IOSR Journal of Dental and Medical Sciences (IOSR-JDMS)
e-ISSN: 2279-0853, p-ISSN: 2279-0861.Volume 23, Issue 1 Ser.3 (January. 2024), PP 22-27
www.iosrjournals.org

Whartin's Tumor: About A Bilateral Parotid Location.

T.W. Chabi Agbassikakou, S.K. M'fa, A. Benbachir, M. K. Fiqhi, J. Hamama, M.K. El Khatib (1) A. Mustapha, M. Oukabli (2)
Department Of Maxillofacial Surgery And Stomatology
Department Of Pathological Anatomy And Cytology
Mohamed V Military Training Hospital, 10500 Rabat Maroc

ABSTRACT :
Warthin's tumor (TW) is the second most common etiology of parotid tumors after pleomorphic adenoma. Currently, this tumor is no longer a reassuring diagnosis for the maxillofacial surgeon considering the risk of cancerization.
An 87-year-old patient with hypertension and chronic smoking history presented with chronic bilateral swelling of the parotid region progressively evolving for more than six years. Magnetic resonance imaging noted the benign nature of the tumor. A right exofacial parotidectomy was performed and histopathological analysis confirmed the diagnosis of Warthin tumour. Post operative follow up was uneventful and no recurrence was observed after six months.
Warthin's tumor typically predominates in smoking men over the age of 50. Cytological puncture is not systematic for establishing the preoperative diagnosis. The majority of authors recommend an exofacial parotidectomy with identification and dissection of the facial nerve. Malignant transformation is exceptional and can occur at the expense of both epithelial and lymphoid components.
KEY WORDS:*parotid swelling – magnetic resonance imaging – Warthin tumor – exofacial parotidectomy – cancerization*

Date of Submission: 01-01-2024 Date of Acceptance: 09-01-2024

I. INTRODUCTION

Warthin's tumor (TW), also called adenolymphoma or papillary cystadenolymphoma, is the second etiology of parotid tumors after pleomorphic adenoma. It represents 5 to 15% of tumors of the parotid gland. [1,2]

This disease owes its name to the American pathologist Alfred Scott Warthin who definedit for the first time in 1929, and its etiopathogenesis is not yet well established. [3.4]

It typically predominates among smoker men over the age of 50. This tumor as it was described almost a century ago is no longer a reassuring diagnosis for the maxillofacial surgeon today because of the risk of cancerization, but especially because there is always the risk of diagnostic confusion, during the preoperative assessment, with oncocytoma, low-grade mucoepidermoid carcinoma or lymphoma. [2,4,5]

The case that we report highlights the major problem of the therapeutic attitude to adopt, either abstentionist or interventionist, and also emphasizes this debate in the light of the literature.

II. OBSERVATION

An 87-year-old patient with high blood pressure and chronic smoking for 18 years consulted the maxillofacial surgery department for chronic bilateral swelling of the parotid region that had been progressively evolving for more than six years.

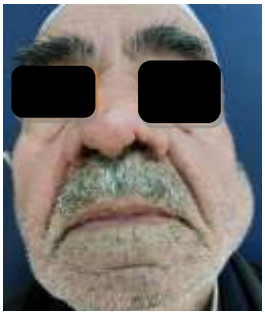

Figure 1: front view showing facial asymmetry by a left jugomyaseterin tumefaction

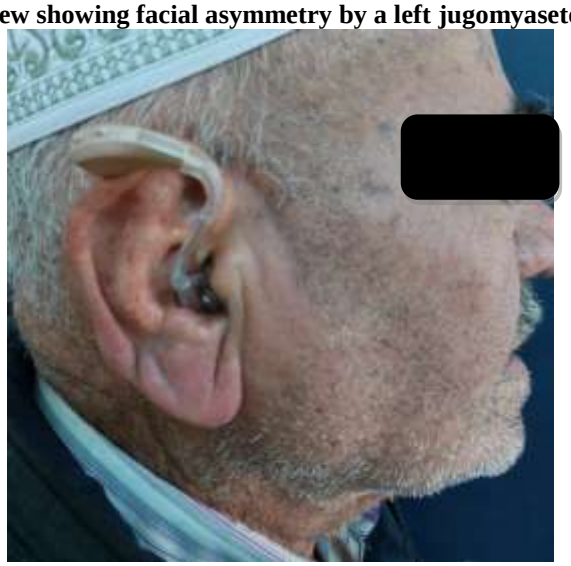

Figure 2: right profile view showing right parotid swelling.

