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Case Report

Oral melanoacanthoma: A new entity in 2022 WHO

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ABSTRACT

Aim: To highlight the occurrence of Melanoacanthoma in oral cavity.
Background: Melanoacanthoma of the oral cavity is rare benign reactive pigmented lesion. It has been recently considered as a distinct entity in the World Health Organization (WHO) classification. Oral melanoacanthomas are reactive and not related to seborrheic keratosis unlike their cutaneous counterparts which are variant of pigmented seborrheic neoplastic in nature. These lesions are characterized by hyperplasia of dendritic melanocytes dispersed throughout an acanthotic epithelium with buccal mucosa being the most common location.
Case Description: An 81-year-old female presented to us with complaint of blackish patch on hard palate. The patient underwent biopsy and the histopathological diagnosis of Melanoacanthoma was given.
Conclusion: To the best of our knowledge this is first such case of Melanoacanthoma on the hard palate in Indian subcontinent.
Clinical Significance: Awareness about these lesions is important for apt diagnosis and preventing under diagnosis.

1. Introduction

Melanoacanthoma (MA) of the oral cavity is a rare reactive lesion that has recently been recognized as a distinct entity in the WHO classification.[1] Its inclusion in the classification is mainly due to its clinical presentation, which initially resembles a rapidly progressing pigmented lesion, mimicking oral mucosal melanoma. In 1927, Bloch first described Melanoacanthoma (MA) as a melanoepithelioma. However, the term "melanoacanthoma" was first used by Mishima and Pinkus in 1960 to refer to a benign pigmented tumor of the skin, consisting of proliferating melanocytes and Malpighian cells (keratinocytes) of the basal and prickle types. In 1978, Tomey and Dorey presented the first case of oral melanoacanthoma involving the buccal mucosa at the Maxillofacial and Oral Pathology Congress of the American Academy.[2–6]

Oral MA is a rare reactive lesion with an unknown etiology, while cutaneous MA is a rare variant of pigmented seborrheic keratosis. These lesions can appear as rapidly growing, brown to brown-black macular, usually solitary lesions that affect younger individuals, with a slight predilection for black females. The most commonly affected site is the buccal mucosa, followed by the gingiva.[4,5,7,8] Therefore, the purpose of this article is to highlight the clinical and immunohistochemical (IHC) features of MA and to report the first case of oral MA affecting the hard palate in an elderly Indian female.

2. Case Description

An 81-year-old Indian female visited our outpatient department with a primary complaint of blackish discoloration on the palate that had been present for

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the past two weeks. The patient had a medical history of hypertension and had been taking Amlodipine medication for the past six years. Upon intra-oral examination, a non-elevated, round, blackish brown lesion measuring 0.5 cm in diameter was observed on the palatal mucosa in the region of the first molar. The adjacent mucosa appeared normal without any notable abnormalities. No other abnormalities were detected in any other part of the oral cavity.

Considering the short history and clinical presentation, a provisional diagnosis of melanocytic nevus was considered. Although less likely, the possibility of malignant melanoma was also taken into account. To obtain a definitive diagnosis, an excisional biopsy of the lesion was planned under local anesthesia, and the excised sample was submitted to our department for histopathological examination.

Microscopic examination of the received tissue sections revealed keratinizing stratified squamous epithelium with prominent acanthosis (thickening of the epidermis) and proliferation of dendritic melanocytes. Melanin pigment was present in the basal layer and suprabasal layers of the epithelium but did not invade the underlying scanty connective tissue. No cytological or architectural atypia (abnormalities) were observed in the epithelium (Figure 1 A-C). Immunohistochemical markers S100 (Figure 2 A), HMB 45 (Figure 2 B), and Melan-A were performed, and they demonstrated strong immunoreactivity in the cytoplasm of scattered dendritic melanocytes throughout the mucosa. Based on these findings, a final diagnosis of Melanoacanthoma was established.

The patient has been regularly followed up for one year without any recurrence of the lesion.

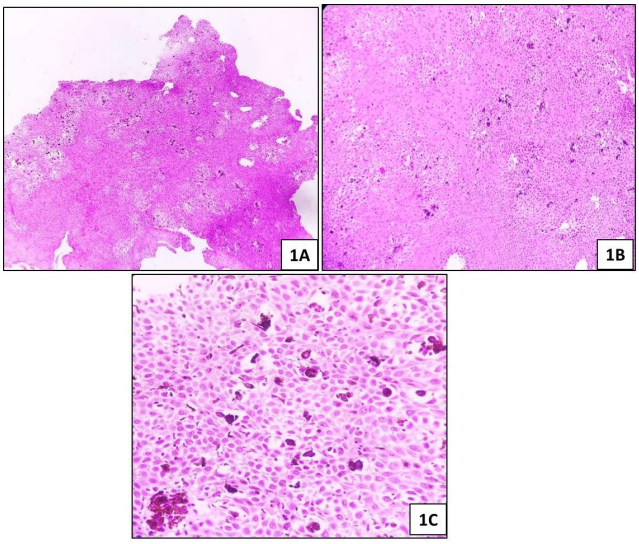

Figure 1: Histopathological findings of Melanoacanthoma; **A)**: Photomicrograph showing stratified squamous epithelium with marked acanthosis (H&E-40x*); **B):** Photomicrograph showing dendritic melanocytes dispersed throughout the epithelium (H&E-100x*); **C)**: Photomicrograph showing dendritic melanocytes within the intercellular spaces lacking any atypia (H&E-400x*). *Hematoxylin and eosin

