

Warthin's tumor of parotid gland: treatment of a retroneural lesion by enucleation

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Abstract. – The papilliferous cystadenomas lymphomatosum or Warthin's tumor is a relatively rare and generally benign neof ormation whose incidence is second only to the pleomorphic adenoma. 84% of all Warthin's tumor strikes the parotid gland.

We report the two cases that come to our observation paying particular attention to the second one in which occurred a synchronous bilateral manifestation, a rarity in the clinical circle.

We want to highlight that enucleation should be the preferred treatment in most cases due to extremely small dimension of Warthin's tumor. Moreover it is possible to perform this kind of surgical treatment even in the case of a retroneural lesion without causing any damage to the facial nerve and its main branches.

As regards complications and sequelae we noticed neither a permanent nor a temporary injury of the facial mimicry and sensibility.

Key Words:

Warthin's tumor, Parotid gland, Enucleation.

Introduction

The papilliferous cystadenomas lymphomatosum or Warthin's tumor is a relatively rare and generally benign neof ormation, characterised by slow growth and still undefined etiology^{1,2}. On account of uncertain histogenesis it is still a topic of debate among authors.

The research conducted by Aldrin Scott Warthin in 1929 on the first 2 cases of papilliferous cystadenomas of the American literature focused some of its still unknown peculiarities and earned him the eponym of Warthin's tumor³.

Among the tumors of the salivary glands, Warthin's tumor is second only to the pleomorphic adenoma and it usually affects people in their fifties and sixties. The parotid gland is the one most frequently struck by this type of neoplasia (84%) (Figure 1).

Warthin's tumor represents the 5-14% of the neoplasias of the parotid and the 2-5% of the submaxillary ones³. It is mainly found in males with a ratio of 6 to 1 although its incidence among women is on the increase given their increasing use of tobacco. Nowadays both sexes show an equal ratio (Figure 2).

In a study dated 1992, Kotwall affirms that smokers are eight times more likely to develop Warthin's tumor than non-smokers^{4,5}. The fact that this neoplasia is found nearly exclusively in the parotid gland can be correlated to the peculiar histological structure of this gland that during its evolution incorporates lymphoid elements. Some of these may subsequently change into true and really lymph nodes that will then be found on the surface of the gland. The ionizing radiations represent a decisive factor in the appearance of this tumor; this theory is confirmed by the high incidence of this tumor among the Japanese who survived the atomic bomb explosions and among subjects irradiated in young age due to ringworm⁴.

It is not our aim to discuss in detail, so we will just briefly present the two theories which are most widely given credit by the medical community. These are confirmed by the particular embryogenesis of the tissues affected by this kind of disease and by the environmental factors that influence its onset.

The first hypothesis maintains that the lesion has an essentially neoplastic nature; while the second asserts that its nature is of an inflammatory type due to the tobacco smoking

