

Lymphoepithelioma-Like Carcinoma of the Vermilion of Lower Lip: A Case Report and Review of Literature

Authors:

¹⁾ Zahra SAYAD¹: corresponding author

Department of Oral and Maxillofacial surgery, Ibn Sina University Hospital center
Faculty of Medicine and Pharmacy of Rabat, Mohammed V University in Rabat, Morocco

²⁾ Olaya HAMIDI¹:

Department of Oral and Maxillofacial surgery, Ibn Sina University Hospital center
City: Rabat
Country: Maroc

³⁾ Hafsa Elouazzani²:

Department of Pathology, Hospital of Specialities,
Ibn Sina University Hospital center
Faculty of Medicine and Pharmacy of Rabat,
Mohammed V University in Rabat, Morocco

⁴⁾ Salma BENZAZZOU¹:

Department of Oral and Maxillofacial surgery,
Ibn Sina University Hospital center
Faculty of Medicine and Pharmacy of Rabat,
Mohammed V University in Rabat, Morocco

⁵⁾ Nadia CHERRADI²:

Department of Pathology, Hospital of Specialities,
Ibn Sina University Hospital center
Faculty of Medicine and Pharmacy of Rabat,
Mohammed V University in Rabat, Morocco

⁶⁾ Malik BOULAADAS¹:

Department of Oral and Maxillofacial surgery,
Ibn Sina University Hospital center
Faculty of Medicine and Pharmacy of Rabat,
Mohammed V University in Rabat, Morocco

Abstract:- Lymphoepithelioma-like carcinoma is a rare neoplasm of the oral cavity. It was first described by Hilderman et al. in 1962 in the parotid gland. In most cases, Epstein-Barr virus (EBV) is associated with this type of carcinoma in endemic regions. We report the observation of a 70-year-old woman presented with chronic vermilion ulceration of the lower lip, a biopsy of which revealed lymphoepithelioma-like carcinoma. The patient underwent a large carcinologic excision. Its management is based on surgery and adjuvant radiotherapy.

Keywords:- Carcinoma, Lymphoepithelioma Like, Lower Lip, Treatment.

I. INTRODUCTION

Lymphoepithelioma-like carcinoma or undifferentiated carcinoma with lymphoid stroma is a rare subtype of neoplasms of the oral cavity, which derives its name from its histological resemblance to lymphoepithelial carcinoma of the nasopharynx. The World Health Organization (WHO) has defined it as poorly differentiated squamous cell carcinoma. (1) Its particularly high incidence in Eskimos and East Asians. In most cases, Epstein-Barr virus (EBV) is associated with this type of carcinoma in endemic regions.

Due to its rarity, there is no established treatment protocol. Treatment strategies are based on surgery and adjuvant therapies, including chemotherapy and radiotherapy. In recent years, significant progress has been made in cancer immunotherapy. (2) The clinical course and prognosis of this tumor after treatment have not been extensively studied in the literature.

We report a case of vermilion lymphoepithelioma-like carcinoma of the lower lip in a 70-year-old woman, and we will discuss the pathogenetic, anatomoclinical and therapeutic aspects of this tumor.

II. PATIENT AND OBSERVATION

A 70-year-old woman with a history of arterial hypertension under dual therapy, coronary artery disease under antiplatelet therapy, who was presented with an ulcerative lesion of the lower lip which has been progressing for 12 months and has been resistant to usual treatments. The clinical examination objectified an ulceration of the vermilion of the lower lip slightly painful, not bleeding on contact, measuring 2 cm long axis. (fig.1) The endooral and ENT examination was normal. Cervical examination did not find cervical lymphadenopathy.

A biopsy was taken, the anatomopathological analysis of which objectified a tumor proliferation consisting of large cells with abundant eosinophilic cytoplasm provided with anisocaryotic nuclei with vesicular chromatin and centered on a prominent eosinophilic nucleolus with the presence of mitotic activity and a few apoptotic bodies. These cells are arranged in clumps and rows in an inflammatory edematous stroma. The immunohistochemical study showed a clear positivity of anti-AE1 / AE3 and anti-P40 in favor of lymphoepithelioma-like carcinoma. (fig.2)

The cervical ultrasound was without abnormalities, while the EBV serology was positive. The patient underwent a carcinological resection of the tumor under local anesthesia, with a W-plasty. (fig.3) The postoperative effects were particularities. Anatomopathological study of the excisional piece confirmed the diagnosis and showed healthy excision margins. The patient was referred for radiotherapy for further treatment. At a follow-up of 16 months, no sign of recurrence was noted.

III. DISCUSSION

Lymphoepithelioma-like carcinoma is a rare malignant tumor of the oral cavity, which is characterized histologically by a non-keratinizing and undifferentiated squamous cell carcinoma with lymphocyte infiltration. (3)

Non-nasopharyngeal lymphoepithelial carcinoma was first described by Hilderman et al. In 1962 in the parotid gland (4). However, can exist in other locations of the head and neck, it has even been described in the cervix, lung, stomach, and thymus. It has been associated with genetically determined susceptibility to exposure to chemical carcinogens at a young age and latent infection with Epstein-Barr virus (5)

Recently, human papilloma virus HPV has also been suggested as a potential etiologic factor for squamous cell carcinomas with similar morphology to EBV-related lymphoepithelial carcinomas. (6) A study is carried out by ALESKI et al (1), which revealed a mean age of diagnosis of 50 years with a male predominance.

A correct diagnosis should therefore include an immunohistochemical examination, looking for the expression of cytokeratin to identify the poorly differentiated epithelial component. (8)

Metastatic lymphoepithelial carcinoma and primary lymphoepithelial carcinoma are histologically and immunophenotypically identical, but are different in their association with EBV, metastatic carcinomas are positive for EBV.

