10.5005/jp-journals-10024-2160

CASE REPORT

Melanotic Macule in Conjunction with a Giant Cell Fibroma

[1]Samantha D Seitz, [2]Thanhphuong N Dinh, [3]Thomas YH Yoon

ABSTRACT

Introduction: The aim of this study is to describe a case of a melanotic macule found in conjunction with a giant cell fibroma (GCF). For oral pigmented lesions without an identifiable etiologic factor, critical factors in determining the differential diagnosis are clinical history, symmetry, and uniformity of the lesions. Potential differential diagnosis includes racial pigmentation, endocrine disturbance, Peutz–Jeghers syndrome, trauma, hemochromatosis, oral malignant melanoma, or idiopathic etiology and melanotic macules. Melanotic macules are the most common solitary pigmented melanocytic lesions in the oral mucosa, corresponding to 86.1% of melanocytic lesions of the mouth. Giant cell fibromas are reactive connective tissue lesions in the oral cavity. They were first described as a distinct entity in 1974 by Weathers and Callihan and make up around 5 to 10% of all oral mucosa fibrous lesions. They are commonly mistaken for other growths, such as pyogenic granuloma and fibroma, and diagnosis is accurately based on its distinctive histopathology.

This article presents the clinicopathologic findings of a 15-year-old Hispanic male presenting for biopsy of a melanotic macule on the mandibular anterior buccal gingiva. Histologic evaluation of the specimen revealed that the lesion also contained a GCF. Pathologic lesions of the mouth should be carefully diagnosed. Conventionally, histologic evaluation is the gold standard to produce a final diagnosis. As evidenced in this article, multiple lesions may exist in a site and may be mistakenly diagnosed as a single entity.

Clinical significance: While each lesion has been reported individually, in reviewing the literature, no cases were reported in which both histopathologic findings of GCF and melanotic macule were present within the same lesion.

[1,2]Department of Restorative Dentistry, Lake Erie College of Osteopathic Medicine, School of Dental Medicine, Bradenton Florida, USA

[3]Department of Periodontology, Lake Erie College of Osteopathic Medicine, School of Dental Medicine, Bradenton, Florida, USA

Corresponding Author: Thomas YH Yoon, Department of Periodontology, Lake Erie College of Osteopathic Medicine School of Dental Medicine, Bradenton, Florida, USA, Phone: +9414051506, e-mail: iyoon@lecom.edu

Keywords: Histopathology, Mucosal lesions, Oral lesions.

How to cite this article: Seitz SD, Dinh TN, Yoon TYH. Melanotic Macule in Conjunction with a Giant Cell Fibroma. J Contemp Dent Pract 2017;18(10):981-985.

Source of support: Nil

Conflict of interest: None

BACKGROUND

Melanotic macules are the most common solitary pigmented melanocytic lesions in the oral mucosa, corresponding to 86.1% of melanocytic lesions of the mouth.[1] Kaugars et al[2] reviewed 353 cases of oral melanotic macule and concluded the mean age to be 43.1 years with a significant predilection for females at a female-to-male ratio of 1.9:1.[3] These lesions are asymptomatic, <1 cm in diameter, and flat and may be brown, black, blue, or gray in color, with occasional occurrences of larger sizes. These are often found on the lips, gingiva, hard and soft palate, and buccal mucosa. Histologically, melanotic macules are characterized by an increase in melanin production by basal melanocytes in the basal cell layer of the epithelium and lamina propria. Melanin pigment is also observed in melanophages in the upper portion of the lamina propria.[4,5]

Other common oral lesions are GCFs, which are reactive connective tissue lesions in the oral cavity. They were first described as a distinct entity in 1974 by Weathers and Callihan.[6] Giant cell fibromas make up around 5 to 10% of all oral mucosa fibrous lesions.[7] Although GCFs are found in patients of all ages, these lesions most often occur in the second and third decades of life, with the second decade predominating, and 90 to 97% of cases are reported in Caucasians.[8] There is also a slight predilection for females, presenting with a female-to-male ratio of 1.2:1.[9,10] 48% of all GCFs occur on the gingival tissue with a higher tendency for the mandibular over the maxillary gingiva at a rate of 2:1.[8,10] Other common sites affected by GCFs in descending order of frequency

are the tongue, palate, buccal mucosa, lips, and floor of the mouth. The lesions are slow growing with size often remaining <1 cm in diameter,[8] although some have been reported with ranges up to 2 cm in diameter.[7]