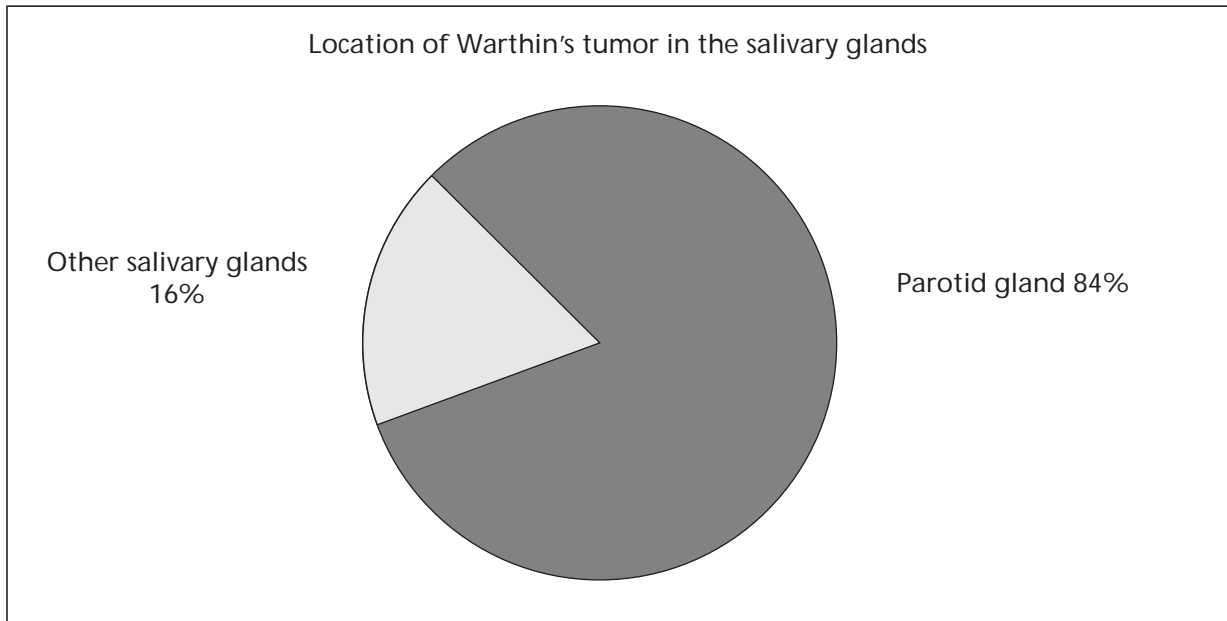


Figure 1.

and the ionizing radiation which cause a metaplasia of the parotid duct.

Macroscopically the cystadenomas lymphomatosum appears roundish or ovoidal, expresses smooth surface and varying consistence from soft to hard elastic. In section it appears partially or predominantly cystic with discharge of a serous, milky or puruloid fluid which may contain cholesterol crystals.

Histologically the two characteristic components of Warthin's tumor are the epithelial parenchyma and the lymphoid stroma splitted

from a thin basal membrane¹. The tumor is surrounded by a thick fibrous capsule and it is usually well circumscribed from the surrounding tissues.

Together with The Department of Surgical Sciences and Applied Medical Technologies, "La Sapienza" University of Rome, Italy, we have recently treated two cases of Warthin's tumor of the parotid gland; the first in a 60-year-old man and the second in a 66-year-old man who came under our observation for a synchronous bilateral manifestation.

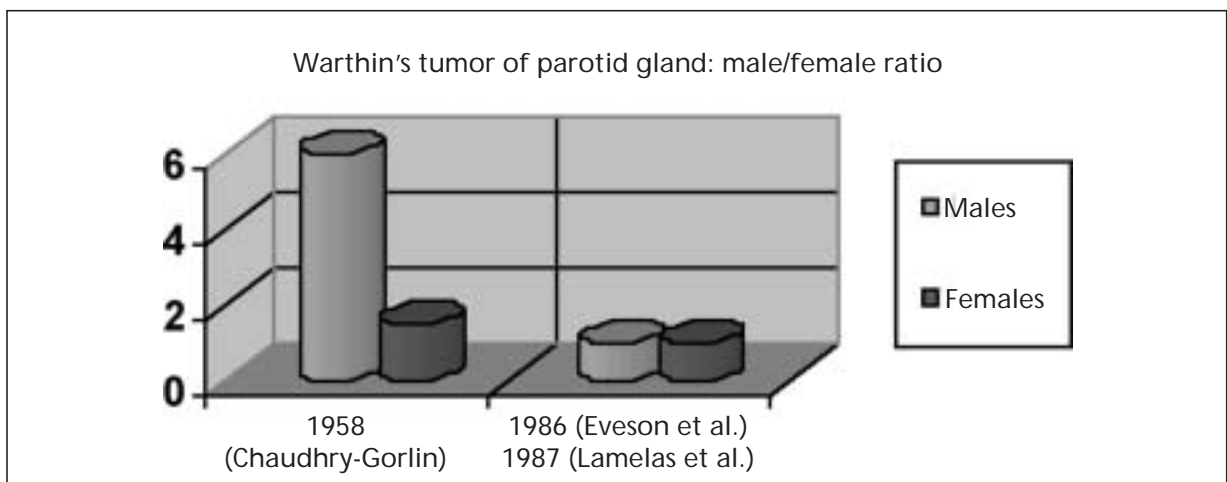


Figure 2.