The examination reveals a bilateral parotid swelling, the largest on the left of which measures about 7 centimeters on the long axis, of soft consistency, slightly sensitive to palpation, mobile in relation to the superficial and deep planes and overflows the mandibular angle at the bottom with a remarkable elevation of the lobule of the left ear. On the right, the curve measures about 5 cm long axis, mobile, sensitive to palpation, with a slight cutaneous infiltration facing giving the appearance of orange peel. There was no motor deficit in the facial nerve territory. Examination of the oral cavity found a totally edentulous patient, with no suspicious mucosal lesions, and the two stenon ostias were without abnormalities. Palpation of the cervical lymph nodes does not reveal lymphadenopathy.

Parotid ultrasound performed as first intention reports a bilateral increase in the size of the glands and the presence in between of well-limited hypoechoic masses with posterior enhancement, non-vascularized on color Doppler. To have a more precise diagnostic radiological contribution, we carried out magnetic resonance imaging (MRI) of the face which confirmed the bilateral parotid hypertrophy, by the presence of two masses in their superficial lobes. On the right, the mass is encapsulated with a lobulated contour, in T1 hyposignal, with an intermediate signal in T2, enhanced earlyafter injection with rapid Wash Out (>30%) and measuring 27x23x30 mm. On the left, the mass is larger in size and measures 35x26x48 mm. It is encapsulated, oval, with lobulated contours with a double component: tissue with an intermediate signal in T2, enhanced early after injection with rapid Wash Out (>30%), and cystic in hypersignal T1 and T2 (rich in proteins).

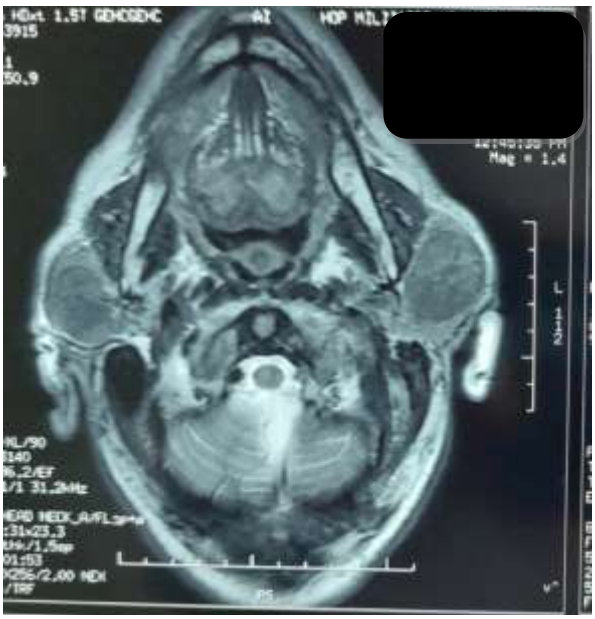

Figure 3: MRI of the parotids, axial section in T2 sequence: On the right, the mass is encapsulated and with a lobulated outline

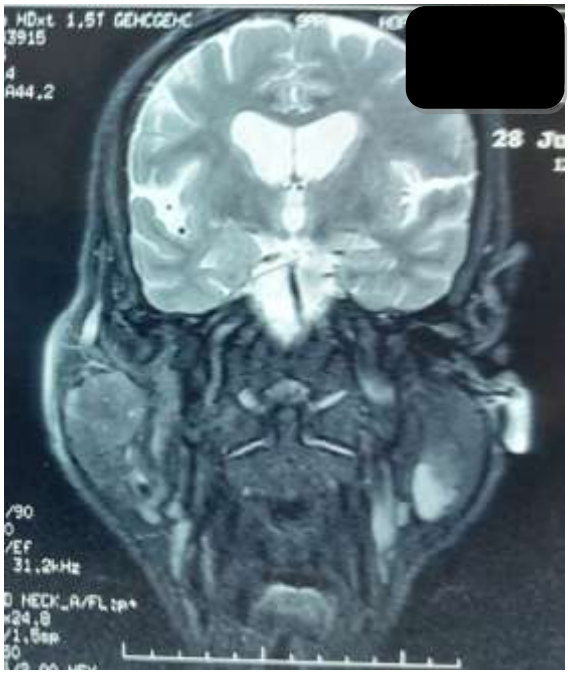

Figure 4: parotid MRI; coronal slice in T2 sequence: On the left, the tumor is encapsulated, oval, with lobulated contours with a double component: tissue and cystic in high signal.

