Granular Cell Tumor Seen in Buccal Sulcus: A Case Report

Bukkal Sulkusta Görülen Granüler Hücreli Tümör: Olgu Sunumu

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ABSTRACT

Granular cell tumor is a rare, benign tumor that usually occurs as a single nodule in the skin, tongue or oral cavity. Granular cell tumor is a rare tumor in the head and neck region and most commonly occurs in the oral cavity, especially in the tongue. Although the lesion is usually seen as a solitary nodule with no boundaries, it can be found in different parts of the body. Abrikossoff thought that the tumor is firstly originated from muscle tissue and was called granular cell myoblastoma. Recent ultrastructural studies and stains such as S100 and neuron specific enolase confirm that this tumor is derived from Schwann cells of the peripheral nerves. A 50 year old female patient was admitted to the Department of Oral and Maxillofacial Surgery of Gazi University Dentistry Faculty due to a yellowish white colored, nodular lesion in the left maxillary buccal sulcus area which has been for about 6 months. Histopathological examination of the excised lesion was revealed a granular cell tumor, which was especially seen on the tongue in the oral cavity. No recurrence was observed at postoperative sixth month.

Key Words: Granular Cell Tumor, Oral Cavity, Buccal Mucosa.

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ÖZET

Granüler hücreli tümör; genellikle ciltte, dilde veya ağız boşluğunda tek bir nodül olarak ortaya çıkan nadir, iyi huylu bir tümördür. Granüler hücreli tümör, baş boyun bölgesinde nadiren görülür ve en sık ağız boşluğunda, özellikle dilde ortaya çıkar. Lezyon genellikle sınırları olmayan soliter bir nodül olarak görülse de vücudun farklı bölgelerinde de bulunabilir. Abrikossoff, tümörün öncelikle kas dokusundan kaynaklandığını düşünmüş ve granüler hücreli miyoblastom olarak adlandırmıştır. S100 ve nöron spesifik enolaz gibi son ultrastrüktürel çalışmalar, bu tümörün periferik sinirlerin Schwann hücrelerinden türetildiğini doğrulamaktadır. 50 yaşında kadın hasta, Gazi Üniversitesi Diş Hekimliği Fakültesi Ağız Diş ve Çene Cerrahisi Anabilim Dalı'na yaklaşık 6 aydır sol maksiller bukkal sulkus bölgesinde bulunan sarımsı beyaz renkli, nodüler lezyon nedeniyle başvurmuştur. Eksize edilen lezyonun histopatolojik incelemesinde özellikle ağız boşluğunda dilde görülen granüler hücreli tümör saptanmıştır. Postoperatif altıncı ayda nüks gözlenmemiştir.

Anahtar Sözcükler: Granüler Hücreli Tümör, Ağız Boşluğu, Bukkal Mukoza.

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INTRODUCTION

Granular cell tumor is a benign soft tissue neoplasm characterized by the accumulation of plump cells with granular cytoplasm in abundance, first described by the Russian pathologist Alexei Ivanovich Abrikossoff in 1926(1,2). Although this lesion is seen anywhere in the body, it is found especially in the head and neck region(45-65%)(2-4), especially on the tongue in oral cavity(1,2,5). It is more common on the dorsal side on the tongue(3). Histopathologically the lesion is well limited but not encapsulated(7). Treatment of this lesion is conservative excision(3,5).

In this literature, we present a case of granular cell tumor in the buccal sulcus of the left maxilla of a 50 year old female patient. The form of the granular cell tumor seen in the buccal sulcus, which is mostly seen in the lateral dorsal part of the oral cavity, is rather rare.

CASE REPORT

A 50 year-old female patient was admitted to the Department of Oral and Maxillofacial Surgery of Gazi University Dentistry Faculty due to a nodular lesion in her left maxillary buccal sulcus region for about 6 months. The lesion is clinically sessile, well-limited yellowish white in color and about 1 cm in size. It was in the form of a solitary nodule and moved in palpation (Figure 1).

