Oral Melanoacanthoma of a Rare Intraoral Site: Case Report and Review of Literature

Kshitiz Rohilla, V Ramesh, PD Balamurali, Namrata Singh

ABSTRACT

Oral melanoacanthoma is rare pigmented mucosal lesion that presents most commonly on the buccal mucosa, characterized by sudden appearance and rapid radial growth, thus clinically mimicking malignant melanoma. It was originally described as a mixed tumor of melanocytes and keratinocytes, but appears to be a reactive process; formed in areas prone to trauma, and regressing after the removal of trauma or incomplete excision. The clinical appearance of oral melanoacanthoma is nondiagnostic, and biopsy is mandatory to rule out malignancy. We report a case of melanoacanthoma of a rarer oral mucosal site in a 12-year-old Asian male. A brief review of the current literature is also presented.

Keywords: Melanoacanthoma, Oral pigmented lesion, Melanocytes.

How to cite this article: Rohilla K, Ramesh V, Balamurali PD, Singh N. Oral Melanoacanthoma of a Rare Intraoral Site: Case Report and Review of Literature. Int J Clin Pediatr Dent 2013;6(1):40-43.

Source of support: Nil
Conflict of interest: None

INTRODUCTION

Melanoacanthoma is an uncommon, benign, mucocutaneous pigmented lesion characterized by dendritic melanocytes dispersed throughout the acanthotic epithelium.1,2 Though it was originally described as a benign skin tumor of keratinocytes and dendritic melanocytes, there is now evidence that the intraoral lesions are unlike those occurring on skin.3

Cutaneous melanoacanthoma was first described in 1927 by Bloch, but the term melanoacanthoma was introduced by Mishima and Pinkus in 1960.1 The first case of oral melanoacanthoma was reported in 1978 by Tomich.4

Melanoacanthoma of the skin is a benign mixed proliferation of keratinocytes and melanocytes and is considered to be a variant of seborrheic keratosis. Most patients are adults, beyond 40 years of age. Sex predominance is not known.5 Most melanoacanthomas are located on the scalp, though lesions have been reported on the scalp, neck and extremities too.5,6 These lesions are almost exclusive to whites in middle to late age, developing slowly over a long period, and usually having a roughened or papillary surface.7

On the contrary, intraoral melanoacanthomas tend to affect a much younger population, occurring almost exclusively in blacks, with a female predilection. These lesions show rapid increase in size and may attain dimensions of several centimeters in a few weeks. Buccal mucosa is the most frequently reported intraoral site, although masticatory mucosa subject to chronic trauma (palate, gingiva) may also be affected.5-11 Involvement of labial mucosa12-14 and alveolar ridge15 has also been reported. Mostly unilateral and solitary,6 these deeply pigmented lesions may have a flat or slightly raised surface. The other end of the spectrum of clinical presentation includes lesions that may be bilateral,17 and even multifocal,1,8,13,18,19 as well as those which even have a proliferative or warty surface. These intraoral hypermelanotic macules or papules are typically brown, black or blue-black in color, with possible variation in the intensity of pigmentation.1,3,13,18,20

Intraoral melanoacanthoma still continues to be a rare entity.21-24 Some of the previously reported cases have been summarized in Table 1.

CASE REPORT

A 12-year-old male patient presented for evaluation of a lesion in the left maxillary gingiva, which was present for the past 6 months. The patient was under medication with valproic acid for the treatment of petit mal seizures, till the age of 8 years, after which it was discontinued. Otherwise, the medical history was noncontributory.

Extraoral examination revealed no clinically significant findings. Intraorally, there was a soft tissue growth in maxillary left quadrant (Fig. 1), involving the attached and
the marginal gingiva on the buccal aspect. The lesion was brownish black in color and had a smooth, slightly raised surface (Fig. 2). The patient denied any association of pain with the lesion. The other three quadrants showed macular pigmentation of the attached and marginal gingivae, which was clinically labeled as racial pigmentation.

The lesion was excised and sent for histopathological examination. Hematoxylin and eosin (H&E) stained sections revealed surface stratified squamous epithelium and underlying fibrous connective tissue. The epithelium exhibited parakeratosis and acanthosis and the rete ridges were irregular in shape. There was a prominence of melanocytes in the basal layer, in a linear fashion (Fig. 3). There was a suspicion of pigmented melanocytes even in the suprabasal layers. The underlying connective tissue appeared normal, showing some evidence of melanophagic activity in the subepithelial zone. Masson-Fontana silver stain supplement the presence of dendritic melanocytes filling up almost the entire epithelium (Fig. 4). The presence of benign appearing melanocytes was salient, and there was no evidence of any cytological atypia, pleomorphism or nuclear hyperchromasia. In light of the history, clinical features and the histopathological picture with H&E and Masson-Fontana stain, the final diagnosis of oral melanoacanthoma was rendered.