Synchronous bilateral Warthin's tumors constitute a rarity in the clinical circle. The statistics taken from international literature, help us to deduce that its frequency varies from the minimum of 1 case out of 104, equal to 0.9% (Ibi et al.) to the maximum of 4 cases out of 122 (Lamelas et al.) equal to 3%³ (Figure 3).

Clinical Cases

Case 1 – M. Antonio 60 y-o living in Rome

The patient records the beginning of his symptomatology approx 4 months before the hospitalization, when he noticed the appearance of a tumefaction in the parotid cavity of the right side that is not painful even when touched. An ecography of the parotid glands was performed, which directed to the diagnosis of pleomorphic adenoma.

The patient is normotype, in good health conditions; B.P. 125/75, regular and rythmical

pulse; integral sensorium, regular breath and temperature. Nothing to say about the neck, thorax and abdomen. Smoker (≈ 10 cigarettes per day).

Cranium examination: in the right parotid gland region is possible to observe a tumefaction as big as a small hazelnut of hard-elastic consistency, mobile, not painful even when touched. It was not possible to feel any latero-cervical lymph node.

Laboratory tests – F.N.A.B. – the cytological report directs to the diagnosis of Warthin's tumor.

Histological diagnosis: Warthin's tumor.

Case 2 – C. Lorenzo, 66 y-o living in Rome

1st hospitalization (Rome April 23 2001)

The patient was admitted to hospital for a tumefaction of the left parotid gland region; approx six year before the hospitalization he reported to have noticed a first growth of such tumefaction. For this reason he went to his doctor who prescribed him an anti-inflammatory therapy which made the tumefaction regress.

