



Blue Nevus of the Hard Palate in a 12-Year Old Male Patient: A Case Report with Review of the Literature

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Abstract

Background and Overview: Nevi are either congenital or developmental pigmented malformations rarely found in the oral cavity. Approximately 30% of reported cases in this anatomical region are of the blue type, a histological variant with the potential for malignant transformation. There is no consensus in the literature regarding biopsy of pigmented lesions with malignant potential. In children, the intraoral blue nevus is rare. This case report describes the clinical features of a blue nevus in a 12-year old Asian male on the right hard palate of the maxilla. A differential diagnosis, rationale for excisional biopsy of this oral lesion and review of the literature are presented. The goal of this case report is to create awareness that such a rare pigmented lesion in the oral cavity can also occur in children.

Case Description: This case report describes the clinical presentation of a 12-year old Asian male who presented with a dark blue-black pigmented lesion on the right hard palate of the maxilla. Because of the fear that the pigmented lesion could represent a malignancy, excisional biopsy for histopathologic diagnosis was performed.

Practical Implications: Although blue nevi are considered rare in the oral cavity, especially in children it is important to differentiate these lesions from the malignant blue nevus or melanoma. To obtain a definitive diagnosis of any suspicious pigmented lesion, excisional biopsy is recommended due to the potential for malignant transformation.

Keywords: Blue nevus; Rare; Child; Differential diagnosis; Biopsy

Introduction

Nevi are congenital or developmental pigmented malformations commonly located on the skin. Depending on the location of the nevus cells, the malformation is classified histologically as intradermal, junctional, compound and blue nevi [1]. The blue nevus is a neoplasm that is composed of pigmented dendritic dermal melanocytic cells in the reticular dermis with the potential for malignant transformation [2]. A blue nevus can develop anywhere on the body, however, approximately 50% of the common blue nevi are usually located on the dorsal surface of the hands and feet, scalp, and buttocks [1,2]. A blue nevus in the oral cavity is considered a rare lesion with important differential diagnoses [3]. Although considered a benign lesion, it may have the potential for malignant transformation in rare cases [4].

In 1959 Scofield described the first two cases of blue nevi which were both located on the hard palate [5]. Since the first two reports, approximately 70 additional cases have been reported [5,6]. Two histological variants were described in these reports: a common blue nevus and a cellular blue nevus [7]. Intraoral blue nevi present as asymptomatic, slightly raised well-circumscribed lesions of variable color, such as gray, blue-black and brown [7-9]. The most common location of the intraoral blue nevus is the hard palate and is just over 5.0 mm in diameter [10]. The present case report describes a blue nevus on the hard palate of a 12-year old Asian male and discusses the differential diagnosis, rationale for excisional biopsy of this pigmented lesion and a review of the literature.

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Figure 1:

Case Presentation

A healthy 12-year old Asian male was referred to the office of one of the authors (CYSL) for evaluation of a blue-black colored pigmented lesion on the right hard palate of the maxilla. The parents stated that the lesion was noticed about two years ago and may be increasing in size. The family was concerned that the pigmented lesion could be a malignant growth. Dental history revealed teeth letters B, I, J, K, L restored with amalgam from 2010 to 2014. Oral examination revealed a complete adult restoration-free dentition, except the third molars that were not present. Examination of the right hard palate of the maxilla revealed a blue-black elliptical shaped macule with distinct margins (Figure 1). With digital pressure, blanching of the lesion was not observed. In addition, no vascular bruit or thrill was appreciated. To obtain a definitive diagnosis, excisional biopsy was completed.

Differential diagnosis

The differential diagnosis of intraoral nevi includes amalgam tattoo, melanotic macule, melanocytic nevi, vascular anomaly and melanoma [11]. Of the five possibilities, the amalgam tattoo is the most difficult to differentiate from a blue nevus and is one of the most common causes of intraoral pigmentation [12]. It presents clinically as a localized flat, blue-gray lesion of variable size and is the result of localized implantation of dental amalgam that appears as a blue, gray or black macule. Blue nevi were most commonly clinically diagnosed as amalgam tattoos. These findings are in agreement with previous studies [8]. No treatment is indicated for removal of the amalgam tattoo, unless a melanocytic neoplasm cannot be excluded. In our patient, the entire dentition was without any restorations, but amalgam tattoo could not be excluded from the differential diagnosis because of the past dental history of primary teeth restored with amalgam.

The melanotic macule is a pigmented oral lesion of unknown etiology that is more commonly observed than the blue nevus [6]. It occurs at any age, but is seen normally in adults with a female predilection. Although these are innocuous lesions, a biopsy is usually warranted for diagnosis because mucosal melanoma can mimic the appearance of a melanotic macule [12]. The classic features include a round, well demarcated, smooth macule that is usually brown in color but may appear blue or black. The vermilion border of the lower lip is the favored site of occurrence, followed by the buccal mucosa, gingiva and palate. Like the amalgam tattoo, no treatment is indicated unless the melanotic macule becomes an aesthetic or malignancy is a concern.