Histological examination of GCFs often reveals a hyperplastic stratified squamous epithelium and stellate-shaped, mononuclear, and multinucleated giant cells loosely arranged in vascular connective tissue.[8,10,11] Sonalika et al[9] reported that although the fibroblastic origin of these giant cells is clear, the reason as to why these giant cells are formed still remains uncertain. The giant cells are usually seen numerous in the connective tissue immediately adjacent to the epithelium, have well-defined cell borders, and show dendritic processes. The characteristic cells are less prominent in the center of the lesion,[10] and the overlying epithelium is hyperplastic with thin-elongated rete ridges.[9]

While usually benign, a melanotic macule should be biopsied to rule out malignancy. Following this general practice, when a pigmented lesion from a patient was differentially diagnosed as possible melanotic macule, we biopsied the lesion. Here, we report the result of the biopsy, which interestingly found the lesion to be a melanotic macule occurring in conjunction with a GCF.

CASE REPORT

A 15-year-old Hispanic male presented to the Lake Erie College of Osteopathic Medicine, School of Dental Medicine, Bradenton, Florida, USA, for an initial examination. His medical history was not significant. During the intraoral examination, a dark, pigmented, hyperplastic area was observed on the interdental papilla between mandibular left canine and mandibular left lateral incisor (Fig. 1). The lesion was asymptomatic with an estimated duration of around 5 years. The lesion was measured less than half of a centimeter in diameter, with no pain on palpation and no bleeding on probing. The patient denied trauma to this area.

A periodontal consultation was attained and a biopsy of the lesion was recommended to reach a diagnosis. Under local anesthesia, an excisional biopsy was performed, measuring 0.4 × 0.4 × 0.3 cm, and perilesional tissue was included in the specimen to allow for histological comparison. The teeth were lightly scaled to remove any subgingival plaque and calculus. The resultant specimen was immediately stored in 10% neutral buffered formalin and sent to the oral and maxillofacial pathology laboratory at the University of Florida, College of Dentistry.

Microscopic examination revealed that the specimen was composed of dense inflamed fibrous tissue surfaced with parakeratinized stratified squamous epithelium (Fig. 2). The epithelium showed thickening of the spinous cell layer with elongated and fused rete ridge formation. The epithelium contained significant increase in the amount of melanin pigment in the basal cell layer (Figs 3 and 4). Furthermore, noted were numerous melanin-engorged macrophages within the superficial lamina propria. The underlying connective tissue, making up the bulk of the specimen, was composed of smudged hyalinized collagen bundles interspersed by plump, active, sometimes multinucleated fibroblasts (Fig. 5). There were also proliferating endothelial cells and fibroblasts present. Additionally seen were congested vascular channels and a dense inflammatory infiltrate, which was patchy and composed of mostly lymphocytes and plasma cells. The biopsy results reported a diagnosis of GCF with melanotic macule and melanin incontinence.

Fig. 1: Dark, pigmented, hyperplastic area on the interdental papilla between mandibular left canine and mandibular left lateral incisor

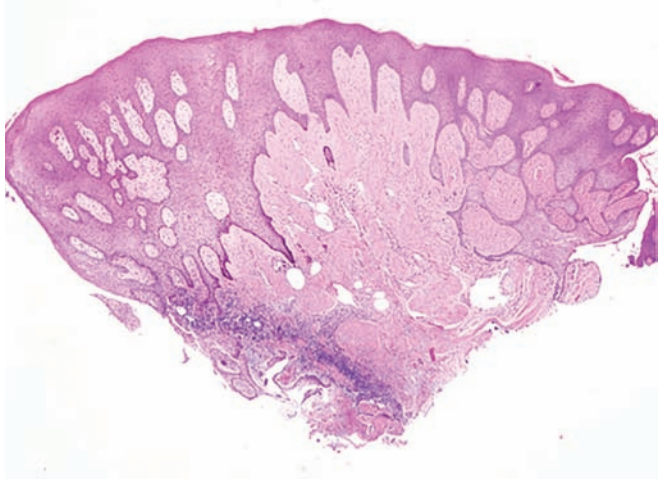

Fig. 2: Low power shows the entire lesion is well excised. Spike-shaped or dagger-shaped rete ridges are noted. The entire specimen is nodular. Melanin pigmentation is seen even in the low-power image (25×)

