

## Extensive intraoral blue nevus - Case report <sup>\*</sup>

### Extenso nevo azul intraoral - Relato de caso

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**Abstract:** Nevus is a congenital pigmented malformation rarely found in the oral mucosa. Around one third of cases located in this anatomical region are of the blue type, a histological variant with considerable tendency to malignancy. This study reports the case of a male patient, chronic smoker, with a blue nevus measuring 5cm in diameter on the hard palate. Since controversy exists in the literature regarding the incisional biopsy of pigmented lesions with malignant or malignant potential, excision without previous biopsy of the lesion was the therapy of choice for this case. The patient was followed-up for two years with no sign of recurrence or malignant transformation.

**Keywords:** Melanins; Nevus, blue; Palate

**Resumo:** O nevo é uma má-formação congênita pigmentada, raramente encontrado na mucosa bucal. Cerca de 1/3 dos casos localizados nesta região anatômica são do tipo azul, uma variante histológica com considerável tendência à malignização. Este artigo relata o caso de um paciente do sexo masculino, tabagista crônico, portador de um nevo azul de 5 cm de diâmetro no palato duro. A excisão da lesão sem biópsia prévia foi a conduta terapêutica de eleição para o caso, uma vez que ainda existe controvérsia na literatura a respeito da realização de biópsia incisional em lesões pigmentadas malignas ou com potencial de malignização. O paciente foi acompanhado por um período de 2 anos, sem sinais de recorrência ou transformação maligna.

**Palavras-chave:** Melaninas; Nevo azul; Palato

### INTRODUCTION

Nevus is a congenital pigmented malformation commonly encountered in skin. Depending on the location of the nevus cells, the malformation is classified histologically as intradermal, junctional, compound and blue, with the latter presenting a considerable tendency to malignancy. Normally found on the

hard palate, the presence of blue nevus in the oral mucosa is nevertheless uncommon.<sup>1</sup> Although the first reports of blue nevus involving the palatal mucosa date back to the 1950s and 1960s, only around 70 cases have been published.<sup>2,3</sup> Two classical forms were described in these reports: common blue nevus and

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cellular blue nevus.<sup>4</sup> The present study reports a case of a large blue nevus on the palate and discusses the diagnosis and treatment of this condition.

**CASE REPORT**

A white 45-year-old male, smoker for 25 years, presented a dark blue spot 5cm in diameter on the hard palate. The spot was asymptomatic and of unknown duration (Figure 1). Panoramic radiographs of the maxilla showed no intra-bone injury. The clinical and radiographic findings suggested blue nevus. Excision without previous biopsy of the lesion was the therapy of choice for this particular case, since there is still some controversy in the literature regarding the incisional biopsy of pigmented lesions with malignant or malignant potential. The resection was performed with electrocautery, preserving the greater palatine artery (Figures 2, 3A and 3B). However, the small bluish pigmentation spots present in the soft palate were not removed given the significant risk of creating a difficult-to-control bucco-nasal communication. A provisional prosthesis was inserted with surgical cement in order to avoid trauma during the healing period, as well as making the postoperative period more comfortable for the patient (Figures 4A and 4B). The specimen removed was sent for histopathological analysis. This revealed the presence of large amounts of thin and elongated melanocytes and dendritic cells in the mucosa, confirming the diagnosis of a common blue nevus subtype (Figures 5A and 5B). The patient was followed up for two years with no signs of recurrence (Figure 6).

**DISCUSSION**

The blue nevus is typically identified between the third and fifth decades of life, although a report dating back to the 1990s describes the problem in the

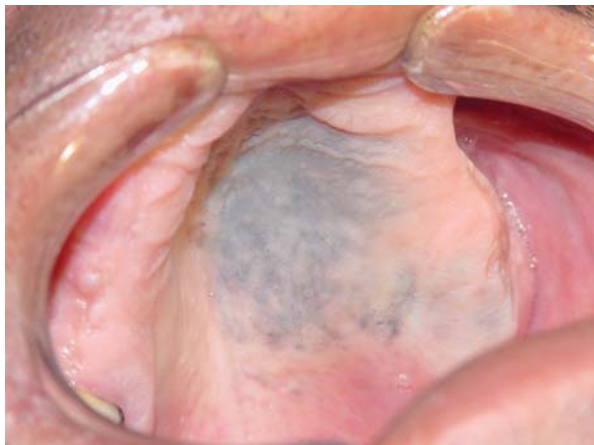


FIGURE 1: Dark blue stain on the hard palate



FIGURE 2: Removal of the lesion with electrocautery

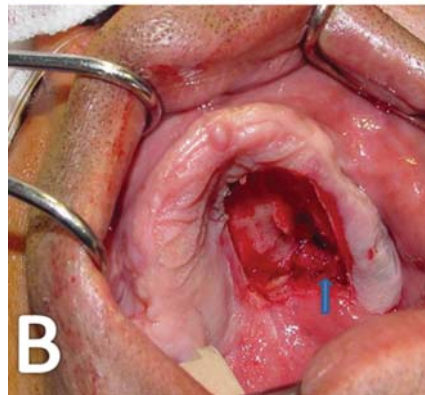


Figure 3: A - Excision of the lesion. B. Appearance of the surgical site after excision of the lesion and preservation of the greater palatine artery (arrow)

palatal region of an 11-year-old child.<sup>5</sup> Blue nevus tends to affect more women than men (male to female ratio of 1:1.5).<sup>16</sup> Around two-thirds of all cases occur in the hard palate, followed by the cheek mucosa.<sup>6,7</sup> Our case coincides with information in the literature regarding patient age and location but not the sex ratio data.

