

Case Report**BLUE NEVUS: A CASE REPORT**LOZACHMEUR Camille^{1,*}, BOURHIS Amélie², FALGUIERE Arthur¹, BOISRAME Sylvie¹¹Department of Dentistry, Oral Medicine and Oral Surgery, Sense Organ Unit, University Hospital Morvan, Brest, France²Department of Anatomy and Pathology, University Hospital Morvan, Brest, France**Abstract**

Oral melanocytic nevi are rare and benign tumors arising from melanocytes located in the oral mucosa. These lesions are clinically asymptomatic and often discovered during a dental consultation. All melanocytic proliferations are not benign and an histological examination have to be performed to rule out malignancy and adjust the treatment and the follow-up. Herein, we presented the case of a 23-year-old- Asian woman with an asymptomatic pigmented lesion on the hard palate discovered during a dental consultation. The lesion was excised and sent to the anatomopathological laboratory and turned out to be a blue nevus, a particular subtype of nevus.

Key Words: Oral melanocytic nevi, mouth diseases, blue nevus, Malignant transformation, Excision, Follow-up

INTRODUCTION

Oral melanocytic nevi (OMNs) are rare and benign melanocytic tumors [2, 5, 7, 11]. Long associated with hamartomas, melanocytic nevi are now considered as neoplasms [7].

OMNs can be congenital or acquired. They are classified histologically as junctional, compound, intradermal, combined and blue. This classification is correlated with the location, morphology of the cells and their distribution [2,7]. By definition, melanocytes are located in the basal layer of the epithelium, originating from the neural crest and are present in the skin and mucous membranes [2, 9, 11]. They are very often embedded between the basal keratinocytes. Therefore, they can be present in all areas of the oral cavity.

OMNs are poorly studied; Buchner et al. reported the weak frequency, about 0,83%, of solitary oral melanocytic lesions in 89,430 cases accessed during a 19-year period at the Pacific Oral and Maxillofacial Pathology Laboratory [4, 5, 11]. It also shows that the most frequent lesions are melanotic macules (665 cases) and that oral malignant melanoma is the least frequent lesion (5 cases). Another study also reported an annual incidence of excised OMN of 4.35 cases per 10 million inhabitants in the Netherlands [5, 11].

Thus, the most common OMN was intramucosal nevus, followed by blue nevus (19-36%) [4, 7, 10, 11].

***Corresponding Author:** LOZACHMEUR Camille*, Department of Dentistry, Oral Medicine and Oral Surgery, Sense Organ Unit, University Hospital Morvan, Brest, France

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Women tend to be more commonly affected than men with a higher frequency between the 3rd and the 5th decade of life [2, 5, 6, 10, 11].

There is no ethnic predominance [5].

The preferential location of the blue nevus is the hard palate (69%) followed by the labial mucosa and vermilion border of the lip [1, 4, 8, 10, 11]. Most lesions are asymptomatic and are discovered incidentally during dental examination.

This following case presents a 23-year-old- Asian woman presented with an asymptomatic pigmented lesion located on the hard palate discovered during a dental consultation. The lesion was completely excised with the histological diagnosis of “blue nevus”. This case report presents a rare lesion of the oral cavity.

CASE REPORT

A 23-year-old Asian woman was referred to our Oral Medicine service following an incidental, asymptomatic discovery of a dark spot on the hard palate.

Anamnesis revealed no medical, surgical or allergy history. The patient was not taking any treatment. She was a non-smoker and non-alcoholic drinker. No similar lesions were detected in the family environment.

Exobuccal examination was unremarkable with no cervical lymphadenopathy alterations (Fig.1). Endo-buccal examination showed an irregular, slightly elevated, nonulcerated, asymptomatic blackened surface lesion located on the hard palate measuring 5mm in diameter

**Figure 1: Blue Nevus**

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with an unknown evolution time.

A diagnostic excision was performed to rule out a potential melanoma. The excision was performed under local anesthesia with lidocaine and epinephrine (1:100 000) and a number 15 scalpel blade. The lesion was excised with a 2mm margin.

Following the excision, post-operative advice and a prescription for painkillers were given to the patient.

The specimen was sent to Histology and Pathology Laboratory for analysis.

