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Case Presentation

Oral compound nevus

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Abstract

The melanocytic nevus is a benign and focal proliferation of nevus cells that can be congenital or acquired. Intraoral lesions are uncommon, and the etiology and pathogenesis are poorly understood. The occurrence rate of oral compound nevus is about 5.9% to 16.5% of all oral melanocytic nevi. A 22-year-old male patient presented with a dark brown macule on the buccal mucosa of the maxilla in the region of tooth 26. The lesion was elliptical, 0.7 x 0.5 cm, well circumscribed, asymptomatic, and the evolution time was unknown. An excisional biopsy was performed and microscopic analysis revealed nests of nevus cells in the epithelium and underlying connective tissue that were compatible with melanocytic compound nevus. Owing to the clinical similarity between oral melanocytic nevus and oral melanoma, a histopathological analysis is mandatory for definitive diagnosis.

Keywords: oral melanocytic nevus, oral compound nevus, oral melanoma

Introduction

Melanocytic nevus is a benign and focal proliferation of nevus cells and can be congenital or acquired. A Caucasian adult shows about 30 cutaneous melanocytic nevi, but intraoral lesions are uncommon and their etiology and pathogenesis are poorly understood [1,2]. In the study of Buchner, Merrell, and Carpenter [2], the oral melanocytic nevi accounted for only 0.1% of 89,430 oral biopsies analyzed over 19 years.

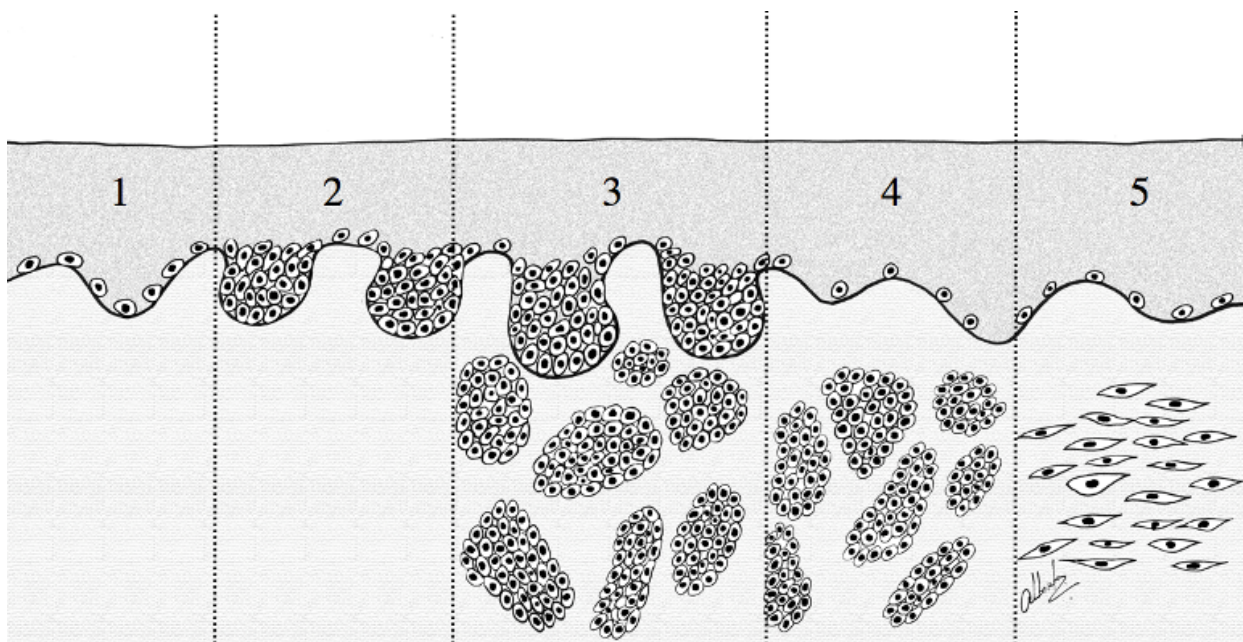


Figure 1. Types of melanocytic nevi according to localization, distribution and morphology of nevus cells: 1. Normal melanocytes, 2. Junctional, 3. Compound, 4. Intradermic or Intramucosal, 5. Blue

The oral melanocytic nevus is classified similarly to their skin counterparts: junctional, compound, intramucosal, blue, and combined [3]. The classification depends on the localization, distribution, and morphology of nevus cells, as shown in Figure 1.