We therefore decided to perform a superficial parotidectomy with conservation of the facial nerve, the choice is made on the left side because the tumor is very large with major morphological repercussions.

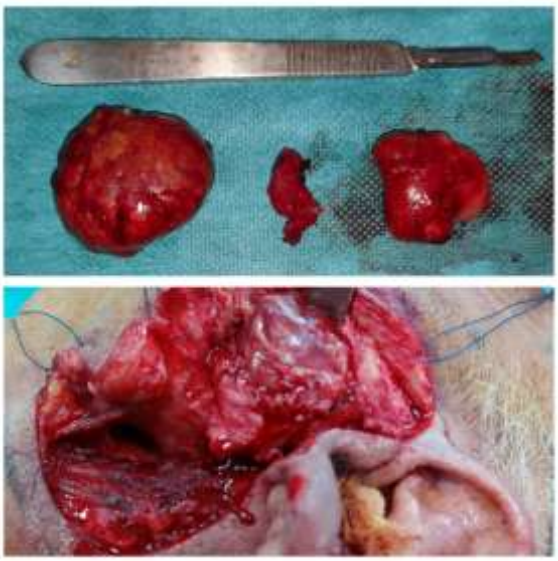

Figure 5: Intraoperative images of the parotidectomy and the anatomical specimen sent to the laboratory.

Histological examination shows a Warthin tumor without histological signs of malignancy. There is therefore a benign tumor proliferation made up of oncocyte cells, organized in papillae bordered by a double layer of regular myoepithelial and oncocyte cells within the same lymphoid stroma. This proliferation is circumscribed by a fibrous capsule which is completely respected.

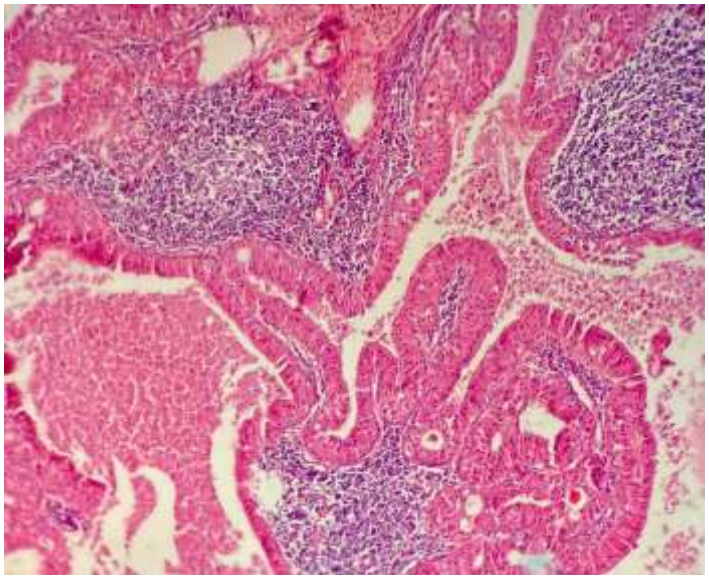

Figure 6: Microscopic view of the slide on pathological examination: there are indeed cystic cavities bordered by papillae with a double cell layer (oncocytes + basal cells) resting on a very abundant lymphoid stroma.

After a follow-up of six months, the postoperative course was simple, we did not note any recurrence on the operated side.

At the level of the right parotid region, we noted no increase in tumor volume and the patient was warned of rigorous self-monitoring.

III. DISCUSSION

Warthin's tumor (TW) typically presents in adults as a mass of a few centimeters, painless, of parotid seat and indolent evolution. [6] It characteristically has oncocytic glandular structures and a lymphoid stroma. It is found almost exclusively in the parotid gland and the peri-parotid lymph nodes [2].

Several hypotheses on the histogenesis of this lymphoid contingent have been proposed to try to understand this pathology.

