OLGU SUNUMU CASE REPORT

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Giant Melanocytic Nevus of Oral Mucosa with Gingival Fibromatosis

Oral Mukozanın Gingival Fibromatozisli Dev Melanositik Nevüsü

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The present case was presented as a poster at the 17th International Congress of Pathology and Medicine (2014, İstanbul).

ABSTRACT The purpose of this case report is to present this rare subtype of oral nevus, which has a rare location and size. A 45-year-old female patient was referred to our clinic for periodontal disease. Clinical examination revealed a large pigmented lesion on the buccal mucosa and gingival hyperplasia was also detected. The differential diagnosis was hemangioma and malignant melanoma. No pulsation was noted on palpation. Multiple biopsies were obtained at different times and sites of the lesion. The specimens were compatible with a melanocytic nevus and gingival fibromatosis. A giant melanocytic nevus with gingival fibromatosis of the oral mucosa is a rare condition. Clinicians should be careful in clinical examination in such cases and make differential diagnosis for appropriate biopsy and treatment.

Keywords: Melanocytes; melanocytic nevus; giant cells; mouth mucosa

ÖZET Bu olgu sunumunda, nadir bir yerleşim yeri ve boyutu olan oral nevüsün bu nadir alt tipinin sunulması amaçlanmaktadır. Kırk beş yaşında kadın hasta periodontal hastalık nedeniyle kliniğimize başvurdu. Klinik muayenede bukkal mukozada büyük pigmente lezyon ve ayrıca diş eti hiperplazisi tespit edildi. Hemanjiyom ve malign melanom ile ayırıcı tanı yapıldı. Palpasyonda nabız yoktu. Farklı zamanlarda ve lezyonun farklı yerlerinden çoklu biyopsiler yapıldı. Örnekler melanositik nevüs ve diş eti fibramatozisi ile uyumluydu. Ağız mukozasında dişeti fibromatozisi ile birlikte dev bir melanositik nevüs nadir görülen bir durumdur. Klinisyenler bu gibi durumlarda klinik muayenede dikkatli olmalı ve uygun biyopsi ve tedavi için ayırıcı tanı yapmalıdır.

Anahtar Kelimeler: Melanositler; melanositik nevüs; dev hücreler; ağız mukozası

Pigmentation of the oral mucosa occurs when one or more pigments accumulate in the mucosa and cause discoloration.[1] Oral pigmented lesions can occur in a variety of clinical conditions, from physiologic pigmentation to malignant melanoma.[2] The color of the pigmentation depends on the depth of the pigment in the mucosa. The color can be seen in various shades from brown to blue or black.[1,3,4] In addition to melanotic macules, solitary pigmented lesions of melanocytic origin (melanoacanthoma, melanoma, oral melanocytic nevus) are rarely seen in the oral mucosa.[5] Oral melanocytic nevi are benign neoplasms composed of cells of the neural crest, often referred to as nevus cells.[6] A melanocytic nevus is a focal proliferation of nevus cells that may be congenital or acquired. These lesions are typically found on the skin but rarely on the oral mucosa.[5] Their etiology and pathogenesis are not fully understood.[7]

They are examined histopathologically in 3 stages: First, a junctional nevus, which is the proliferation of nevus melanocytes throughout the submucosal-mucosal junction; second, a compound nevus, which involves migration of nevus cells into the underlying mesenchymal tissue, and third, a subepithelial or intramucosal nevus, which involves loss of the submucosal-mucosal junction so that all remaining

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nevus cells are located in the subepithelial connective tissue stroma.[8,9]

Intramucosal nevus is the most common type of nevi in the oral cavity and occurs in about 63-80.6%. The second most common type is blue nevus with an incidence of 8.3-32%. Oral compound nevus is one of the less rare types of oral mucosal nevi, with an incidence of only 5.9%-16.5%.[5]

Fibromatosis (fibrous hyperplasia) can be described as an inflammatory reaction or fibroblastic proliferation without neoplasia. In general, it may involve various organs or be confined to the gingiva.[10]

This case report demonstrates this unusual subtype of oral nevus that has an unusual region and size.

CASE REPORT

A 45-year-old female patient approached our clinic because of periodontal disease. No abnormality was noted on extraoral examination. Clinical examination revealed a large pigmented lesion (right buccal mucosa) with the largest diameter of about 6 cm, irregular margins, and painless on palpation in the right buccal mucosa and gingival hyperplasia (Figure 1).

