

Double Warthin's Tumor: Case report and our 4-year experience

Ermal Pashaj ¹, Fabio Luciani ^{1,*}, Carla Sgroi ², Bora Kerpi ³, Filadelfo Coniglione ^{1,4} and Ernesto Bruno ⁴

¹ Department of Surgical Sciences, Catholic University "Our Lady of Good Counsel" – Tirana, Albania.

² Faculty of Medicine, School of Dentistry, University "Our Lady of Good Counsel" – Tirana, Albania.

³ Department of Medical Sciences, Catholic University "Our Lady of Good Counsel" – Tirana, Albania.

⁴ Department of Medical Science and Translation Medicine, University of Rome "Tor Vergata", Roma, Italy.

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Abstract

The purpose of this study was to determine the clinical and histological features, as well as the treatment approach, of Warthin's Tumor. Albrecht-Artz-Warthin Tumor is a benign neoplasm of the salivary glands, historically known as papillary cystadenoma lymphomatosum. It accounts for approximately 0.6–11% of all salivary gland neoplasms and is more common in men during the 6th and 7th decades of life. This tumor almost exclusively occurs in the parotid gland and can present as bilateral or multifocal lesions, and it manifests as a painless swelling, usually within the lower portion of the parotid gland.

The aim of this work was to describe the clinical and histological characteristics of Warthin's Tumor, providing useful information for the differential diagnosis with other salivary gland neoplasms. Data from the literature were compared with ours, collected over a ten-year clinical activity in the Oral and Maxillofacial Surgery Unit of "Our Lady of Good Counsel Hospital".

The prognosis of Warthin's Tumor (W.T.) is certainly favorable, with nearly 100% *restitutio ad integrum*. Between May 2022 and November 2025, we reported 3 diagnoses of Warthin's Tumor. The incidence of W.T. in our patient sample was 0.181%.

In conclusion, Warthin's Tumor is a benign salivary gland tumor that requires accurate diagnosis, complete surgical excision, and adequate follow-up, as malignant transformation, although rare, remains possible.

Keyword: Warthin Tumour; Salivary Glands Tumour; Histological Features

1. Introduction

Warthin's tumor was first described in literature by Dr. Hildebrand in 1895 as a papillary cystadenoma. It represents the most frequent histotype among monomorphic adenomas, with an incidence of approximately 10% of all major salivary gland neoplasms. Males are more commonly affected, with a prevalence ratio of 1,6:1 compared to females. In 87% of cases, the involved gland is the parotid, specifically within its prenervous portion.

From an anatomopathological standpoint, this neoplasm originates from Neisse–Nicholson rests, clusters of ectopic glandular epithelial tissue located within intraglandular lymph nodes [1,2]. A review of the literature shows that the manifestations of papillary cystadenoma lymphomatosum may vary; indeed, both unilateral and bilateral presentations

* Corresponding author: Fabio Luciani

can be distinguished; the latter further subdivided into synchronous and metachronous forms depending on their time of onset [1,3].

Several studies have demonstrated a strong correlation between the proliferation of Neisse–Nicholson rests and smoking, with an approximately eight times increased risk of onset in individuals with a positive smoking history. The pathogenic factor involved appears to be the disruption of glandular parenchyma caused by nicotine-mediated secretory stimulation.

In recent years, an increase in the incidence of Warthin's tumor in women has been observed, likely due to the rising prevalence of smoking among females [2,4].

Another finding emerging from the literature review is that multiple presentations are very rare or even exceptionally uncommon. When observed (with approximately 20/30 cases reported), they have been classified into two major groups: multiple tumor manifestations with the same histological classification, and multiple tumor manifestations with different histological classifications [3]. Among the latter, the most frequently encountered synchronous multifocal unilateral neoplasm is undoubtedly Warthin's tumor [2,3,5].

Multiple neoplastic lesions of the salivary glands must also be distinguished based on their specific nomenclature [4, 5]: Biphasic neoplasms (a mixture of two cellular clones within the same tumor); Collision tumors (two neoplasms arising in different topographic areas that subsequently invade the same anatomical region); Hybrid tumors (neoplasms composed of different cellular clones within the same topographic area) [5, 6].

2. Materials, methods and case report

The patient, M.S., a 63-year-old male, presented to our observation at the Oral and Maxillofacial Surgery Unit of the University Hospital "Our Lady of Good Counsel" in Tirana, reporting a swelling in the left parotid region that had been present for a long time.

