Warthin's tumor of the parotid gland: study of 70 cases

Tumor de Warthin da glândula parótida: estudo de 70 casos

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A B S T R A C T

Objective: To report the clinical characteristics, treatment and complication of Warthin tumors. **Methods**: we conducted a retrospective study of 70 patients undergoing resection of papillary lymphomatous cystadenoma. Variables: age, sex, ethnicity, presence or absence of smoking, primary site of tumor, tumor size in cm, presence of bilateral tumor, type of operation, multicentricity, treatment complications, recurrence and follow up. **Results**: Forty-four patients (62.8%) were male and 26 (37.2%) female, with a mean age of 56.7; smoking was present in 55 (78.6%) patients; 66 (94.3%) tumors were unilateral and four (5.7%) bilateral, with an average size of 4.1 cm on physical examination. The complications were 14 (19%) cases of facial paresis and / or paralysis, seroma in 10 (13.8%), Frey's syndrome in three (4%), hematoma in two (2.7%) and wound infection in one (1.5%) case. **Conclusions**: Warthin tumors have a higher incidence in male smokers and predominantly unilateral location of the parotid gland. Superficial parotidectomy with preservation of facial nerve was effective in 100% cases. Complications were transient paresis or paralysis, seroma, Frey syndrome, hematoma and wound infection.

Key words: Adenolymphoma. Salivary glands. Neoplasm of the salivary glands. Parotid region.

INTRODUCTION

The Warthin tumor is a benign neoplasm of salivary glands, being initially reported in 1929 by the pathologist Aldred Scott Warthin, with a location predominantly in the parotid[1]. The term Warthin tumor was used as a synonym for lymphomatous papilliferum cistoadenoma and for cystadenolymphoma and adenolymphoma, and so they are found in the literature.

The Warthin tumor presents peculiar characteristics by virtue of its pathophysiology. The most accepted theory among various authors is the development of the tumor from salivary ducts imprisoned in intraparotid lymph nodes, during embryogenesis or from heterotopic salivary glands[2,3]. Other theories credit its origin to the presence of lymphocytic Infiltration in a pre-existing adenoma[4]. Recent molecular evidence demonstrated the presence of a polyclonal epidermal component and absence of allele losses in cell clones, suggesting an entity that is not a true neoplasm[5].

These tumors have a predilection for the parotid gland and are located in its superficial portion in 90% of cases. When in the parotid region, the lymph nodes display oncocytic and papillary changes and the tumor, when present, displays epithelial differentiations similar to those observed in Warthin tumors present in intraparotid lymph nodes and absence of a stromal lymphoid component[6].

It occurs between the sixth and seventh decades of life, with predominance in the male gender and is associated with smoking[7]. The ideal treatment is parotidectomy with complete dissection of the facial nerve and its branches, the total parotidectomy being advocated by some authors due to the multicentricity of the tumor[8].

The objective of this study is to report clinical features, treatment and complications of Warthin tumors located in the parotid glands.

METHODS

We retrospectively analyzed 83 records of patients with lymphomatous papilliferum cistoadenoma of major salivary glands admitted in the Head and Neck Surgery and Otolaryngology Department of the Hospital Heliopolis – Hosphel, in the period from January 1979 to December 2007 with sufficient information for the collection of the

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variables studied. All cases undergoing surgical treatment and with histopathologic evidence of Warthin tumor were eligible, totaling 70 patients.

The variables analyzed were: age, gender, ethnicity, smoking, primary tumor site, tumor size in centimeters, presence of bilaterality, type of surgery, multicentricity, complications of treatment, presence of relapse and time of follow-up.

The statistical analysis used in the study was descriptive.

RESULTS

On the characterization of the series we noted that 44 (62.8%) patients were male and 26 (37.2%) female. Sixty-one patients were Caucasian (87%) and nine (13%) were non Caucasians (13.3%). The average age group was 56.7 years, ranging from 35 to 80 years. The primary site of the tumor was the parotid gland in all cases (Figures 1 and 2).