Small vascular anomalies of the oral cavity, such as the hemangioma or varices may look similar to the blue nevus. A hemangioma is a common congenital vascular lesion that presents

as a blue-purple colored fluctuant nodule [13]. Blanching with digital pressure is a characteristic feature and can be consistently used to rule-out a vascular lesion. Another vascular anomaly to rule-out is the varix that has a nodular appearance that is blue-purple in color [14]. With digital pressure, like the hemangioma, it will blanch with digital pressure. The lip, tongue and floor of the mouth are common sites for this vascular anomaly. Varices are not commonly observed in children.

Pigmented nevi are rarely the etiology for focal oral pigmentation [8]. They often present as either brown or blue lesions. Histologically, nevus cells are observed in the basal epithelial layers, the connective tissue or both. Therefore, they are classified as junctional, intradermal, intramucosal and compound nevi [1,2]. Of the different types of pigmented nevi, blue nevi are characterized by a proliferation of dermal melanocytes deep within the lamina propria, which accounts for the surface blue color [2,3,7-14]. It may be difficult to clinically diagnose a nevus from a mucosal melanoma. This is especially if the lesion is on the palate, as this is the most common site for both lesions [15]. Although transformation of oral pigmented nevi to melanoma is questionable, nevi may represent a precursor to oral mucosal melanoma [15]. Excisional biopsy is therefore recommended for histopathologic examination.

The anatomic location of oral melanoma is most frequently observed on the hard palate and maxillary gingiva. Malignant melanoma develops from malignant melanocytes [16]. They exhibit a very poor prognosis when discovered within the oral cavity [17]. Melanoma usually occurs over the age of 50 years and the incidence is greater in males. Oral melanoma can occur in any racial and ethnic group, but the highest incidence is observed in Japanese patients [18]. Therefore, melanoma should always be considered in the differential diagnosis as it cannot be clinically distinguished from a melanotic nevus [19].

In our patient, histopathology revealed pigmented spindle-shaped cells with branching dendritic extensions deep in the lamina propria (Figure 2). High power view revealed slender and elongated melanocytes aligned parallel to the surface epithelium (Figure 3). Based on the histopathologic findings, the diagnosis was negative for a malignancy and consistent with a common blue nevus of the hard palate.

Discussion

The blue nevus is an asymptomatic, benign melanocytic lesion first described by Tièche in 1906 as small sharply defined blue to blue-black spots commonly observed on the face and extremities [1,2]. It is the second most common type of nevus of the oral cavity, accounting for 19-36% of all cases [10,20]. The blue color of the nevus is due to

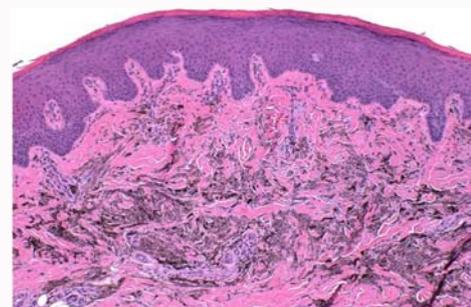


Figure 2:

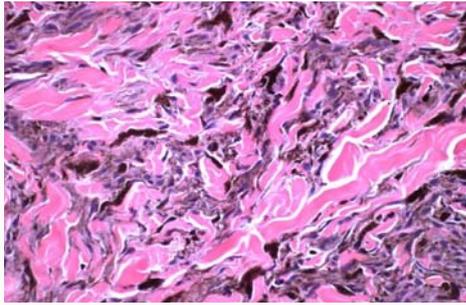


Figure 3:

the presence of melanin deep within the dermal melanocytes and the Tyndall effect [1,2]. The variation in color is related to the depth of the melanocytic cells in the dermis, the amount of melanin present, and the presence or absence of melanin in the cells of the overlying epidermis.

In the oral cavity, blue nevi are rare lesions with a prevalence of 0.1% in the general population [1,7,8,16]. The predominant intraoral site is the hard palate of the maxilla. Intraoral lesions have a predilection for females in the third to fourth decades of life. In a literature review by Fistarol et al. [21] from 1959 to 2005, 64 reports of blue nevus were identified in the oral cavity. Six patients under the age of 18 had biopsy proven blue nevi located on the hard palate.

Although blue nevi are considered rare in the oral cavity it is important to differentiate these lesions from the malignant blue nevus or melanoma [3,4,22-29]. Malignant transformation of blue nevi has been reported in children in the head and neck region, but no intraoral case reports have been reported [28,29]. To obtain a definitive diagnosis of any suspicious pigmented lesion, excisional biopsy is recommended due to the potential for malignant transformation [19,24-29].

Conclusion

Accurate diagnosis of all pigmented lesions of the oral cavity is challenging and the decision to biopsy the lesion is not without controversy. Histopathological evaluation of oral pigmented lesions is required for a definitive diagnosis. We are of the opinion that biopsy of any pigmented lesion will allow the clinician to obtain a definitive diagnosis that will allow timely surgical management, if indicated.

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