The medical history revealed a fair general health status, with no systemic diseases and no chronic pharmacological therapy. The patient reported smoking >40 cigarettes per day for at least 40 years.

The extraoral physical examination showed the presence of two apparently distinct swellings involving the left parotid gland, one located in the infero-posterior portion and the other, more superficial, in the antero-superior area. [7, 8] Both lesions were non-tender and not painful on palpation, with a firm-elastic consistency, both mobile over the fascial planes, and were covered by apparently normotrophic and normochromic cutaneous tissue. The intraoral examination did not reveal any dento-skeletal alterations [9].

A magnetic resonance imaging (MRI) scan of the facial skeleton and neck was therefore requested, with particular attention to the parotid space, performed with and without contrast medium (Gadolinium), using axial (Fig.1), coronal (Fig.2), and sagittal sequences weighted in T1 and T2.

The MRI examination revealed, within the left parotid gland, two solid and slightly inhomogeneous masses, lacking intralesional vascularization and with well-defined margins that clearly separated them from the surrounding tissues. The first lesion measured approximately 25.3 mm and was located in the parapharyngeal extension, while the second, measuring approximately 19.5 mm, was situated in the anterior pole of the gland. The MRI did not show significant alterations of the lymph node stations of the circular ring of Poirier and Cuneo, nor of the lateral cervical transverse system.

After undergoing routine blood chemistry tests, the patient underwent total Parotidectomy Surgery: "Following monitoring of vital functions and oro-tracheal intubation, a preauricular and pretragal incision (Fig.3) was made under general anesthesia according to *Redon*; elevation of the skin flap and s.m.a.s; access to the parotid compartment by lateral dissection of the external auditory canal (Fig.4), isolating the anatomical boundaries of the Parotid Gland (Fig.5).

Visualization and palpation of the styloid process in the anterior portion of the homonymous compartment were performed, with careful preservation of the white and red fibers of *Riolano*. Identification and preservation of the main trunk of the Facial Nerve (VII cranial nerve) and its cervicofacial and temporofacial branches, including all minor rami, were carried out according to Conley's tunneling technique [8, 10].

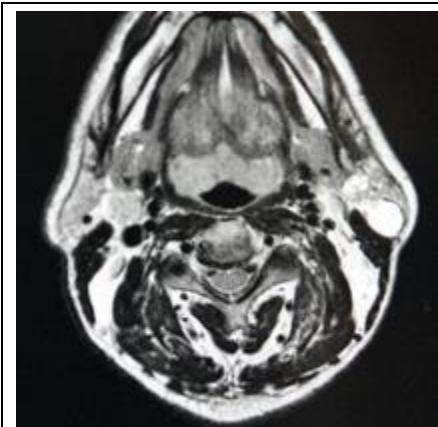


Figure 1 Axial sequence

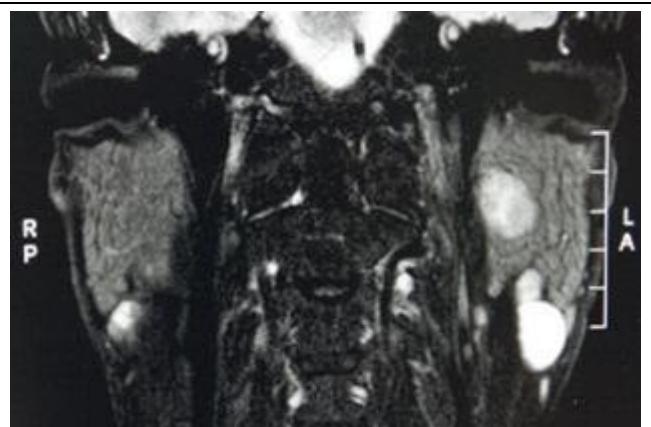


Figure 2 Coronal sequence



Figure 3 Preauricular and pretragal incision made under general anesthesia, according to Redon.

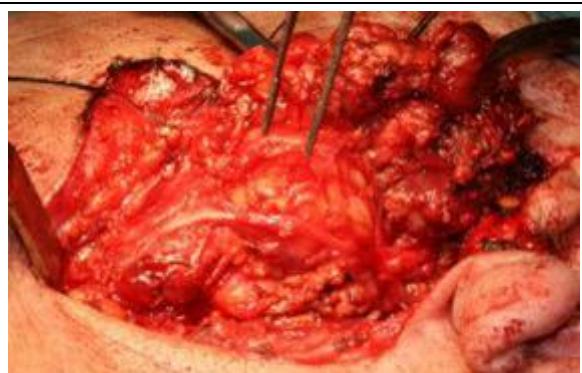


Figure 4 Access to the parotid compartment by lateral dissection of the external auditory canal



Figure 5 Isolation of the anatomical boundaries of the Parotid Gland.