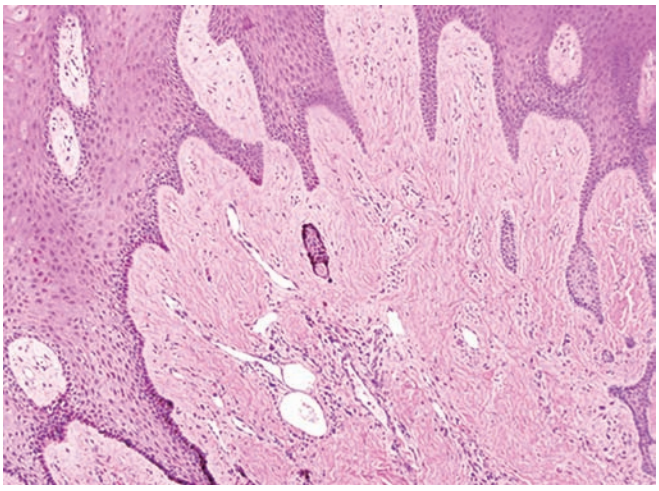

Fig. 3: Melanin pigmentation is clearly evident. Deep areas of melanin aggregates are noted in the basal cell layer of the epithelium (100×)

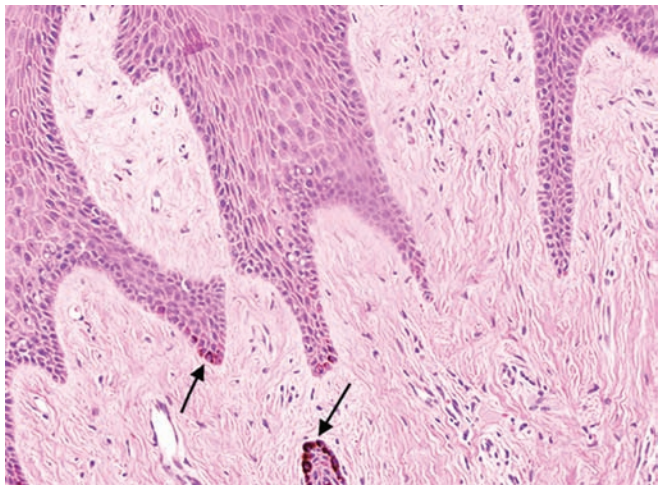

Fig. 4: Melanin pigmentation is clearly evident. Deep areas of melanin aggregates are noted in the basal cell layer of the epithelium (200×)

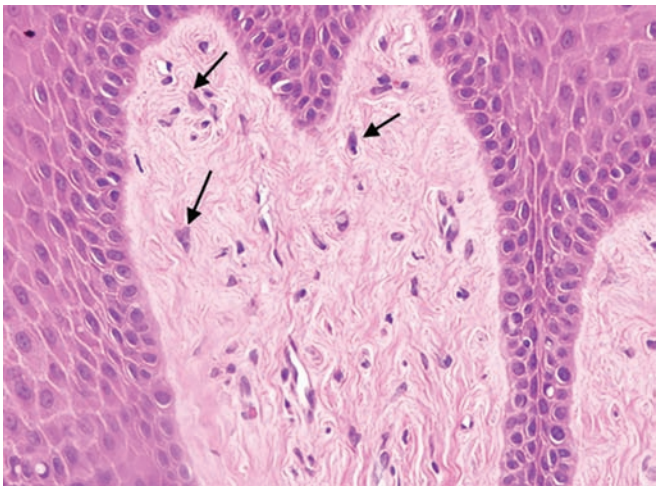

Fig. 5: Arrows point to stellate-shaped fibroblasts, some are multinucleated and exhibit dendritic processes, hence the name giant cell fibroma (400×)

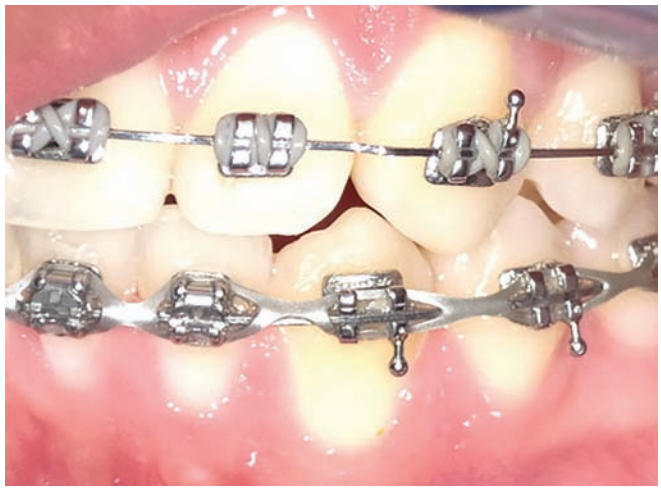

Fig. 6: Patient presents 16 months postbiopsy. No gross signs of recurrence observed

The patient was scheduled for a suture removal appointment; however, he did not present for this initial postoperative care. At a subsequent visit, healing was found to be uneventful and the patient did not report any complaint. The biopsy site was reevaluated over a year later, and there was no recurrence of either lesion type (Fig. 6).

