CASE REPORT

Oral melanoacanthoma: A rare case of diffuse oral pigmentation

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ABSTRACT
The clinical presentation of diffuse pigmentation can be alarming to the patient as well as the clinician. A histopathologic examination of a pigmented lesion is necessary in most of the cases in the oral cavity. Oral melanoacanthoma is a very rare diffuse pigmentation with no specific treatment required. It shows increased number of dendritic melanocytes in an acanthotic epithelium. We present a rare case of diffuse pigmentation in the oral cavity whose diagnosis was done on the basis of clinical presentation and histopathology. Also immunohistochemistry was done.

Key words: Diffuse pigmentation, melanocytes, oral melanoacanthoma

INTRODUCTION

Healthy oral mucous membrane is normally of varying shades of red. When either the patient or the clinician notices areas of pigmentation, there is often an element of increased concern.[¹] Focal lesions usually need an in-depth examination to exclude a melanoma, while diffuse lesions often have no specific histological features and do not generate prognostic perplexity. However, diagnosis of these lesions is important because they could be a sign of diseases with systemic implications such as Peutz-Jeghers syndrome or adrenal insufficiency.[²] Melanin is produced by melanocytes in the basal layer of the epithelium and is transferred to adjacent keratinocytes via membrane-bound organelles called melanosomes.[³] The term melanoacanthoma 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. Matsuoka et al. in 1979 reported the first case in the oral cavity.[⁴] Oral melanoacanthoma (OMA) is a rare, benign pigmented lesion, brown to brown-black, well circumscribed, characterized by hyperplasia of spinous keratinocytes and dendritic melanocytes.[⁵] We present a rare case of diffuse oral pigmentation which led us to diagnose it as OMA.

CASE REPORT

A 22 year old female patient came to our institution Vaidik Dental College and Research centre, Daman with a complaint of black pigmentations in the mouth since 2 months. Patient reported that she was apparently alright 2 months back when she noticed black pigmentations in the mouth. There was no pain. It progressively went on increasing in size. The patient was concerned whether it was normal or some disease and so the patient reported to our institute. Patient gave negative history of any medication and tobacco consumption. Extraoral examination revealed nothing conclusive.

Intraoral examination revealed brown black macular pigmentations widespread along the posterior part of Left and Right Buccal mucosa [Figure 1]. It also showed involvement of retromolar areas, hard palate, soft palate and fauces. There were no nodular growths. An incisional biopsy from buccal mucosa was taken for histopathologic examination. A routine Hematoxyllin and Eosin staining was done. The section showed non-keratinized stratified squamous epithelium. There was marked acanthosis with diffuse distribution of dendritic melanocytes in suprabasal layers of epithelium [Figure 2]. There was no cytologic atypia. Diagnosis of OMA was made. Confirmatory immunohistochemical analysis was done with S-100 protein. Immunohistochemistry revealed diffuse nuclear and cytoplasmic immunoreactivity with for S-100 protein [Figure 3].

DISCUSSION

The differential diagnosis of diffuse oral pigmentation include Peutz-Jeghers syndrome, Addison’s disease, pigmentation due
Oral melanoacanthoma

Gupta, et al. 442

Peutz-Jeghers syndrome is an inherited, autosomal dominant disorder with variable inheritance, characterized by hamartomatous polyps in the gastrointestinal tract, mostly in the small bowel, and pigmented mucocutaneous lesions.[5] In the present case, the patient had no previous or family history of any kinds of pigmented lesions or gastric intestinal polyposis.

Hematological examination revealed normal levels of ACTH. This ruled out the possibility of Addison’s disease.

Diffuse pigmentation of the oral mucosa and/or skin secondary to systemic drug administration is a well-recognized phenomenon.[6] The patient did not give any history of medications including oral contraceptive and anti-malarial drugs.

Laugier-Hunziker syndrome is an idiopathic macular hyperpigmentation of skin characterized by brownish black spots on oral mucosa including lips associated with longitudinal melanonychia of nails.[7] Although the pigmentation was diffuse and macular, the nails did not show any abnormalities. Also this syndrome is more common in white population.

