Head and Neck Pathol (2008) 2:289–292
DOI 10.1007/s12105-008-0092-z

MEETING REPORT

14th International Congress, International Association of Oral Pathologists and 62nd Annual Meeting, American Academy of Oral & Maxillofacial Pathology, CPC Case 4

Yoichi Tanaka · Julia Yu Fong Chang

Received: 2 September 2008 / Accepted: 13 October 2008 / Published online: 9 November 2008

Clinical Presentation

A 53-year-old female presented with a history of a black pigmented lesion in the left buccal mucosa associated with mild pain, first noticed 18 months previously. The patient had previously been prescribed an ointment for pain by her dentist that had brought about pain relief, however the pigmentation persisted. The patient, therefore, sought another opinion.

Clinically, a few black spots were observed in the buccal mucosa with some redness around the lesion, however, there was no swelling or ulceration (Fig. 1). There were numerous yellow spots around the black spots. The oral surgeon performed a wide local excision.

Differential Diagnosis

Oral mucosa is normally pink, although the color varies depending on the location, function and underlying tissue under both physiologic and pathologic conditions [1, 2]. The final color of the oral mucosa is determined by the accumulated materials on the epithelial surface, thickness of the keratin layer or epithelium, numbers and melanogenic activity of the melanocytes, the vasculature, and the composition of the submucosal tissues. Discoloration of the oral mucosa can be commonly seen and is known to be associated with a variety of endogenous and exogenous etiologic factors [2]. Pigmented lesions of the oral cavity include a group of different entities, including melanocytic lesions, lesions due to foreign material deposition, vascular lesions, and salivary gland lesions [2, 3]. In addition to the complexity of multiple etiologic factors, various stimuli can evoke the same tissue response and different conditions might share the same histopathologic features. The color and shape of the lesions in the same disease varies according to the location and duration of the lesions [4]. For these reasons, the differential diagnosis of pigmented lesions in the skin and mucosa is sometimes difficult. A thorough examination of oral and extraoral tissues, especially the skin, an evaluation of both local and systemic symptoms and signs, and a complete medical, dental, drug, and oral habit (such as smoking or betel nut chewing habit) history are required for making a definitive diagnosis of pigmented lesions [2, 5]. In general, the information on the number, distribution, size, shape, color, consistency, and texture of the lesions will guide the differential diagnosis in the right direction. Moreover, some clinical tests or examinations like diascopy, radiography, laboratory investigations (blood tests), and biopsy of the lesion might be useful in making the differential diagnosis. Combining all the information accumulated until now [1–3, 6], we present a flow-chart as a guide to the differential diagnosis of a pigmented lesions (Fig. 2).

This adult female patient presented with an 18-month-history of a localized, flat pigmented lesion with an irregular shape and various colors ranging from brown to black. The mucosa showed some redness and the patient felt mild

Y. Tanaka (✉)
Division of Surgical Pathology, Clinical Laboratory, Tokyo Dental College, Ichikawa General Hospital, Chiba, Japan
e-mail: tanaka@tdc.ac.jp

J. Y. F. Chang
School of Dentistry, National Taiwan University College of Medicine, Taipei, Taiwan

J. Y. F. Chang
Division of Oral Pathology and Diagnosis, Department of Dentistry, National Taiwan University Hospital, Taipei, Taiwan