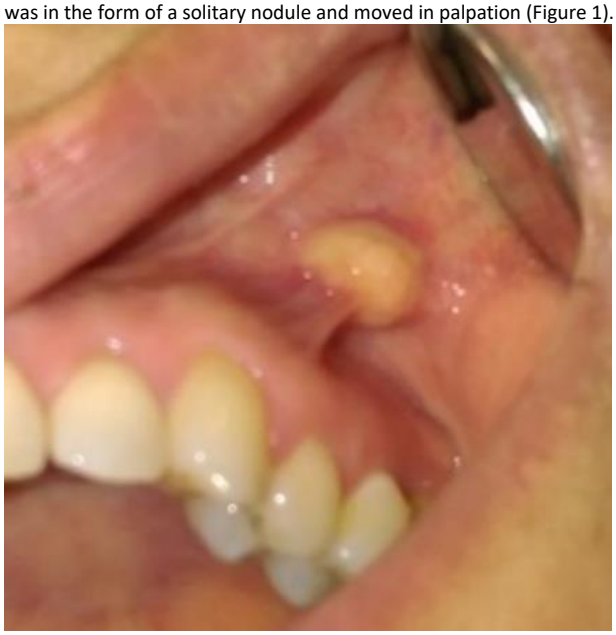

Figure 1. Intraoral photograph of the patient

Patient did not mention the presence of pain, ulceration and bleeding. The patient stated that such a lesion occurred in her mouth for the first time but she received lipomas from her back. No other disease has been detected that could cause this lesion and no abnormal findings were found on the panoramic radiograph taken (Figure 2).

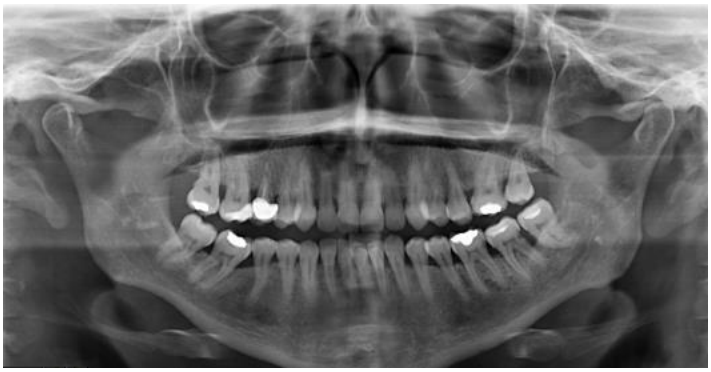

Figure 2. Panoramic radiography of the patient

According to the clinical appearance of the lesion, clinical diagnosis of fibroma or lipoma was made. The lesion was planned to be excised under local anesthesia for histopathological examination (Figure 3,4).

Figure 3. Image of after lesion excision

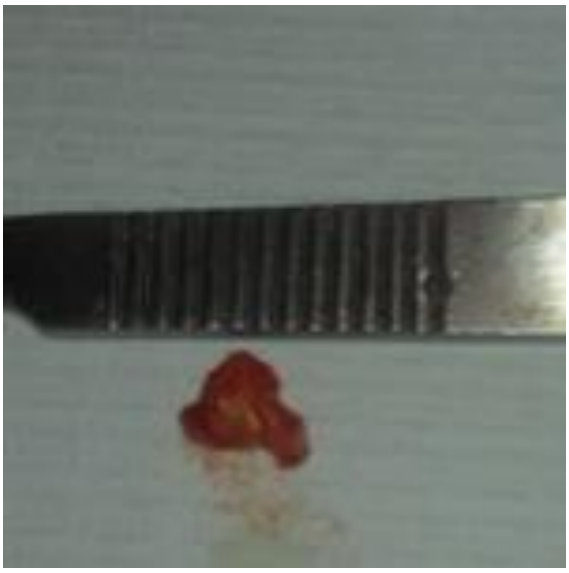

Figure 4. Excised lesion material

Histological sections revealed a lobulated benign tumor composed of the cells with granular cytoplasm beneath the oral mucosa epithelium. The lesion was diagnosed as a granular cell tumor (Figure 5).