DISCUSSION

For oral pigmented lesions without an identifiable etiologic factor, critical factors for determining the differential diagnosis are clinical history, symmetry, and uniformity of the lesions. In regard to melanotic macules, potential differential diagnosis includes racial pigmentation, endocrine disturbance (i.e., Addison's disease), Peutz–Jeghers syndrome, trauma, hemochromatosis, oral malignant melanoma, or idiopathic etiology.[4] While the lesion found in our patient may be benign, an excisional biopsy is recommended to rule out more serious differential diagnoses, such as malignant melanoma. Biopsy result also confirms a definitive diagnosis for the lesion and offers proper treatment options to the patient.

The first lesion to be ruled out by the biopsy result was oral malignant melanoma. Although this melanoma accounts for <1% of all oral malignancies[3] and generally affects adults between fourth and seventh decades of life, case reports of oral malignant melanoma in children exist.[12,13] Since oral mucosal melanomas carry a 5-year estimate survival rate of 40%, early diagnosis is of utmost importance.[14] If excision is not performed, the oral melanotic macule should at least be monitored at frequent intervals for any change in shape, size, or color.[15]

Initially, the differential diagnoses for this case included strictly pigmented lesions and combined with the generalized clinical appearance, the lesion did not suggest a

GCF. Conventionally, GCFs comprised a fibrous mucosal mass of hyperplastic connective tissue, asymptomatic, typically of normal mucosal color, and often described as pedunculated, papillary, or bosselated growths.[10] They are commonly mistaken for other growths, such as pyogenic granuloma and fibroma, and diagnosis is accurately based on its distinctive histopathology. The etiology of GCF is unknown. Irritation and trauma are noted in some cases; however, GCF does not appear to be associated with chronic irritation.[16] A possible viral origin of the tumor has also been postulated.[9] The origin of giant multinucleated cells is a matter of discussion. Souza et al[17] found that mononucleated, binucleated, and multinucleated cells from fibrous hyperplasia, GCF, and fibroepithelial polyp originated from fibroblasts lineages. Since GCF is clinically similar to other nonneoplastic lesions, the final diagnosis depends on microscopic findings.

The treatment of choice for this lesion is conservative surgical excision, with a rare reoccurrence rate.[10,11,18] Electrosurgery is an alternative option for treatment and has the advantage of direct tissue hemostasis without the need for sutures. Recall visits are necessary to ensure the absence of recurrence. If the lesion is left untreated, it may continue to proliferate, but there is certified limited growth potential due to its benign nature.[8] Fortunately, for this patient, the selected treatment of conservative excision was sufficient for both the melanotic macule as well as the GCF.

A factor to consider in regards to gingival biopsies is the risk of gingival deficiencies. The removal of interdental papilla carries a risk of altered or minimal regeneration.[19] In this particular case, the entire papilla was removed to obtain both lesional and perilesional tissues. Although a significant amount of tissue was excised for biopsy, the patient was able to successfully regenerate the entire papilla. The patient's orthodontic therapy could have helped with interdental papilla regeneration. According to Kokich,[20] insufficient interdental papilla can be successfully regenerated through controlled orthodontic movement. Therefore when faced with a situation that a biopsy may yield esthetic deficiencies, an orthodontic consultation should be considered.

The current case presents a dilemma in regards to determining a clinical diagnosis since the initial lesion was concluded through biopsy to be two different lesions. These lesions often present with few common characteristics. The patient case meets the usual criteria for GCF considering the location of the lesion within the mandibular buccal gingiva. The case also meets the common conditions for the melanotic macule with the size and clinical appearance of the lesion. At the age of 15 years, this patient is significantly younger than the average age for an occurrence of melanotic macule and slightly younger than the average age for a GCF. GCF and melanotic macule both have a predilection for females, therefore making his gender also uncommon. Moreover, his Hispanic race is significantly unusual, as the majority of reported cases of GCF have been in Caucasian patients.[8]

The most unique characteristic of the lesion in the case study is the appearance of both the GCFs in conjunction with the melanotic macule on the same biopsied lesion. To the authors' knowledge, while each lesion has been reported individually, in reviewing the literature, no cases were reported in which both histopathologic findings of GCF and melanotic macule were present within the same lesion.