The patient has been on a regular follow-up (Fig. 5), and the lesion was observed to be healing well 10 months postoperatively.

**DISCUSSION**

The credit for the first fully documented case of oral melanoacanthoma goes to Matsouka (1979). Since then, there has been an addition of more than 65 cases to the literature. Table 1 summarizes the previously reported cases.

| Authors            | Years | Number of cases | Affected oral site                                      |
|--------------------|-------|-----------------|--------------------------------------------------------|
| Tomich et al⁴      | 1978  | 1               | Buccal mucosa                                          |
| Matsouka et al¹⁴    | 1979  | 1               | Labial mucosa                                          |
| Schneider et al²²   | 1981  | 1               | Buccal mucosa                                          |
| Wright et al²⁰      | 1983  | 2               | Buccal mucosa                                          |
| Goode et al¹⁵       | 1983  | 10              | Buccal mucosa, palate, labial mucosa, alveolar ridge, attached gingiva |
| Frey et al²³        | 1984  | 1               | Buccal mucosa                                          |
| Sexton and Maize¹³  | 1987  | 3               | Labial mucosa                                          |
| Wright³             | 1988  | 1               | Buccal mucosa                                          |
| Whitl et al²⁴       | 1988  | 1               | Buccal mucosa                                          |
| Horlick et al²¹     | 1988  | 2               | Buccal mucosa                                          |
| Heine et al¹⁷       | 1996  | 1               | Buccal mucosa                                          |
| Chandler et al¹     | 1997  | 1               | Palate, tonsillar fossae, upper nasopharynx             |
| Landwehr et al¹⁶    | 1997  | 1               | Buccal mucosa                                          |
| Fialitz¹¹           | 2000  | 1               | Attached gingiva                                       |
| Fatahzadeh et al¹⁸  | 2002  | 1               | Buccal mucosa, palate                                  |
| Fornatora et al⁹    | 2003  | 10              | Buccal mucosa, gingiva, hard palate, lower lip, floor of mouth, retromolar pad |
| Kauzman et al¹⁹     | 2004  | 1               | Buccal mucosa, labial mucosa, tonsillar pillars        |
| Carlos-Bregni et al¹⁰ | 2007 | 4               | Gingiva, buccal mucosa, hard palate                    |
| Marocchio et al⁸    | 2009  | 1               | Buccal mucosa, lips, gingiva, tongue                    |

*Fig. 2: The lesion covering the entire buccal surfaces of the teeth and also partially covering the occlusal surface of the first molar*

*Fig. 3: Photomicrograph showing acanthosis and parakeratosis of the surface epithelium as well as linear melanocytic hyperplasia in the basal layer. Benign melanocytes are seen in parabasal layers, and there is evidence of melanophagic activity*
available literature. The pathogenesis still remains obscure, though some authors have ascribed the potential role of chronic trauma in these cases.

The intraoral melanoacanthoma is essentially reactive in nature—a fact supported by the clinical course of the lesion. It is characterized by a tendency to affect the mucosal sites that are exposed to trauma, and typically shows rapid growth and observable regression of the lesion—spontaneously or following incomplete removal or elimination of local irritants. The histologic picture of subepithelial inflammatory cell infiltrate and slightly increased vascularity further add evidence to this concept. To differentiate these lesions from cutaneous melanoacanthoma and to emphasize their reactive nature, several terms have been suggested, including melanoacanthosis, reactive melanocytic hyperplasia and mucosal melanotic macule. Some authors opine that in contrast to the other pigmented lesions, the melanin in oral melanoacanthoma is restricted mainly to melanocytes, the adjacent keratinocytes being devoid of melanin. Interestingly, in our case, the histopathological picture showed the presence of ‘dusty’ melanin in the basal as well as parabasal keratinocytes.

Melanoacanthoma is a reparative lesion with no malignant potential. The treatment should be directed toward removing all local causes of trauma and excluding any other causes of oral pigmentation, particularly malignant melanoma. The authors advocate the replacement of the misnomer ‘melanoacanthoma’ with a more appropriate description ‘melanoacanthosis’, a term which gives due credit to the clinical behavior and histopathological picture of this rare and interesting lesion.

SUMMARY
We present a case of a rare entity, oral melanoacanthoma, occurring at a rarer oral mucosal site, that is, gingiva, in a 12-year-old Asian male. The diagnosis was based mainly on the histologic findings with H&E and Masson-Fontana stains. The patient is on a regular follow-up and the lesion has regressed completely after the initial surgery.