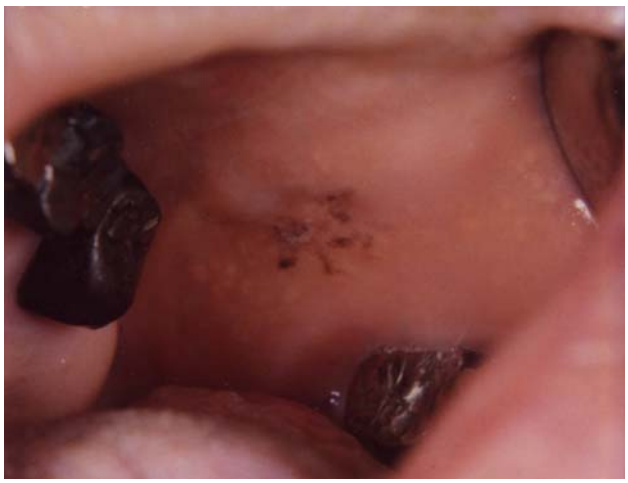

Fig. 1 Clinical presentation

soreness which resolved following topical treatment. The redness, painful sensation, and the response to treatment indicated an inflammatory process, either primary or secondary to trauma or irritation. Discolored prostheses on the left maxillary teeth were noted opposite to the left buccal mucosa. The possibility of a foreign body tattoo from the discolored prostheses was considered, especially if there was a history of trauma. Careful examination revealed a few white patches admixed with the pigmented lesion in this patient. A mucocutaneous disease, such as lichen planus with post-inflammatory pigmentation was considered as a reasonable differential diagnosis. This patient was not taking any medications and did not have any endocrine or other systemic diseases. According to the clinical findings, extraoral tissue examination, and the medical, dental, oral habit, and medication histories, the possibilities of a salivary gland or hematologic/vascular lesion, non-melanotic systemic diseases, early-onset pigmentations or syndromes, or pigmentations due to systemic diseases (especially endocrine disorders) were excluded. Finally, the most possible differential diagnoses of this localized, flat, pigmented lesion included a foreign-material tattoo from the discolored prostheses with or without occlusal trauma (post-inflammatory pigmentation), a chronic inflammatory condition, for example, lichen planus with post-inflammatory pigmentation, or a melanoacanthoma induced by a local irritation.

Diagnosis and Discussion

Histopathologic observation showed the lesion to be surfaced by a thin parakeratotic epithelium with a proliferation of basaloid tumor cells arranged in small and large nests without any extension into the underlying adipose tissue (Fig. 3). The peripheral basal cells showed palisading of the nuclei (Fig. 4). The tumor islands were composed of

