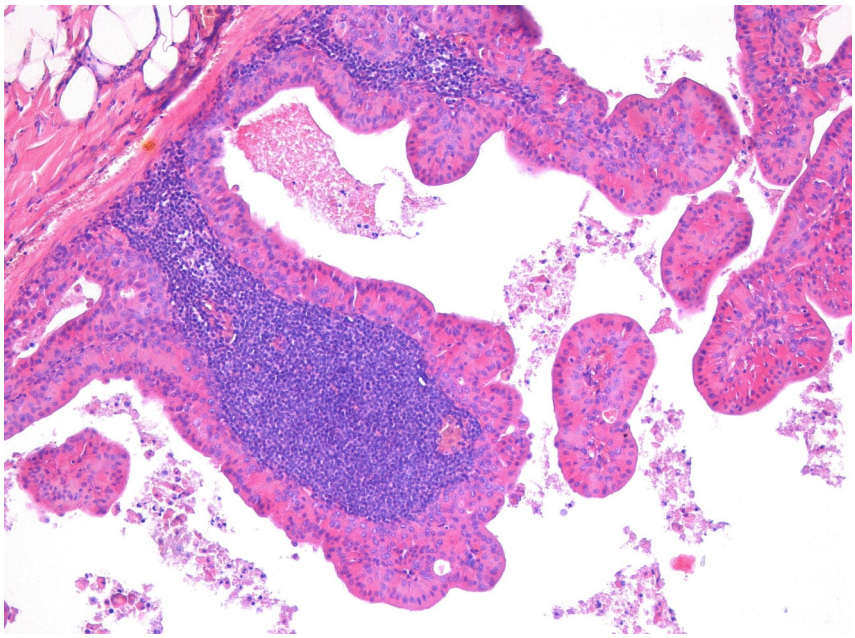

Figure 3: Second hystologicval examination of case 2: whartin tumour

cult the study and the identifying of a cause especially of genetic type. Anyway we may conclude that the beginning of the WT in the same family may be linked to the familiarity as exposure to the same risk factors and also to genetic abnormalities.

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