## **OLGU SUNUMU**

# Çocuk hastada primer hiperparatirodizmin ağız içi belirtisi olarak periferal dev hücreli granülom: Birolgu sunumu

# Peripheral giant cell granuloma as an oral manifestation of primary hyperparathyroidism on a pediatric patient: A case report

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#### **SUMMARY**

Peripheral giant cell granuloma (PGCG) occurs exclusively on the gingiva, presenting as a red or bluish-purple nodular mass. Histologically, PGCG shows proliferation of multinucleated giant cells within a background of mesenchymal cells similar to brown tumors, a characteristic feature of hyperparathyroidism. In this case report, clinical, histological findings and periodontal treatment approach were presented of a 11 year-old girl who had localized gingival overgrowth interdentally between the teeth #43 and #44. Radiographic findings were normal. Initial periodontal treatment consisted of oral hygiene instructions, scaling and root planning. Subsequently, the lesion (0,9X0,6X3 cm) was excised and examined histologically. Immediately after excision of the lesion, soft tissue was reconstructed using free gingival graft obtained from palate. Hematoxylin-eosin stained sections revealed bluish-purple colored nodular proliferation that had multinucleated giant cells and fibroblasts under the squamous epithelium, all of which were consistent with PGCG. This finding led to the further evaluation of the patient for hyperparathyroidism, which was diagnosed along with hypercalcemia as a result of biochemical analyses. Treatment was initiated for hyperparathyroidism by the pediatric endocrinologist. Gingival healing was uneventful without any recurrence at 6 months follow-up period.

**Keywords**: Biopsy, giant cell granuloma, hyperparathyroidism

#### ÖZET

Periferal dev hücreli granülom (PDHG) çoğunlukla dişetinde kırmızı ya da mavimsi mor renkli nodüler kitleler şeklinde görülür. PDHG histolojik olarak mezenkimal hücrelerden yoğun bağ dokusu içinde çok çekirdekli dev hücre proliferasyonu ile karakterizedir. Bu histolojik görüntüsü hiperparatiroidizmin karakteristik özelliklerinden olan "kahverengi tümör" ile benzerlik gösterir. Bu olgu raporunda, 43 ve 44 numaralı dişlerin interdental dişetinde lokalize dişeti büyümesi bulunan 11 yaşında kız çocuğunun klinik ve histolojik bulgularıyla periodontal tedavisi sunuldu. Radyografik bulgular normaldi. Hastaya ağız hijyeni eğitimi, diş ve kök yüzeyi temizliği ve kök yüzeyi düzleştirmesi işlemlerini içeren başlangıç periodontal tedavi uygulandı. Boyutları 0,9X0,6X3 cm olan lezyon eksize edilerek çıkarıldı ve takiben damaktan elde edilen serbest dişeti grefti ile yumuşak dokunun rekonstrüksiyonu gerçekleştirildi. Hematoksilen-eozin boyamayla incelenen kesitlerde histolojik olarak PDHG ile uyumlu, çok katlı epitel tabakasının altında çok çekirdekli dev hücreler ve fibroblastlardan zengin bağ dokusu içeren mavimsi mor renkli nodüler



proliferasyon izlendi. Bu bulgu hastanın ayırıcı tanısının hiperparatiroidizm açısından yapılması gerektiğini gösterdi. Pediatrik endokrinolog ile konsülte edilen hastaya yapılan biyokimyasal tetkikler sonucunda hiperkalsemi bulundu ve hastaya hiperparatiroidizm tanısı konuldu. Hasta hiperparatirodizim tedavisi için pediatrik endokrinolog tarafından kontrol altına alındı. Dişeti dokusu 6 aylık takip süresi boyunca herhangi bir rekürrans olmaksızın sorunsuz iyileşme gösterdi.

