ANNALI ITALIANI DI CHIRURGIA

Digital Edition
e-publish on-line
ISSN 2239-253X

Direttore Nicola Picardi

Incidental finding of upper lip Warthin tumor

Ann Ital Chir, Digital Edition 2017, 6
pii: S2239253X17027578 - Epub, November 14
free reading: www.annitalchir.com

Marzia Petrocelli*, Carolina Sbordone**, Giovanni Salzano**, Giovanni Dell'Aversana Orabona**
Francesco Maria Cassandro°, Stefano Fusetti °°, Luigi Califano**, Ettore Cassandro*

**Otolaryngology Unit Department of Medicine, Surgery and Odontoiatrics, University of Salerno, Fisciano, Salerno, Italy*
***Maxillofacial Surgery Unit, Department of Neurosciences, Reproductive and Odontostomatological Sciences, University "Federico II", Naples, Italy.*
°Dentistry Unit, Department of Neurosciences, Reproductive and Odontostomatological Sciences, University "Federico II", Naples, Italy
°°Department of Maxillofacial Surgery, University of Padova, Padova, Italy

Incidental finding of upper lip Warthin tumor

AIM: *This report shows an incidental finding of Warthin tumor in upper lip mucosa during hospitalization for a biting lesion of cheek mucosa*
MATERIALS AND METHODS: *A 32-year-old male affected by a biting lesion of cheek mucosa was presented at Maxillo-Facial Unit of Federico II University. Clinical examination showed as an incidental finding a solid mass in the superficial layer of upper lip mucosa. We performed mini-invasive surgical treatment to obtain a radical excision of the cheek lesion at the same time as excision of Warthin tumor.*
Results: a follow up of 12 months was performed. The complete healing of the two wounds was achieved, with no recurrence of any of the pathologies.
DISCUSSION: *The location of this Warthin tumor of minor salivary glands is very unusual. The role of imaging in diagnosis of Warthin tumor of minor salivary glands is to define localization, shape and dimension, contour, malignant features, nodal involvement. The role of fine needle aspiration cytology (FNAC) is critical in the diagnosis and therapy of minor salivary gland tumors. The surgical treatment in patients affected by Warthin tumour of minor salivary glands is local excision with a wide tumor free margin to prevent potential recurrence.*
CONCLUSIONS: *Warthin tumor of minor salivary glands is a rare disease. We report a singular case of Warthin tumor localized in the upper lip mucosa, found as an incidental finding during a recovery for a biting lesion of cheek mucosa.*

KEY WORDS: Incidental finding, Minor salivary glands, Warthin tumor

Introduction

Papillary cystadenoma lynphomatosum, better known as Warthin tumor, is a benign salivary gland tumor and represents about 13% of all salivary gland tumors [1].

It predominantly occurs in males. The mean age at diagnosis is sixth decade of life and it rarely presents before the forthieth decade [2,3].
It is a well-defined histological entity with a dual component: oncocytic epithelium and basaloid epithelium-forming cystic structures, papillae and glands; and a lymphoid stroma with germinal centers consisting of small T and B-lymphocytes [4,5].
It was first described by Hildebrand in 1895 as a variant of a lateral cyst of the neck (hild) [6]. Albrech and Arzt in the 1910 described an upper neck tumor as papillary cystadenomas in limphonode [7]. Warthin in 1929 described a parotid benign tumor with similar findings

Pervenuto in Redazione Luglio 2017. Accettato per la pubblicazione Settembre 2017
Correspondence to: Giovanni Salzano, University "Federico II", Department of Neurosciences, Reproductive and Odontostomatological Science, Maxillofacial Surgery Unit, Via Pansini 5, 80131 Naples, Italy (e-mail: giovannisalzanomd@gmail.com)

of previous tumor that was therefore renamed after him[8]. The Warthin tumor commonly arises in the parotid gland (84%), less frequently in the submandibular glands (7%) or in cervical lymphonodes (8%), rarely arises in minor salivary glands (1%) [9].
Diagnosis of Warthin tumor of salivary glands is based on clinical evidence, fine needle aspiration cytology (FNAC), ultrasound examination, MRI or CT scan examination in cases not accessible by conventional ultrasound [10-12]. This report shows an incidental finding of Warthin tumor in upper lip mucosa during hospitalization for a biting lesion of cheek mucosa. A mini-invasive surgical treatment with local anesthesia allowed to perform a contemporary radical excision of the cheek lesion and the Warthin tumor.