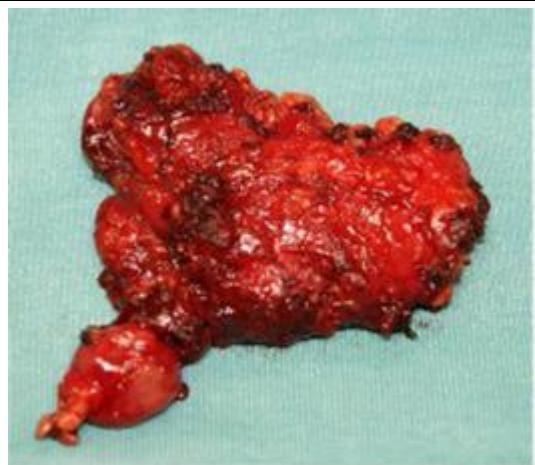


Figure 6 The entire left Parotid Gland was removed as a whole with the neoformation.

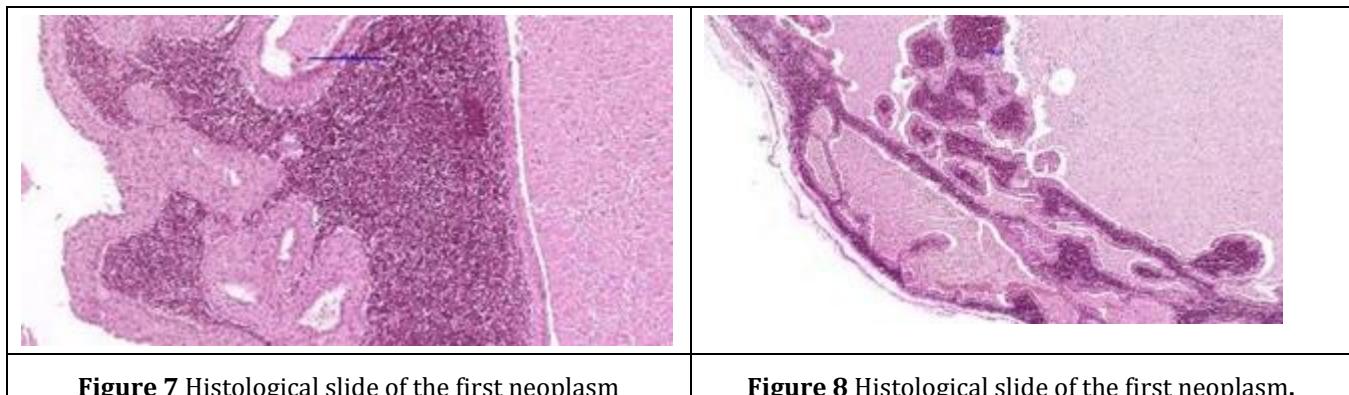


Figure 7 Histological slide of the first neoplasm

Figure 8 Histological slide of the first neoplasm.