Experimental evidence about the origin of blue nevus is lacking. It is believed however that the blue nevus is the result of melanocytes being trapped in the dermis of the neural crest during migration

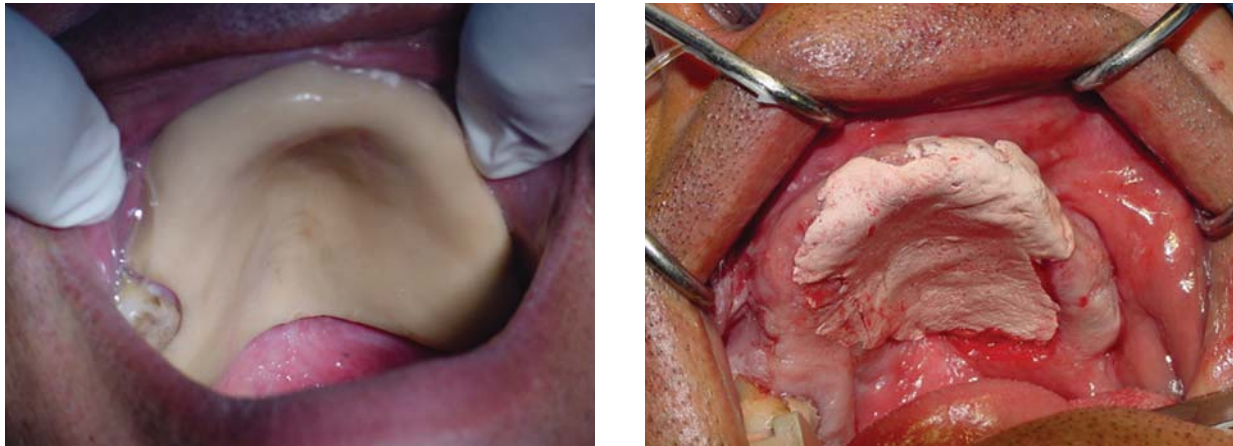


FIGURE 4: A - Postoperative prosthesis. B. Surgical cement

towards the embryonic epidermis. Groups of melanocytes can be found in fetal dermis and evolve during pregnancy. Genetic predisposition has been suggested, given the varied occurrence of blue nevus in different populations. However, familial cases of blue nevus are extremely rare and are not associated with chromosomal aberrations.<sup>8</sup> The patient reported no similar lesion in close relatives.

The blue nevus is a benign melanocytic lesion, typically asymptomatic. It is the second most common type of nevus of the oral cavity, accounting for 19-36% of all cases. It is characterized by a variety of histological subtypes (plate, epithelioid, cellular and 'common').<sup>9</sup> The common subtype which affects the back of hands and feet is either flat or elevated, brownish-blue or dark blue, usually with a diameter of between 2-10 mm.<sup>7</sup> Despite being the most common subtype, it rarely occurs in the oral cavity, which explains our interest in studying this case. Of significant interest

also is the fact that this blue nevus is probably the largest ever reported, given that all the other cases in the literature have involved nevi of less than 1cm in diameter.

Histopathologically, the common blue nevus subtype is composed of spindle-shaped melanocytes grouped in elongated and slender fascicles arranged in parallel under the epithelium. The melanocytes have dendritic processes that normally contain a large amount of melanin. Abundant tissue fibrosis with pigmented cells is found between the fascicles.<sup>4</sup> This description is in line with the histopathological findings in our case.

The blue color of the lesion can be explained by the "Tyndall" effect, which is related to the interaction of light with particles in a colloidal suspension. In the blue nevus, the melanin particles are deep, causing the reflected light to pass through the overlying tissue. Long wavelength colors tend to be more readily