Case Report

In February 2015, a 32-year-old male was presented at Maxillo-Facial Unit of Federico II University, Naples, Italy, for an oral mucosa lesion. At clinical examination of oral cavity, the patient had a biting lesion on lower right cheek mucosa, covered with whitish and frayed mucosa of 2,5 x 1,5 cm in size. The patient also had swelling of the right upper lip mucosa. The swelling was palpable at the intraoral examination and a submucosal cystic formation of about 3 x 4 cm in size was identified, floating on deep and superficial layers, with soft elastic consistency, painless to superficial and deep palpation, covered with normotrophic and normochromic mucosa (Fig. 1a). The ultrasound examination of the neoformation were prescribed and performed during the first access to the hospital. The ultrasound in our case showed a well-defined cystic cavity, filled with hypoechoic solid mass of about 3 x 4 cm in size. In February 2015, under local anesthesia with mepivacaina 1%, we outlined the cheek lesion of the right geniene mucosa with macroscopic safe margins of 1 cm and we performed the total excision of the lesion. After that, in the same surgical time, a Warthin neoformation of the right upper lip mucosa was identified and exposed through an horizontal incision of 3 cm. An accurate blunt dissection was performed with preservation of the right Stenone salivary duct and of the facial artery and vein (Fig. 1b). The neoformation was isolated and total excised in whole with its capsule (Fig. 1c); an accurate haemostasis was performed. Wound was then closed by vycril 3/0 stitches. The two lesions were sent for histopathological examination. The definitive histopathological diagnosis of the cheek lesion was of leucocherathosis of oral mucosa. The definitive histopathological analysis of the upper lip neoformation diagnosed a Warthin tumor. It described an oncocytic epithelium and basaloid epithelium that formed cystic structures and a lymphoid stroma with germinal centers consisting of small T and B-lymphocytes. A follow up of 12 months was performed (Fig. 1d). A clinical examination and an ultrasound control were performed at 3 and 6 month after surgery. The complete healing of the two wounds was achieved, with no recurrence of any of the pathologies.