CONCLUSION

Pathologic lesions of the mouth should be carefully diagnosed. Conventionally, histologic evaluation is the gold standard to produce a final diagnosis. As evidenced in this article, multiple lesions may exist in a site and may be mistakenly diagnosed as a single entity. Thus, practitioners should be encouraged to biopsy suspicious lesions to ensure the overall health and well-being of patients.

CLINICAL SIGNIFICANCE

While each lesion has been reported individually, in reviewing the literature, no cases were reported in which both histopathologic findings of GCF and melanotic macule were present within the same lesion.

REFERENCES

1. Buchner A, Merrell PW, Carpenter WM. Relative frequency of solitary melanocytic lesions of the oral mucosa. J Oral Pathol Med 2004 Oct;33(9):550-557.
2. Kaugars GE, Heise AP, Riley WT, Abbey LM, Svirsky JA. Oral melanotic macule. A review of 353 cases. Oral Surg Oral Med Oral Pathol 1993 Jul;76(1):59-61.
3. Gondak RO, da Silva-Jorge R, Jorge J, Lopes MA, Vargas PA. Oral pigmented lesions: clinicopathologic features and review of the literature. Med Oral Patol Oral Cir Bucal 2012 Nov;17(6):e919-e924.
4. Pais S, Hegde SK, Bhat SS. Oral melanotic macule – a case report. J Indian Soc Pedod Prev Dent 2004 Jun;22(2):73-75.
5. Kumar C, Sharma D, Pandita A, Shastri S. Congenital lingual melanotic macule: rare entity in infants. Sultan Qaboos Univ Med J 2015 Aug;15(3):e440-e441.
6. Weathers DR, Callihan MD. Giant-cell fibroma. Oral Surg Oral Med Oral Pathol 1974 Mar;37(3):374-384.
7. Campos MS, Domaneschi C, Nunes FD, Weinfeld I. Giant cell fibroma of the maxillary gingiva in children: a case report. J Dent Child (Chic) 2010 Sep-Dec;77(3):174-176.
8. Nikitakis NG, Emmanouil D, Maroulakos MP, Angelopoulou MV. Giant cell fibroma in children: report of two cases and literature review. J Oral Maxillofac Res 2013 Jan-Mar;4(1):e5.
9. Sonalika WG, Sahu A, Deogade SC, Gupta P, Naitam D, Chansoria H, Agarwal J, Katoch S. Giant cell fibroma of

tongue: understanding the nature of an unusual histopathological entity. Case Rep Dent 2014 Jan;2014:864512.
10. Swan RH. Giant cell fibroma. A case presentation and review. J Periodontol 1988 May;59(5):338-340.
11. Mello-Moura AC, Santos AM, Bonini GA, Del Conte Zardetto CG, Moura-Netto C, Wanderley MT. Giant cell fibroma in a two-year-old child. Case Rep Dent 2016 Sep; 2016:7058356.
12. Kumar SV, Swain N. Oral malignant melanoma: a case report. Int J Oral Maxillofac Pathol 2011 Jul-Sep;2(3):50-54.
13. Krenz RM, Matusik J, Csanaky G. Amelanotic oral malignant melanoma in a 16 year-old girl. Rep Pract Oncol Radiother 2004 Aug;9(5):183-186.
14. Rapidis AD, Apostolidis C, Vilos G, Valsamis S. Primary malignant melanoma of the oral mucosa. J Oral Maxillofac Surg 2003 Oct;61(10):1132-1139.
15. Lamichhane NS, An J, Liu Q, Zhang W. Primary malignant mucosal melanoma of the upper lip: a case report and review of the literature. BMC Res Notes 2015 Sep;8:499.
16. Gnepp, DR.; editor. Diagnostic surgical pathology of the head and neck. Philadelphia (PA): Saunders, Elsevier; 2009. p. 2.
17. Souza LB, Andrade ES, Miguel MC, Freitas RA, Pinto LP. Origin of stellate giant cells in oral fibrous lesions determined by immunohistochemical expression of vimentin, HHF-35, CD68 and factor XIIIa. Pathology 2004 Aug;36(4):316-320.
18. Reddy VK, Kumar N, Battepati P, Samyuktha L, Nanga SP. Giant cell fibroma in a paediatric patient: a rare case report. Case Rep Dent 2015 Oct;2015:240374.
19. Holmes CH. Morphology of the interdental papillae. J Periodontol 1965 Nov-Dec;36(6):455-460.
20. Kokich VG. Esthetics: the orthodontic–periodontic restorative connection. Semin Orthod 1996 Mar;2(1):21-30.