

Multifocal diffuse oral melanoacanthoma: a case report

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Abstract: Oral melanoacanthoma (OMA) is a rare benign lesion characterized by colonization of acanthotic epithelium by dendritic melanocytes. Although its pathogenesis remains uncertain, its clinical behavior and spontaneous remission suggest a non-neoplastic nature. Clinically, it may present as a solitary or multifocal lesion; however these two variants exhibit different features. The clinical appearance of OMA is not pathognomonic and biopsy is mandatory. OMA requires no treatment or periodic observation. Here, we report a case of OMA with diffuse lesions also affecting the tongue in a 74-year-old black woman, whose diagnosis was based essentially on clinical and histological features. The immunohistochemical profile is also presented. (J Oral Sci 51, 463-466, 2009)

Keywords: oral melanoacanthoma; oral pigmented lesion; melanocyte.

Introduction

The term melanoacanthoma (MA) was first used by Mishima and Pinkus in 1960 to describe a benign mixed skin tumor composed of basal and prickle cell keratinocytes and pigment-laden dendritic melanocytes (1). Matsuoka et al. in 1979 reported the first case in the oral cavity (2).

Oral melanoacanthoma is a rare pigmented lesion that may present as a solitary or multifocal lesion (3-5) characterized by sudden appearance and rapid radial

growth (4,5). Oral lesions occur almost exclusively in black females, affecting a wide age range (from the first to the eighth decades) (5). OMA is a distinct entity not related to the cutaneous MA, which is considered to be a variant of seborrheic keratosis (5). It may present as a solitary or multifocal lesion; however, these two variants exhibit different demographic features (5). According to Yaron et al., only about 53 patients with 72 cases of oral MA have been reported in the literature (43 solitary and 10 multifocal) (5). Although a female predominance has been observed in both variants, multifocal OMA presents an equal gender distribution and tends to occur more frequently on the palate (5). In contrast, buccal mucosa is usually the most affected site in the solitary cases (4,6). Solitary or multifocal, they are thought to have a reactive nature and usually they regress spontaneously or after incomplete removal, such as an incisional biopsy (3,5,6). Clinically, OMA presents as a black or brownish lesion with a flattened surface (5,6). Histologically, OMA is characterized by the proliferation of keratinocytes and pigmented dendritic benign melanocytes (5,6). An incisional biopsy is mandatory to rule out the possibility of melanoma (5-7).

This article documents an unusual case of oral multifocal and diffuse melanoacanthoma, affecting the whole oral mucosa, including the tongue.

Case Report

In August 2005, a 74-year-old black woman, came to the Stomatology Department of São Paulo Public Health Center of Jardim Peri Peri, for treatment of multiple and diffuse black/brownish intraoral lesions. The lesions were asymptomatic. Although the patient was unaware of their onset, she reported that the lesions had rapidly increased in size.

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The patient's medical history was uneventful. She avoided sun exposure, the use of drugs (medication), smoking and alcohol. Excluding the long term use of upper and lower complete dentures, no other initiating factors could be associated. Intraoral examination revealed multiple black and brownish lesions with heterogeneous pigmentation and width. These maculae were distributed throughout the buccal mucosa, lips, gingiva and tongue, and presented a smooth but not ulcerated surface (Figs. 1A and B).

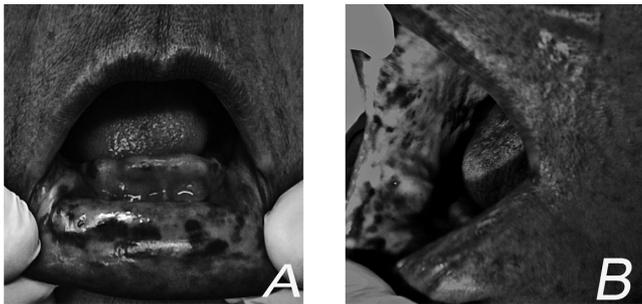


Fig. 1 OMA. Multiple maculae are seen throughout the oral mucosa: (A) lips, (B) buccal mucosa and lateral border of the tongue.

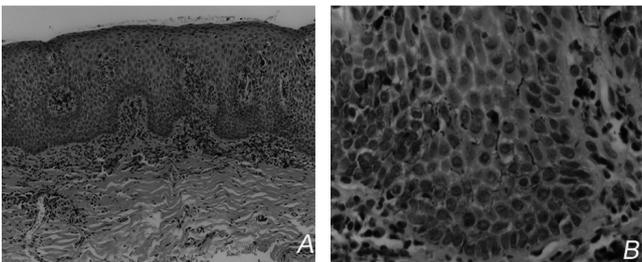


Fig. 2 (A) Occasional dark-staining melanocytes are present at various levels within the stratified squamous epithelium; original magnification $\times 100$; (B) melanocytes with pigment-laden dendritic processes are present among epithelial cells – H–E staining, original magnification $\times 400$.

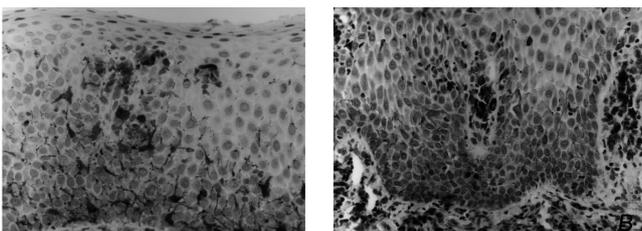


Fig. 3 (A) Melan-A; (B) HMB-45; Streptavidin-biotin, original magnification $\times 400$.