The first, the most widely accepted, would be that the tumor arises from an ectopic ductal epithelial tissue located in the normal parotid lymphoid tissue. [6,7] Indeed, the parotid is the only salivary gland to physiologically contain lymph nodes.

The second hypothesis would be that this adenoma initially develops on normal parotid epithelial tissue and that it induces a significant lymphoid stromal reaction. [8]

This theory could explain the frequent location of Whartin's tumors in the parotid gland, the bilaterality and the multiplicity of these tumors. [9]

Several studies have shown that more than 94% of patients with Warthin's tumor are smokers [10,11].

In the MNEJJA series, a high frequency of smoking was observed among affected patients (82% were smokers and 67% were heavy smokers). [12]

Yu Gy also showed in his study that the frequency of smoking was higher in patients with Whartin's tumor (96.3% patients smokers in TW) than in those with Pleomorphic Adenoma AP (26.4% of patients smokers in patients with OA) and compared to the general population (25.5%). [13]

4–7.5% of parotid Warthin tumors are bilateral. [14] In a series of 71 cases, Tveterås and Kristensen, found only seven bilateral and synchronous localizations. [15] For some authors, the high incidence of Whartin tumors in men suggests possible hormonal dependence because sex hormone receptors have been identified in tumor cells. [16]

The exact role of sex hormones and their impact on the salivary glands remains unclear. On the other hand, the incidence of Whartin's tumors in the black race is extremely rare, probably implying a participation of genetic factors [16,17]

Many studies have tried to show the performance of magnetic resonance imaging in the histopathological approach of parotid tumors. This examination brings back with a very appreciable precision, an evaluation of the characteristics of the tumor and the peri-tumoral environment. It is of remarkable performance in the distinction between the benign and malignant nature of the tumor process, but also in the histological typing of benign tumors since the advent of new dynamic diffusion and perfusion sequences. [18.19]

In a retrospective study of a series of 50 patients with parotid tumors who underwent MRI and echo-guided cytopuncture for histological confrontation with the etiology mentioned after the radiological evaluation, we retain for the correct diagnoses, an MRI sensitivity of 92.8% and a specificity of 97.2%. [19]

The echo-guided fine needle aspiration biopsy will therefore be performed as a second intention by a radiologist or a cytologist who is well trained and experienced enough to provide the histological proof necessary for the final therapeutic decision. Its diagnostic performance is quite variable. While it generally has good specificity for malignant tumours, the sensitivity is far too low, hence the high risk of missing serious tumours, not to mention the significant percentage of non-contributory examinations. [18,19] There is little work on fine needle aspiration cytology (CAf) which would have a diagnostic sensitivity ranging from 75% to 89.2% with in particular false positives between the diagnosis of Whartin tumor and the squamous cell carcinoma. [2]

The cytological puncture is therefore not systematic for the establishment of the preoperative diagnosis and the therapeutic strategy, and its association with imaging does not seem to significantly improve the results at the expense of the greater cost induced by this examination. [20]

the PubMed database, numerous cohorts of several hundred patients report malignant transformation. They described lymphomas developing within the lymphoid component of Warthin's tumor and authentic "cancerizations" in the form of mucosquamous cell carcinomas, adenocarcinomas, salivary duct carcinomas, oncocyte cell carcinomas and squamous cell carcinomas. (most published transformation). One of these clinical cases even detailed the area where the benign epithelium of the Warthin tumor transformed into a squamous cell carcinoma, leaving no doubt about the reality of this phenomenon. [21]

This exceptional malignant transformation can occur at the expense of both epithelial (carcinoma) and lymphoid (lymphoma) components. Nagao et al reported a malignant transformation frequency of 0.1%. [22]

The occurrence of a lymphoma on a known Warthin lymphoepithelial tumor or the synchronous discovery of the two tumors is exceptional (less than 30 cases reported in the Anglo-Saxon literature. [23] MALT lymphomas are the forms most often reported, but cases of Hodgkin's lymphoma or T-cell lymphoma have also been described [24, 25, 26, 27].