When we took the anamnesis, we learned that the lesion had been present for 30 years. On palpation of the lesion, no pulsation was felt. Multiple biopsies were performed at different times and from 6 different sites of the lesion. On histologic examination, the nevus cells appeared as nondendritic oval melanocyte nests with cytoplasmic polygonal melanin pigmentation in the chorion, and there was no fusion activity. There was no cellular atypia or invasion of the epidermal area, and no mitotic figures suggestive of melanoma were observed, and the lesion did not spread to the lingual region (Figure 2a). No vascular permeation or cellular necrosis was observed. A papillomatous structure appeared in the linea alba region. Fibrous tissue was seen only in the gingival specimen. There were no Ki-67-positive melanocytes, and human melanoma black-45 immunostaining was also negative (Figure 2b). The histopathological appearance was shown to be inhomogeneous pigmentation (Figure 2a, Figure 2b). The specimens were compatible with melanocytic nevus and generalized gingival fibromatosis (Figure 3).

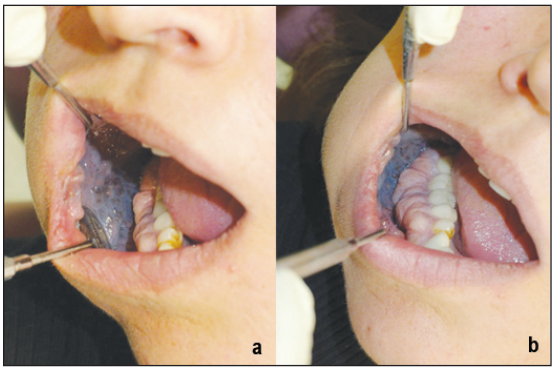

FIGURE 1: a: View of the buccal mucosa, **b:** View of the gingiva, gingival fibromatosis.

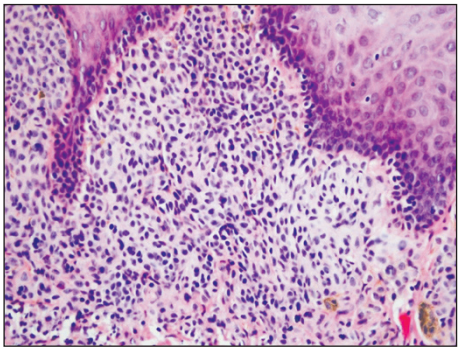

FIGURE 2a: Appearance of non-mitotic, ovoid, non-invasive melanocytes nests, subepithelial area (Hematoxylin-eosin, mag. x200).

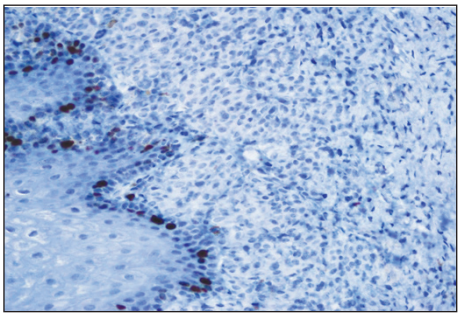

FIGURE 2b: Histopathological view of Ki-67. There were no K-67 (+) melanocytes (Ki-67 immunohistochemistry and HMB-45 mag. x200).

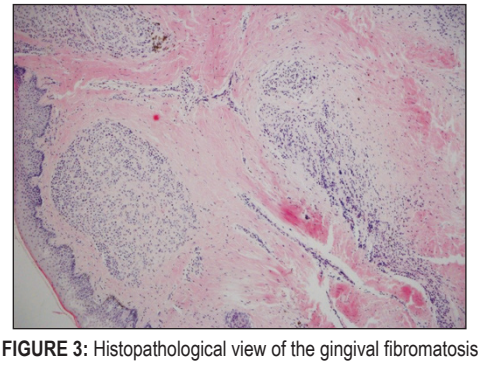

FIGURE 3: Histopathological view of the gingival fibromatosis (Hematoxylin-eosin, mag. x40).

Nevus cell clusters were detected at the epithelial-connective tissue junction and in the underlying lamina propria and submucosa, and the histopathologic diagnosis was made as a compound melanocytic nevus. Treatment was planned for the area in question, but the patient could not be followed up because she refused treatment.

This case report was approved by the Local Research Ethics Committee (protocol: 2021-280, 27.01.2021). Informed consent was obtained from the patients before the examinations by the principles of the Declaration of Helsinki, which includes all changes and revisions.

