

A Rapidly Enlarging Pigmented Lesion of the Buccal Mucosa

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The following Case Challenge is provided in conjunction with the American Academy of Oral and Maxillofacial Pathology.

Case Summary

A 39-year old African American male noticed the presence of a brown, slightly rough lesion on the inside of his left cheek. It was asymptomatic and was discovered by the patient three weeks ago. He could not recall a history of trauma, such as cheek biting, undue force to the side of his face, or any recent dental treatment. He was unaware if the lesion had changed size or color during the several weeks prior to presentation.

After you have finished reviewing the available diagnostic information, make the diagnosis.

Diagnostic Information

Clinical Findings

On inspection, a 1 x 1 cm lesion was identified on the left buccal mucosa adjacent to the mandibular first molar. (Figure 1) The lesion showed uniform pigmentation with a dark-brown center and a lighter brown periphery. Although the lesion was fairly well circumscribed, in some areas the margins blended with the surrounding mucosa. No nodules or areas of induration could be palpated, but the mucosa was slightly roughened in texture. Examination of the rest of the oral cavity revealed some physiologic gingival pigmentation. His dentition was in fairly good repair; an old occlusal amalgam restoration was present in the mandibular left first molar. Physical examination of the head and neck revealed no other pathology.

The patient's medical history was noncontributory, although he was being evaluated for carpal tunnel syndrome. He did not currently take any medications, and he was allergic to penicillin. He did not smoke or drink alcohol.

The patient was scheduled to return in two weeks for re-examination of the lesion. If the pigmentation was still present, a biopsy of the area was planned.

Additional Clinical History

Because of local complications related to surgery for his carpal tunnel syndrome, the patient failed to return in two weeks for follow-up. Two months later, he presented for continuation of his dental treatment. The pigmented lesion on the left buccal mucosa had greatly increased in size to 6 x 2 cm, extending from the first bicuspid back to the third molar region. (Figure 2) In addition, a 2 x 1 cm darkly pigmented lesion had appeared on the right buccal mucosa. (Figure 3) An incisional biopsy of the lesion on the left buccal mucosa was obtained.

Photomicrographs

The lesion on the left buccal mucosa was incised in an elliptical fashion under local anesthesia and submitted for biopsy. Microscopic examination revealed a wedge of mucosa that was covered by acanthotic stratified squamous epithelium. (Figure 4)

Scattered dendritic melanocytes could be seen migrating through the entire epithelial thickness. (Figures 5 & 6)



Figure 1. Relatively well-circumscribed pigmented lesion on the left buccal mucosa.



Figure 2. Clinical photograph showing dramatic enlargement of the lesion two months later.



Figure 3. Two months following the initial visit, a similar lesion developed on the right buccal mucosa.

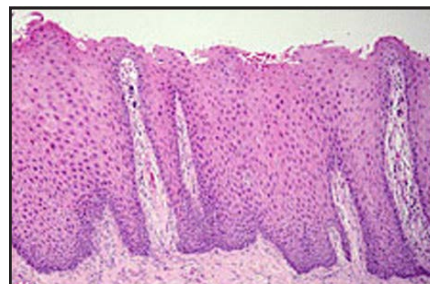


Figure 4. Low power photomicrograph showing thickening (acanthosis) of the epithelium.

These cells were highlighted by special stains (Fontana-Masson) for melanin. (Figure 7) No atypical melanocytes were identified. Focal

incontinent melanin was noted in the superficial lamina propria along with scattered lymphocytes and occasional eosinophils.

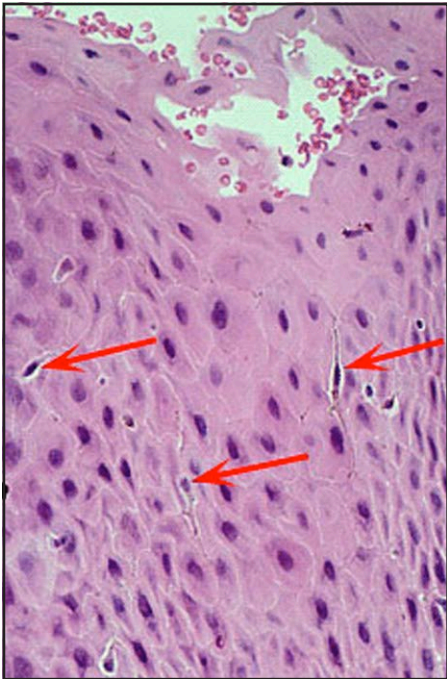


Figure 5. Medium power photomicrograph showing scattered dendritic melanocytes (arrows) in the upper spinous layer of the epithelium.