In general, the risk of malignant transformation is around 0.3%. [28] This risk of malignant mutation seems close to that associated with basal cell carcinoma, and well below the percentage of malignant transformation of pleomorphic adenoma, which is 5 to 15%. The question of the absolute benignity of this previously mentioned tumor thus becomes a legitimate concern, and consequently questions about the aggressiveness to adopt in the therapeutic attitude become legitimate. [29.20]

If the therapeutic indication often depends on the functional and aesthetic impact of the swelling, we would be tempted to operate on some patients after the multidisciplinary consultation meeting. Parotidectomy will be discussed in the event of diagnostic doubt or a large lesion responsible for aesthetic damage. The majority of authors recommend an exofacial parotidectomy with identification and conservation of the facial nerve. This attitude is the most adequate to avoid recurrences and reduce postoperative incidents [3].

Proponents of simple tumor enucleation maintain that it is an encapsulated tumor for which enucleation increases neither the risk of recurrence nor that of facial paralysis and does not compromise exofacial parotidectomy in the event of recurrence but the frequency of multifocality is against this attitude. [3O]

MNEJJA in his series of 11 cases performed lower polar exofacial parotidectomies with fewer complications and without tumor recurrence with an average follow-up of 5 years. [12]

IV. CONCLUSION:

Since the risk of cancerization has been scientifically established, this has considerably changed the view of surgeons and the therapeutic attitude to adopt vis-à-vis Whartin's tumour.

From a medico-legal point of view, this data reinforces the absolute necessity of a discussion of the medical file of these tumors in a multidisciplinary consultation meeting before any therapeutic decision and this must change the information delivered to the patient on the oncological particularities of the tumor of Warthin.

ACKNOWLEDGEMENT : The authors declare that there is no conflict of interest.

REFERENCES :

[1]. Gao M, Hao Y, Huang MX, Et Al. Salivary Gland Tumours In A Northern Chinese Population: A 50-Year Retrospective Study Of 7190 Cases. Int J Oral Maxillofac Surg 2017 ;46 :343–9

[2]. Paris J, Facon F, Chrestian MA, Giovanni A, Zanaret M. Diagnostic Et Traitement Des Tumeurs De Warthin : Présentation Clinique, Ponction Cytologique Et IRM. Rev Laryngol Otol Rhinol (Bord) 2004 ;125 :65–9.

[3]. HM. Chedid, A. Rapoport, Kf. Aikawa, Ad. Menezes, And Al. Warthin's Tumor Of The Parotid Gland: Study Of 70 Cases. Rev Col Bras Cir. 2011 ;38(2) :90-4

[4]. A.Teymoortash, Y.Krasnewicz, JA.Werner. Clinical Features Of Cystadenolymphoma (Warthin's Tumor) Of The Parotid Gland : A Retrospective Comparative Study Of 96 Cases. Oral Oncol.2006 ;42 :569-73.

[5]. Yu Gy, Liu XB, Li ZL, Peng X. Smoking And The Development Of Warthin's Tumour Of The Parotid Gland. Br J Oral Maxillofac Surg. 1998 ; 36 :183-5

