ORAL MELANOTIC NEVI: A CASE REPORT AND REVIEW OF LITERATURE

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Abstract:

Oral melanotic nevi are uncommon oral lesions causing focal pigmentation. Melanotic nevi are benign melanocytic tumours originating from defective melanoblasts of the neural crest. Clinically, it is an asymptomatic, flat or slightly elevated lesion of brown or brown-black color. It is usually located on the palate and buccal mucosa and rarely on the gingiva and lips. Based on histological criteria, intraorally four types of nevi have been described: the intramucosal, junctional, compound and blue.

In this article, we are reporting a case of oral pigmented lesion in a young female with an unusual history of associated occasional pain which was histopathogically diagnosed as intramucosal nevus.

Key words: *oral melanotic nevi, pigmentation, melanoblasts*

Introduction:

Oral pigmentation may be exogenous or endogenous in origin. Exogenous pigmentation is commonly due to foreign-body implantation in the oral mucosa. Endogenous pigments include melanin, hemoglobin, hemosiderin and carotene. Pigmented lesions caused by increased melanin deposition may be brown, blue, grey or black; depending on the amount and location of melanin in the tissues.[1] Pigmented lesions of the oral cavity are quite common and have multiple etiology. Such lesions represent a variety of clinical entities, ranging from physiologic changes (e.g., racial pigmentation) to manifestations of systemic illnesses (e.g., Addison's disease) and malignant neoplasms (e.g., Melanoma and Kaposi's sarcoma).[2]

Melanotic nevi of the oral mucosa are benign melanocytic tumours originating from defective melanoblasts of the neural crest and causes focal oral pigmentation. Although nevi are common lesions that are seen on the skin in the large majority of the population, they are rare intraorally. They can be seen in persons of all ages and are usually less than 5 mm in diameter. When seen intraorally, they are most commonly observed on the hard palate.[3] Clinically, pigmented nevi is an asymptomatic, flat or slightly elevated spot or plaque of brown or brown-black color.[4]

This paper presents a case of oral melanotic nevus of a young female presenting with an unusual history of occasional pain in the same area.

Case Report:

A 22-year-old female presented with a pigmented lesion on the labial mucosa in

relation to lower right canine and premolar. Patient noticed this lesion one year back associated with occasional pain. Intraoral examination showed a well-demarcated, oval- shaped lesion on the right labial mucosa, measuring 5 mm in diameter. The lesion was black in color and slightly raised [Figure 1]. There were no other pigmented lesions on the oral mucosa.

The differential diagnosis of this focal, raised pigmented lesion included amalgam tattoo, hematoma, oral melanotic macule, pigmented nevus, melanoacanthoma and melanoma. Oral melanotic macule was less likely because the lesion was raised. The colour of the lesion was not consistent with amalgam tattoo or a vascular lesion. The long duration without change in size favoured pigmented nevus over melanoacanthoma and melanoma.

Excisional biopsy showed hyperplastic parakeratinized stratified squamous epithelium with areas of atrophy in between. The underlying connective tissue stroma contained collections of nevus cells in the form of islands and sheets. Of these superficial nevus cells were large and contained melanin pigment. At areas nevus cells also showed cellular atypia. The intervening scanty connective tissue stroma was delicately collagenous with mild chronic inflammatory infiltrate. [Figure 2]

As we had seen some cellular atypia in the superficial nevus cells, bleaching procedure was done to clearly observe cellular and nuclear details after removing the melanin pigment. [Figure 3]

The histopathological diagnosis was suggestive of intramucosal nevus.

Fig1. A pigmented lesion on right labial mucosa

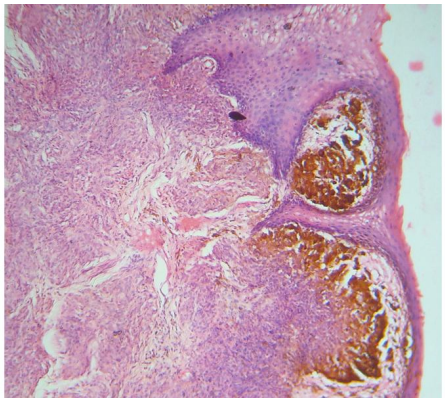

Fig2. H & E photomicrograph (10x) showing intramucosal nevus.

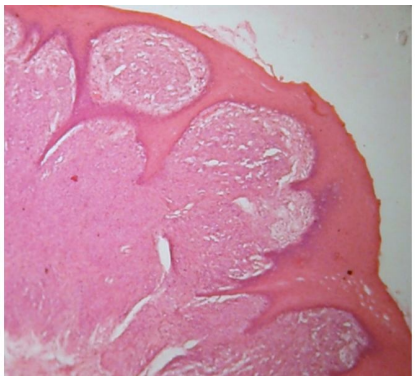

Fig3 H & E photomicrograph (10x) of intramucosal nevus after bleaching procedure.