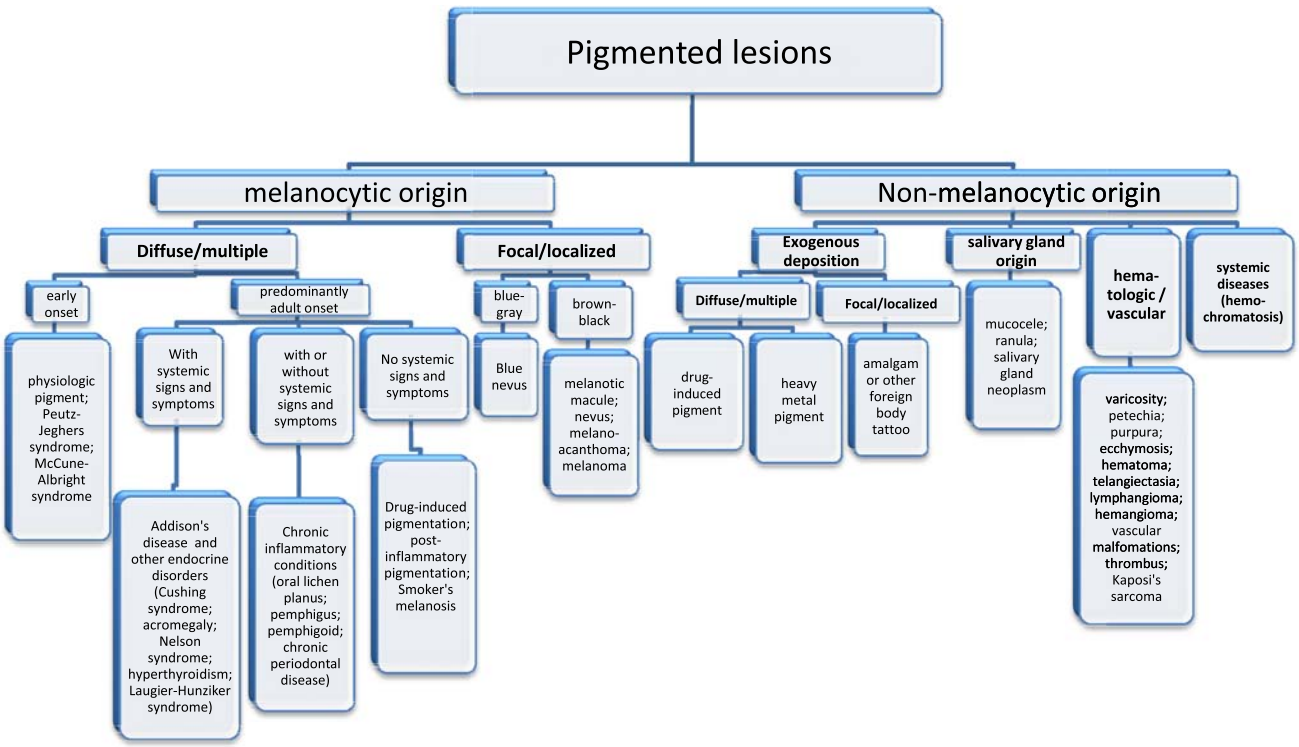

Fig. 2 The flow chart to guide the differential diagnosis of pigmented lesions

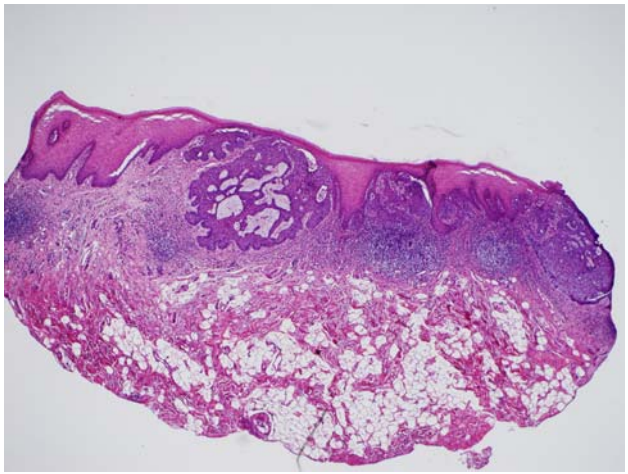

Fig. 3 Low power photomicrograph of intraoral basal cell carcinoma (×25)

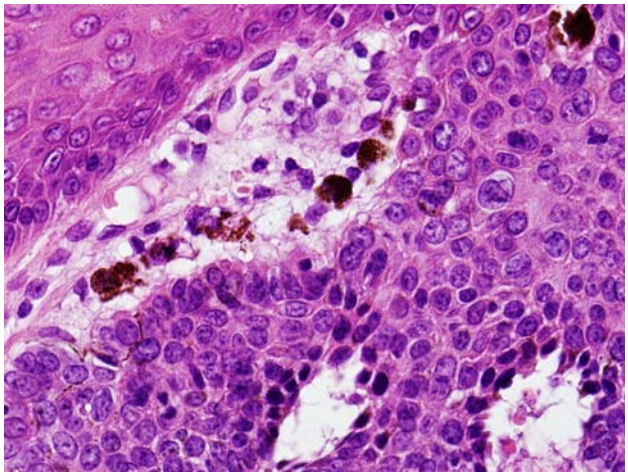

Fig. 5 Melanophages and variable amounts of melanin pigment within the tumor cells (×200)

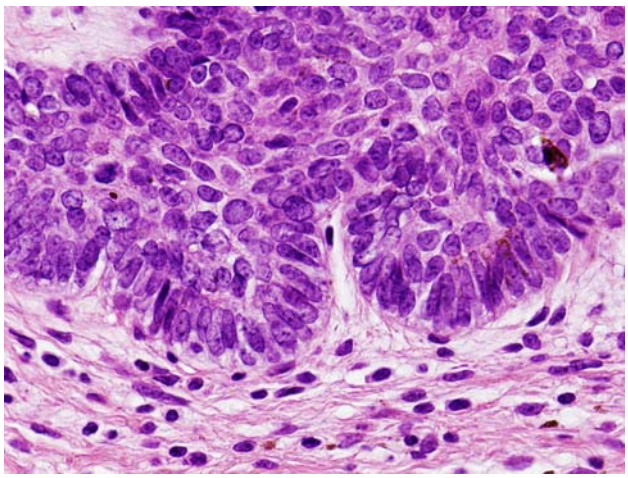

Fig. 4 Peripheral palisading of the nuclei on basal layer (×200)

