CASE SEIRES AND REPORTS

Simultaneous nasopharyngeal and parotid gland Warthin's tumour: a case report

Un caso raro di tumore di Warthin sincrono della ghiandola parotide e del rinofaringe

S. PELUCCHI[1], C. BIANCHINI[1], A. CIORBA[1], F. STOMEO[1], A. FERRON[2], A. PASTORE[1]

[1] Otorhinolaryngology Department, and [2] Section of Anatomy Histology and Pathological Cytology, University Hospital of Ferrara, Italy

SUMMARY

Herein, a rare case of synchronous cystoadenolymphoma (Warthin's tumour) of the right parotid gland and the nasopharyngeal space is described. Although Warthin's tumour (WT) of the parotid gland is a common benign pathology, the occurrence of extra-parotid cystoadenolymphoma is rare. Extra-parotid WT have been mainly localised in the submandibular gland, periparotid region and occasionally in other sites, such as the oral cavity, hard palate and nasopharynx. The simultaneous occurrence of an intra-parotid and extra-parotid WT localisation, as in the case presented, is extremely uncommon.

KEY WORDS: Warthin's tumour • Parotid Gland • Nasopharynx

RIASSUNTO

Obiettivo del presente lavoro è descrivere un raro caso di cistoadenolinfoma (tumore di Warthin) sincrono della ghiandola parotide destra e del rinofaringe. Il tumore di Warthin della ghiandola parotide è una neoplasia benigna relativamente comune; più raro è il suo riscontro in ambito extra-parotideo (per lo più, a livello della ghiandola sottomandibolare, del cavo orale o del rinofaringe). Molto più raro è poi il riscontro di due localizzazioni simultanee, una intra-parotidea ed una extra-parotidea, come nel caso qui descritto.

PAROLE CHIAVE: *Tumore di Warthin • Ghiandola Parotide • Rinofaringe*

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Introduction

Warthin's tumour (WT) is a benign salivary gland tumour, almost exclusively located in the parotid gland and periparotid region [1]. From 12-19% of patients develop more than one WT, frequently with a bilateral parotid involvement (incidence: 5-14%) [1]. WT has been rarely reported to occur at multiple sites and at the same time; to the best of our knowledge, in the literature there is only another case report of a simultaneous intra-parotid and extra-parotid WT [2].

Case report

A 63-year-old man was referred to the ENT department of the University Hospital of Ferrara for the assessment of a right, non-painful, parotid mass. The lesion had been present for 4-6 months, but was increasing in size very slowly. He was also complaining the onset of snoring since 6 months with occasional nasal discharge.

Apart from a 40-year habit of smoking 20 cigarettes a day and the onset of a non-insulin dependent diabetes mellitus two years ago, his medical history was unremarkable.

ENT examination revealed a mobile 3.5 × 2.0 cm mass just behind the angle of the jaw (right side). Facial nerve function was normal. Nasopharyngeal endoscopy revealed an oval mass (1.0 cm) with a smooth surface in the left side of the nasopharynx. There were no other notable findings on physical examination.

Fine needle aspiration of the right parotid mass was performed. Histological examination revealed mixed lymphoid cells (lymphocytes and macrophages). The exact anatomical location of both lesions was confirmed by MRI (Fig. 1A-B).

Under general anaesthesia, the parotid mass was excised, via a superficial parotidectomy; at the same time, the nasopharyngeal mass was removed under FESS guidance. At histological examination, a synchronous parotid and nasopharyngeal WT was diagnosed (Figs. 2, 3). There have been no signs of tumour recurrence at 16 months postoperatively.