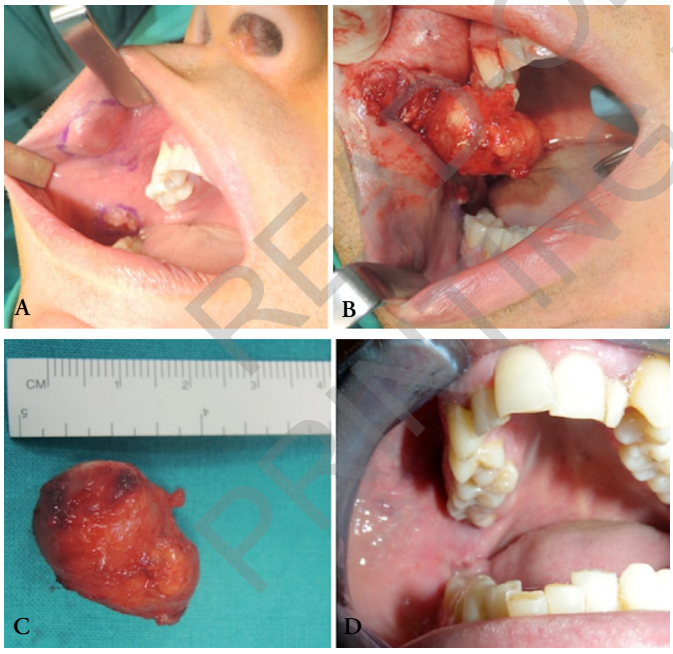

Fig. 1: A) PREOPERATIVE IMAGE: Intraoral view of swelling in the right upper lip buccal mucosa and of biting lesion of the lower cheek mucosa; B) INTRAOPERATIVE IMAGE : Intraoral view of the tumor that was about 3 x 4 cm of size; C) WARTHIN TUMOR: Macroscopic aspects of warthin tumor that was isolated and total excised in whole with its capsule; D) POSTOPERATIVE IMAGE: Intraoral view of the right upper lip mucosa after 12 months from surgery.

Discussion and Comments

The location of this Warthin tumor of minor salivary glands is very unusual. It generally arise from parotid glands (84%), less frequently in the submandibular glands (7%), in the cervical limphonodes (8%), rarely arises in minor salivary glands (1%) [1,2]. Hendrick et al. in the 1964 described 1 case of Warthin tumor of the palate[12] . Eveson et al. in the 1986 described 2 cases of submaxillary glands Warthin tumor on a data of 278 patients affected by Warthin tumor of salivary glands[13]. In 2009, Parraga-Linares et al. described a case of Warthin tumor of palate[2]. In 2012 Iway et al. reported a case of Warthin tumor of buccal mucosa [1].
The Warthin tumor have a male propensity with a ratio of 5:1. Tipically. it appears in 35-80 years old patients and major incidence is reported between 55-65 years[14-17]. Etiological origin is multifactorial, it depends on immunological reaction during the tumor development; genetic

and environmental factors seem to play a role in the origin of this tumor. Allergic diathesis stimulated the reactivity of germinal centres in the limphoid stroma and induced epithelial proliferation [18-21].

Macroscopically, Warthin tumor (WT) presents as an ovoidal mass with a capsule filled with a viscous dark material.

Microscopically, WT is a circumscribed cystic entity composed by dual component: oncocytic epithelium and basaloid epithelium-forming cystic structures, papillae and glands; and a lymphoid stroma with germinal centres consisting of small T and B-lymphocytes. The accredited hypothesis on the pathogenesis of this tumor is that it arises from proliferative salivary gland ductal cells entrapped and developed within lymphonodes involvement with a secondary lymphoid reactive response in the stroma [2,22,23].

Clinically, the Warthin tumour usually presents as a well circumscribed neoformation, as a fluctuant mass, with a size ranging from few millimiters to centimeters, with a slow growing. The differential diagnosis should be performed with pleomorphic adenoma and cystoadenoma through histological analysis [3].

The role of imaging in diagnosis of Warthin tumor of minor salivary glands is to define localization, shape and dimension, contour, malignant features, nodal involvement. The ultrasound analysis is an ideal tool for initial assessment. The MRI or CT scans are mandatory, mostly in cases not accessible by conventional ultrasound, to evaluate tumor extent and its radiographic features [9]. The role of fine needle aspiration cytology (FNAC) is critical in the diagnosis and therapy of minor salivary gland tumors. The value of FNAC is to obtain a specific diagnosis with a specificity of 95%, sensitivity of 97.6% and diagnostic accuracy of 96.3%; it should be considered in the first line investigation salivary gland tumors [10,11]. However, it is essential to evaluate FNAC results in the context of all clinical and instrumental information [24-30].

The surgical treatment in patients affected by Warthin tumour of minor salivary glands is local excision with a wide tumor free margin to prevent potential recurrence. In our case, in accordance with surgical protocol about the management of WT, we performed a radical excision of the tumor. We did not perform a FNAC examination because there was an incidental finding of Warthin tumor, so we preferred to perform an excisional biopsy. In fact we removed the tumor from right upper lip mucosa in whole, with its capsule, through an accurate blunt dissection with preservation of the right Stenone salivary duct and of the facial artery and vein. An accurate hemosthasis and wound closure were performed. According with WHO classification of Warthin tumor, the definitive histopathological analysis described an oncocyte epithelium and basaloid epithelium that formed cystic structures and a lymphoid stroma with germinal centers consisting of small T and B-lymphocytes [4,5].