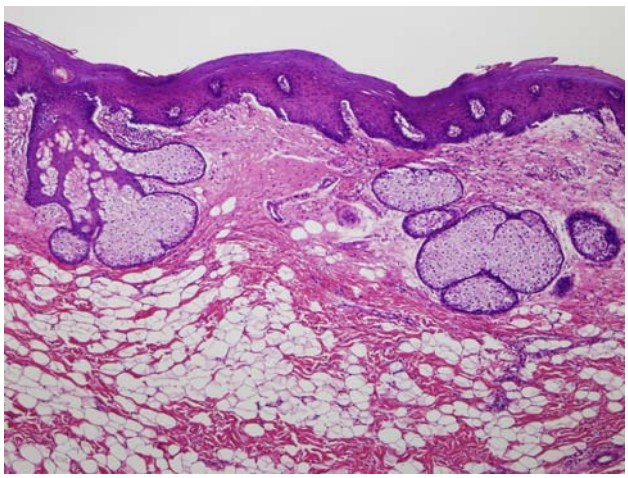

Fig. 6 Adjacent to the tumor, typical sebaceous glands are noted (Fordyce granules)

myxomatous connective tissue. A lace-like pattern of the tumor was apparent, besides a luminal architecture (there were no spindle or stellate cells in the center of the lesion). Melanocytes and melanin pigment were observed in the tumor cells, and large melanophages were seen in the surrounding stroma between the tumor islands (Fig. 5). This lesion was associated with adjacent ectopic sebaceous glands (Fig. 6). Based on the histopathology, the diagnosis of basal cell carcinoma (BCC), adenoid pigmented subtype arising from the buccal mucosa, with associated Fordyce granules was made.

True BCC involving the oral mucous membrane is extremely rare [7]. In the English literature, 14 cases of BCC have been reported to date, including 5 of oral mucosal origin and 2 arising from the lip. The male-to-female ratio was 5:3 and the patients ranged in age from 44 to 75 years. The lesions varied from 2 mm to 4.5 cm in diameter [8].

BCC is considered as a low-grade malignant tumor due to its propensity for only local recurrence and absence of association with distant metastasis; only one case with metastasis to the cervical lymph node has been reported in the literature [9]. BCC is the most common type of skin cancer, but because of the rarity in the oral mucosa, it becomes important to differentiate from peripheral ameloblastoma (PA), which shares some common microscopic features. Immunohistochemical staining has been used for the differential diagnosis of BCC and PA; while BCC shows positive staining for Ber-EP4 and negative staining for KL-1 antibodies, PA shows positive staining for KL-1 antibodies and negative staining for Ber-EP4 antibodies [8, 10]. In this case report, we found that the lesion showed positive immunohistochemical staining for Ber-EP4 and negative staining for KL-1.

BCC often arise in areas of the skin with abundant pilosebaceous units and it has been postulated that BCC may originate from multipotential cells in either the basal cell layer of the skin epithelium or in the pilosebeceous units. Interestingly, in the present case, Fordyce granules were noted in the same histologic field as the BCC mimicking what is often noted in cutaneous BCC.

References

1. Meleti M, Vescovi P, Mooi WJ, et al. Pigmented lesions of the oral mucosa and perioral tissues: a flow-chart for the diagnosis and some recommendations for the management. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2008;105(5):606–16.
2. Kauzman A, Pavone M, Blanas N, et al. Pigmented lesions of the oral cavity: review, differential diagnosis, and case presentations. J Can Dent Assoc. 2004;70(10):682–3.
3. Hatch CL. Pigmented lesions of the oral cavity. Dent Clin North Am. 2005;49(1):185–201. ix–x.
4. Gaeta GM, Satriano RA, Baroni A. Oral pigmented lesions. Clin Dermatol. 2002;20(3):286–8.
5. Eisen D. Disorders of pigmentation in the oral cavity. Clin Dermatol. 2000;18(5):579–87.
6. Jabbour SA. Cutaneous manifestations of endocrine disorders: a guide for dermatologists. Am J Clin Dermatol. 2003;4(5):315–31.
7. Edmondson HD, Browne RM, Potts AJ. Intra-oral basal cell carcinoma. Br J Oral Surg. 1982;20:239–47.
8. Del Rosario RN, Barr RJ, Jensen JL, Cantos KA. Basal cell carcinoma of the buccal mucosa. Am J Dermatol. 2001;23:203–5.
9. Nishimura N, Sakurai K, Noguchi K, Urade M. Keratotic basal cell carcinoma of the upper gingival with cervical lymph node metastasis: report of a case. J Oral Maxillofac Surg. 2001;59:677–80.
10. Lentini M, Simone A, Carrozza G. Peripheral ameloblastoma: use of cytokeratin 19 and Ber-EP4 to distinguish it from basal cell carcinoma. Oral Oncology Extra. 2004;40:79–80.