The early reports of oral compound nevus are from the 1960s [6,7] and few cases were reported. The rare occurrence of intraoral compound nevus represents from 5.9% to 16.5% of all oral melanocytic nevi [2,3,4,5]; its clinical similarity to melanoma justifies the interest in describing this case report.

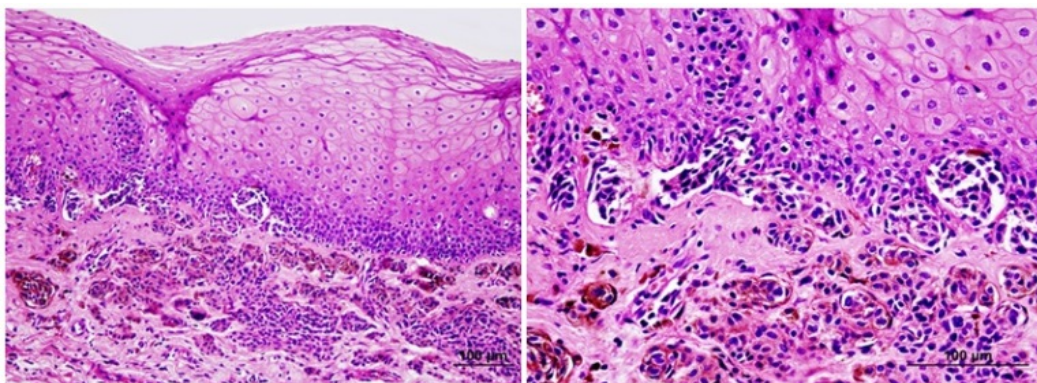
Case synopsis

A 22-year-old epileptic man, Fitzpatrick skin type V, presented with gingival hyperplasia (grade 2) related to phenytoin use and a dark brown macule in the buccal mucosa of the maxilla in the region of tooth 26. The lesion was elliptical, 0.7 x 0.5 cm, well circumscribed, asymptomatic, and the evolution time was unknown (Figure 2). The patient didn't showed any nevi in other mucous membranes of the body (including anogenital area). He was not a smoker and had a negative history of alcohol use. He was unaware if anyone in his family had nevi in the same anatomic area.



Figure 2. Dark brown macule, well circumscribed, in the buccal mucosa of the maxilla

An excisional biopsy was performed and microscopic analysis revealed the presence of clusters of cells with clear cytoplasm and densely basophilic nuclei. The islets and cords of nevus cells were distributed at the tips of the epithelial ridges and randomly in the lamina propria (Figure 3). The histopathological diagnosis was compound melanocytic nevus.



Discussion

In most cases of oral compound nevus (Table 1), the lesion affects mainly the hard palate (33.3% to 57.1%), but in this case, it was located in the buccal mucosa, the second most commonly affected site (28.6% to 41.7%). It can occur at any age, but is most prevalent between the second and third decade of life with more than 50% of patients in this age group, similarly to this case [2,3,4,5].

Table I – Location and age distribution of compound nevi in various studies

Location (%)	Study			
	Buchner and Hansen, 1987 (12 cases)	Buchner et al., 1990 (12 cases)	Buchner et al., 2004 (15 cases)	Melete et al., 2007 (7 cases)
Hard palate	4 (33,3%)	5 (41,7%)	5 (33,3%)	4 (57,1%)
Buccal mucosa ^a	5 (41,7%)	4 (33,3%)	5 (33,3%)	2 (28,6%)
Gingiva ^b	1 (8,3%)	1 (8,3%)	2 (13,3%)	1 (14,3%)
Labial mucosa	1 (8,3%)	1 (8,3%)	3 (20%)	-
Retromolar pad	1 (8,3%)	1 (8,3%)	-	-