Conclusion

In conclusion we can state that Warthin tumor of minor salivary glands is a rare disease. We report a singular case of WT tumor localized in the upper lip mucosa, found as an incidental finding during a recovery for a biting lesion of cheek mucosa. We performed a mini-invasive surgical treatment and achieved a radical excision of the cheek lesion contemporary to total excision of Warthin tumor, with a complete healing without recurrence of pathology.

Riassunto

Presentiamo un caso di Tumore di Warthin delle ghiandole salivari minori in un maschio adulto caucasico affetto da una lesione da masticamento della mucosa della guancia. L'esame clinico mostrava un riscontro accidentale di una massa solida situata nello strato superficiale della mucosa del labbro superiore. L'esame ecografico ha evidenziato una massa solida ipoecogena ben definita di circa 3x4 cm. Abbiamo eseguito un trattamento chirurgico mini-invasivo nel febbraio 2015 per ottenere una escissione radicale della lesione guancia e contemporaneamente del tumore di Warthin del labbro superiore senza recidiva della patologia.

References

1. Iwai T, Baba J, Murata S, Mitsudo K, Maegawa J, Nagahama K, et al.: *Warthin tumor arising from the minor salivary gland.* J Craniofac Surg, 2012; 23:374-76.

2. Párraga-Linares L, Aguirre-Urízar JM, Berini-Aytés L, Gay-Escoda C: *Papillary cystoadenoma lynphomatosum (Warthin like) of minor salivary glands.* Med Oral Patol Oral Cir Bucal, 2009; 14:597-600.

3. Dell'Aversana Orabona G, Abbate V, Piombino P, Romano A, Schonauer F, Iaconetta G, et al.: *Warthin's tumour: Aetiopathogenesis dilemma, ten years of our experience*. Journal of Cranio-Maxillo-Facial Surgery, 2015; 43:427-31.

4. Barnes L, Eveson JW, Reichart P, Sidransky D, (eds): *Pathology and genetics of head and neck tumours*. Lyon: IARC Press. WHO classification of tumours series 2005; 306.

5. Ellis GL: *Lymphoid lesions of salivary glands: malignant and benign*. Med Oral Patol Oral Cir Bucal, 2007; 12: E 479-85.

6. Hildebrand O: *Ber angeborene epitheliale Cysten und Fistlen des Halses*. Arch Klin Chir, 1895; 49:167-92.

7. Albrecht H, Arzt L: *Zur Frage der Gewebsverirrung. I. Papillare Cystadenome in Lymphdrusen.* Frankf Z Pathol, 1910; 4:47-69.

8. Warthin AS: *Papillary cystadenoma lymphomatosum: A rare teratoid of parotid region*. J Cancer Res, 1929; 13:116-125.

9. Yoo GH, Eisele DW, Askin FB, Driben JS, Johns ME: *Warthin's tumour: A 40-year experience at the Johas Hopkins Hospital.* Laryngoscope, 1994; 104:799-803.

10. Lee YY, Wong KT, King AD, Ahuja AT: *Imaging of salivary gland tumours*. Eur J Radiol, 2008; 66:419-36.

11. Nag D, Biswas PK, Mandal PK, Bhattacharyya NK, Gautam D, Mukhopadhyay S: *Tumours of minor salivary glands. A clinicopathologic study*. J Indian Med Assoc, 2012; 110:567-69, 577.

12. Hendrick JW: *Papillary cystadenoma lymphomatosum of the palate. Report of a case*. Arch Otolaryngol, 1964; 79:15-7.

13. Eveson JW, Cawson RA: *Warthin's tumor (cystadenolymphoma) of salivary glands. A clinicopathologic investigation of 278 cases*. Oral Surg Oral Med Oral Pathol, 1986; 61:256-62.

