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A Problematic Oral Pigmentation: Deep Penetrating Nevus

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Abstract: This case report is an interesting pigmented lesion presenting as a melanoma like appearance, with synchronous occurrence of calcifying hyperplastic dental follicle. A 13-year-old boy was referred to Oral Medicine Department of Mashhad Dentistry School complaining of a dark growing pigmentation lasting from two years ago. Recently rapid growth and pain was emerged. Plaques like, gray-black pigmentation (1.2×1 cm diameter) on maxillary gingiva overlying semi-erupted right permanent canine with firm consistency and non-homogenous color and irregular margins was observed in clinical examination. Periapical radiography showed no abnormal finding. Oral malignant melanoma was notified as the first diagnosis and biopsy was performed. On histopathologic examination a nevus like lesion penetrating to deep portions (e.g., reduced enamel epithelium of semi-erupted canine) beside a calcifying component (type A and B calcifications) was seen. A diagnosis of synchronous occurrence of deep penetrating nevus and single calcifying hyperplastic dental follicle was established.

Key words: Deep penetrating nevus, oral pigmentation, case report, melanoma, calcifying, dental follicle

INTRODUCTION

There are much different oral pigmentations with different demographic and clinical characteristics, helpful in differential diagnosis. A spectrum exists from simply oral physiologic pigmentation, oral melanotic maculae and oral melanocytic nevus to Oral Malignant Melanoma (OMM) (Meleti *et al.*, 2008).

OMM is probably the most ominous malignancy of oral cavity with a 5-year survival of 15% that can arise denovo from normal mucosa. In about 30% of cases OMM arises from previous oral pigmentation, lasting from months to years, although the accuracy of this finding has been doubted recently. The most common oral sites for OMM are palate and maxillary gingivae. The age of reported patients ranges from 20 to 80 years with a reported male predilection (Meleti *et al.*, 2007b). When slightest doubt exists about OMM to be considered in differential diagnosis of any oral pigmentation, a biopsy must be performed (Meleti *et al.*, 2008).

Here we report a problematic oral pigmentation with unusual clinical and Histopathological features. The clinical pattern of this lesion led us to consider OMM as the first diagnosis. Further examination revealed Deep

Penetrating Nevus (DPN) of the gingivae. Although there are many cases of dermal DPN (Flauta *et al.*, 2006; Edwards and Blessing, 2000) with a great diagnostic challenge between DPN and OMM, there is not any case report about intraoral DPN. Only one report exists in the field of oral medicine about DPN originating from cheek mucosa (Cheng *et al.*, 2007).

CASE REPORT

A 13-year-old boy was referred to Oral Medicine Department of Mashhad Dentistry School at the Mashhad University of Medical Sciences in June 2008. This case complained of an exophytic black-gray lesion on right maxillary gingiva, with a history of gradual growth pattern from two years ago. Pain was a recent complaint from two months ago coinciding with a sudden increase in size of the lesion due to parents report. There was no abnormal finding (e.g., cutaneous pigmentation) in extra oral sites. Medical and social histories were not contributory. On clinical examination there was a plaque like lesion with a 1.2×1 cm diameter on maxillary gingivae and alveolar mucosa overlying semi-erupted right permanent canine. On palpation the lesion was firm. There was a gray-black

pigmentation overlying this nodule with non homogenous color and irregular margins.(Fig. 1). Some satellite pigmentations were observed around the main pigmented lesion. The cusp of the right canine was exposed in the oral cavity but eruption was not complete. In comparison to left canine there was a 10 months delay in eruption. The patient reported pain especially on palpation. Periapical radiography showed no abnormal finding (Fig. 2).

Although the patient was teenager and the pigmentation had two year duration but some features was on the side of melanoma as the first diagnosis. Due to sudden growth pattern (in a two months period) non homogenous color, irregular margins, satellite pigmentations, unusual texture and recent pain, the possibility of an atipic presentation of pigmented



Fig. 1: Unusuall clinical appearance of the lesion; look at the satellite pigmentation, un-uniform color and size (>1 cm) of the lesion



Fig. 2: Periapical radiograph is normal with no evidence of calcification

malignancy (e.g., Oral Malignant Melanoma (OMM)) was expressed. Other solitary pigmented lesions were considered in differential diagnosis (DDx). So the Ddx was ranked as follows: (1) OMM, (2) Plaque type blue nevus, (3) Oral melanotic maculae, (4) Melanoacanthoma, (5) Amalgam tattoo and (6) Nevus of Ota. Except for OMM, all mentioned lesions need no treatment.

