

CASE REPORT

# Synchronous bilateral Warthin's tumours of the parotid glands: a case report

## *Tumore di Warthin bilaterale sincrono della parotide: case report*

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### SUMMARY

Warthin's tumour is the most frequent monomorphic adenoma of the major salivary glands, representing about 2-15% of all parotid tumours. Most of the multifocal Warthin's tumours are unilateral, whereas bilateral Warthin's tumours are far less common; bilateral Warthin's tumours are metachronous with few synchronous cases having been described in the literature. The Authors present an interesting case of simultaneously occurring bilateral Warthin's tumours growing in the parotid glands.

KEY WORDS: Parotid gland • Salivary gland tumours • Warthin's tumour • Multifocal tumours • Bilateral tumours

### RIASSUNTO

*Il tumore di Warthin è il più frequente adenoma monomorfo delle ghiandole salivari maggiori, rappresentando dal 2 al 15% di tutti i tumori parotidici. La maggior parte dei tumori di Warthin multifocali sono unilaterali, mentre i tumori di Warthin bilaterali sono molto meno frequenti; i tumori di Warthin bilaterali sono in genere metacroni con solo pochi casi di tumori bilaterali sincroni descritti in letteratura. Gli Autori presentano un interessante caso di tumore di Warthin insorto contemporaneamente in entrambe le ghiandole parotidi.*

PAROLE CHIAVE: Parotide • Tumori delle ghiandole salivari • Tumore di Warthin • Tumori multifocali • Tumori bilaterali

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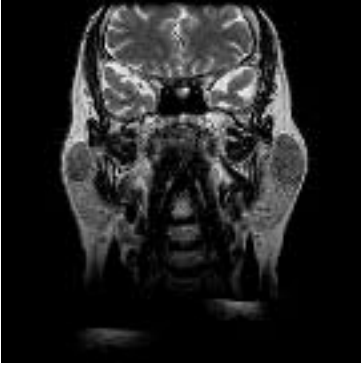
## Introduction

Salivary gland tumours represent approximately 3% of all head and neck tumours. About 70-80% of these neoplasms occur in the major salivary glands, with the parotid gland being the site most often affected<sup>1-3</sup>. Warthin's tumour is the most frequent monomorphic adenoma of the major salivary glands, representing about 2-15% of all parotid tumours<sup>3,4</sup>. Salivary gland neoplasms are usually solitary while multiple tumours occurring simultaneously in an individual are uncommon. Albeit, there is evidence that multicentric Warthin's tumours are more common than any other salivary gland tumour; in fact, about 12% of patients develop multiple tumours<sup>5</sup>. They may display identical or different morphological features and may occur unilaterally or bilaterally, metachronously or synchronously<sup>6</sup>. Most of the multifocal Warthin's tumours are unilateral, whereas bilateral Warthin's tumours are much more uncommon; bilateral Warthin's tumours are metachronous with few synchronous cases having been described in the literature<sup>4,6,7</sup>. However, the true incidence and implications of multiple synchronous and metachronous tumours remain to be defined.

In the present report, an interesting case of a simultaneously occurring bilateral Warthin's tumour growing in the parotid gland is described.

## Case report

A 62-year-old female patient was admitted to the Maxillofacial Surgery Unit with bilateral swellings of the parotid gland which had been present for one year. Apart from the habit of smoking about 20 cigarettes a day, for 30 years, her medical history was normal. Clinical examinations confirmed bilateral mobile, non-tender swelling, with normal intra-oral examination. Fine-needle aspiration biopsy (FNAB), under ultrasonography (US) guidance, was carried out on both nodules and was indicative of Warthin's tumour. The magnetic resonance imaging (MRI) examination revealed two bilateral nodular lesions of the parotid glands, well defined and rounded in appearance (Fig. 1). The right nodule measured 3 cm in diameter, while the left nodule measured 3.5 cm. On the basis of these data the patient underwent surgical treatment: local excision (enucleation) of the two neoplasms at the same time. Meticulous intra-operative inspection and pal-



**Fig. 1.** Pre-operative magnetic resonance image.

pation of the glandular tissues and peri-glandular lymph nodes were performed in order to exclude the presence of other small neoplasms undetected at MRI. The histological examination confirmed the pre-operative diagnosis of Warthin's tumour, with no evidence of malignancy. The patient has been regularly monitored at follow-up and there have been no recurrences in 5 years.

## Discussion

Warthin's tumour is an adenoma comprising 4-11% of all tumours, up to 27% of which are benign, and as many as 34% of parotid salivary gland tumours, making it the second most common after pleomorphic adenomas. Most of these tumours occur in patients over the age of 60. Smoking seems to be a significant risk factor for development of Warthin tumours. Some Authors reported that the risk of bilateral Warthin tumours correlated significantly with the amount of nicotine intake<sup>8</sup>.

In patients with bilateral parotid tumours, the histopathology is primarily Warthin's tumour. More than 100 cases have been reported in the literature<sup>6</sup> and most of the bilateral Warthin's tumours are metachronous. Multiple occurrence of Warthin's tumour seems to be related to its histological origin. There is no intra-glandular compartmentalization between the epithelial and lymphoid components in the early stages of embryogenesis of the pa-

rotid gland. Epithelial cells, the precursors of the salivary duct-acinar system, may remain embedded within the lymphoid component. These lymphoid components will later constitute the intra-glandular lymph nodes. These epithelial inclusions may give rise to Warthin's tumour following an unknown tumourigenic event<sup>4</sup>.

Multiple lymph nodes are present in the tail of the parotid gland, therefore, bilateral or unilateral multiple Warthin's tumours are more likely to develop in this part of the gland.

Since Warthin's tumours can be multifocal, a pre-operative diagnosis by means of FNAB is mandatory and complete bilateral screening of the gland by MRI is needed to programme surgery.

The treatment for bilateral tumours is the surgical approach, similar to that indicated for solitary tumours, i.e., partial parotidectomy when it occurs in the superficial lobe, or total parotidectomy with the identification, dissection, and preservation of facial nerve branches when located in a deep lobe. An alternative for superficial parotidectomy, in the form of extra-capsular tumour dissection (ECD), can be performed, as in the patient described here. This surgical modality is based on meticulous dissection immediately outside the tumour capsule with preservation of the facial nerves<sup>9</sup>.

Some Authors have pointed out that the incidence of multifocal Warthin's tumour is probably underestimated since multiple Warthin's tumour with small extensions may escape histo-pathological evaluation if sampling is not adequate<sup>4,5</sup>. This is the reason why a meticulous macroscopic examination of the parotid is indicated, being focused on the tail of the gland, when the gland is completely removed.

In view of the possible association of Warthin's tumour with extra-salivary neoplasms, extensive work-up of the patients harbouring multiple Warthin's tumours is, therefore, indicated and long-term follow-up is mandatory, due to the possible occurrence of metachronous salivary and extra-salivary tumours even after prolonged time intervals<sup>10,11</sup>.

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