Its management should include a thorough ear, nose, and throat examination to rule out a metastatic form of the nasopharynx, which is known as a radiosensitive tumor. However, the standard treatment for this tumor with an extra nasopharyngeal location is surgical, consisting of a large excision, to be completed with adjuvant radiotherapy. (9, 10)

IV. CONCLUSION

Non-nasopharyngeal lymphoepithelial carcinomas of the head and neck are rare epithelial tumors, the pathogenesis of which remains poorly understood and appears to be multifactorial. Treatment strategies are based on surgery and other therapies, including chemotherapy and radiation therapy.

Competing interests

The authors declare no competing interests.

REFERENCES

- [1]. Aleks E. Rytko nen • Pasi P. Hirvikoski • Tuula A. Salo. Lymphoepithelial Carcinoma: Two Case Reports and a Systematic Review of Oral and Otherwiseal Cases. *Head and Neck Pathol* (2011) 5: 327–334 DOI 10.1007 / s12105-011-0278-7
- [2]. Zhengwu Tanga, Rui Fangb, Guihui Tongc, Peng Liud, Zhu'an Oud, Yong Tangd. Overcoming resistance to anti-PD-1 immunotherapy in lymphoepitheliomalike carcinoma: A case report and review of the literature. *Lung Cancer* 146 (2020) 335–340
- [3]. TL Chow 1, TK Chow, YH Lui, WM Sze, NWF Yuen, SPY Kwok. lymphoepithelioma like carcinoma of oral cavity: report of three cases and literature review. *Int J Oral Maxillofac Surg* 2002 Apr
- [4]. Hilderman WC, Gordon JS, Large HL, Carrol CF. Malignant lymphoepithelial lesion with carcinomatous component apparently arising in parotid gland. A malignant counterpart of benign lymphoepithelial lesion? *Cancer* 1962; 15: 606–10.
- [5]. Zong Y, Liu K, Zhong B, Chen G, Wu W. Epstein-Barr virus infection of sinusal lymphoepithelial carcinoma in Guangzhou. *Chin Med J (Engl)* 2001; 114: 132–6
- [6]. Singhi AD, Stelow EB, Mills SE, et al. Lymphoepithelial-like carcinoma of the oropharynx: a morphologic variant of HPV-related head and neck carcinoma. *Am J Surg Pathol.* 2010; 34: 800–5.
- [7]. Loren E Clarke 1, Michael D Ioffreda. lymphoepithelima like carcinoma with spindle cell differentiation. *J Cutan Pathol.* 2005 Jul; 32 (6): 419-23
- [8]. M. Khouchania, B. Elmorabita, A.Elomrania, A.Tahria, A.Moutaouakilb, B. Belabidiac. Lymphoepithelial carcinoma of the conjunctiva: an exceptional location. *Cancer / Radiotherapy* 16 (2012) 219–221
- [9]. NEKTARIOS I. LOUNTZIS, MD, y WILLIAM B. TYLER, MD, y AND VICTOR J. MARKS, M. Primary Cutaneous Lymphoepithelioma-Like Carcinoma Treated Using Mohs Micrographic Surgery and Zinc Chloride Fixative. *Dermatol Surg* 2010; 36: 564–567
- [10]. Y HSIUNG, MD, 2C-C HUANG, MD, 1C-J WANG, MD, 1E-Y HUANG, MD and 2H-Y HUANG, MD. Lymphoepithelioma-like carcinoma of salivary glands: treatment results and failure pattern. *The British Journal of Radiology*, 79 (2006), 52–55 E 2006 The British Institute of Radiology

Figures



Figure 1: Clinical view of the ulcerated lesion on the right side of vermilion of lower lip.

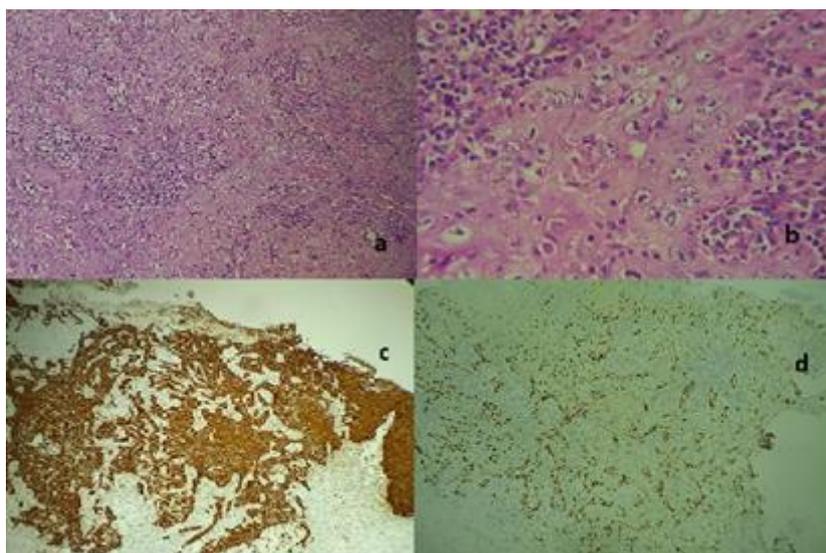


Figure 2: Photomicrographs showing: **a-** carcinomatous proliferation made up of syncytial clumps of tumor cells developed in an abundant lymphoid stroma (H&E stain, original magnificationX20). **b-** the tumor cells have an eosinophilic cytoplasm with imprecise boundaries and provided with nucleolus and mitotic vesicular nuclei. (H&E stain, original magnificationX40). **c-** cytokeratin AE1/AE3 positivity highlighting the malignant cells; **d-** express p40.



Figure 3: clinical view of the patient 1 week after surgery.