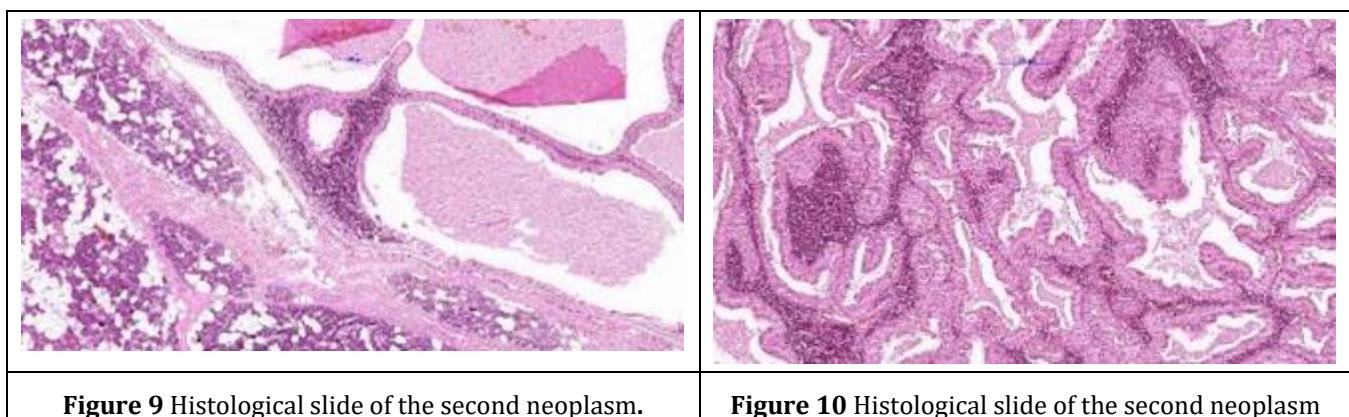


Figure 9 Histological slide of the second neoplasm.

Figure 10 Histological slide of the second neoplasm

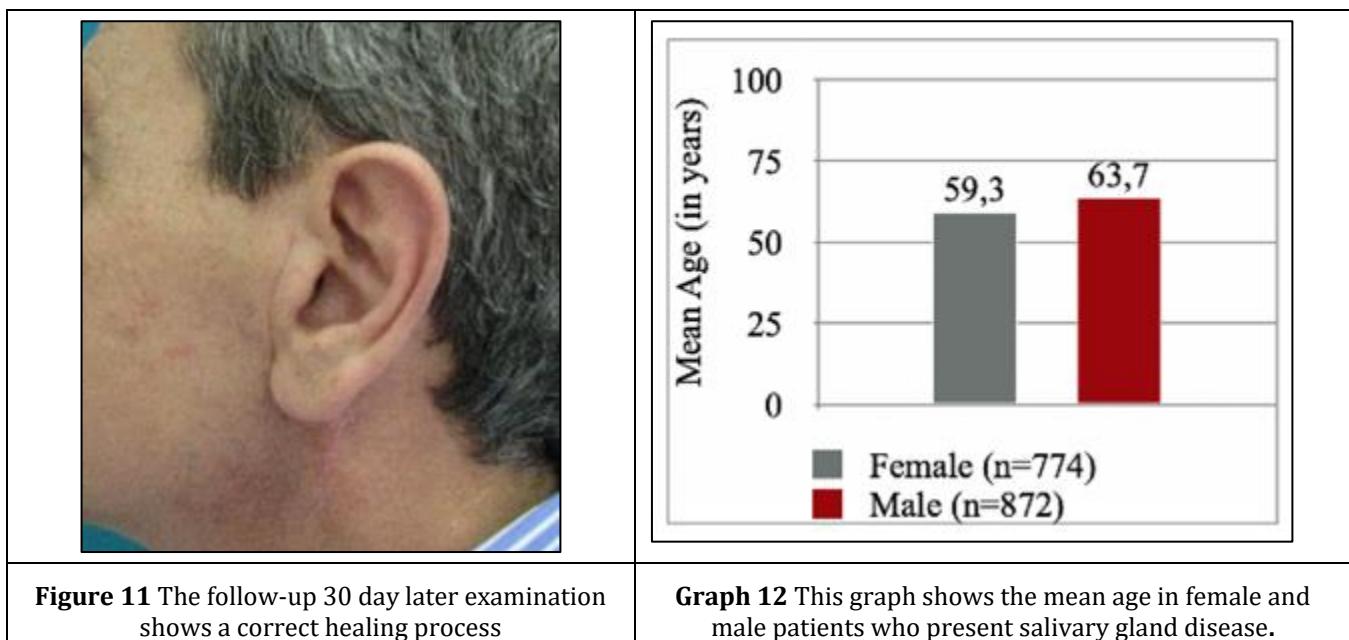
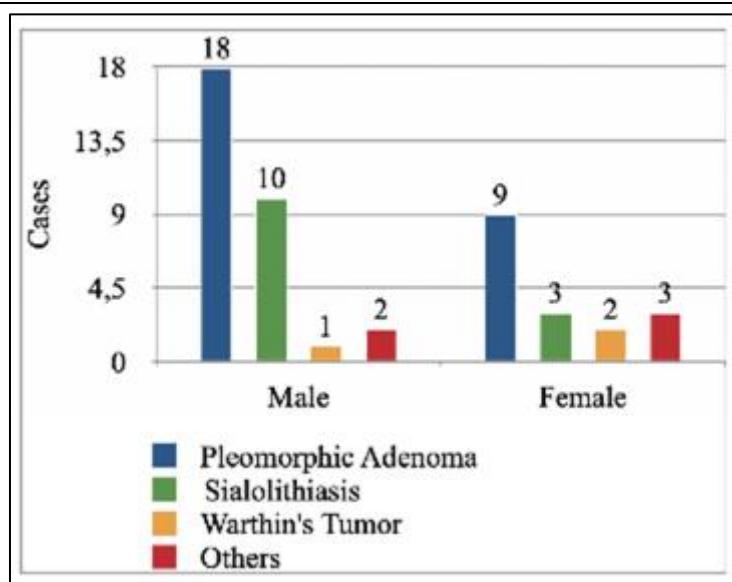