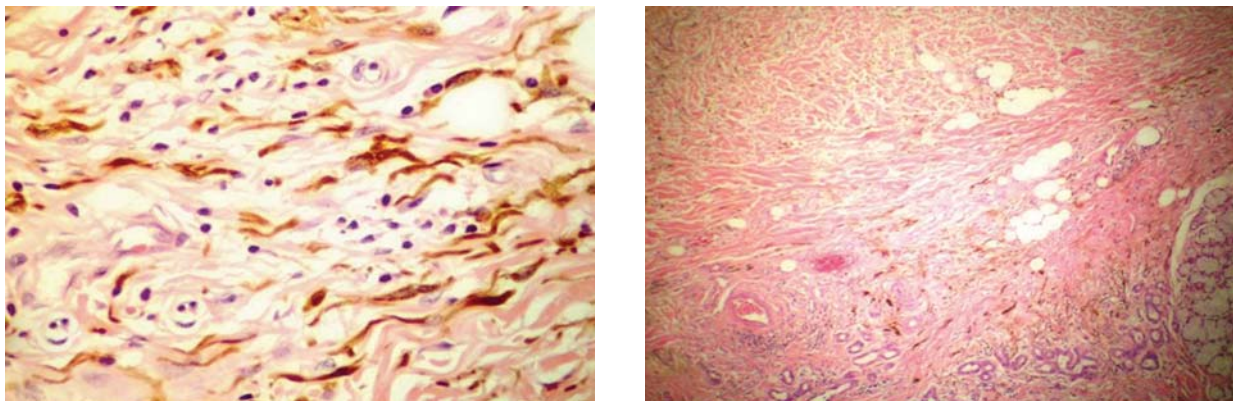


FIGURE 5: A. Large number of slender and elongated melanotic and dendritic extensions (hematoxylin and eosin, 100x). B. Elongated dendritic melanocytes and extensions located beneath the mucosa (hematoxylin and eosin, 100x)

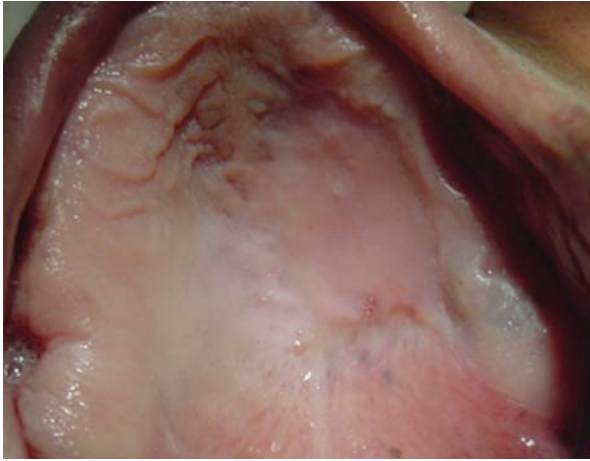


FIGURE 6: No signs of recurrence after two years monitoring

absorbed by the tissues, while the shorter wavelength light such as light-blue is more likely to be reflected back to the eye of the beholder.<sup>7</sup> In contrast to the literature, our case involved a dark-blue nevus, suggesting the reflection of a longer wavelength.

The 'plate' subtype of the blue nevus is a rare variant, especially when it occurs in the oral mucosa. It was first described by Fistarol and Itin in 2005.<sup>4</sup> Its histopathological features are similar to the common subtype, usually possessing focal points of hypercellularity. It is large flat or raised lesion which, in view of its clinical features, can be easily confused with metastatic melanoma.<sup>4,10</sup> Because of its large size our case could have been diagnosed as being of the 'plate' subtype. Histopathology however showed no focal hypercellularity. The distribution of melanocytes was homogeneous, which fitted the description of the 'common' type nevus.

The palate is very susceptible to physical and chemical risk factors for carcinogenesis, such as tobacco use. Changes such as chronic sialadenitis due to heavy smoking (particularly pipe-smoking) can be observed on microscopic examination. The literature indicates surgical removal of melanocytic nevus skin when exposed to chronic irritation due to the potential risk of malignant transformation.<sup>11</sup> Controversy however exists about the risk of malignant transformation of the blue nevus. While some authors agree

with its potential for malignancy,<sup>2,3,9,11,12</sup> some studies point to the lack of scientific evidence to support this theory,<sup>7,13,14</sup> while others recommend excisional biopsy of the areas delimited by pigmented oral lesions.<sup>2,13</sup> Other authors are of the opinion that that incisional biopsy can be performed without risking the spread of cancer cells.<sup>15</sup> In our specific case we decided not to perform the previous biopsy. Given our view that the lesion would be diagnosed as blue nevus, we justified its removal by the fact that the patient was a chronic smoker and had failed to attend routine monitoring sessions to assess the potential for malignant transformation of the blue nevus when exposed to chronic irritation (a controversial subject in the literature). In the event, histopathology revealed no malignancy.

Due to the rarity of blue nevus occurring in the oral cavity and the clinical and microscopic similarities of the blue nevus cellular subtype with melanoma, accurate identification and diagnosis are crucial.<sup>15</sup> The malignant blue nevus is an extremely rare form of melanoma in that the main precursor is the cellular subtype. Notwithstanding the controversy about its origin, some studies report that when it originates from a 'common' blue nevus, the cellular subtype is often transformed.<sup>4,10</sup>

The definitive treatment of oral melanoma of the palate is palatotomy or maxillectomy. The clinical correlation associated with the histopathological diagnosis is essential in order to avoid unnecessarily aggressive procedures, since the common blue nevus runs a benign course and only removal of the lesion is indicated,<sup>7,14</sup> as was done in this case.

In the case described above the patient was followed up over two years after surgery, with no signs of recurrence or malignant transformation. The observation that the patient was a heavy smoker, the large size of the lesion and the warnings in the literature about the potential for malignant transformation, were important factors underscoring our decision to undertake surgical treatment. □

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