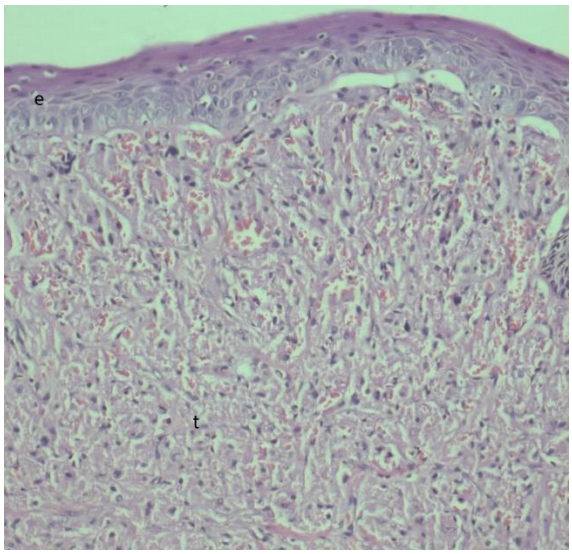

Figure 5. The tumor (t) composed with granular cells beneath the oral mucosa epithelium (e) (HE x200)

No recurrence was detected at postoperative 3rd and 6th months (Figure 6,7).

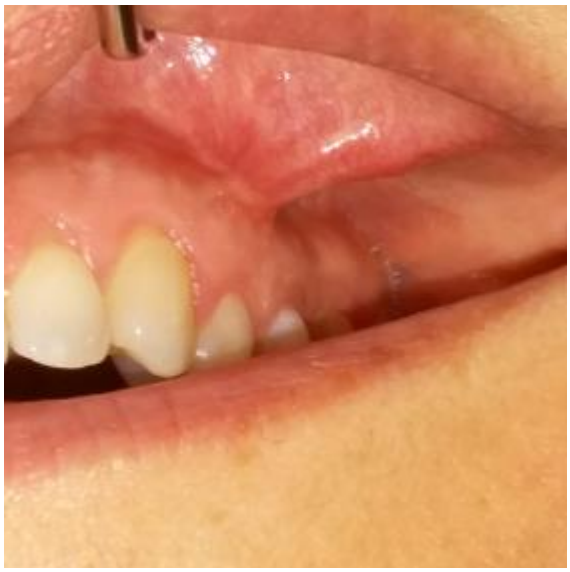

Figure 6. Postoperative 3rd month

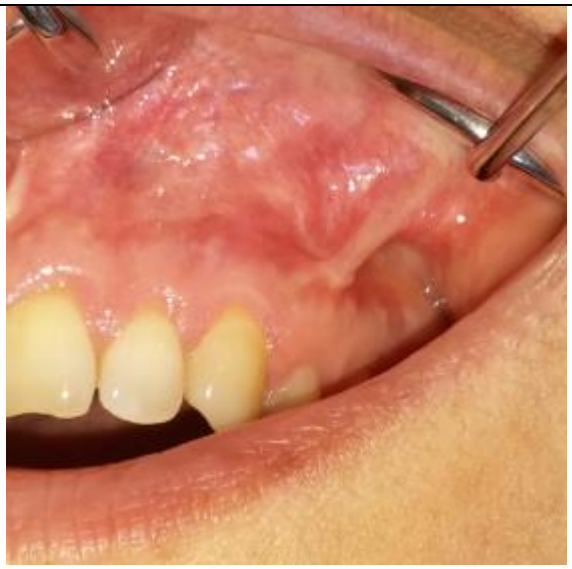

Figure 7. Postoperative 6th month

DISCUSSION

GCT, also known as Abrikossoff's tumor, is a rare benign neoplasm that occurs at various areas of the body preponderance to the oral cavity(6,7). The histogenesis of GCT remains mysterious despite numerous immunohistochemical and ultra-structural studies(7). It has been discussed for a long time whether the tumor is caused by muscle, fibroblast, histiocyte, nerve crest or nerve sheath. However, S-100 protein is considered to be of neural sheath origin, as demonstrated by peripheral nerve myelin protein, such as P2 and PO protein positivity(6). This is based on the close anatomical relationship of GCTs with the peripheral nerve fiber; ultrastructural representation of myelin shapes; on axon-like structures and immunohistochemical reactivity with S-100 protein, neuron-specific cells and myelin proteins(7,8). In our case; cytoplasmic granules were positive with PAS and DPAS (+), in immunohistochemical studies, the cells forming the lesion were positive with S100 and negative with SMA (-).

