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## Case Report

### Blue nevus of the hard palate: A case report



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#### ABSTRACT

Oral pigmentation ranges from physiologic pigmentation to malignant neoplasms. Blue nevi (BN), benign melanocytic lesions that rarely develop in the oral cavity, can be associated with malignant melanoma. Although most oral pigmented lesions are benign, long-term follow-up/histopathologic evaluation eliminate oral malignant melanoma (OMM) when lesions develop in the most frequent sites (palate and maxillary gingiva).

A 58-year-old Japanese man with a BN presented with a 5-year history of a blue-black macule on the hard palate. The patient had no malignant signs, no abnormal radiologic findings in the maxillofacial region, or abnormal laboratory values. Histopathology showed parallel spindle-shaped melanocytes under the epithelium with fibrosis. A BN was diagnosed. The 2-year follow-up and clinical outcome were satisfactory, without recurrence or complications.

Close monitoring and long-term follow-up or early biopsy to exclude OMM in the oral cavity are important in cases in which the diagnosis is not definitive clinically.

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## 1. Introduction

The oral cavity is a common site for development of diversified pigmented lesions resulting from, for example, physiologic pigmentation, systemic diseases, oral mucosal insults, drug-induced pigmentation, and pigmented neoplasms [1]. Among the pigmented neoplasms, oral melanocytic nevi (OMNs) are rare benign tumors of the melanocytes [2]. Melanocytes are located in the basal layer of the epithelium, originate from the neural crest, and are found in skin and mucosa [3]. They are interspersed regularly between the basal keratinocytes and are also present in any region of the oral cavity.

The incidence and prevalence of OMNs have not been studied systematically. Buchner et al. reported the relative frequency of solitary oral melanocytic lesions in 89,430 cases accessed during a 19-year period at the Pacific Oral and Maxillofacial Pathology Laboratory [4]. The most common lesions were melanotic macules (665 cases), followed by OMNs (91 cases). Oral malignant melanoma (OMM) was the least common lesion (5 cases). Another study also reported an annual incidence of excised OMNs of 4.35 cases per 10 million populations in the Netherlands [5].

OMNs usually are discovered during routine dental examinations because they are asymptomatic. Diagnosis of OMNs can be challenging because up to 15% of the lesions are nonpigmented, and in these cases, inexperienced clinicians may fail to consider nonpigmented OMNs in the differential diagnosis [2]. Because the clinical appearance of OMNs is not specific, a biopsy usually is warranted to eliminate other pigmented lesions, most importantly, early-stage OMM. The most common of the OMNs is the intramucosal nevus followed by the blue nevus (BN) [6]. Although several hundred cases have been reported in the literature, the pathogenesis and etiology of OMNs are poorly understood.

The first case of an intraoral BN was reported in 1959 [7]. The literature indicates that oral BNs affect all age groups, but patients

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in the third to fifth decades of life are generally affected [8]. Women tend to be more commonly affected than men [6]. The hard palate is the most common site for development of BNs, with 69% identified in this area followed by the labial mucosa and vermillion border [9]. The data from the current patient agrees with information in the literature regarding patient age and location but not sex. The palate is susceptible to continuous frictional, thermal, chemical, and physical risk factors that promote carcinogenesis. Patients who use tobacco and alcohol may be at even higher risk. BNs are characterized by a variety of histologic subtypes; the most frequently occurring are the combined and cellular types [2,6]. Many pigmented lesions can be diagnosed clinically based on the shape, size, or color, along with the clinical features. However, in some cases, immunohistochemical assessment may be required to achieve a correct diagnosis.

We report the case of a man with a BN on the hard palate and present a review of the literature.

## 2. Case report

A 58-year-old Japanese man was referred to our hospital with a 5-year history of a blue-black macule without clinical symptoms on the hard palate (Fig. 1). The medical history indicated that the patient was implanted with a cardiac pacemaker for complete heart block but was otherwise unremarkable. Clinical examinations showed that the largest diameter of the blue-black pigmented lesion was 5 mm and it had distinct margins. The surface was regular and the same texture as the palatine. Clinical symptoms were absent, and no palpable lymph nodes were detectable in the neck. Panoramic radiography and computed tomography showed no radiologic maxillary abnormalities. The results of routine blood examinations were within the normal limits. The patient had no systemic symptoms or diffuse pigmentation affecting multiple sites. The oral pigmentation was not similar to disease-associated pigmentation such as that in Addison's disease, Peutz-Jeghers syndrome, McCune-Albright syndrome, and Laugier-Hunziker syndrome.



Fig. 1. Black macule with a definite border on the hard palate.

In the current case, we excised the lesion and examined it pathologically based on the patient's wishes. The excisional biopsy was performed under general anesthesia. The palatine torus also was removed. The mucosal defect was covered with a polyglycolic acid sheet (Neoveil, Gunze Co., Ltd., Tokyo, Japan) and fibrin glue (Bolheal, Chemo-Sero-Therapeutic Research Institute, Kumamoto, Japan) (Fig. 2). Microscopic examination of tissue sections showed proliferation of spindle melanocytes in the lamina propria (Fig. 3A–C). Some spindle melanocytes have long branching

dendritic processes and no hyperchromatic and pleomorphic nuclei. The nucleoli were small and inconspicuous. Immunohistochemically, melanocytes were positive for Melan-A (Fig. 3D) and S-100 protein, and negative for HMB-45 (Fig. 3F). There were fewer MIB-1-positive nuclei (Fig. 3E). Based on the histologic and immunohistochemical findings, a diagnosis of common BN was established.



Fig. 2. Appearance of the surgical site covered with a polyglycolic acid sheet after excision of the pigmented lesion.

The postoperative follow-up was uneventful and the patient has been followed closely for 2 years without recurrence (Fig. 4).

## 3. Discussion

Discoloration of the oral mucosal surface occurs under many different conditions, which may have clinical relevance because they frequently show signs of either local or systemic disease. Multiple etiologies of pigmented oral mucosa range from iatrogenic mechanisms, such as amalgam tattoos to systemic disorders such as Peutz-Jeghers syndrome. Oral pigmented lesions result from cellular hyperplasia that can range from benign nevi to OMM.

Commonly, two groups of pigmented lesions of the oral cavity are recognized. The first are melanin-associated lesions, including racial pigmentation, melanocytic nevi, melanotic macules, systemic disorder-related oral pigmentation, and malignant melanoma. The second are nonmelanin-associated lesions, including endogenous and exogenous pigments and drug-associated pigmentation [1]. Among them, OMNs are uncommon benign melanocytic proliferations that can be congenital or acquired and that are classified similarly to the skin counterparts: junctional, compound, intradermic (intramucosal), combined, and blue [6]. This classification depends on the distribution, localization, and morphology of the nevus cells. Tische first reported BNs in 1906 [10]. They mainly affect young women and are found predominantly in the sacral area and scalp. Most BNs are on the skin; however, in rare cases, they have been observed in a variety of mucosal sites, including the oral mucosa.

BNs can become malignant in some cases. However, this is a controversial theory and most often malignancy occurs in the cellular types [11,12].

Histologically, the diagnostic cell of the BN is a variably pigmented, spindle-shaped dendritic melanocyte with a slender, branching network of dendritic processes [13]. The nuclei are small, elongated, and hyperchromatic. The dendritic cells of BNs do not display any significant cytologic atypia or mitotic figures. A previous study reported that BN cells frequently are immunoreactive to

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