In sixty-six patients (94.3%) tumors were unilateral, being bilateral in four (5.7%). Three of the four patients with bilateral tumors were submitted to resection in distinct dates. Distribution of tumors between genders was similar, in a 2:2 proportion. The average size of lesions at clinical examination was 4 cm in diameter, with variation from 2 to 10 cm in the largest diameter, 43 cases (59%) presenting with injuries from 4 to 6 cm. With regard to surgery, a patient was subjected to total parotidectomy (deep pole tumor) with preservation of the facial nerve and the remaining operations were partial parotidectomies with preservation of the facial nerve. Multicentric lesions were not observed during the operative act or upon histological examination.

Fifty-five patients were smokers (78.6%), 34 men (61.8%) and 21 women (38.2%). Forty-three (63%) parotidectomies evolved without postoperative complications. The occurrence of complications, in frequency, were 14 (19%) cases of paresis and/or temporary paralysis of facial nerve branches, seroma in 10 (13.8%), Frey Syndrome in three (4%), hematoma in two (2.7%) and wound infection in one (1.5%) case (Table 1).

We did not observe any relapse during the follow-up, which varied from one to 146 months, averaging 37.4.

DISCUSSION

The lymphomatous papilliferum cistoadenoma is the second most frequent benign neoplasm of salivary glands, with incidence of 2% to 6% of all parotid tumors. It occurs in virtually all cases in the parotid and the presence in other salivary glands is rare and controversial[9]. Eveson and Cawson[10] reported a series of 278 cases, 99.2% being of parotid location and submandibular in two. In some series of tumors in minor salivary glands, the incidence of Warthin tumor was not reported[10,11].

Its occurrence is observed in adult age, particularly between 58 and 70 years. There are studies that suggest incidence of neoplasm in less than 6% of individuals under the age of 40[12]. For some authors, the incidence between man and woman is similar, 1.6:1, while other authors have reported male dominance, a ratio of 5:1[8,13]. Approximately 5% of cases are bilateral, reaching 10%, and in 10% to 15% there is a multicentric clinical manifestation[10]. The multicentricity is frequently observed in Warthin tumors when compared to other salivary gland neoplasms, an argument in favour of development of more than one tumor inside lymph nodes[14].

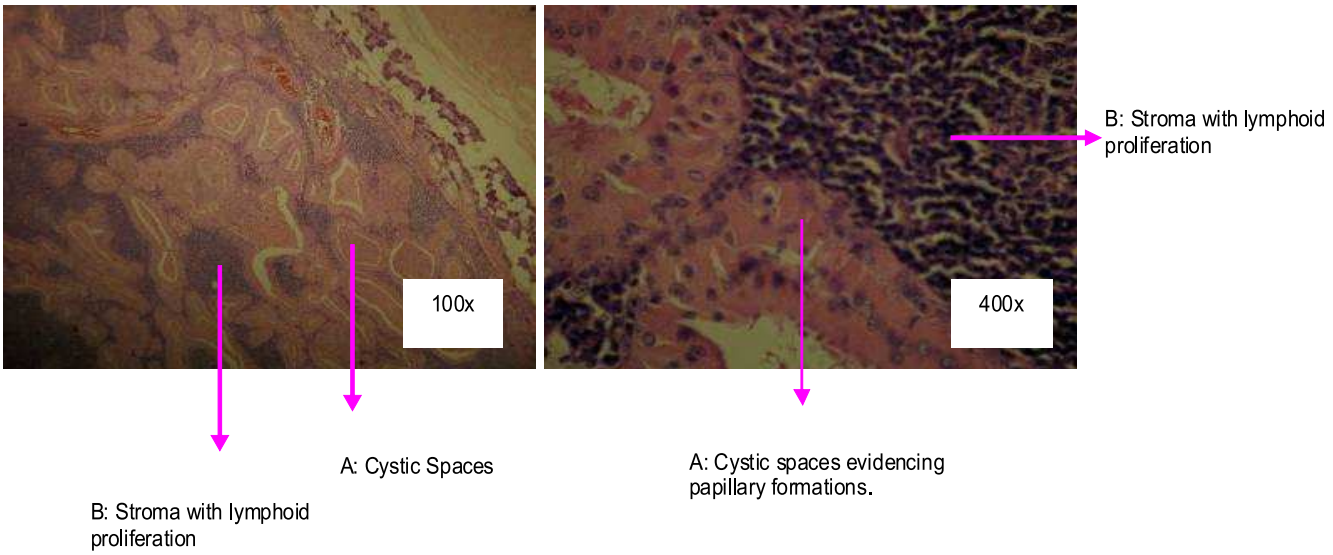

Figure 1 - *Histological cut depicting epithelium of Warthin tumor forming cystic spaces with papillae (A) and lymphoid tissue (B) through HE (hematoxylin-eosin) staining.*