14. Gallo O, Bocciolini C: *Warthin's tumour associated with autoimmune diseases and tobacco use.* Acta Otolaryngol, 1997; 117:623-27.

15. Dos Santos Almeida A, Costa Hanemann JA, Tostes Oliveira D: *Warthin tumor of the upper lip: an unusual location of a benign salivary gland tumor.* Int J Surg Pathol, 2011; 19:366-68.

16. Chaudhry AP, Gorlin RJ, Mitchell DF: *Papillary cystadenoma of minor salivary gland origin. Report of a case.* Oral Surg Oral Med Pathol, 1960; 13:452-54.

17. Friedlander AH, Thorn R: *Warthin's tumor in an intra oral minor salivary gland.* Quintessence Int Dent Dig, 1977; 8:9-11.

18. Fantasia JE, Miller AS: *Papillary cystadenoma lymphomatosum arising in minor salivary glands.* Oral Surg Oral Med Pathol, 1981; 52:41-6.

19. Friedlander AH, Thorn R: *Warthin's tumor in an intra oral minor salivary gland.* Quintessence Int Dent Dig. 1977; 8:9-11.

20. Tilakaratne WM, Jayasooriya PR, Tennakoon TM, Saku T: *Epithelial salivary tumors in Sri Lanka: A retrospective study of 713 cases*. Oral Surg Oral Med Oral Pathol Oral Radiol Endod, 2009; 108:90-8.

21. Ito FA, Ito K, Vargas PA, de Almeida OP, Lopes MA: *Salivary gland tumors in a Brazilian population: a retrospective study of 496 cases*. Int J. Oral Maxillofac Surg, 2005; 34:533-36.

22. Salzano FA, Guastini L, Mora R, Dellepiane M, Salzano G, Santomauro V, Salami A: *Nasal tactile sensitivity in elderly.* Acta Otolaryngol, 2010; 130(12):1389-393.

23. Catalfamo L, Nava C, Lombardo G, Iudicello V, Siniscalchi EN, Saverio de PF: *Tongue lymphangioma in adult*. J Craniofac Surg, 2012; 23(6):1920-922.

24. Dell'aversana Orabona G, Salzano G, Petrocelli M, Iaconetta G, Califano L: *Reconstructive techniques of the parotid region*. J Craniofac Surg, 2014; 25(3):998-1002.

25. Iaconetta G, Friscia M, Dell'Aversana Orabona G, de Biasi S, Romano A, Piombino P, Graziano P, Abbate V, Salzano G, Maglitto F, Califano L: *Castleman's disease mimicking a parotid gland tumor: report of a case and review of the literature*. Eur Rev Med Pharmacol Sc, 2014; 18(8):1241-246.

26. Dell'Aversana Orabona G, Salzano G, Abbate V, Piombino P, Astarita F, Iaconetta G, Califano L: *Use of the SMAS flap for reconstruction of the parotid lodge*. Acta Otorhinolaryngol Ital, 2015; 35(6):406-11.

27. Dell'Aversana Orabona G, Iaconetta G, Abbate V, Piombino P, Romano A, Maglitto F, Salzano G, Califano L: *Head and neck myxofibrosarcoma: A case report and review of the literature*. J Med Case Rep, 2014; 29; 8:468.

28. Naz S, Hashmi AA, Khurshid A, Faridi N, Edhi MM, Kamal A, et al.: *Diagnostic role of fine needle aspiration cytology (Fnac) in the evaluation of salivary gland swelling: An institutional experience.* BMC Research Notes, 2015; 8:101.

29. Sood S, Mc Gurk M, Vaz F: *Management of salivary gland tumours: United Kingdom national multidisciplinary guidelines*. The Journal of laryngology and Otology, 2016; 130:142-49.

30. Guerra G, Testa D, Montagnani S, Tafuri D, Salzano FA, Rocca A, et al.: *Surgical management of pleomorphic adenoma of parotid gland in elderly patients: Role of morphological features.* Int J Surg, 2014; 12:S12-16.