**Anahtar kelimeler:** Biyopsi, dev hücreli granülom, hiperparatiroidizm

### INTRODUCTION

Peripheral giant cell granuloma (PGCG) is a localized, red or bluish-purple nodular mass that occurs exclusively on the gingiva<sup>1</sup>. Pain is not a common clinical characteristic for the lesion except for the presence of trauma. PGCG rarely affects the underlying bone and radiological examination may reveal slight crestal alveolar bone loss<sup>2</sup>. These lesions may be seen over a wide age range<sup>3</sup>, even in paediatric patients<sup>1,4</sup>. The frequency of PGCG in the mandible is approximately two or three-fold higher than in the maxilla<sup>5</sup>.

Histologically, PGCG shows proliferation of multinucleated giant cells within a background of mesenchymal cells, which is similar to brown tumors<sup>6,7</sup>, a characteristic feature of primary hyperparathyroidism<sup>7</sup>. Primary hyperparathyroidism is caused by hypersecretion of parathyroid hormone due to idiopathic hyperplasia of parathyroid tissue, generally and less commonly parathyroid adenoma and carcinoma<sup>8</sup>. Primary hyperparathyroidism is diagnosed by parathormone and calcium levels in serum<sup>3</sup>.

## **CASE REPORT**

In this case report, clinical, histological findings and periodontal treatment approach were presented of a 11 year-old girl who had localized, painless gingival overgrowth interdentally between teeth #43 and #44. The lesion had initially been noticed by the patient 3 months ago. Bluish-red colored firm structured sessile lesion had no ulceration on its surface (Figure 1). The patient was not using any medication or over the counter remedies that cause gingival overgrowth. There were no local aetiological factors, such as caries, abrasion, food impaction, occlusal trauma in the region related with the lesion. Radiographic findings were normal and there was no alveolar bone destruction (Figure 1).

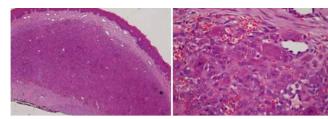
Initial periodontal treatment consisted of oral hygiene instructions, scaling and root planing were applied in 3 sessions with one-week intervals. Four weeks after initial periodontal treatment, the lesion (0,9X0,6X3 cm) was



**Figure 1.** Clinical, radiographical images and excision of the lesion and reconstruction of the wound site with free gingival graft obtained from the hard palate.

excised (Figure 1). Immediately after excision, soft tissue of the wound site was reconstructed using free gingival graft obtained from hard palate (Figure 1). The donor site was covered with a periodontal wound dressing agent and uneventful secondary wound healing was completed at the donor site.

Excised lesion was fixated with 10% formaldehyde solution, embedded into paraffin for histopathological examination and routinely processed <sup>9</sup>. Hematoxylin-eosin stained sections revealed bluish-purple colored nodular proliferation which had multinucleated giant cells and



**Figure 2.** Bluish-purple coloured nodular proliferation which had multinucleated giant cells and fibroblasts under the squamous epithelium. (HE X40, X400)

fibroblasts under the squamous epithelium, all of which were consistent with PGCG (Figure 2). This finding led to the further evaluation of the patient for hyperparathyroidism, which was diagnosed along with hypercalcemia as a result of biochemical analyses in blood sample (Table 1). Elevated parathormone levels triggered alterations in bone remodeling thus, caused hypercalcemia. The patient started to be monitored and a medication was prescribed to reduce serum calcium level by the paediatric endocrinologist.



Due to convenient healing of free gingival graft on the wound site, there was no aesthetical problem. Gingival healing was uneventful without any loss of keratinized tissue width (Figure 3). No recurrence was observed during the 6 month follow-up period.



Figure 3. Clinical image at postoperative 6 months.

	Parathormone (pg/mL)	Calcium (mg/dL)	Phosphorus (mg/dL)
Patient	143	10,5	2,9
Reference Interval	11-67 pg/mL	8,4-10,2 mg/dL	2,7-4,5 mg/dL

**Table 1.** Serum levels of parathormone, calcium and phosphorus

#### **DISCUSSION**

Clinically, PGCG presents as a firm, soft, pedunculated or sessile nodular mass that may be observed in various sizes and colour from dark red to bluish-purple<sup>1,5</sup>. Pain is not a clinical feature of PGCG, as in our case. PGCG is a soft tissue lesion and the underlying bone is rarely affected 1,4. In our case, localized, red or bluish-purple nodular mass in the interdental gingiva between teeth #43 and #44 did not cause interdental alveolar bone loss radiographically.

