Incidental finding of upper lip Warthin tumor

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

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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 Maxillofacial 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 lymphomatous, 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.
of previous tumor that was therefore renamed after him. 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%).

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. 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 neoforation 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 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.

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%). Hendrick et al. in the 1964 described 1 case of Warthin tumor of the palate. 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. In 2009, Parraga-Linares et al. described a case of Warthin tumor of palate. In 2012 Iway et al. reported a case of Warthin tumor of buccal mucosa.

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. 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 lymphoid 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: oncocyte 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 hemostasis 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 ipoeccogena ben definita di circa 3x4 cm. Abbiamo eseguito un trattamento chirurgico mini-invasivo nel febbraio 2015 per ottenere una esecizione radicale della lesione guancia e contemporaneamente del tumore di Warthin del labbro superiore senza recidiva della patologia.

References