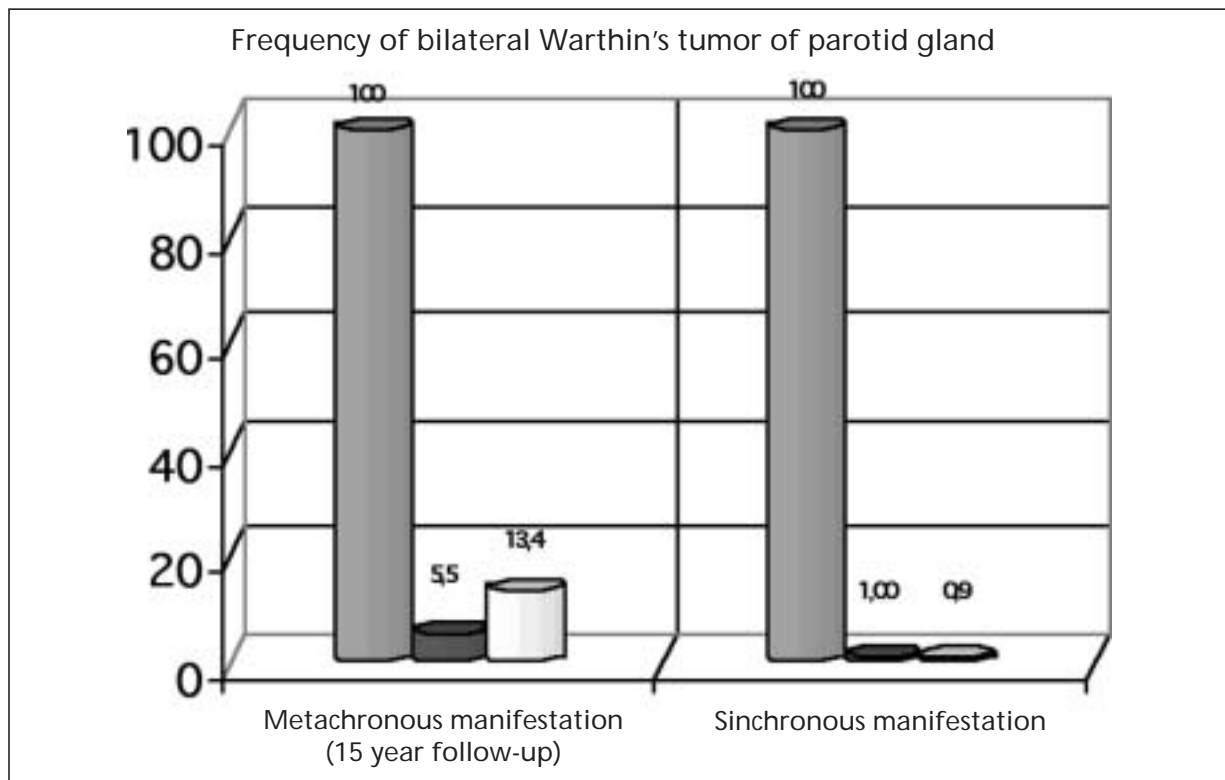


Figure 3.

In the past years he doesn't report any kind of problem until five months before the hospitalization when he noticed the same tumefaction had grown again. His personal doctor sends him for a check-up.

The NMR of cranium (Figure 4) performed on 19/02/2001 refers:

"...there is fair volume increase of both parotid glands. It is possible to notice areas of altered signal intensity, of oval morphology one of 23 mm set in the front-left side, one of 20 mm in the rear-right side..."

The patient is normotype, in good health conditions; B.P. 130/80, regular and rythmical pulse; integral sensorium, regular breath and temperature. Nothing to say about the neck, thorax and abdomen. Smoker (\approx 15 cigarettes per day).

Cranium examination: in the left parotid gland region is possible to observe a tumefaction of 2 cm of diameter of hard-elastic consistency, mobile, painful to touch. In the right parotid gland region is possible to observe a tumefaction not finely appreciable of 2.5 cm of diameter, of hard consistency, mobile as to the superficial layers and fixed as to the deep layers, that is not painful even when touched.

Laboratory tests – nothing remarkable.

Surgical operation – 27/4/2001: enucleation of the tumor from the left parotid gland.

Histological diagnosis (left parotid gland):

Macroscopical – nodular neoformation of max. 3 cm of diameter, capsuleless, of hard-elastic consistency of grey-ish colour.

Microscopical – Warthin's tumor.

The patient had been discharged from hospital three days after the surgical operation.

Subsequently, waiting for the second hospitalization, another NMR of cranium was performed in order to study properly the connections between the lesion and the two main branches of the facial nerve.

2nd hospitalization (Rome July 4 2001)

The NMR performed on 08/06/2001 reported: "...it is present in the deep portion of the right parotid gland a lesion of 20 mm of diameter, adjacent to the main trunk of the facial nerve. The foresaid nerve is pushed laterally and develops itself in depth without involving the superficial portion of the gland..."

Surgical operation – 6/7/2001: enucleation of the tumor from the right parotid gland.

Histological diagnosis (Right parotid gland):

Macroscopical – neoformation of $2 \times 1 \times 0.5$ cm of dimension partly cystic and partly papillar with an epithelial coating set up in a duoble layer made of cylindrical cells in continuity with abundant lymphoid stroma. Presence of a pseudocapsule.

Microscopical – Warthin's tumor.