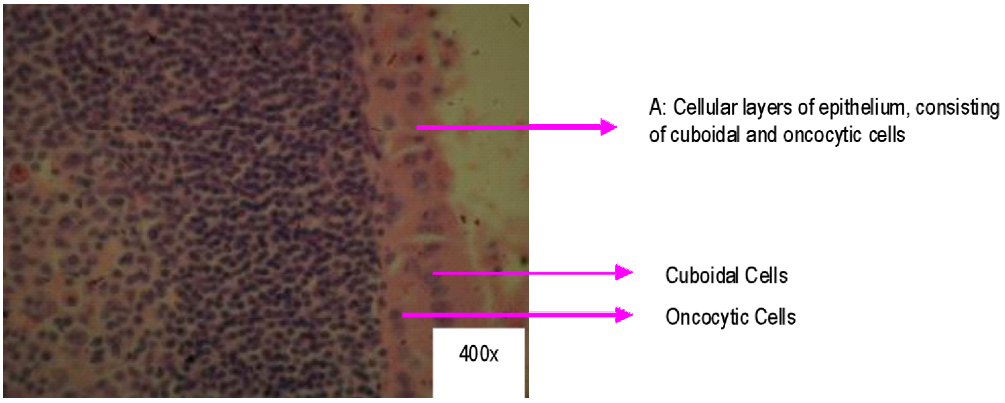

Figure 2 - *Histological cut showing the cellular layers of epithelium of Warthin tumor forming cystic spaces with papillae (A).*

Table 1 - Distribution of postoperative complications.

	n	%
Facial paresis/paralysis	14	19.2%
Seroma	10	13.7%
Frey Syndrome	3	4.0%
Hematoma	2	2.7%
Wound infection	1	1.4%
Without complications	43	59%
Total	73	100%

n = 73 parotidectomies.

The Warthin tumor is usually a nodular lesion and is painless or slightly painful. The patient may complain of tinnitus, pain in the ear and deafness. The size is variable, from a few millimeters to centimeters, averaging 2 to 4 cm in diameter, with a preferred location in the lower pole of the gland (in the jaw angle). According to a study of 278 tumors, 56% had 1 to 3 cm in diameter, 40% from 4 to 6 cm and one exceeded 10 cm[9].

The ideal treatment is parotidectomy with safety margin, with the purpose of preventing potential relapse and because of the high incidence of multicentricity. It is important to stress that there are reports of low incidence of relapse, which may develop after the first, third, up until the tenth year after resection[10]. There are authors who advocate regular semi-annual ambulatory postoperative returns for 10 years with the purpose of diagnosing relapse and/or malignant transformation. Total parotidectomy is advocated by some authors because of the relapse rates of 25% after superficial parotidectomy[15]. We believe that the precaution of regular follow-up with goal of early diagnosis of recurrence and malignant transformation is valid in the case of Warthin tumors associated with other neoplasms with proven potential to malignant transformation as the adenoma in the same gland and treated on the same occasion. We indicate superficial parotidectomy as the ideal and minimum procedure and we do not advocate the periodic return of operated patients, except in situations as described or when there is association of the tumor with malignant neoplasms in minor salivary glands.

Complications must be unusual and of low frequency for the surgical resection of a Warthin tumor, including some complications considered of minor importance, such as paresis of the ear lobe resulting from manipulation and/or section of the *auricularis magnus* branch of superficial cervical plexus. The *auricularis magnus* nerve, in its path toward the ear lobe, may pass through the tumor, hampering the dissection. Another complication of lesser importance is the change of facial contour due to resection of a large portion of the parotid gland.

Frey Syndrome can be considered a sequel expected in parotidectomy, with appearance in a few months, even years, after surgery, by virtue of section of the auriculotemporal nerve, which is accompanied on his path by parasympathetic and sympathetic branches also severed during the procedure. The most accepted hypothesis is the anomalous reinervation of sweat glands of the skin in the area of the external ear, lacrimal gland and nasal mucosa by parasympathetic branches that cause hyperemia and hyperhidrosis of the operated area upon gustatory stimulation. Clinically, it is observed in 35% to 70% of parotidectomies with facial nerve dissection, being present in almost 100% of cases upon application of the iodine test. Preoperative patient knowledge and facilitated postoperative management, considering the pathophysiology of the event, can minimize it with the adoption of surgical and preventive measures, such as the use of antiperspirants in the affected area.

