Oral Melanocytic Nevus (OMN) with papillomatous presentation-A case report

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ABSTRACT

Introduction: Oral melanocytic nevi are rare benign melanocytic tumors which present as papules, plaques or flat lesions. The authors are reporting this case as it was interesting due to its rare occurrence in oral cavity and unusual papillomatous presentation, which is not seen in oral cavity. **Case presentation:** A 42 year old female patient presented with a solitary pigmented papillomatous lesion on anterior palate. Microscopic analysis of excisional biopsy revealed nests of nevus cells in superficial lamina propria. The diagnosis of an oral compound nevus was given. **Conclusion:** Histopathological analysis should be mandatory for such lesions to rule out oral melanomas.

Keywords: Oral melanocytic nevus, oral compound nevus, oral nevi

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Conflict of interest: None

INTRODUCTION

Oral melanocytic nevi (OMN) are benign melanocytic tumors which are composed of nevus cells.[1] They clinically present as papules, plaques or flat lesions and may be pigmented or non-pigmented.[2] Nevus cells are similar to melanocytes, but differ by arranging themselves in clusters and nests.[1] They may not show dendritic processes in compact nests.[1]

The occurrence of Oral melanocytic nevi is rare.[3] Meleti et al. have mentioned that only a small series of cases of OMNs have been reported and there is a need to systematically study the prevalence and incidence of OMNs.[4] Buchner et al reported OMNs to represent

0.1% of the 90,000 biopsies considered in their study.[5] The authors hereby report a rare case of oral melanocytic nevus with papillomatous presentation, with special emphasis on clinical and pathological features of this lesion.

CASE PRESENTATION

A 42 year old female reported to a private practitioner with the chief complaint of a painless swelling in the palate. The patient was not sure of the duration but had noticed the lesion eight months before consulting the doctor. There was no history of trauma and patient had no adverse habits like smoking, tobacco or alcohol. The patient did not suffer from any other systemic diseases and drug history was insignificant.

On examination, a lesion measuring approximately 2cmX1cm with papillomatous surface and brownish melanotic patch in the centre was seen on the left side of anterior palate, just adjacent to the midline (Figure 1). It was asymptomatic and not associated with any discharge or induration. The lesion was solitary and not associated with any other melanotic patches or other lesions throughout the body.

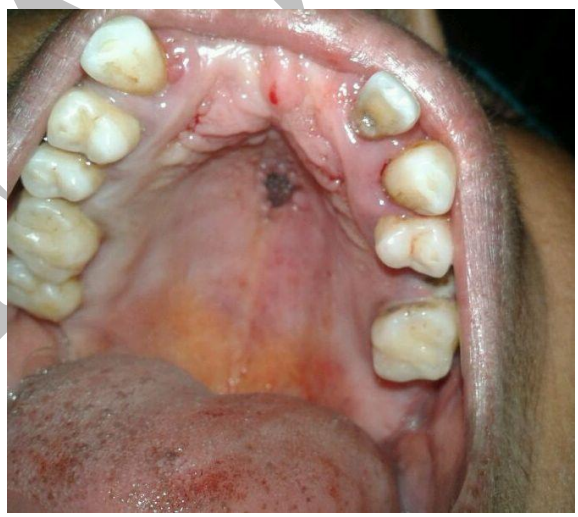

Figure 1: Lesion on anterior palate with a papillomatous surface and brownish melanotic patch in the centre.

The clinical differential diagnosis of oral melanocytic nevus, hematoma, oral melanotic macule, melanoacanthoma and oral melanoma was considered. Excisional biopsy of the lesion was carried out and submitted for histopathological analysis.

Gross examination of the specimen revealed one bit of soft tissue measuring 1cmX1.4cmX0.4cm in greatest dimensions. It had a pebbled surface with some raised globules around an umbilicated area showing the brownish discoloration (Figure 2).

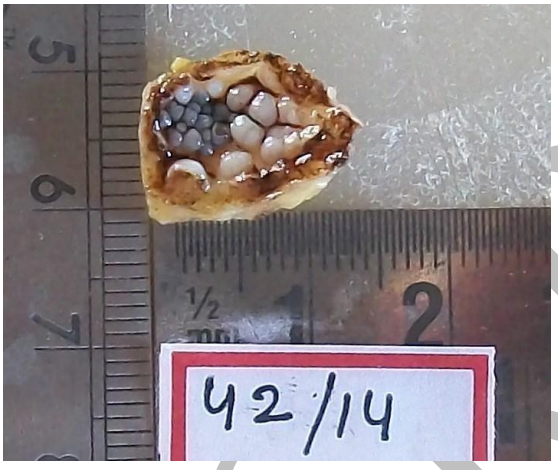

Figure 2: Gross examination revealed a soft tissue having pebbled surface with some raised globules around an umbilicated area showing brownish discoloration.

