

Oral mucous keratoacanthoma secondary to CO₂ laser vaporization of a dysplastic lesion of oral cGVHD

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Abstract – Introduction: The oral mucosa is involved in 80% of patients with chronic Graft Versus Host Disease (cGVHD). Oral mucous keratoacanthoma (KA) has never been reported in these patients. **Case report:** We report the case of a patient with gingival KA that developed after laser CO₂ therapy of an oral lichenoid cGVHD lesion. **Discussion:** In spite of its exceptional character, this case poses the question of etiology, differential diagnosis and treatment of oral mucous KA in patients with cGVHD.

Mots clés :

cGVHD / kératoacanthome / laser

Résumé – Kératoacanthome muqueux secondaire à une lésion dysplasique de GVHD traitée par laser CO₂. **Introduction :** La muqueuse orale est touchée dans 80 % des cas de maladie du greffon contre l'hôte (cGVHD). Aucun cas de kératoacanthome muqueux n'a été décrit dans les lésions muqueuses de cGVHD. **Observation :** Le premier cas de kératoacanthome muqueux secondaire à un traitement laser CO₂ d'une lésion lichénoïde est décrit. **Discussion :** Ce cas exceptionnel pose les questions de l'étiologie, du diagnostic différentiel et du traitement des kératoacanthomes chez les patients présentant une cGVHD orale.

Case report

A 43-year-old man presented for evaluation of a recurrent gingival lichenoid lesion. This lesion appeared following allogeneic hematopoietic stem cell transplant (HSCT) for acute myeloid leukemia in 2003. In 2005, clinical examination revealed a keratinized gingival lesion at teeth 36 and 37. Surgical excision was performed and histology revealed moderate dysplasia. Three years later, the same lesion with moderate dysplasia recurred. Excision was done with CO₂ laser. Three months later, a keratinized lesion developed at the same site. Because of rapid growth and the circular keratinized volcano-like shape, a mucous KA was suspected (Fig. 1a). Total excision was performed. Histology revealed a keratin-filled crater associated with cellular atypia (Fig. 1b). Twelve months after the excision, there was no sign of recurrence and no cervical lymphadenopathy. The patient is still on regular follow-up. The clinical presentation, histology and clinical course allowed us to make the diagnosis of mucous KA.

Discussion

KA was first described in 1889 by Jonathan Hutchinson. Men are affected about 3 times more often than women. Mucous KA is an extremely rare lesion [1]. The incidence of KA is unknown because many lesions are treated as squamous cell carcinoma (SCC) and others are not treated because of their tendency to

regress spontaneously. The etiology of KA is still a subject of debate. Cutaneous KA has been documented after inflammatory skin disorders, trauma, immunosuppression, UV exposure, thermal burns, radiation therapy, fractional photothermolysis, surgery and laser treatment [2, 3]. CO₂ laser resurfacing is known to be an inciting event causing traumatic KAs on the skin and on the lip [4]. The laser produces a wound with significant inflammation and can lead to the development of KA [4]. Cutaneous KAs have been reported in postoperative healing wounds or surgical scars after the removal of skin cancer [5]. In the present case, the previous surgical excision of the moderate dysplasia as well as the inflammation associated with the lichenoid cGVHD lesion and laser CO₂ treatment may have favored the development of KA. The difference in diagnosis between KA and SCC may carry therapeutic implications, making it important to discriminate between the two [5]. In our case, the dysplasia in the lesion raises the issue of the differential diagnosis between KA and well-differentiated oral SCC. Clinically, KA differs from SCC by its history of rapid growth and its volcano-like shape, but the only true test of a distinction is spontaneous involution of the KA [5]. Sometimes lesions regarded as KA have to be reclassified as SCCs on the basis of their subsequent clinical course. In addition to a mistaken diagnosis, other explanations are the combination of KA and SCC, as well as transformation of KA into SCC. In our case, the oral lesion had self-limited, rapid growth and a keratinized volcano-like shape consistent with a KA [6]. KA and SCC can be differentiated histologically in typical cases

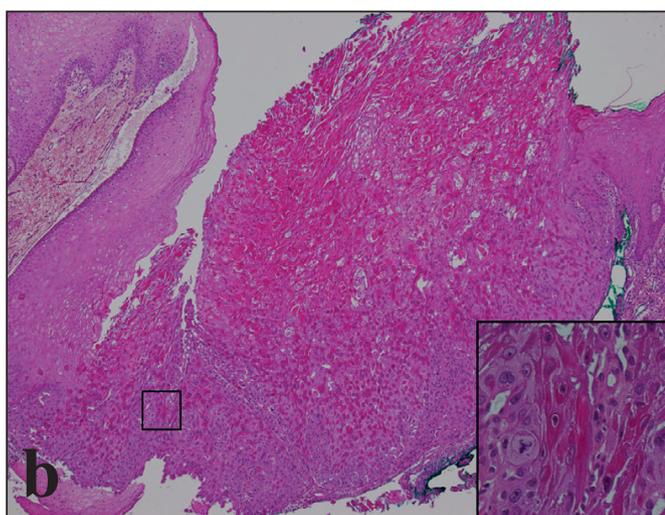


Fig. 1. a) Macroscopic view of the gingival lesion: firm, circular dome-shaped nodule with a keratin-filled crater in the center. b) Histological section of the lesion: central well-demarcated, keratin-filled, crater surrounded by asymmetrical, shallow, cup-shaped invaginations of thin atrophic epithelium with overhanging edges (hematoxylin-eosin stain; original magnification $\times 4$). At higher magnification, scattered atypical and hyperchromatic cells along with keratinizing cells can be seen (hematoxylin-eosin stain; original magnification $\times 20$).

Fig. 1. a) Vue macroscopique de la lésion gingivale : papule kératinisée. b) Coupe histologique mettant en évidence un cratère rempli de kératine, bordé par un fin épithélium asymétrique (coloration hématoxyline-éosine ; grossissement $\times 4$). À plus fort grossissement ($\times 20$), à la base de la lésion, des cellules hyperchromatiques et atypiques sont observées.

but this is much more difficult in borderline cases. In our case, the lesion had a keratin-filled crater surrounded by a proliferative atypical squamous epithelium consistent with a gingival KA. However, because of the asymmetrical cup-shaped

invaginations of the epithelium on histological section, and because of the enlarged hyperchromatic and atypical cells at the base of the lesion, it had to be differentiated from oral SCC. The clinical presentation and the lack of recurrence after surgical excision were in favor of the diagnosis of mucous KA. A dilemma exists regarding the management of KA. Because it can regress spontaneously, some advocate an observational approach. Others maintain that it is a malignant neoplasm – a variant of SCC – and therefore should be treated like SCC [7]. Alternative treatments such as systemic retinoids, radiotherapy, curettage, electrodesiccation, 5-fluorouracil combined or not with Er:YAG laser, CO₂ laser, intralesional methotrexate, intralesional interferon alpha-2b and topical imiquimod have also been used, but less frequently. In our case, the history of dysplasia in the lesion was an indication for surgical treatment. Because of the difficulties in distinguishing between KA and SCC, it is important to carry out regular follow-up after surgical treatment, particularly in potentially malignant disorders like oral lichenoid cGVHD lesions.

Conflicts of interests: none declared

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