OMM was the most likely diagnosis in present case: (1) there was a history of recent growth in two months before referral. (2) The clinical feature was consistent with OMM: Un-uniform color, ill-defined margins, satellite pigmentations, site of lesion, size (>1 cm) (Aguas *et al.*, 2009). (3) Presence of pain is not an usual complaint in benign oral pigmentations and it is considered as one of rare symptoms of OMM (Meleti *et al.*, 2007a; Aguas *et al.*, 2009). Pain was the main complaint of the patient, which resulted in seeking consultation. (4) Delayed eruption of canine tooth: Although it is not mentioned in the literature but logically presence of unexplained proliferating component can result in delayed eruption.

Plaque-type Blue Nevus (PBN) was the second probability. Although PBN is a rare intra-oral pigmentation, because of multiple adjacent pigmentations (satellite pigmentation) and large size (more than 1 cm) is a better diagnosis than intra-oral blue nevus. PBN is usually congenital but some times it appears later in life. In histopathologic examination it has characteristics very similar to intra-oral blue nevus but hyper cellularity is encountered (Fistarol and Itin, 2007).

Oral melanotic maculae is a common lesion caused by increased production and deposition of melanin within the basal layer, lamina propria and both. Lower lip and anterior maxillary gingivae are the most common intra-oral sites. It is more frequent among females. Clinically a single well circumscribed blue or brown-to-black homogenous pigmentation less than 1 cm diameter is observed (Meleti *et al.*, 2008).

Melanoacanthoma is a reactive lesion with mixed origin: Both keratinocytes and melanocytes are responsible for this lesion. It almost exclusively occurs in young black and has a rapid course. Buccal mucosa is the main intra-oral location. Spontaneous regression, specially after incision biopsy is common (Meleti *et al.*, 2008).

Amalgam tattoo is usually seen when a history of amalgam filling exists. A traumatic event can push amalgam into muco-gingival tissues. Our patient had not any dental filling or aggressive dental procedure (Meleti *et al.*, 2008) in addition no opacity was observed in preiapical radiograph. No history

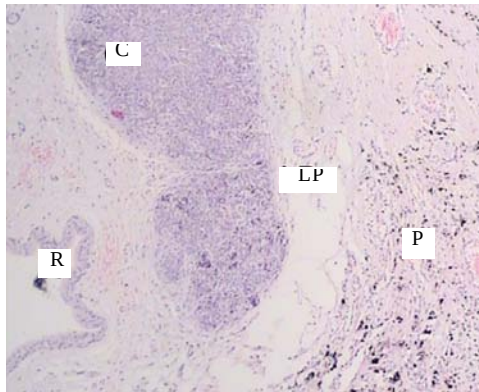


Fig. 3: Two parts can be considered for the lesion: first part from gingiva (offside the right part of this view) to Lamina Propria (LP) which Pigmented Component (P) is lied within this part and second part from LP to Reduced enamel epithelium (R), Calcifying component (C) is within this part and (Hematoxiline and Eosin, original magnification x100)

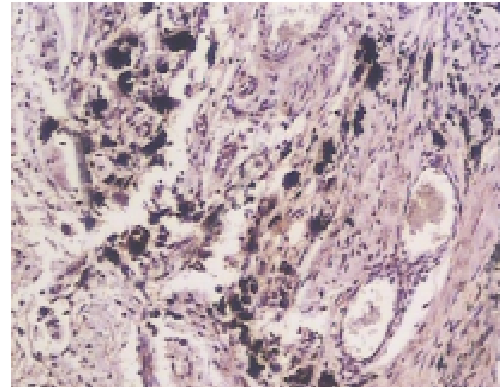


Fig. 4: Melanocytes and Melanophages (Hematoxiline and Eosin, original magnification x400)

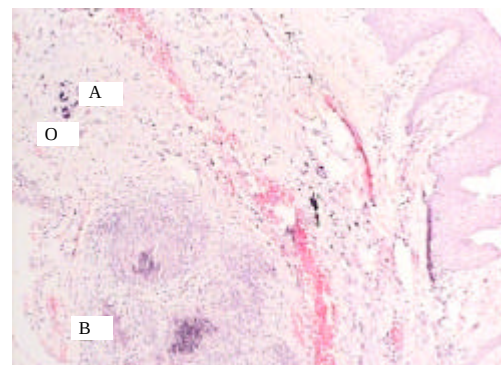


Fig. 5: Calcifying hyperplastic dental follicle with type (A) and type (B) calcification and odontogenic epithelium (O) (Hematoxiline and Eosin, original magnification x400)

of graphite embedding was revealed. History of rapid growth and pain set these entities low in DDx.