Hematomas, infections and seromas are potentially avoidable complications. Parotid surgery, as well as other major salivary glands procedures, must obey strict principles of surgical hemostasis in the field, facilitating the identification of branches of the facial nerve. In the approach of the deep pole one should employ careful hemostasis of the pterygoid plexus, which may be responsible for larger bleedings. In the review of the surgical field at the end of

the procedure, saline solution should be poured to the removal of clots. In addition to this care, attention to the parameters of the patient's blood pressure should be elevated at the end of the procedure.

The incidence of seroma in this series deserves consideration because of its occurrence in 10 cases (13.7%). Traditionally, the parotidectomies are drained with a Penrose drain associated with local compressive dressing with elastic bandage tape. As a general rule in our service, after the withdrawal of the Penrose drain the bandage is kept for 24 hours and in various situations the patient is discharged with it. Routinely, the drains are kept until the second postoperative day. In resections of larger tumors, over the past 10 years we give preference to the use of continuous suction drains. The permanence of the drain is compulsory until its debt is less than 15-20 ml a day, achievable, on average, between the second and third postoperative day. We can credit the high incidence of seroma to early drain withdrawal and/or absence of compressive dressing after drain removal. However, by the time of the withdrawal of the drain, given its debt, the effectiveness of the compressive dressing may be questioned. Another measure would be the routine adoption of field drainage with continuous suction (negative pressure) drain.

In the presence of the accumulation of fluid under the flap puncture with needle must be carried out and the material sent to the laboratory for amylase measurement. After the puncture, the dressing with elastic bandage tape is necessary. Usually, more than one puncture is needed for resolution of seroma.

Rigor in drainage care and/or dressing is essential in order to avoid complications with a resection for benign disease, made usually in young, adult patients in a tertiary hospital.

Paralysis/paresis of the facial nerve has significant functional and emotional impact in patients submitted to parotidectomy. The reports are ptosis, dysphagia, chewing difficulty and xerophthalmia, and social exclusion resulting from aesthetic sequels. The incidence depends on several factors, including the will of the physician to identify minimum postoperative motor deficits. Some series report 16% to 47% temporary facial palsy and zero to 9% permanent facial paralysis from parotidectomies[16,17]. Paralysis of one or more branches of the facial nerve is more common during the realization of total parotidectomy. In the presence of paralysis, the marginal nerve is the most commonly impaired, the time expected to full postoperative recovery being two to three months. A study of variables related to facial nerve paralysis, such as the patient's age, sex, type of resection, duration of operation, pathology of tumor and size of the lesion showed that age was the variable associated with higher incidence of paralysis. When comparing the experience of the surgeon to cases of facial nerve paralysis there has not been correlation[17].

Traditionally, the surgeon relies on anatomical knowledge, on clinical experience and on operative technique for identification and preservation of the facial nerve during a parotidectomy procedure. In recent years, intraoperative monitoring showed lower indices of temporary paralysis when compared to cases not subject to nerve monitoring[18]. However, intraoperative facial nerve monitoring has well-established indications and we do not deem its use necessary to reduce the number of paresis/paralysis, besides our impossibility to utilize this type of monitoring.

In the study of 70 Warthin tumor cases operated, we observed a higher incidence in males, in smokers and unilateral location prevalent in the Parotid gland. Superficial parotidectomy with preservation of facial nerve proved effective in 100% of cases. The complications encountered were paresis or transitional paralysis, Frey Syndrome, seroma, hematoma and wound infection.