Melanoacanthoma of the oral mucosa is a rare condition indicative of a reactive process. OMA is a benign pigmented disorder of the oral mucosa, characterized by simultaneous proliferation of both melanocytes and keratinocytes. To emphasize the non-neoplastic nature of the disease, Tomich and Zunt suggested the term melanoacanthosis while reserving the designation “melanoacanthoma” for cutaneous tumors.[8] The intra-oral site most commonly affected in OMA is the buccal mucosa but involvement of other sites such as the mucosa of the lip, palate, gingiva and alveolar mucosa has also been reported.[9] The clinical presentation is a brown to brown-black macular lesion, predominantly solitary, encountered in the younger age group with a distinct female predilection.[9] Our case had a diffuse presentation which is all the more rare. OMA is considered to be a reactive phenomenon and it has been suggested that masticatory and frictional trauma may play an etiological role.[10] Microscopically OMA is characterized by the presence of numerous benign appearing dendritic melanocytes scattered throughout an acanthotic and mature squamous cell epithelium. Basal layer melanocytes may be increased in number and spongiosis is a common finding. In addition inflammatory cells with eosinophils may be present in the subjacent connective tissue stroma.[10] The diagnosis of OMA can be done with proper history and routine histopathological examination. An immunohistochemical analysis with melanocytic marker (S-100 protein) is an additional way to confirm. It is a benign condition and once the diagnosis is confirmed no treatment may be required.

CONCLUSION

OMA is a rare benign condition which must be distinguished from other diffuse pigmentations like Peutz-Jeghers syndrome, Addison’s disease, pigmentation due to medication and smoking and medications, Laugier-Hunziker syndrome and OMA.

Peutz-Jeghers syndrome is an inherited, autosomal dominant
Laugier-Hunziker syndrome. There is usually no treatment required in such cases and there have not been any cases of malignant transformation reported till date.

REFERENCES

1. Hatch CL. Pigmented lesions of the oral cavity. Dent Clin North Am 2005;49:185-201.
2. Montebugnoli L, Grelli I, Cervellati F, Misciali C, Raone B. Laugier-hunziker syndrome: An uncommon cause of oral pigmentation and a review of the literature. Int J Dent 2010;2010:525404.
3. Gondak RO, da Silva-Jorge R, Jorge J, Lopes MA, Vargas PA. Oral pigmented lesions: Clinicopathologic features and review of the literature. Med Oral Patol Oral Cir Bucal 2012. [In Press].
4. Marocchio LS, Júnior DS, de Sousa SC, Fabre RF, Raitz R. Multifocal diffuse oral melanoacanthoma: A case report. J Oral Sci 2009;51:463-6.
5. Kopacova M, Tacheci I, Rejchrt S, Bures J. Peutz-Jeghers syndrome: Diagnostic and therapeutic approach. World J Gastroenterol 2009;15:5397-408.
6. Rangwala S, Doherty CB, Katta R. Laugier-Hunziker syndrome: A case report and review of the literature. Dermatol Online J 2010;16:9.
7. Sachdeva S, Sachdeva S, Kapoor P. Laugier-hunziker syndrome: A rare cause of oral and acral pigmentation. J Cutan Aesthet Surg 2011;4:58-60.
8. Geetha T, Rani GG, Krishnaram AS. Bilateral oral melanoacanthoma in an Indian boy. Indian J Dermatol Venereol Leprol 2011;77:210-2.
9. Lakshminarayanan V, Ranganathan K. Oral melanoacanthoma: A case report and review of the literature. J Med Case Rep 2009;3:11.
10. Contreras E, Carlos R. Oral melanoacanthosis (melanoacanthoma): Report of a case and review of the literature. Med Oral Patol Oral Cir Bucal 2005;10:11-2; 9-11.

How to cite this article: Gupta AA, Nainani P, Upadhyay B, Kavle P. Oral melanoacanthoma: A rare case of diffuse oral pigmentation. J Oral Maxillofac Pathol 2012;16:441-3.

Source of Support: Nil. Conflict of Interest: None declared.

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