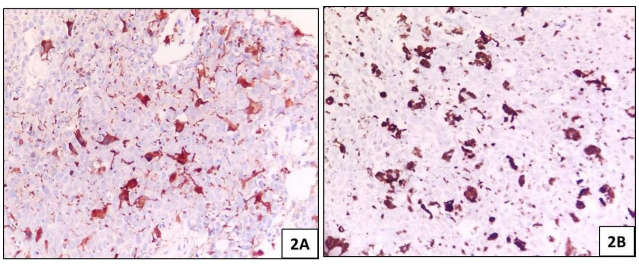

Figure 2: Immunohistochemical findings in Melanoacanthoma; **A):** Photomicrograph showing strong immunoreactivity forS-100 protein in dendritic melanocytes. (IHC- 400x*); **B)**: Photomicrograph showing strong immunoreactivity for HMB-45 in dendritic melanocytes. (IHC- 400x*) *Immunohistochemistry

3. Discussion

Melanoacanthoma (MA) of the oral cavity has been officially recognized as a distinct entity in the recent WHO classification for head and neck pathologies. The earliest reference to this particular lesion can be traced back to 1927 when Bloch described it as a "benign non-nevoid melanoepithelioma of the skin." Bloch further categorized these cases into type I lesions, characterized by the presence of dendritic melanocytes and keratinocytes, and type II lesions, which featured dendritic cells. Subsequently, Mishima and Pinkus introduced the term Melanoacanthoma (MA), referring specifically to lesions resembling Bloch's type I classification. However, the occurrence of MA in the oral cavity is exceedingly rare, and its oral presentation was initially discussed by Tomey and Dorey. [1–4]

The precise etiology of these lesions remains unknown to date, although many authors suggest that they may be preceded by a history of trauma. Simon et al proposed three variants of melanocytic seborrheic keratosis: irritant, non-irritating, and nested. They postulated that oral MA represents an irritant seborrheic keratosis. It should be noted that while cutaneous MA is considered a rare variant of pigmented seborrheic keratosis, oral MA is not directly associated with seborrheic keratosis. Moreover, several other distinctions exist between cutaneous MA and oral MA. Cutaneous lesions are true neoplasms resembling seborrheic keratosis, while oral MA is categorized as a reactive lesion. Oral MA predominantly affects young individuals of African descent, whereas cutaneous MA tends to be more common in elderly Caucasian patients. Oral MA exhibits a slight preference for females, typically appearing in the third decade of life, although cases have been reported in individuals ranging from the second to the seventh decade of life. In the oral cavity, MA typically manifests with a sudden onset and presents as a flat, brown to black pigmented surface. In contrast, cutaneous MA

develops slowly and is characterized by a rough, papillary surface.[1,3–5,7–9] In our particular case, the patient is an elderly Indian female in her eighth decade of life. She presented with a recently developed lesion that exhibited a brown to black flattened appearance, without any immediate history of associated trauma or irritation.

Oral MA predominantly manifests on the buccal mucosa, although it can also occur on the labial mucosa, gingiva, and, rarely, the palate. Some authors have even reported cases of bilateral lesions in the oral cavity. Additionally, Chandler et al have documented an increased incidence of oral MA in individuals with Acquired Immunodeficiency Syndrome (AIDS).[4,6,9,10]

In the case we are currently discussing, a solitary MA lesion was observed on the hard palate of an elderly Indian female. To the best of our knowledge and based on an extensive review of the literature, this appears to be the first reported case of MA on the hard palate in an elderly Indian female.

Melanoacanthoma has been recently added in the 2022 WHO due to the pigmented sudden onset lesion raising concern for oral mucosal melanoma.[1]

Histologically, MA exhibits characteristic features including epithelial acanthosis (thickening of the epithelium) and the presence of dendritic melanocytes within the intercellular spaces. These dendritic melanocytes can be identified by their strong immunoreactivity for S-100 protein, HMB-45, or Melan-A. One of the most crucial distinguishing factors from mucosal melanoma is the absence of atypia (abnormal cellular characteristics) and invasive melanocytic nests, which are indicative of malignancy.[1,7,8]

In our case, the microscopic examination displayed marked acanthosis and the presence of dendritic melanocytes throughout the mucosa, consistent with the histological features of MA. Immunostaining using S-100, HMB-45, and Melan-A antibodies resulted in strong staining of the dendritic melanocytes. Importantly, there were no atypical cells or foci of invasion observed in our case, further supporting its differentiation from mucosal melanoma, which is known to frequently occur on the hard palate.

The treatment of oral MA that has been conventionally used is surgical removal to exclude the possibility of mucosal melanoma. Andrews et alused Argon Plasma Coagulation to ablate the lesion with successful removal and no recurrence.[9] The patients have reported no recurrence or malignant transformation in the past literature.[3,5,6] Our patient underwent in toto removal of the lesion for diagnosis and is under follow up for last one year with no local recurrence or malignant transformation.

4. Clinical Significance

Oral MA is a newly recognized entity that has been included in the WHO 2022 classification. While it is considered a benign reactive lesion, its rapid development necessitates the need to differentiate it from oral mucosal melanoma. The objective of this article is to bring attention to this rarely encountered lesion and emphasize its consideration in the differential diagnosis of pigmented lesions affecting the oral mucosa. To the best of our knowledge, this is the first reported case in the Indian subcontinent of Oral Melanoacanthoma on the hard palate in an Indian female in her eighth decade of life.

5. Source of Funding

None.

6. Conflict of Interest

None.

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