R E S U M O

Objetivo: *Relatar as características clínicas, tratamento e complicações.dos tumores de Warthin.* ***Métodos:*** *Estudo retrospectivo com 70 pacientes submetidos à ressecção de cistoadenoma papilífero linfomatoso,. Variáveis analisadas: idade, sexo, etnia, presença ou não de tabagismo, sítio primário do tumor, tamanho do tumor em centímetros, presença de bilateralidade, tipo de operação, multicentricidade, complicações do tratamento, presença de recidiva no seguimento e tempo de seguimento..* ***Resultados:*** *Quarenta e quatro pacientes (62,8%) eram do sexo masculino e 26 (37,2%), do sexo feminino, com média etária de 56,7 anos, O tabagismo estava presente em 55 (78,6%) pacientes, (94,3%) dos tumores eram unilaterais e quatro (5,7%) bilaterais, com tamanho médio de 4,1cm ao exame físico. As complicações foram 14 (19%) casos de paresia e/ou paralisia facial, seroma em 10 (13,8%), síndrome de Frey em três (4%), hematoma em dois (2,7%) e infecção de ferida operatória em um (1,5%) caso.* ***Conclusão: O*** *tumor de Warthin tem maior incidência no sexo masculino, em tabagistas e localização unilateral predominante na glândula parótida. A parotidectomia superficial com preservação do nervo facial mostrou-se eficaz em 100% casos. As complicações encontradas foram paresias ou paralisias transitórias, seroma, síndrome de Frey, hematoma e infecção de ferida operatória.*

Descritores*: Adenolinfoma. Glândulas salivares. Neoplasia de glândulas salivares. Região parótida.*

REFERENCES

1. Warthin AS. Papillary cystadenoma lymphomatosum. A rare teratoid of the parotid region. J Cancer Res 1929; 13:116-25.
2. Chapnik JS. The controversy of Warthin's tumor. Laryngoscope 1983; 93(6):695-716.
3. Thompson AS, Bryant HC Jr. Histogenesis of papillary cystadenoma lymphomatosum (Warthin tumor) of the parotid salivary gland. Am J Pathol 1950; 26(5):807-49.
4. Barnes L, Eveson JW, Reichart P, Sidransky D, editors. World Health Organization classification of tumors. Lyon: IARC Press; 2005. Pathology and genetics, head and neck tumours; p.209-81.
5. Arida M, Barnes EL, Hunt JL. Molecular assessment of allelic loss in Warthin tumors. Mod Pathol 2005; 18(7):964-8.
6. Faur A, Lazăr E, Cornianu M, Dema A, Vidita CG, Gălu°can A. Warthin tumor: a curious entity—case report and review of literature. Rom J Morphol Embryol 2009; 50(2):269-73.
7. Yoo GH, Eisele DW, Askin FB, Driben JS, Johns ME. Warthin's tumor: a 40-year experience at The Johns Hopkins Hospital. Laryngoscope 1994; 104(7):799-803.
8. Heller KS, Attie JN. Treatment of Warthin's tumor by enucleation. Am J Surg 1988; 156(4):294-6.
9. Párraga-Linares L, Aguirre-Urízar JM, Berini-Aytés L, Gay-Escoda C. Papillary cystoadenoma lymphomatosum (Warthin-like) of minor salivary glands. Med Oral Patol Oral Cir Bucal 2009; 14(11):e597-600.
10. Eveson JW, Cawson RA. Warthin's tumor (cystadenolymphoma) of salivary glands. A clinicopathologic investigation of 278 cases. Oral Surg Oral Med Oral Pathol 1986; 61(3):256-62.
11. Jaber MA. Intraoral minor salivary gland tumors: a review of 75 cases in a Libyan population. Int J Oral Maxillofac Surg 2006; 35(2):150-4.
12. Yih WY, Kratochvil FJ, Stewart JC. Intraoral minor salivary gland neoplasms: review of 213 cases. J Oral Maxillofac Surg 2005; 63(6):805-10.
13. Ellis GL, Auclair PL. Atlas of Tumor Pathology. Washington: Armed Forces Institute of Pathology; 2007. Tumors of the salivary glands. p.1-30.
14. Aguirre JM, Echebarría MA, Martínez-Conde R, Rodriguez C, Burgos JJ, Rivera JM. Warthin tumor. A new hypothesis concerning its development. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1998; 85(1):60-3.
15. 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(3):183-5.
16. Gant TD, Hovey LM, Williams C. Surgical management of parotid gland tumors. Ann Plast Surg 1981; 6(5):389-92.
17. Langdon JD. Complications of parotid gland surgery. J Maxillofac Surg 1984; 12(5):225-9.
18. Mra Z, Komisar A, Blaugrund SM. Functional facial nerve weakness after surgery for benign parotid tumors: a multivariate statistical analysis. Head Neck 1993; 15(2):147-52.

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