Figure 11 The follow-up 30 day later examination shows a correct healing process

Graph 12 This graph shows the mean age in female and male patients who present salivary gland disease.



Graph 13 This graph shows the incidence in male and female patients for different salivary gland disease.

Subsequently, after locating the surgical landmarks, the entire left Parotid Gland was removed as a whole with the neoformations following standard operative steps and techniques (Fig.6); intraoperative tests were positive for evaluating the functionality of the VII cranial nerve; meticulous and prolonged hemostasis was achieved, a No. 15 Blake drain was placed in the parotid compartment, layered closure was performed with subcutaneous *Polyglactin 910 4/0* interrupted Donati sutures and skin closure with 6-0 *Nylon* intradermal stitches. A compressive medication and alternating cryotherapy were applied.

The excised tissue was sent for definitive histopathological examination. The patient was awakened according to protocol and remained under observation by the anesthesiology team.

Dexamethasone 8 mg in slow infusion with 500 cc of saline, Paracetamol 1000 mg twice a day, and intravenous Ceftriaxone 1g for five days were administered.

The postoperative course was regular, and cutaneous sutures were removed seven days after surgery. Follow-up examinations at 7, 10, 30, 60, and 90 days (Fig.11) showed no residual deficit of the facial nerve (VII cranial nerve), neither in the main trunk nor in the minor branches.

The definitive histopathological examination of the excised tissue reported: “Completely excised parotid gland with two synchronous neoplasms composed of glandular, cystic, and papillary structures lined by stratified epithelium consisting of eosinophilic columnar cells and granular basal cells. The stroma contains a variable amount of lymphoid tissue with germinal centers in active phase. *The diagnosis is compatible with a double Warthin's Tumor of the left Parotid Gland*” (Fig.7-8-9-10).

3. Results and discussion

Most salivary gland neoplasms present as single lesions involving only one of the glands in humans. Multiple tumors of the major salivary glands represent only 1.1% of international case series, and those of the parotid gland account for approximately 0.7%, with fewer than 20/30 cases reported in the literature [3, 5, 7]. The neoplasm with the most frequent multicentric development and synchronous involvement is Warthin's Tumor, followed by Pleomorphic Adenoma, Acinic Cell Carcinoma, and Oncocytoma [7, 9]. The aim of our work was to retrospectively evaluate W.T.'s incidence over four years of clinical activity in the Oral and Maxillofacial Surgery Unit of our hospital. During this period, we analyzed 1,646 patients, 53% were male (872 subjects) and 47% were female (774 subjects); 48 of whom had salivary gland disease (66% were female and 34% were male). The mean age (SD) was 59,3 years in females and 63,7 years in males (Figure. 12). Between May 2022 and November 2025, we reported 3 diagnoses of W.T., with an incidence in our patient sample of 0.181%. Our data are in agreement with the international literature, confirming that W.T. is a neoplasm of rare clinical occurrence. The histopathological diagnosis was “papillary cystadenoma lymphomatosum”.

4. Conclusion

We can therefore conclude that the case we examined of a “Double Warthin’s Tumor within the same parotid gland with synchronous development” represents an exceptional clinical finding and is furthermore one of the few cases reported in the literature. Between May 2022 and November 2025, in the Maxillofacial Surgery Unit of the “Our Lady of Good Counsel” observed a 2.91% incidence of salivary gland pathologies. Of particular clinical significance is the finding of a rare case of synchronously growing Warthin’s tumor, representing 0.06% of the total cases seen (Figure. 13). As consistently confirmed, multidisciplinary collaboration among specialists, such as the dentist, the maxillofacial surgeon, and the anesthesiologist-intensivist, using a multimodal and holistic approach to the patient represents the gold standard for accurate diagnosis and therapeutic success.

Compliance with ethical standards

Disclosure of conflict of interest

The authors have nothing to disclose.

Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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