DISCUSSION

Oral melanocytic nevi are usually asymptomatic. Clinically, compound nevi present as slightly raised papules or plaques on the skin or, less commonly, as well-defined macules; they are usually smaller than 1-2 cm.[7] In this case, clinical examination revealed a large pigmented lesion (right buccal mucosa) and gingival hyperplasia. The surface of the lesion is smooth and contains dark brown areas. It is a lesion on the right buccal mucosa with a diameter of about 6 cm, with irregular margins, and painless on palpation.

Most cases of oral compound nevus are localized to the hard palate (33.3% to 57.1%), but in this case, it is located in the buccal mucosa (28.6% to 57.1%), an area less commonly affected than the hard palate. There is no specific age range, but it is most common between the ages of 20 and 30, similar to the case described in more than 50% of patients.[5,11]

It has been noted that biopsy of compound nevus is more common in women.[5] It is likely that the prevalence is higher in women because they have greater health awareness and better referral to medical care. This also supports this. However, blue nevi have been observed more frequently in men than in women.[5,11]

Buncher et al. found that the most common intramucosal nevus (55-80.6%) was observed in the oral mucosa, followed by blue nevi (8.3-32%) and compound nevus (5.9-16.5%) in third place.[5,11]

There are insufficient data on malignant transformation of oral melanocytic nevi.[12] Approximately one-third of oral melanomas originate from pigmented lesions, but the nature of these precursor lesions has not been extensively reported.[12,13] Although there is no conclusive evidence of their malignant potential, their clinical similarity to early-stage oral melanomas, their infrequent occurrence, and the small size of the lesion prompt the clinician to perform an excisional biopsy.[5,11]

In the presence of intraoral pigmentation, the patient should receive a comprehensive medical and dental history accompanied by intraoral and extraoral examinations. Many pigmented lesions can be diagnosed clinically based on size, shape, or color, which are accompanied by clinical clues. Therefore, effective clinical maneuvers against pigmented lesions of the oral mucosa are critical in ruling out possible malignancies.[8] The differential diagnosis of intraoral nevi can include melanotic macules, smoking-related melanosis, physiologic pigmentation, melanoacanthoma, amalgam tattoo, and melanoma.[11]

The frequency of pigmented lesions in the oral cavity has not been adequately addressed in the literature.[5]

In their study, Ferreira et al. collected data on the size of melanocytic nevi in 45 patients.[14] They found that the greatest length of the lesion varied from 0.1 cm to 2.5 cm. In the study, 86.7% of lesions were measured as smaller than 1.0 cm. These data show that this case, with the largest diameter of 6 cm, was significantly larger than the other cases.

It is significantly larger than the cases in the literature, it is not seen very often according to the data in the literature, and it is located in a different location than the most common localization and is seen at the same time. The fact that it is associated with gingival fibromatosis makes this situation interesting.

The main reasons for treating melanocytic nevus are the risk of malignant transformation and esthetic reasons. Surgical excision is recommended as the first treatment option.[15] In this case, the patient was offered treatment, but it was not performed because the patient did not accept the treatment.

A giant melanocytic nevus with gingival fibromatosis in the oral mucosa is a rare condition. Clinicians should be cautious in clinical examination in such cases and make a differential diagnosis for appropriate biopsy and treatment. When a large pigmented lesion is detected in the oral mucosa, multiple biopsies from different parts of the lesion and a detailed evaluation of the lesion are essential for accurate diagnosis and exclusion of malignancy.

Source of Finance

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Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: *Hilal Peker Öztürk;* ***Design:*** *Hilal Peker Öztürk, Burak İncebeyaz, Ömer Günhan;* ***Control/Supervision:*** *Hilal Peker Öztürk, İsmail Hakan Avsever, Buğra Şenel;* ***Data Collection and/or Processing:*** *Hilal Peker Öztürk, İsmail Hakan Avsever, Ömer Günhan;* ***Analysis and/or Interpretation:*** *Hilal Peker Öztürk, Aslıhan Aslan Balcı, Burak İncebeyaz, Buğra Şenel, Mehmet Hakan Kurt;* ***Literature Review:*** *Hilal Peker Öztürk, Burak İncebeyaz, Buğra Şenel, Mehmet Hakan Kurt;* ***Writing the Article:*** *Hilal Peker Öztürk, Burak İncebeyaz;* ***Critical Review:*** *Ömer Günhan;* ***References and Fundings:*** *İsmail Hakan Avsever.*

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