Histopathological examination revealed a hyperplastic parakeratinized stratified squamous epithelium showing papillomatous morphology (Figure 3).

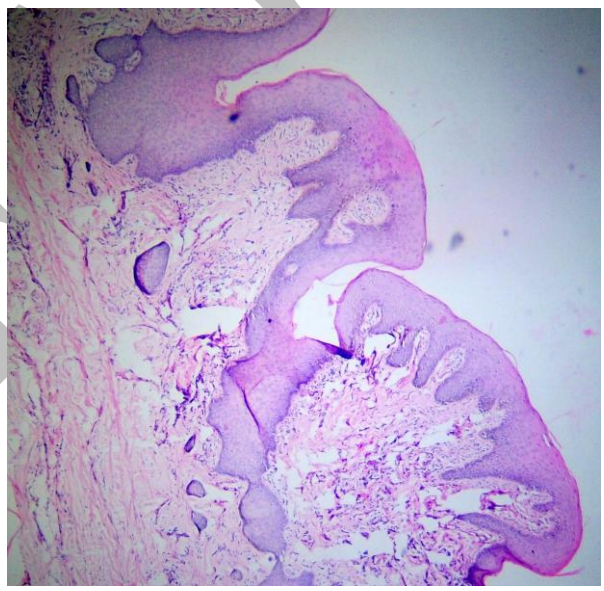

Figure 3: Hyperplastic parakeratinized stratified squamous epithelium showing papillomatous morphology (H&E 40X)

Melanin pigmentation was seen in basal layer of epithelium. Some groups of nevus cells arranged in clusters were seen in the epithelium and superficial lamina propria. Some of these cells showed melanin pigments (Figure 4).

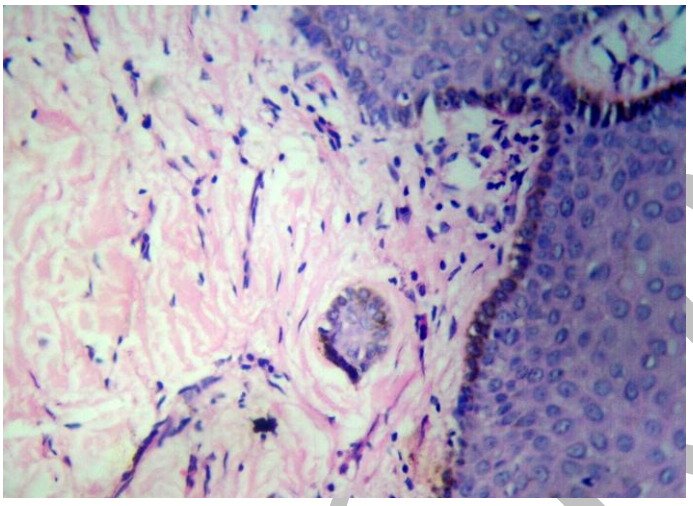

Figure 4: Nevus cells containing melanin pigments in basal cell layer and superficial lamina propria (H&E 400X)

Cellular atypia was absent. Connective tissue contained focal areas of chronic inflammatory cells, mainly lymphocytes and plasma cells. Few melanophages were also seen. Epithelium appeared to be eroded in few areas. Reoriented and deeper sections were taken to rule out the artefactual presence of clusters of nevus cells in the connective tissue. The final diagnosis was given as a Compound nevus.

DISCUSSION

Any form of oral pigmentation can be physiological or pathological; exogenous or endogenous; congenital or acquired and focal or diffuse.[3] Important parameters to be considered for differential diagnosis may include color, location, distribution, duration, drugs use, family history, type of growth and change in pattern.[3]

Oral pigmented lesions can be melanocytic and non-melanocytic.[2] Melanocytic lesions include physiologic (ethnic) pigmentation, smoking associated melanosis, oral melanotic macules, pigmented neuroectodermal tumor of infancy, melanocytic nevus, melanoacanthoma and melanomas.[2] Whereas, non-melanocytic lesions include focal argyrosis (amalgam tattoo), drug

induced pigmentations and heavy metal pigmentations.[2]

Oral melanocytic nevi are rare, usually asymptomatic and thus, most of the times, they are discovered when the patient visits a dentist or doctor for some other chief complaint.[3] Most commonly, these lesions are found on the hard palate in the oral cavity.[2] It can also be found at other less common sites like buccal mucosa, labial mucosa, gingiva, alveolar ridge and vermilion.[2] In the present case, the lesion was present on the commonest intraoral site, although the papillomatous presentation was very unusual for this location.