Figure 6. High power photomicrograph of dendritic melanocytes between spinous epithelium cells.

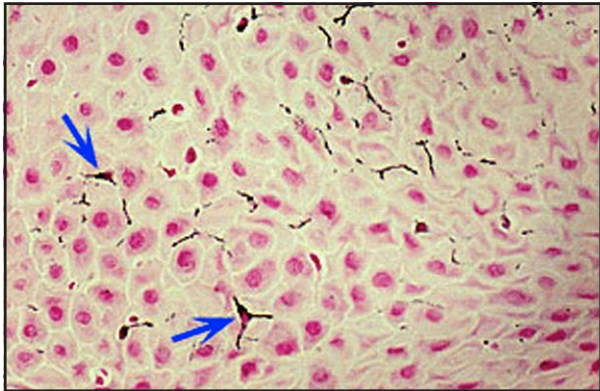


Figure 7. The Fontana-Masson stain highlights numerous dendritic melanocytes throughout the epithelium.

Can you make the diagnosis?

A 39-year old African American male noticed the presence of a brown, slightly rough lesion on the inside of his left cheek.



Select the Correct Diagnosis

- A. Racial Pigmentation
- B. Melanocytic Nevus
- C. Melanotic Macule
- D. Amalgam Tattoo
- E. Melanoacanthosis
- F. Melanoma

Racial Pigmentation

Choice A. Sorry, this is not the correct diagnosis.

Physiologic melanin pigmentation of the oral mucosa in individuals with deeply pigmented skin most commonly involves the attached gingiva and

is generally diffuse and symmetrical. Sudden and dramatic changes, as noted in this case, would not be expected. Microscopically, the increased melanin production is typically limited to the basal cell layer of the epithelium.

Please re-evaluate the information about this case.

Melanocytic Nevus

Choice B. Sorry, this is not the correct diagnosis.

Intraoral melanocytic nevi are rare, occurring in approximately 0.1% of the general population.¹ The palate is a common site for oral nevi and they are most frequently diagnosed during the third and fourth decades of life. Clinically, more than 80% of intraoral nevi are less than 1 cm in

diameter, with half of all cases measuring from 0.1 to 0.3 cm.² Neither the rapid growth of the left buccal lesion nor the sudden appearance of the right buccal lesion would be consistent with a melanocytic nevus. In addition, the biopsy specimen did not show the characteristic nested arrangement of nevus cells within or beneath the epithelium.

Please re-evaluate the information about this case.

Melanotic Macule

Choice C. Sorry, this is not the correct diagnosis.

Melanotic macules are usually solitary, well circumscribed, brown to black macules that measure less than 1 cm.^{3,4} These lesions are most commonly located on the vermillion border of the lower lip, followed by the gingiva, buccal mucosa, and palate.⁵ Microscopically, they are

characterized by increased melanin production in the basal cell layer with incontinent melanin in the lamina propria. The presence of large, dendritic melanocytes throughout the entire epithelial thickness would not be expected. The rapid development and progression of the lesions in this case would not be consistent with a melanotic macule.

Please re-evaluate the information about this case.

Amalgam Tattoo

Choice D. Sorry, this is not the correct diagnosis.

Foreign materials (silver amalgam or graphite) introduced into the tissues iatrogenically or secondary to trauma may produce pigmented macular lesions. Amalgam tattoos are most common on the gingiva and alveolar ridge,

although almost any oral mucosal site can be affected.⁶ They may appear to enlarge slowly and radially as the pigment gradually diffuses through the tissues.⁷ However, the rapid enlargement seen in this case would not be consistent with an amalgam tattoo. Histopathologically, amalgam may appear as dark, discrete, fine granules or as irregular solid fragments in the connective tissue.

Please re-evaluate the information about this case.

Melanoacanthosis

Choice E. Congratulations! You are correct.