The differential diagnosis of PGCG must be done with central giant cell granulomas which have very similar clinical and histological features<sup>2</sup>. Differential diagnosis is made on the basis of radiographical examination. Central giant cell granulomas are located in jaw bones and cause aggressive type bone destruction.

The treatment of PGCG includes surgical excision with the elimination of the basement of the lesion<sup>5,10</sup>. Moreover,

local aetiological factors, if present, must be eradicated to avoid recurrence of the lesion<sup>10</sup>. Bone resection and reconstruction need to be performed in cases with alveolar bone destruction<sup>1</sup>. In the present case, the lesion was totally excised from the basement. Immediately after excision, a free gingival graft obtained from hard palate was placed onto the wound site to cover the exposed bone surface and to avoid loss of keratinized tissue width. In this manner, the wound site was primarily closed to reduce postoperative complications like hemorrhage and pain.

Histopathological evaluation of the lesion demonstrated multinucleated giant cells and fibroblasts under the squamous epithelium and these findings were consistent with PGCG. Since giant cell granulomas could be seen as an oral manifestation of hyperparathyroidism<sup>6,7</sup>, and no clinical and radiographic local aetiological factors were present, the patient was consulted with a pediatric endocrinologist for further evaluation of the risk for hyperparathyroidism. As a result of blood sample analyses and clinical examination, primary hyperparathyroidism was diagnosed by the paediatric endocrinologist and treatment was initiated with a prescribed medication.

#### **CONCLUSION**

As reported in this case, manifestations and symptoms of some systemic conditions, diseases and disorders may be observed primarily in the oral cavity. Therefore, the dentists should be aware of oral manifestations of systemic conditions, diseases and disorders like hyperparathyroidism for early diagnosis and refer the patient to appropriate medical departments.

# REFERENCES

- Flaitz CM. Peripheral giant cell granuloma: a potentially aggressive lesion in children. Pediatr Dent 2000; 22:232-3.
- 2. Abu Gharbyah AZ, Assaf M. Management of a peripheral giant cell granuloma in the esthetic area of upper jaw: a case report. Int J Surg Case Rep 2014;5:779-82.
- Alagaratnam S, Kurzawinski TR. Aetiology, diagnosis and surgical treatment of primary hyperparathyroidism in children: new trends. Horm Res Paediatr 2015;83:365-75.
- Pandolfi PJ, Felefli S, Flaitz CM, Johnson JV. An aggressive peripheral giant cell granuloma in a child. J Clin Pediatr Dent 1999;23:353-5.
- Katsikeris N, Kakarantza-Angelopoulou E, Angelopoulos AP. Peripheral giant cell granuloma. Clinicopathologic study of 224 new cases and review of 956 reported cases. Int J Oral Maxillofac Surg 1988;17:94-9.
- Wilson JJ, Schwartz HC, Tehrany GM. Brown tumor of the posterior maxilla as initial manifestation of primary hyperparathyroidism: case report. J Oral Maxillofac Surg 2013;71:886-90.
- Gulati D, Bansal V, Dubey P, Pandey S, Agrawal A. Central giant cell granuloma of posterior maxilla: first expression of primary hyperparathyroidism. Case Rep Endocrinol 2015;2015:170412.
- 8. Spiegel AM. Pathophysiology of primary hyperparathyroidism. J Bone Miner Res 1991;6: 15-7.
- Meseli SE, Agrali OB, Peker O, Kuru L. Treatment of lateral periodontal cyst with guided tissue regeneration. Eur J Dent 2014;8:419-23.
- 10. Kfir Y, Buchner A, Hansen LS. Reactive lesions of the gingiva. A clinicopathological study of 741 cases. J Periodontol 1980;51:655-61.