REFERENCES
1. Chandler K, Chaudhry Z, Kumar N, Barrett A, Porter SR. Melanoacanthoma: A rare cause of oral pigmentation. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1997;84(5):492-494.
2. Neville BW, Dampos D, Allen CM, Bouquot J. Oral and maxillofacial pathology. 3rd ed. Philadelphia: WB Saunders Company; 2009. 984 p.
Oral Melanoacanthoma of a Rare Intraoral Site: Case Report and Review of Literature

3. Wright JM. Intraoral melanoacanthoma: A reactive melanocytic hyperplasia: Case report. J Periodontol 1988;59(1):53-55.
4. Tomich CE. Oral presentation. Paper presented at: 32nd Annual Meeting of the American Academy of Oral Pathology 1978; 23-28.
5. LeBoit PE, Burg G, Weedon D, Sarasin A, editors. Pathology and genetics of skin tumours (IARC WHO Classification of tumours). Oxford: Oxford University Press. 2009. 355 p.
6. Tomich C, Zunt S. Melanoacanthosis (melanoacanthoma) of the oral mucosa. J Dermatol Surg Oncol 1990;16(3):231-236.
7. Buchner A, Merrell PW, Hansen LS, Leider AS. Melanocytic hyperplasia of the oral mucosa. Surg Oral Med Oral Pathol 1991;71(1):58-62.
8. Marocchino LS, Junior DS, da Sousa SC, Fabre RF, Raitz R. Multifocal diffuse oral melanoacanthoma: A case report. J Oral Sci 2009;51(3):463-466.
9. Formotora ML, Reich RF, Haber S, Solomon F, Freedman PD. Oral melanoacanthoma: A report of 10 cases, review of the literature, and immunohistochemical analysis for HMB-45 reactivity. J Dermatopathol 2003;25(1):12-15.
10. Carlos-Bregni R, Contreras E, Netto AC, Mosqueda-Taylor A, Vargas PA, Jorge J, Leon JE, de Almeida OP. Oral melanoacanthoma and oral melanotic macule: A report of 8 cases, review of the literature, and immunohistochemical analysis. Med Oral Patol Oral Cir Bucal 2007;12(5):374-379.
11. Flaitz CM. Oral melanoacanthoma of the attached gingiva. Am J Dent 2000;13(3):162.
12. Maize JC. Mucosal melanosis. Dermatol Clin 1988;6(2):283-293.
13. Sexton FM, Maize JC. Melanotic macules and melanocanthomas of the lip: A comparative study with census of the basal melanocyte population. Am J Dermatopathol 1987;9(5):438-444.
14. Matsouka LY, Glasser S, Barsky S. Melanoacanthoma of the lip. Arch Dermatol 1979;115(9):1116-1117.
15. Goode R, Crawford B, Callihan M, Neville B. Oral melanocanthoma: Review of the literature and report of ten cases. Oral Surg Oral Med Oral Pathol 1983;56(6):622-628.
16. Landwehr DJ, Halkias LE, Allen CM. A rapidly growing pigmented plaque. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1997;84(4):332-334.
17. Heine B, Drummond JF, Damm DD, Heine RD 2nd. Bilateral oral melanoacanthoma. Gen Dent 1996;44(5):451-52.
18. Fatahzadeh, Siros DA. Multiple intraoral melanoacanthomas: A case report with unusual findings. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2002;94(1):54-56.
19. Kauzman A, Pavone M, Blana S, Bradley G. Pigmented lesions of the oral cavity: Review, Differential diagnosis, and case presentations. J Can Dent Assoc 2004;70(10):682-683.
20. Wright JM, Binnie WH, Byrd DL, Dunsworth AR. Intraoral melanoacanthoma. J Periodontol 1983;54(2):107-111.
21. Horlick HP, Walther RR, Zegarelli DJ, Silvers DN, Eliezri YD. Mucosal melanotic macule, reactive type: A simulation of melanoma. J Am Acad Dermatol 1988;19(5):786-791.
22. Schneider LC, Mesa ML, Haber SM. Melanoacanthoma of the oral mucosa. Oral Surg Oral Med Oral Pathol 1981;52(3):284-287.
23. Frey VM, Lambert WC, Seldin RD, Schneider LC, Mesa ML. Intraoral melanoacanthoma. J Surg Oncol 1984;27(2):93-96.
24. Whitt JC, Jennings DR, Arendt DM, Vinton JR. Rapidly expanding pigmented lesion of the oral buccal mucosa. J Am Dent Assoc 1988;117(5):620-622.

ABOUT THE AUTHORS
Kshitiz Rohilla (Corresponding Author)
Demonstrator, Department of Oral Pathology, Postgraduate Institute of Dental Sciences, Rohtak, Haryana, India, e-mail: dr.k.rohilla@gmail.com

V Ramesh
Dean, Professor and Head, Department of Oral Pathology and Microbiology, Mahatma Gandhi Postgraduate Institute of Dental Sciences, Puducherry, India

PD Balamurali
Professor, Department of Oral Pathology and Microbiology, Mahatma Gandhi Postgraduate Institute of Dental Sciences, Puducherry, India

Namrata Singh
Ex-Senior Lecturer, Department of Orthodontics and Dentofacial Orthopedics, Indira Gandhi Institute of Dental Sciences, Puducherry, India