Nevus of Ota can be discovered at birth or later in life. Many patients are female Asians. Clinically a macular, patchy brown, slate blue or blue-black pigmentation confined to 1st and 2nd branches of trigeminal nerve, affecting large parts of oral mucosa is apparent (Fistarol and Itin, 2007).

Due to all above the most appropriate differential diagnoses were OMM and PBN. A biopsy was performed in order to exclude OMM. During biopsy, a similar pigmentation was observed on the bone surface of alveolar bone overlying the tooth crown. Unfortunately, this part was shaved by the periodontist until fading this color and no specimen was sent from this site. The crown was exposed and the entire pigmented soft tissue lesion with a 1.5 mm margin was sent for histopathologic evaluation.

In low magnification the lesion consisted of two separate parts. First part (superficial part) was considered from superficial oral mucosa (alveolar mucosa and gingivae) to deep portion of Lamina Propria (LP) and second part (deep part) was considered from LP to reduced enamel epithelium of semi-erupted tooth (Fig. 3).

In histopathologic examination of the superficial part (first part), there was a parakeratinized stratified squamous epithelium on the surface with short rete pegs extending to LP. Greatly elongated, slender, slightly wavy melanocytes with long branching

dendritic processes lied grouped in irregular bound lesion in the LP. A large plenty of macrophages were interspersed within LP with abundant melanin granules. These cells were fulfilled with melanin granules to the extent that their nuclei were obscured (Fig. 4). Elongated melanocytes were oriented parallel to the epithelium. There was no mitotic figure but some degree of pleomorphism was observed. Nuclear vacuoles and pseudo-inclusions were not observed. Under this nevus like lesion, in the deep section, a mass of spindle type cells with droplet calcifications (Type A) and dentinoid calcifications (Type B) were seen (Santiago *et al.*, 1998) (Fig. 5). These calcifications were interspersed within thickened remnants of dental follicles of semi-erupted tooth. An interesting finding was presence of melanocytes adjacent to reduced enamel epithelium of semi-erupted tooth. Fontana and

DISCUSSION

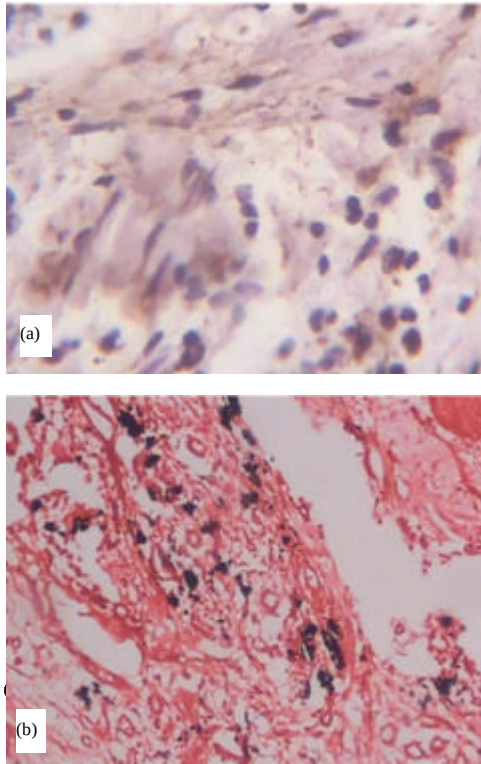


Fig. 6: (a) S-100 positive nevus cells (S-100 staining, original magnification x400), (b) Melanin and melanophages (Fontana staining, original magnification x400)

S100 staining were both positive and HBM45 was negative (Fig. 6a, b). The calcified component was simply a single calcifying hyperplastic dental follicle hamartoma (Santiago *et al.*, 1998).

Synchronous occurrence of an atypical pigmented lesion with a calcified lesion in a semi-erupted tooth developed a significant diagnostic challenge for this case. Due to deep penetration of melanocytes toward reduced enamel epithelium of tooth, the most appropriate diagnosis for the melanotic component was Deep Penetrating Nevus (DPN).

It seems that it is the first time to report such a synchronous findings in oral cavity. It is also the first intraoral DPN.

Since excisional biopsy was performed with a safe margin and there was no defect to reconstruct by free gingival graft, no treatment was necessary. Patient is under close follow up and no recurrence has been occurred after two years (from June 2008 until June 2010).