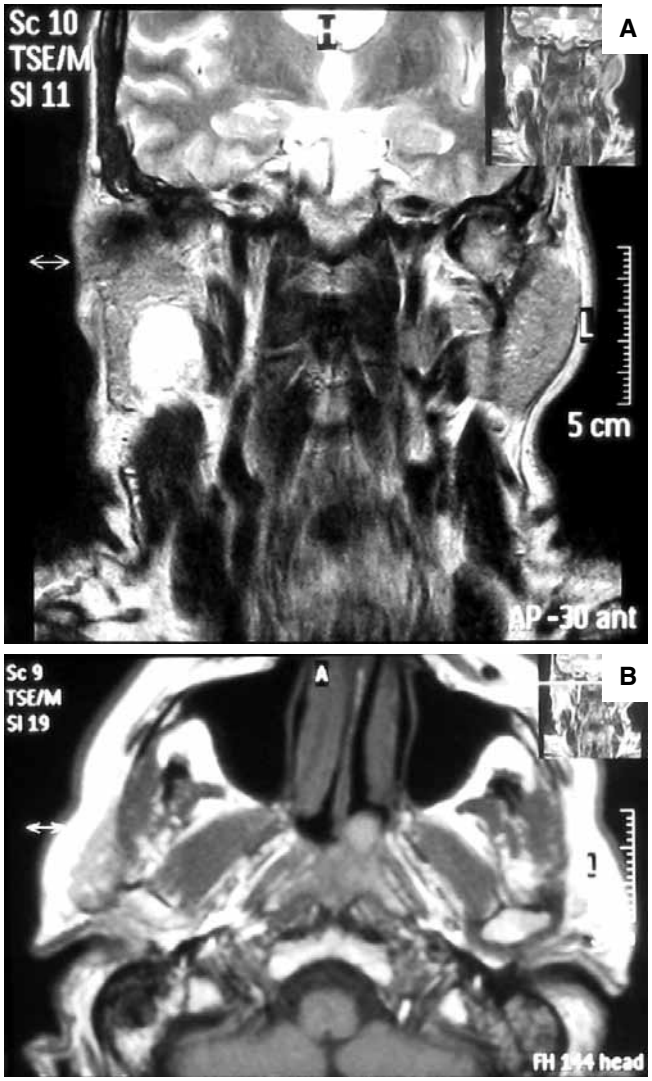

Fig. 1. A. T1 weighted MRI scan, coronal section: a 3.5 × 2.0 cm mass is located in the superficial lobe of the right parotid gland. B. T1 weighted MRI scan, axial section: an oval mass of about 1.0 cm in diameter is located in the left side of the nasopharynx.

Discussion

Warthin's tumour (adenolymphoma, papillary cystadenoma lymphomatosum, cystoadenolymphoma) is one of the most common benign salivary gland tumours, generally involving the parotid gland [1-3]. Even if benign tumours of the salivary glands are more common in women, WT, on the other hand, is found more frequently in men between the ages of 55 and 70 years. An association with cigarette smoking has been described [3-5].

Although some cases have been reported in extra-parotid locations such as the cervical lymph nodes, submandibular gland, lip, cheek, tongue and hard palate, WT of the nasopharynx are extremely rare [2-10]. Only few isolated cases of primitive nasopharyngeal involvement have been described so far, and we could find (through a PubMed database search performed in January 2011) only one published case of a nasopharyngeal WT with a simultaneous associated parotid tumour [2]. A synchronous intra-parotid and extra-parotid WT, as in our case, can therefore be considered exceptional.

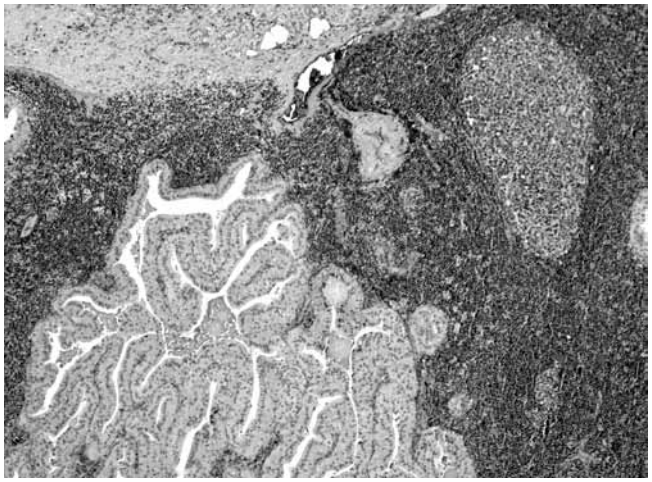

Fig. 2. Parotid lesion showing the classic histological pattern of Warthin's tumour: bilayered oncoctic epithelium and lymphoid stromal tissue. Haematoxylin-eosin, magnification 20×.