Discussion:

Diagnosis of pigmented lesions of the oral cavity and perioral tissues is challenging. Clinicians should evaluate and diagnose all alterations in pigment. But definitive diagnosis usually requires histopathological evaluation. There is an algorithm suggested by Kauzman A. *et al* to guide the assessment of pigmented lesions of the oral cavity on the basis of history, clinical examination and laboratory investigations.[2] [Table 1]

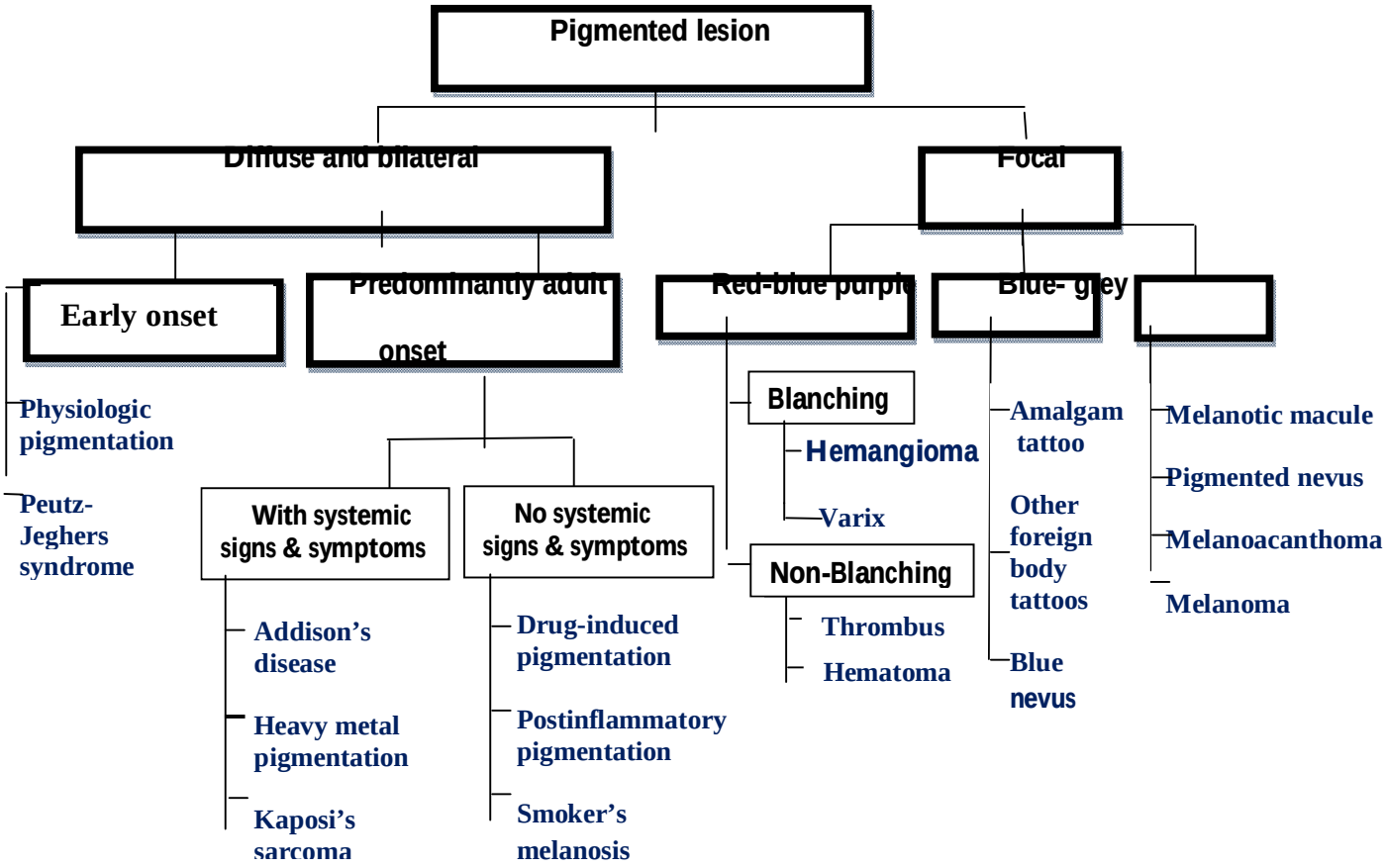

Table 1: An algorithm for evaluation of pigmented lesions of the oral cavity

Oral melanocytic nevi (OMN) are rare benign tumours of melanocytes. Epidemiological data are scanty regarding OMN to predict its etiology and pathogenesis.