Discussion

Oral melanoacanthosis is a rare, benign acquired pigmentation of the oral mucosa characterized by dendritic melanocytes dispersed throughout a thickened epithelium. The term “melanoacanthoma;” was first used by Mishima and Pinkus in 1960 to describe an epidermal tumor composed of basal cells, prickle cells, and highly dendritic melanocytes.⁸ Tomich and Dorey presented the first case of intraoral melanoacanthoma at a meeting of the American Academy of Oral Pathology in 1979.⁹ Since then, over 25 intraoral cases have been reported.¹⁰⁻²¹

Despite its microscopic similarity to melanoacanthoma of the skin, the oral counterpart exhibits clinical differences which indicate that it represents a distinct and separate entity. Because oral examples are not considered to be neoplasms, the suffix “-oma” should not be used; therefore, “oral melanoacanthosis” has been proposed as a better term for intraoral lesions.²⁰ Horlick and associates¹⁷ suggested using the term “mucosal melanotic macule, reactive type;” however, this term would be confused too easily with the conventional oral melanotic macule, a much more common and distinctly different lesion. Zemtsov and associates¹⁹ proposed using a descriptive term such as “oral melanodendritic mucositis.”

Cutaneous melanoacanthoma is a benign epithelial neoplasm which generally affects elderly Caucasians with an average age of 62 years.¹³ The lesion typically develops slowly over several years and has a rough or papillary surface. In contrast, oral melanoacanthosis is not neoplastic, but has characteristics of a reactive condition.^{13,20} It is seen almost exclusively in blacks in the third and fourth decades of life (average age = 23 years) and has a female predilection (3:1 female:male gender ratio).^{13,20} Two reported cases have occurred in whites.^{13,16}

The lesion usually involves lining mucosa which is susceptible to trauma, especially the buccal mucosa (57% of reported cases) and labial mucosa (22%).²⁰ Approximately 50% of the patients with oral lesions recall a history of preceding trauma.¹⁷ Masticatory mucosa subject to chronic trauma (i.e., palate, gingiva, and alveolar mucosa under dental prostheses) also may be involved. The lesion may be flat or slightly raised, ranging from dark-brown to black in color. On occasion, bilateral lesions have been described.^{15,21} Lesions often demonstrate a rapid increase in size and may reach a diameter of several centimeters within a short period of a few weeks. All reported cases with follow-up information resolved spontaneously following biopsy or removal of irritants. No case of oral melanoacanthosis has been associated with malignant transformation.

Histopathologically, oral melanoacanthosis is characterized by a thickened (acanthotic) epithelium with benign dendritic melanocytes scattered throughout the spinous layer. Melanocytes are also present in increased numbers in the basal cell layer but, unlike melanoma, no atypical melanocytes are identified.¹⁶ The spinous layer often appears spongiotic with widened intercellular bridges.¹⁹ In addition, eosinophils and a mild to moderate chronic inflammatory cell infiltrate are usually seen within the underlying connective tissue.¹³

The etiopathogenesis of oral melanoacanthosis remains unknown. Because oral melanoacanthosis is difficult to distinguish from other pigmented lesions and may demonstrate an alarming growth rate, submission of a biopsy for microscopic evaluation is usually necessary to rule out the possibility of melanoma. Once the histopathologic diagnosis has been established, no further treatment is necessary. In most reports, lesions have undergone spontaneous resolution after incisional biopsy. In the current case, both lesions showed resolution approximately three months following the biopsy of one side. (Figures 8 & 9) No additional lesions have developed.



Figure 8. Three months following the biopsy, the lesion on the left buccal mucosa has disappeared.



Figure 9. Three months later, the lesion on the right buccal shows only faint, diffuse pigmentation remaining.

Melanoma

Choice F. Sorry, this is not the correct diagnosis.

The clinical presentation of a rapidly expanding pigmented lesion would create concern about the possibility of melanoma. This malignancy rarely occurs intraorally and most cases involve the maxillary alveolar mucosa and hard palate. Although some melanomas cover a large

surface area, a bilateral distribution (as illustrated in this patient) is not a feature of melanoma. Microscopically, melanomas may demonstrate pigmented dendritic cells in all levels of the epithelium. However, the dendritic cells in the present case did not demonstrate pleomorphism. No abnormal melanocytes were seen proliferating along the basal layer of the epithelium or invading into the connective tissue.

Please re-evaluate the information about this case.

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