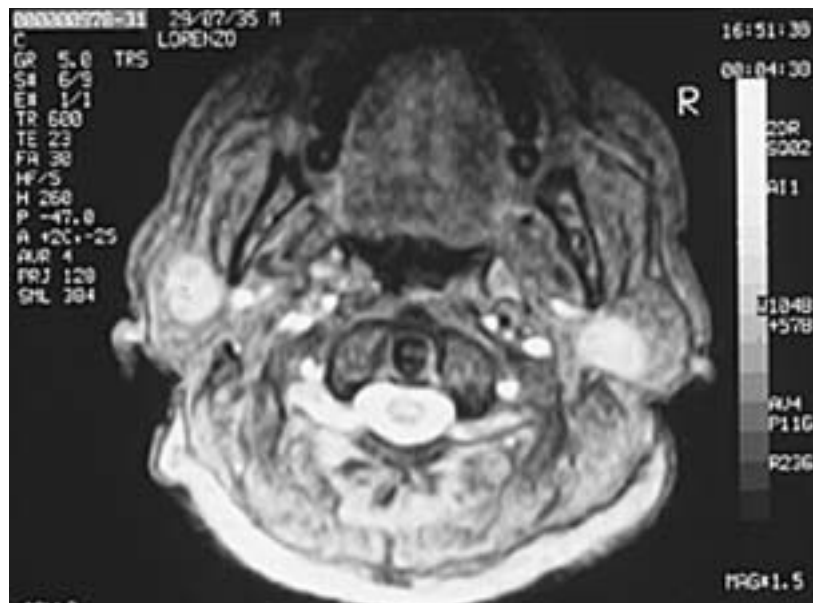


Figure 4.

Discussion

In the first case the lesion was studied with echography investigation which allowed the check up of morphostructural characteristics. This investigation could itself be already enough for a diagnostic definition. The technique of the needle biopsy may eventually be added. The lesion was in fact very evident and it was characterised by a superficial localization, easily accessible to manual exploration.

After that a F.N.A.B. was performed but as it turned out, it wasn't the most useful technique as we didn't discover anything more from it. This procedure should in fact be restricted to dubious cases that would possibly require a change in the surgical therapeutic strategy.

We proceeded in a totally different way with the second case which showed two synchronous lesions: one was superficial and placed on the right, the other was deeper and placed in the left parotid gland. The investigations carried out did not leave any doubt on the benignity of the lesions, so we consid-

ered it unnecessary to employ the F.N.A.B. technique for the histological definition.

We therefore preferred to divide our operation into two different phases. We initially performed an enucleation of the superficial lesion of the right parotid with respect for the surrounding structures and we postponed the operation on the contralateral parotid for a second hospitalization.

The absence of a painful symptomatology and the deep and not palpable localization of the lesion suggested us to take time in order to study through a NMR the relationships that the lesion was developing with the branches of the facial nerve.

The images showed the retroneural localization of the tumefaction and its close relationships with the facial nerve. The pre-operative investigations set the surgical indication for a total parotidectomy as typical for deep retroneural localizations (Figure 5).

The surgical indication for a total parotidectomy seemed excessive given the elevated risk of iatrogenic lesion for the facial nerve and its main branches. This consideration was supported by the relative absence of