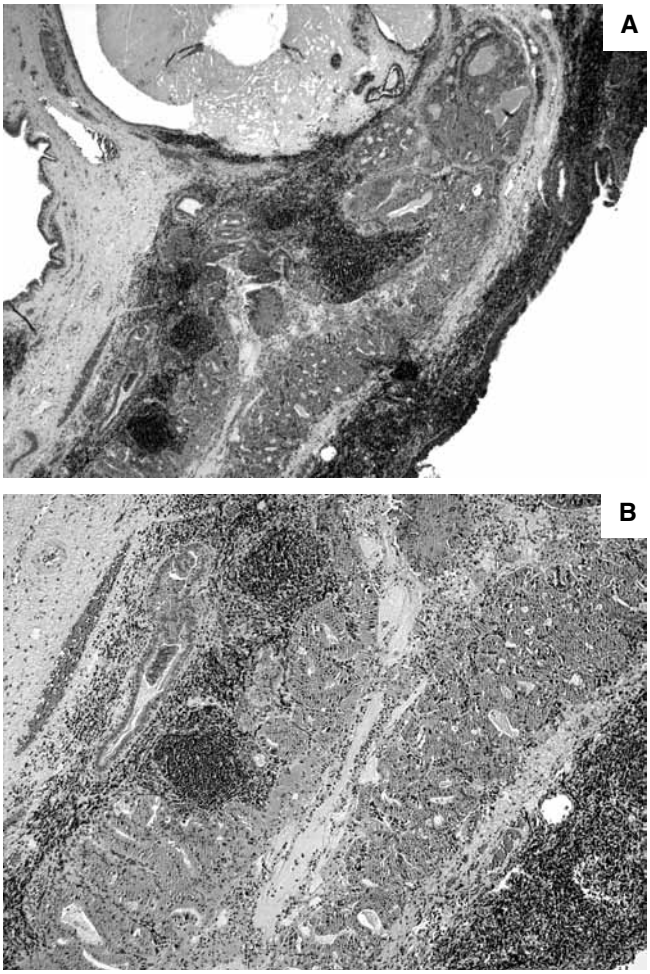

Fig. 3. The nasopharyngeal lesion shows the same histological pattern of Warthin's tumour. A. Haematoxylin-eosin, magnification 10×. B. Haematoxylin-eosin, magnification 20×.

Histologically, WT is an adenoma in which bilayered columnar and basaloid oncocytic epithelium forms multiple cysts with multiple papillae and accompanied by a proliferation of lymphoid tissue [1]. Sometimes, oncocytic cells can also form in nodal tissue. Our case was judged to meet these criteria and the lesion was diagnosed as WT.

It is difficult to explain the occurrence of a synchronous intra-parotid and extra-parotid WT. To date, there is no evidence in either the previously reported case [2] or our patient that one or more systemic or local factors might have influenced or induced the development of WT in two different sites: the hypothesis that we can make is of a multiple and simultaneous origin.

The origin of WT is still controversial and has been much debated. The most accepted hypothesis suggests that parotid WT could arise from salivary duct epithelium inclusions in the parotid gland lymph nodes, during ontogeny [2 11]. Nonetheless, other authors propose that the lymphocytic component is the result of an immunological reaction to the epithelial component, or could arise as an inflammatory response [2 3].

At the same time, the pathogenesis of nasopharyngeal WT also remains unclear. By using monoclonal antibodies, Fantozzi et al. found the ratio of T to B cells in extraparotideal WT to be similar to that of a normal lymph node, thus strengthening the theory of salivary organogenesis rather than that of reactive proliferation or hypersensitivity [12]. Extraparotideal WT may then arise from components of the minor salivary glands that are engaged in a preexisting lymphoid stroma, and chronic inflammation in the nasopharynx could induce the formation of oncocytic metaplasia of glandular tissues in the stroma [2 9].

The question also arises as to whether cigarette smoking, a chronic inflammatory stimulus, as well as a reported risk factor for the onset of WT [3-5], could have acted as a trigger for the simultaneous occurrence of WT in this case.

In conclusion, WT is a predominantly benign lesion almost exclusively found in the parotid, which can exceptionally appear simultaneously in other areas, such as the nasopharynx. Head and neck surgeons should always be aware of extra parotideal WT and consider performing MRI in highly suspicious cases during initial workup. Available data [2 6] support a surgical, conservative, approach to the management of WT, even in case of synchronous lesions, as in the patient presented.

It is likely that once the details of the pathogenesis of WT are better clarified, it will be possible to understand the occurrence of synchronous and/or multifocal lesions.

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Address for correspondence: Chiara Bianchini, Otorhinolaryngology Department, University Hospital of Ferrara, corso Giovecca 203, 44100 Ferrara, Italy. Tel. +39 0532 236383. Fax +39 0532 247709. E-mail: chiara.bianchini@unife.it