Other papillomatous lesions of oral cavity can be viral papillomas (focal epithelial hyperplasia, squamous papilloma, verruca vulgaris, condyloma acuminatum), verruciform xanthoma, fibroepithelial polyps, papillary hyperplasia, pyostomatitis vegetans, acanthosis nigricans, darier's disease, verrucous hyperplasia, sialadenoma papilliferum papillary dysplasia, proliferative leukoplakia, papillary carcinoma, verrucous carcinoma and carcinoma cuniculatum.[6]

A predilection for females is reported in studies by Buchner et al and Kaugars et al. [5,7] Melanocytic nevi are mostly seen in adolescence and early childhood, although rarely seen at birth.[1] Meleti et al, after analysis of 119 cases reported that combined and junctional nevi are found most commonly in young individuals.[4] 75% of the lesions in their analysis were from patients below the age group of 40 years.[4] They also mentioned that most subepithelial nevi are diagnosed in the fifth decade.[4] The authors supported the idea that melanocytic nevi follow the sequence of junctional phase, compound phase and finally intramucosal phase.[4] In our case the patient was above 40 years and not showing any intramucosal nests of nevus cells.

The three main types of nevi, depending on the location of nests of nevus cells, are junctional nevi, compound nevi and intradermal (or intramucosal in oral mucosa) nevi.[1] Special variants include balloon cell nevus, halo nevus, spitz nevus, pigmented spindle cell nevus, congenital melanocytic nevus and dysplastic nevus.[1] If melanocyte proliferation occurs in basal layer, it is a junctional nevus.[8] The presence of this

melanocytic proliferation in superficial lamina propria along with the basal layer are a feature of compound nevus. Finally, in intramucosal nevus, nevomelanocytes can be found only in lamina propria.[8] In Butchner's study, Intramucosal nevi were the most common (64%), followed by compound nevi (16.5%), common blue nevi (16.5%) and Junctional nevi being uncommon (3.0%).[5]

Clinically, most compound nevi on skin exist as slightly elevated lesions whereas some are papillomatous.[1] According to Kauzman et al, Compound nevi are typically light brown, dome-shaped lesions.[9] Regezi et al quote that 'most oral melanocytic nevi present as small (0.5cm) elevated papules or nodules that are often non-pigmented.'[2] Compound nevi have very rarely presented as papillomatous lesions with slight central depression and brownish pigmentation, as was seen in our case.

A compound nevus may show features of both junctional and intradermal/intramucosal nevus.[1]

In this type, nevus cells may be seen in the epidermis or epithelial covering of the mucosa, as well as dropping off into the dermis or into the superficial lamina propria.[1,3] In the upper dermis or superficial lamina propria, nevus cells are usually cuboidal with abundant cytoplasm and containing varying amounts of melanin granules.[1] Melanosomes may be spotted in the surrounding stroma.[1] Cells in the mid-dermis or deeper lamina propria are distinctly smaller than those in upper dermis or superficial lamina propria, lie in well defined aggregates and contain less cytoplasm and less melanin.[1]

Nevus cells are derived from neural crest cells, can synthesize melanin and are found in skin and mucosa.[9] They differ from melanocytes not only in their arrangement as clusters and nests, but also in their cell morphology.[10] Nevus cells are spindle to ovoid-shaped and do not have dendritic processes.[10] They are arranged in clusters because contact inhibition has been lost.[10] Melanosomes produced are not transferred to adjacent keratinocytes, as seen in melanocytes.[10] Also they have the ability to migrate from the basal layer of epithelium into the underlying submucosa.[11] Nevus cells lack pleomorphism, cytologic atypia, and rarely show mitotic activity, as can be seen in case of malignant cells of oral melanoma.[10] Melanoma cells can invade

the connective tissue and spread in pagetoid manner, and also metastasize to the draining lymph nodes as well as distant sites.[10]

The etiology and pathogenesis of Melanocytic nevi are not clearly understood. Cutaneous melanocytic nevi are believed to result from BRAF or NRAS oncogenic mutation in melanocytes.[3,11] This mutation may result in excessive cellular proliferation and thus formation of nevi. A cell senescence ability in nevus cells differentiates them from the melanoma producing ones.[11] In view of the histologic similarities between cutaneous and oral melanocytic nevi, it seems possible to assume that the pathogenesis of oral nevi will be similar to that of the cutaneous nevi.[4]

Malignant transformation of intraoral nevi has not been reported, but they may be considered to represent precursor lesions to oral mucosal melanomas.[10,12] Biopsy is advisable for any new oral pigmented lesion or suspected oral melanocytic nevus to rule out an early melanoma.[12] Mostly surgeons prefer to go for excisional biopsy due to small size of lesion. Similar treatment planning was followed in our case due to presence of a solitary lesion with small diameter.

If present at other extraoral sites, malignant transformation may occur in 3-6months, and will be accompanied by alterations in size, color, topography and formation of ulcers.[3]

CONCLUSION

Diagnosis of Oral nevi can be challenging, but awareness about them is a must. In order to appropriately treat these patients, clinicians must be able to rule out the differential diagnosis of malignant melanoma, clinically and histologically.

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