Age (%)	Study			
	(11 cases)	(12 cases)	(14 cases)	(7 cases)
0-9	2 (18,2%)	3 (25%)	4 (28,6%)	-
10-19	3 (27,3%)	4 (33,3%)	6 (42,8%)	2 (28,6%)
20-29	3 (27,3%)	4 (33,3%)	1 (7,1%)	2 (28,6%)
30-39	1 (9,1%)	-	1 (7,1%)	1 (14,3%)
40-49	1 (9,1%)	-	2 (14,3%)	2 (28,6%)
50-59	1 (9,1%)	-	-	-
60-69	-	1 (8,3%)	-	-

^aIncluding mucobuccal fold

^bIncluding alveolar ridge

Oral melanocytic nevi are usually discovered during routine intraoral examination because they are asymptomatic [5]. Clinically, compound nevus manifests as slightly raised papules or plaques and, less often, as well-defined macules; they are usually small in size (<1.2cm).

The color varies from light brown to dark brown, but occasionally presents as a non-pigmented lesion. Most have a smooth surface, although some may have a rough, papillomatous, or warty surface [3,4]. In this case the macule was asymptomatic, well delineated, 0.7 x 0.5 cm in size, and dark brown in color with a smooth surface,

The biopsy of a compound nevus is performed more often in females than in males [2,3,5] and it may be a more cosmetic concern among women compared to men [8].

In a cutaneous junctional melanocytic nevus, the nevus cells are islets at the epithelium-connective tissue junction, especially in the tips of the epithelial ridges. When located along the junctional area and in the underlying connective tissue, the lesion is classified as a compound melanocytic nevus, as in this case report. However, in an intramucosal melanocytic nevus, nevus cells are located only in the underlying connective tissue [5]. Fibrosis is often abundant among the cells and in the presence of melanophages [9,10,11].

In the oral mucosa, the intramucous nevus is the most common (63% to 80.6%), followed by the blue nevus (8.3% to 32%) and compound nevus (5.9% to 16.5%) [2,3,4,5]. This highlights the rarity of the described case (Table 2).

Table II – Histopathologic type of oral nevi in various studies

Type (%)	Study			
	Buchner and Hansen, 1987 (191 cases)	Buchner et al., 1990 (130 cases)	Buchner et al., 2004 (91 cases)	Melete et al., 2007 (119 cases)
Intramucosal	105 (55%)	82 (63%)	58 (63,7%)	96 (80,6%)
Compound	12 (6%)	12 (9%)	15 (16,5%)	7 (5,9%)
Junctional	10 (5%)	7 (5%)	3 (3,3%)	5 (4,2%)
Common blue	61 (32%)	24 (19%)	15 (16,5%)	10 (8,3%)
Combined	3 (2%)	5 (4%)	-	1 (0,8%)

Most compound oral nevi present as elevated lesions. However, one-third appear as macules, making it difficult to differentiate from flat pigmented lesions, including melanoma in situ [4]. The differential diagnosis also includes flat pigmented lesions, such as oral melanotic macule, racial pigmentation, and amalgam tattoo. It is important to rule out the diagnosis of oral melanoma, which has a poor prognosis. In general, the median survival is slightly over 2 years from time of diagnosis. One reason for this fact is the generally advanced stage of mucosal melanoma at initial diagnosis [5,12].

Oral melanocytic nevi have a predilection for the upper portion of the mouth; oral melanoma predominates in the same anatomical region [2,3,4,5]. This suggests that some oral melanocytic nevi become malignant [5]. The preferred site for oral melanoma is the palate with over 40% occurring either on the hard or soft palate. The maxillary gingiva is the second most affected site (16%). The buccal mucosa, the site where the macule was located in this case, the incidence is 7% [12].

There is no data about the malignant potential of oral melanocytic nevi [3,13]. Approximately one-third of oral melanomas are preceded by pigmented lesions for months or years, but the types of these microscopic precursor lesions have not been reported in detail [13,14]. The report of Meleti et al [5] contained 119 patients with oral melanocytic nevus and none developed melanoma during the 8.6 years of follow-up. However, all lesions were removed and the question of whether these lesions could have turned into melanoma remains unanswered.

Despite the lack of evidence for its real potential for malignancy, the clinical similarity of oral melanocytic nevus with oral melanoma at an early stage, its rare occurrence, and the usually small size of the lesion justify the recommendation for excisional biopsy [2,3,4].

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