Histologic variants of OMN corresponds to melanocytic proliferation at various levels: 1) proliferation of benign neoplastic melanocytes along the epithelial-mesenchymal junction (junctional nevus); 2) migration of these cells into the mesenchymal compartment (compound nevi); and 3) loss of the junctional component of the nevus, so that all remaining nevomelanocytes are located within the subepithelial compartment (intramucosal nevi) [5, 6]

Ordie H. *et al* in 1967 reported that pigmented nevi of the oral cavity had been found in 0.1 per cent of the Negro patients examined at his medical centre. They also found in their study that pigmented nevus of the oral cavity was a relatively common lesion which was usually overlooked because of its small size and innocuous behaviour. The most common intraoral location was buccal mucosa and intramucosal nevi the most common type.[7]

Buchner A. *et al* did a clinicopathologic study of 32 new cases and reviewed 75 cases from the literature and found that pigmented nevi were much less common in the oral cavity than on the skin. They also supported the earlier reports that the most common type of nevus was intramucosal, followed in decreasing order by the common blue nevus, compound nevus, and junctional nevus.[8]

Again in 1980, Buchner A. *et al* reviewed and analysed data of seventy-five cases from the literature, together with an additional thirty-two new cases and revealed that nevi of the oral mucosa were not rare and also stated that oral nevi (especially those that were clinically nonpigmented) were often misdiagnosed, indicating that they were far more common than they would seem from the reported cases.[9]

In another literature review by Buchner and Hansen, hard palate was the most frequent location for the blue nevus, whereas the buccal mucosa was the most frequent site for the intramucosal nevus.[10]

Buchner and Hansen in still another review and analysis of data on 191 cases of oral pigmented nevi from the literature and from two studies at the University of California, San Francisco, found that 55% of the pigmented nevi were of intramucosal type, 32% common blue nevi, 6% compound nevi, 5% junctional nevi, and 2% combined. Sitewise, 41% of all nevi were found on the hard palate, 20% on the buccal mucosa, 12% on the vermilion border, and 11.5% on the gingiva. They were rarely found on the soft palate, tongue, and retromolar pad. They also reported that oral nevi were small, most being between 0.1 and 0.6 cm and most of them were raised, which could be of help in the differential diagnosis.[11]

In 1990, Buchner A. and Leider A.S. *et al* presented a paper on analysis of data on 130 cases of oral melanocytic nevi from the files of the University of the Pacific, San Francisco and the University of California, San Francisco reporting that intramucosal type were the most commonest (63%) followed by the common blue nevus (19%), compound nevi were uncommon (9%) and junctional nevi were rare (5%). Combined nevi were the rarest type (4%).[12]

Biesbrock A.R. *et al* in 1992 documented the unusual occurrence of multiple intraoral junctional nevi in a patient[13] and a case of

unpigmented intramucosal nevus occurring in the palatal mucosa has been reported by George Laskaris *et al* in 1994. [14]

Later in 2004, Buchner A. *et al* analysed 773 cases of solitary pigmented melanocytic lesions in the oral mucosa and revealed that oral melanocytic nevi comprised 11.8% of the entire melanocytic group with mean age at diagnosis being 30.5 years and palate the most common site.[15]

Takata and Saida identified different patterns of genetic alterations among different kinds of cutaneous melanotic nevi but still their role in OMN is poorly understood.[16]

A report from the Netherlands during the period 1980–2005 by Meleti *et al* revealed an annual incidence of excised OMN around 4.35 cases per 10 million populations per year. According to them there was no concrete support for the idea that the presence of an oral melanocytic nevus indicates a risk of future development of oral malignant melanoma (OMM).[17]

Though to date there are no reported cases of malignant transformation of intramucosal type of OMN, all OMN should be surgically excised as a prophylactic measure because of the constant chronic irritation of the mucosa in nearly all intraoral sites occasioned by eating, toothbrushing etc.

On reviewing the literature, there are no reports of ocassional pain associated with OMN but in our case we reported the same which could be attributed to trauma or of psychological origin. Even the presence of cellular atypia in the superficial nevus cells in our case have not been reported previously and hence significance not known.

Conclusion:

Pigmented lesions of the oral mucosa range from the extremely common and harmless (eg, amalgam tattoo) to the rare and deadly (eg, malignant melanoma). Various pigmented lesions can have similar clinical presentations, posing a diagnostic dilemma for the dentist.

In the present paper, we have reported a case of OMN in lower labial mucosa which is not so common a site of presentation and also with an unusual history of associated pain. Though the reason of associated pain could not be elicited, the lesion had healed completely after biopsy. Since there are no reported cases of symptomatic nevi, the most probable reason for occurrence of pain in our case could be trauma due to its location or it could be psychological.

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