Although it is common in the third and fourth decades of life, GCT may occur at any age. It has been reported that two-thirds of cases are reported in women(6) and it is quite rare in children(9). Approximately 40% of cases are in the tongue, while skin and subcutaneous tissue share one third of the cases. Other areas such as esophagus, stomach, larynx, bronchus, uvea, muscle and pituitary stem can also be affected(6). Multiple GCTs are most commonly found in the intradermal or subcutaneous tissue, but in the oral mucosa, gastrointestinal and genital tract(10). In our patient, the lesion was detected in the left maxilla buccal mucosa, this is a very rare localization.

GCT usually offers a smooth, rough, or verrucous surface with a uniform, single, raised, rigid nodule of 0.5-3.0 cm in diameter. It usually remains asymptomatic, but sometimes it may be associated with mild pruritus or tenderness(6). Our case was clinically sessile, well-limited yellowish white in color and about 1 cm in size asymptomatic.

Histologically, GCT is characterized by the proliferation of large polygons cytoplasmic granular neoplastic cells, cytoplasma, and undefined cytoplasmic boundaries, which are small and eccentric nuclei placed in eosinophilia. In some cases, the tumor invades epithelium pseudoepitheliomatous hyperplasia(8).

Granular cell tumor is most commonly seen as a painless solitary nodüle(1,2,9). Recurrence may occur if the lesion is not completely removed. The risk of malignant transformation of the granular cell tumor is quite low(<2%)(5,9). Histopathologically these lesions present with round or polygonal cells with abundant granular cytoplasm and small nucleus that is eccentrically placed. The cells are arranged in the form of unencapsulated sheets, cords or nests with a syncitial appearance(1).

CONCLUSION

Granular cell tumor is a neoplasm that develops in the soft tissues, mainly in the skin, oral cavity and gastrointestinal tract, but the tumor is relatively rare. In this article, we present a 50-year-old female patient who was admitted to Gazi University Faculty of Dentistry, Department of Oral and Maxillofacial Surgery because of the lesion in the left maxillary buccal mucosa for 6 months. Histopathological examination of the excised lesion revealed a granular cell tumor, which was especially seen on the tongue in the oral cavity. No recurrence was observed at postoperative sixth month.

Conflict of interest

No conflict of interest was declared by the authors.

REFERENCES

1. Suchitra G, Tambekar KN, Gopal KP. Abrikossoff's tumor of tongue: Report of an uncommon lesion. J Oral Maxillofac Pathol 2014;18:134–6.
2. Ferreira JCB, Leite AFO, Guidi R, Mendonça EF. Granular cell tumor mimicking a squamous cell carcinoma of the tongue: a case report. Ferreira et al. BMC Res Notes 2017;10:14.
3. Tamborini F, Cherubino M, Scamoni S, Valdatta LA. Granular Cell Tumor of the Toe: A Case Report. Hindawi Publishing Corporation Dermatology Research and Practice. Volume 2010, Article ID 184125, 3 pages.
4. Daniels JSM. Granular cell tumour of tongue: A case report. The Saudi Dental Journal. 2009;21:75-8.
5. Loo SV, Thunnissen E, Postmus P, Waal I. Granular cell tumor of the oral cavity; a case series including a case of metachronous occurrence in the tongue and the lung. Med Oral Patol Oral Cir Bucal. 2015;20:30-3.
6. Gayen T, Das A, Shome K, Bandyopadhyay D, Das D, Saha A. Granular Cell Tumor: An Uncommon Benign Neoplasm. Indian J Dermatol. 2015;60:322.
7. Dive A, Dhobley A, Fande PZ, Dixit S. Granular cell tumor of the tongue: Report of a case. J Oral Maxillofac Pathol 2013;17:148.
8. Costa NCS, Bertini F, Carvalho YR, Almeida JD, Cavalcante ASR. Granular cell tumor presenting as a tongue nodule: two case reports. Journal of Medical Case Reports. 2012;6:56.
9. Akkaya H, Toru HS, Ayva ES, Karabulut Z, Durusoy Ç. Metachronous Occurrence of Granular Cell Tumor in Breast Skin and Scalp: Diagnostic Challenging Differentiating Benign from Malignant and a Literature Review. Hindawi Publishing Corporation Case Reports in Pathology Volume 2016, Article ID 8043183, 6 pages.
10. Sirera BV, Zabala P, Mira CA, Sempere FJV. Multiple granular cell tumors with metachronous occurrence in tongue and vulva. Clinicopathological and immunohistochemical study. J Oral Maxillofac Pathol 2014;18:437–41.