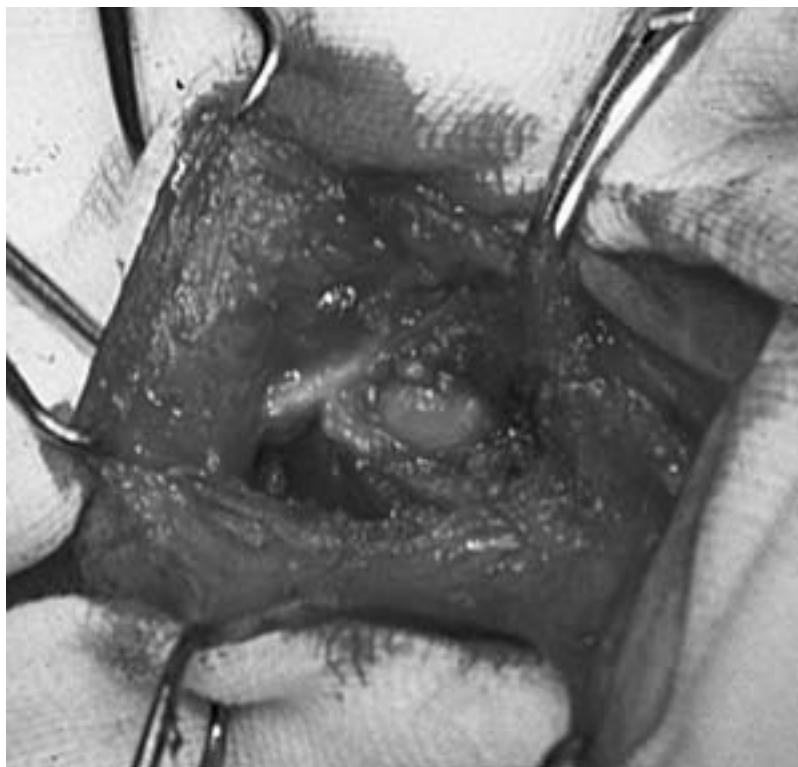


Figure 5.

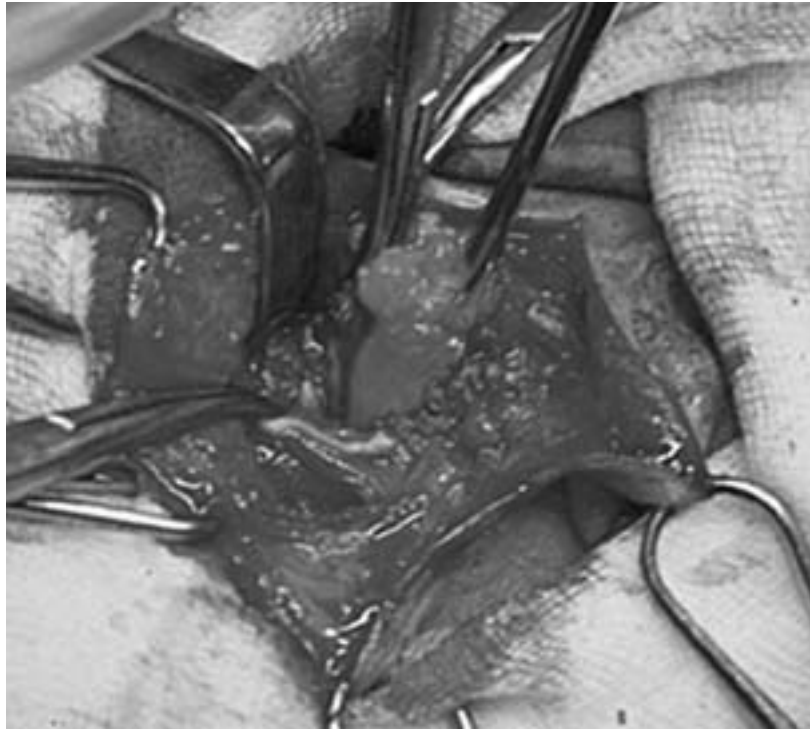


Figure 6.

symptomatology and the benignity of the lesion.

During the operating action, on the basis of the data previously collected, we prepared the facial nerve with the direct technique following it up to its two main branches (cervico-facial and temporo-facial).

Only after that, lacerating the underlying glandular parenchyma, we were able to see the presence of the lesion: subsequently, opening wide and very gently the two nervous branches, the lesion appeared in the plan delimited by them.

Only when the lesion became superficial, it was possible to gain access to the lesion and to perform its extirpation (Figure 6).

This kind of treatment did not cause any neurological deficit of the facial mimicry and sensibility.

The histological examination confirmed the suspected nature of the lesion.

Under no circumstances was any multifocal origin of the lesion pointed out.

In conclusion, we want to highlight the relative inadequacy of a total parotidectomy together with its complications and sequelae due to the benign nature of the lesion. This is a very important point as it is always in our

interest to salvage these delicate structures through extreme precision in our surgery. In addition, we can always affirm without hesitation that an enucleation should be the preferred treatment in these cases.

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