There are much different oral pigmentations with different demographic and clinical characteristics, helpful in differential diagnosis. A spectrum exists from simply oral physiologic pigmentation, oral melanotic maculae and oral melanocytic nevus to Oral Malignant Melanoma (OMM). Here, we report a problematic oral pigmentation with unusual clinical and Histopathological features.

Diagnosis of pigmented lesions of the oral cavity and peri oral tissues is challenging (Meleti *et al.*, 2008). Rapidly growing and symptomatic pigmentations should always be biopsied to rule out OMM (Gonzalez-Garcia *et al.*, 2005). Although exclusion of melanoma is sometimes difficult in case of DPN. There are several differential diagnoses for present case: (1) OMM, (2) Plaque-type blue nevus, (3) Oral melanotic maculae, (4) Melanoacanthoma, (5) Amalgam tattoo and (6) Nevus of Ota. In this case, there was a one-year history of appearance in oral cavity but the patient complained of rapid growth and pain since two month ago. The lesion was observed in maxillary alveolar mucosa and marginal gingiva of a semi-erupted canine tooth, where the main differential diagnosis is between Amalgam tattoo and melanoma (Gonzalez-Garcia *et al.*, 2005; Meleti *et al.*, 2008).

The growing nature, anterior position with no history of amalgam filling and absence of opacity in periapical radiographs ruled out the amalgam tattoo in clinical diagnosis and in histopathologic examination, Fontana staining confirmed the melanin origin of pigmentation. Graphit tattoo was also excluded due to this finding (Meleti *et al.*, 2008).

Malignant Melanoma was clinically suspected due to various observations mentioned in differential diagnosis section.

In histopathologic examination there was no malignant proliferation of melanocytic cells. No mitotic figure or hyperchromatism was observed and a little pleomorphism was found. Positive S100 confirmed the neural crest origin of melanocytic cells but negative HMB45 shows that melanocytes did not express all melanogenic activities (Gonzalez-Garcia *et al.*, 2005; Meleti *et al.*, 2008).

Blue nevus may seem an appropriate diagnosis. There is a proliferation of dendritic melanocytes, with an increased fibrous tissue and melanophage accumulation, with no junctional activity. But in blue nevus the cells are confined to dermis and penetration to deep structures (e.g., gingival sulcus, beyond reduced enamel epithelium) is not usual (Cheng *et al.*, 2007).

Nevus of Ota is an uncommon oral pigmentation, which is described as irregularly shaped flat macule with blue, brown, black color. This nevus usually is near the midline of palate but isolated manifestation of this entity without involvement of the skin has not been described from the histopathologic point of view, this lesion has lesser concentration of dendritic melanocytes and melanophages are uncommon (Fistarol and Itin, 2007).

Considering all clinical and histopathologic findings DPN seems the best diagnosis for this case. DPN is essentially a cutaneous pigmented lesion, first reported by Flauta *et al.* (2006) and Cheng *et al.* (2007) as a lesion that clinically and histopathologically can mimic thick melanoma (Cheng *et al.*, 2007).

DPN is characterized by a pigmented plaque less than 1 cm in head and neck and upper extremities of young individuals (Cheng *et al.*, 2007).

Histopathologically DPN penetrate deeply into reticular dermis, dermal appendage structures, vessels and muscles (Edwards and Blessing, 2000) nerves and satellite islands of nevus cells may be found. Cells of DPN have nuclear pleomorphism but mitoses are rare (Edwards and Blessing, 2000).

Although some authors consider DPN as a variant of blue nevus, Plexiform Spindle Cell Nevus (PSCN) or clonal nevus. The deep penetrating nature of DPN is not a feature in these entities. Cooper also suggested that DPN is a variant of combined nevus (Cheng *et al.*, 2007). In present case no junctional component was seen. Cheng *et al.* (2007) postulated that frequency of DPN is under estimated because many nevi are removed incompletely by shave biopsy, without sampling of the deeper dermal tissues. Considering characteristics of dermal DPN, present results are in agreement with previous findings about DPN of the skin.

If we consider this lesion as oral compartment of DPN, this is the first report of DPN in oral cavity. Synchronous occurrence of a pigmented lesion with a hyperplastic calcifying dental follicle is not mentioned before in the literature.

CONCLUSION

Here, we report an unusual oral pigmentation with features similar to oral malignant melanoma. Histopathologic examination revealed this lesion to be a

Deep Penetrating Nevus (DPN) of the oral cavity with synchronous occurrence of calcifying hyperplastic dental follicle. It is the first report of *intraoral mucosal* DPN.

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