Although our differential diagnosis included lesions where melanocytic benign hyperplasia is present, an incisional biopsy was performed on both the gingiva and buccal mucosa.

The gross specimen was fixed in 10% buffered formalin and then submitted for histopathologic examination. Sections were stained with hematoxylin and eosin and revealed a segment of oral mucosa in which the surface lining stratified squamous epithelium exhibited parakeratinization, mild acanthosis and broadening of rete ridges. Benign melanocytes with well-defined nuclei and pigment-laden dendritic processes were seen at various levels within

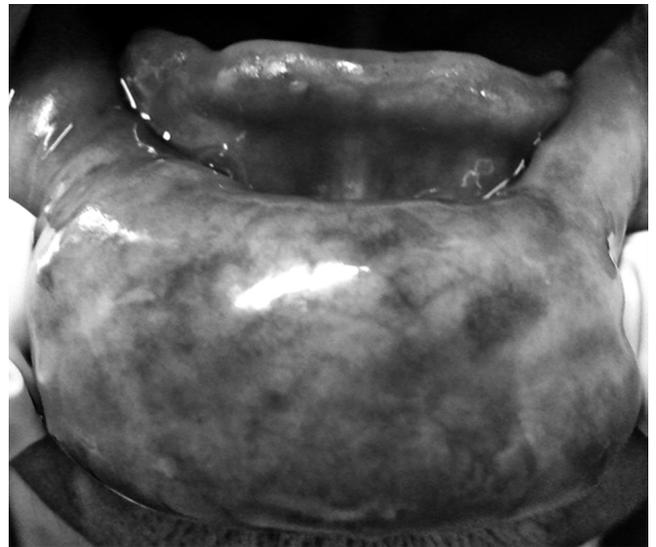


Fig. 4 After one year, the lip lesions faded away.



Fig. 5 After one year, the lesions in the tongue had almost completely disappeared.

the epithelium (Fig. 2A). They were dark stained and surrounded by a clear peripheral zone and did not exhibit any cytologic atypia, nor any tendency to aggregate (Fig. 2B). The diagnosis was consistent with oral melanoacanthoma.

As many of the dendritic melanocytes were densely pigmented, a red chromogen – fast red – was used to differentiate the immunohistochemistry reaction product from the background melanin. The dendritic melanocytes exhibited diffuse cytoplasmic expression of HMB-45 and melan A (Figs. 3A and B)

After the biopsy, the patient was followed up and one year later, the lesions disappeared almost completely (Figs. 4 and 5).

Discussion

The first fully documented case of OMA was published in 1979 by Matsuoka (2). Since then, only 53 additional cases have been reported in English language literature (5). Commonly, OMA occurs in dark-skinned females within a wide age range. Most of these cases involved solitary lesions, and multifocal OMA had been reported only in 10 cases (5).

A review of the literature revealed that multifocal OMA presents an equal gender distribution, occurring more frequently in the palate with the number of lesions ranging between two to five (5). In the present case, the preference for ethnicity was confirmed. However, it is somewhat unusual because she had a large number of lesions, throughout the oral mucosa, and the tongue was also affected. Previously, two cases in which the tongue was affected were reported, but they were solitary OMA (4,5). To our knowledge, this is the first case of multifocal OMA in which the tongue was also affected.

In the present case, the lesions mimicked the radial growth phase and heterogeneous dark pigmentation of an intraoral melanoma, although the lesions were spread all over oral mucosa (Fig. 1). To exclude the possibility of a melanoma, an incisional biopsy was performed. This procedure is mandatory (5-8).

The etiopathogenesis of OMA remains uncertain, but it has been associated with a traumatic process (5,9). In the present case, the only irritant factor that could act as an etiologic agent was the prosthesis. However, during the follow-up period, the patient continued using her prosthesis and spontaneous regression was observed before the first year (Figs. 4 and 5). This fact supports the non-neoplastic nature of OMA. Also, to emphasize the reactive nature of this lesion and in order to differentiate it from cutaneous melanoacanthoma, several terms have been suggested, including “melanoacanthosis”, “reactive melanocytic

hyperplasia”, and “mucosal melanotic maculae” (9,10).

The differential diagnosis of OMA includes any pigmented disorder usually linked to the activity of melanocytes. The most common are melanocytic nevus, melanotic maculae and melanoma (5,7,9). Its diagnosis is based on the clinical and histologic features (3,5,6.). In contrast to the other pigmented lesions, in which melanin is transferred from dendritic epidermal melanocytes to the epidermal keratinocytes that are associated with the epidermal melanocyte unit, the melanin in OMA is restricted mainly to melanocytes, the adjacent keratinocytes being devoid of melanin (9). In the present case, microscopic examination showed typical histological features of a benign lesion that was compatible with melanoacanthoma.

The immunohistochemical profile of these lesions is essentially limited to the melanocytic markers, but it is not necessary for the diagnosis (2,3).

In summary, this report presents a multifocal OMA affecting the whole oral mucosa, including the tongue and corroborates with the possible reactive and also reversible nature of OMA. After one year, the lesions had almost disappeared.

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