Macroscopically, it was a non-oriented skin flap of 4*7*2mm with a raised, heterochromatic lesion of 4*6mm. Histologically, it was a melanocytic proliferation located in the chorion, small, well limited, symmetrical, ovoid in shape and parallel to the surface. Cells were regular, containing a variable amount of melanin, without atypia or mitosis and were isolated or organized in small fascicles. The stromal collagen showed a sclerotic appearance around the lesion. (Fig.2)

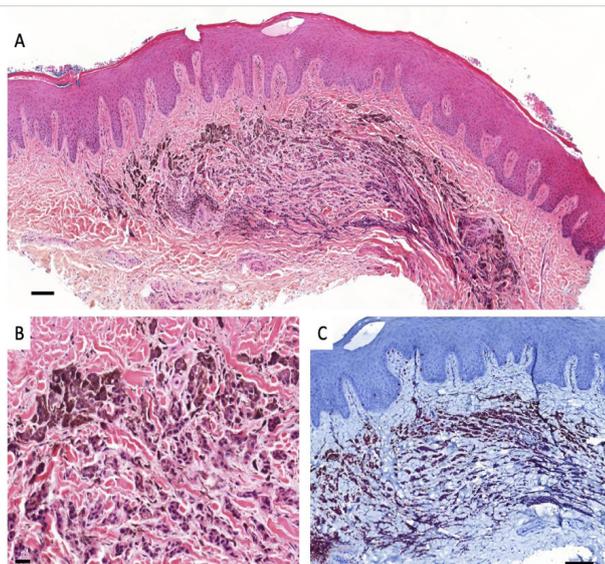


Figure 2: Histopathology.
A: Low magnification view of the HES staining of the blue naevus presented as a well-circumscribed chorionic lesion composed of dendritic melanocytes and sclerotic stromal collagen (scale bar 200µm).
B: High power view of the same lesion, Melanocytes were isolated or grouped in small fascicles. They had a spindle shape and contained a variable amount of melanin which give them a brown color (scale bar 20µm).
C: melanocytes were strongly stained red in immunohistochemistry with SOX 10 antigen (monoclonal- SP 267 - ROCHE - 760 4968 - pre-diluted) (scale bar 100µm).



Figure 3: Follow-up

The epithelium was unremarkable without inflammatory or neoplastic features. Immunohistochemical examination showed a positive staining for SOX10 (monoclonal- SP 267 - ROCHE - 760 4968 - pre diluted - CC1 standard) confirming the melanocytic origin and a negative staining for PRAME (Prame-clone QR005- rabbit monoclonal antibody-Quartett) favoring a benign lesion rather than a melanoma. These aspects made the diagnosis of blue nevus and the excision was complete.

Upon follow-up, the patient remained asymptomatic, and the surgical wound was fully healed with no signs of clinical recurrence. (Fig.3)

DISCUSSION

Oral melanocytic nevi (OMNs), like blue nevus, are relatively rare, pigmented lesion in the oral cavity. Moreover, due to its asymptomatic nature, it is frequently discovered incidentally during a dental examination.

Clinically, oral melanocytic nevi (OMNs) are small, well-circumscribed, circular, flat or slightly elevated. The color is variable: it can be brown, blue or sometimes black [1, 5, 8].

The blue nevus questions the clinicians about the malignant character. Thus, the ABCDE rule, first introduced in 1985 in the form ABCD [12, 13], then extended in 2004 to the ABCDE rule [13, 14], allows them to be guided by the different forms of melanoma, including Asymmetry, Border irregularity, Color Variation, Diameter greater than 6mm and Evolving.

Malignant transformation or recurrence of blue nevus, though extremely rare, can be, in fact, develop [2, 11].

Indeed, oral nevi and oral malignant melanomas have several similarities. Both conditions occur most often on the palate; although rare with an incidence of 0,07%, oral malignant melanoma occurs 80% on the palate [17]. So, it remains insufficient to make the diagnosis with clinical impression and most pigmented lesions are excised or biopsied [2, 8, 11].

In this case, the pigmentary lesion was located on the palate and the ABCD criteria were present, raising the suspicion of an oral malignant melanoma.

Excision and pathological examination are, therefore, mandatory to obtain the diagnosis but also, to rule out any differential diagnosis including oral malignant melanoma. The histologic examination of HES staining is usually sufficient to make the diagnosis of the different melanocytic tumors: nevi, particular subtypes of nevi like blue nevi or melanoma. However, in some complicated cases immunohistochemistry can be useful to confirm the melanocytic origin or to help to distinguish malignant from benign lesions.

Melanocytic markers Melan A (MART1), Homatropine Methyl Bromide-45 (HMB-45), SOX10 and Microphthalmia associated Transcription Factor (MITF) confirmed the melanocytic origin. Fatty acid synthase or preferentially expressed antigen in melanoma (PRAME) has been identified as a useful marker to differentiate oral melanoma from OMN [7, 15, 16].

All these techniques are particularly useful to confirm the diagnosis of blue nevus and rule out a melanoma.

It turns out that OMNs have been suspected of being a precursor to oral malignant melanoma. However, no cases of dysplasia's OMNs have been described [1]. Moreover, the potential for malignant trans-

formation of OMNs has never been determined [1, 5, 8].

CONCLUSION

The following clinical case reported a pigmented lesion on the hard palate on a young Asian woman. An excision was performed to rule out malignancy. The positive diagnosis revealed a blue nevus.

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