[6]. Barnes L, Eveson JW, Reichart P, Sidransky D. Pathology Andgenetics Of Head And Neck Tumours. 1st Ed. Lyon: IARC; 2005. Swerdlow S, Campo E, Harris NL, Jaffe ES, Pileri SA, Stein H, Et Al. WHO Classification Of Tumours Of Haematopoietic And Lym-Phoid Tissue. 4th Ed. Lyon : IARC ; 2008
[7]. A Boutin, L Chaput, C Lecointre, R Kerdraon, Gi Metrard, P Michenet. An Unexpected Warthin's Tumor. Annales De Pathologie (2015) 35, 270—272
[8]. (J. Lamelas, JH.Terry, E.Antonio. AE.Alfonao. Warthin's Tumor : Multicentricity And Increasing Incidence In Women. Am J Surg. 1987 ;154 : 347-51.)
[9]. 1,4 Vories AA, Ramirez SG. Warthin's Tumor And Cigarette Smoking. South Med J 1997;90: 416–8.
[10]. Cadier M, Watkin G, Hobsley M. Smoking Predisposes To Parotid Adenolymphoma. Br J Surg 1992 ;79 :928–30.
[11]. M.MNEJJA, B.Hammami, L.Bougacha, A. Chakroun, I.Charfeddine, A.GHORBEL J. LA TUMEUR DE WARTHIN DE LA PAROTIDE : A PROPOS DE 11 CAS. Tun ORL . 2012 ; 28Juin :28-31.
[12]. Yu Gy, Liu XB, Li ZL, Peng X. Smoking And The Development Of Warthin's Tumour Of The Parotid Gland. Br J Oral Maxillofac Surg. 1998 ; 36 :183-5
[13]. Lefor AT, Ord RA. Multiple Synchronous Bilateral Warthin's Tumors Of The Parotid Glands With Pleomorphic Adenoma. Case Report And Review Of The Literature. Oral Surg Oral Med Oral Pathol 1993; 76:319–24.
[14]. Tveterås K, Kristensen S.Warthin's Tumour With Bilateral Synchronous Presentation. Survey Of The Literature And A New Case. J Laryngol Otol 1986; 100:487–492
[15]. A.Teymoortash, Y.Krasnewicz, JA.Werner. Clinical Features Of Cystadenolymphoma Warthin's Tumor) Of The Parotid Gland: A Retrospective Comparative Study Of 96 Cases. Oral Oncol.2006 ;42 :569-73
[16]. Yk.Chung, ML.Khoo, Mk.Heng, Gs.Hong, Kc.Soo. Epidemiology Of Warthin's Tumour Of The Parotid Gland In An Asian Population. Br J Surg. 1999 ; 86 : 661-4
[17]. Espinoza S, Felter A, Malinvaud D Et Al. Warthin's Tumor Of Parotid Gland : Surgery Or Follow-Up ? Diagnostic Value Of A Decisional Algorithm With Functional MRI. Diagn Interv Imaging 2016 ;97(1) :37-43
[18]. Mohamed Masmoudi Et Al. Performance De L´Imagerie Par Résonance Magnétique Dans L´Approche Histopathologique Des Tumeurs Parotidiennes, PAMJ - 39(10). 04 May 2021
[19]. El-Naggar AK, Chan JKC, Grandis JR, Takata T, Slootweg PJ, Editors. WHO Classification Of Head And Neck Tumours. 4th Ed. Lyon: International Agency For Research On Cancer; 2017.
[20]. Skalova A, Nathanskÿ MM, Epidermoid Carcinoma Arising In Warthin's Tumour : A Case Study. J Oral Pathol Med 1994; 23:330-333
[21]. T.Nagao, I.Sugano, Y.Ishida, Y.Tajima, And Al. Mucoepidermoid Carcinoma Arising In Warthin's Tumour Of The Parotid Gland: Report Of Two Cases With Histopathological, Ultrastructural And Immunohistochemical Studies. Histopathology 1998; 33:379-86.
[22]. [Park CK, Manning JT, Battifora H, Medeiros LJ. Follicle Center Lymphoma And Warthin Tumor Involving The Same Anatomic Site Report Of Two Cases And Review Of The Literature. Am J Clin Pathol 2000 ;113 :113—9
[23]. Swerdlow S, Campo E, Harris NL, Jaffe ES, Pileri SA, Stein H, Et Al. WHO Classification Of Tumours Of Haematopoietic And Lym-Phoid Tissue. 4th Ed. Lyon: IARC; 2008.
[24]. Liu Y, Tang Q, Wang LL, Liu Q, Fan S, Li H. Concomitantlymphocyte-Rich Classical Hodgkin's Lymphoma And Warthin'stumor. Oral Surg Oral Med O 2013; 116:117—20.
[25]. Giaslakiotis K, Androulaki A, Panagoulias G, Kyrtsonis MC, Laza-Ris AC, Kanakis DN, Et Al. T-Cell Lymphoblastic Lymphoma Inparotidectomy For Warthin's Tumor: Case Report And Review Ofthe Literature. Int J Hematol 2009; 89:359—64.
[26]. A Boutin, L Chaput, C Lecointre, R Kerdraon, Gi Metrard, P Michenet. An Unexpected Warthin's Tumor. Annales De Pathologie (2015) 35, 270—272
[27]. Thangarajah T, Reddy VM, Castellanos Arango F, Panarese A. Current Controversies In The Management Of Warthin Tumour. Postgrad Med J 2009 ;85 :3–8.
[28]. O. Laccourreyea, ∗ C. Lépine B Warthin's Tumour : Not So Benign ! Https://Doi.Org/10.1016/J.Anorl.2020.05.003
[29]. Ks.H Eller , JN.Attie . Treatment Of Warthin's Tumor By Enucleation. Am J Surg. 1988 ; 156 ; 4 : 294-6.)