

Oral Hemangioma with Multiple Phleboliths

Çok Sayıda Flebolitle İzlenen Oral Hemanjiyom

¹Merve Hacer DURAN^a, ²Sümeyye COŞGUN BAYBARS^a

^aDepartment of Oral and Maxillofacial Radiology, Firat University Faculty of Dentistry, Elazığ, Türkiye

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ABSTRACT Hemangioma is a rare, non-reactive, benign vascular tumor, characterized by endothelial cell proliferations and results from vascular malformation. Hemangioma mostly occurs on head and neck skin and regresses spontaneously within 5-10 years in 50-90% of cases. However, oral lesions show less regression than skin lesions. The most commonly affected area is tongue and palatal mucosa, and rarely observed in jaw bones. Diagnosis is based on anamnesis and clinical features. Blanching of the hemangioma under pressure is characteristic clinical finding. Phlebolith is calcified thrombus in veins, venules and sinusoidal walls of hemangiomas. Head and neck phleboliths almost always indicate the soft tissue hemangioma. Phleboliths observed with hemangioma in head and neck region have distinctive appearance on panoramic radiographs. Cone-beam computed tomography provides useful information about location and distribution of phleboliths. In this case report, clinical and radiological features of oral hemangioma and many associated phleboliths are presented.

Keywords: Hemangioma; phlebolith;
cone beam computed tomography

ÖZET Hemanjiyom, endotelial hücre proliferasyonu ile karakterize, non-reaktif, vasküler malformasyon sonucu oluşan iyi huylu ve nadir bir vasküler tümördür. Hemanjiyomlar çoğunlukla baş boyun bölgesinde görülür ve vakaların %50-90'ında 5-10 yıl içinde kendiliğinden geriler. Ancak oral lezyonlar deri lezyonlarına göre daha az gerileme gösterir. En sık etkilenen bölge dil ve damak mukozasıdır ve çene kemiklerinde nadirdir. Tam anamnez ve klinik özelliklere dayanır. Hemanjiyomun basınç altında beyazlaması karakteristik bir klinik bulgudur. Flebolit, venlerde, venüllerde ve hemanjiyomların sinüzoidal duvarlarında bulunan kalsifiye trombüstür. Baş ve boyundaki flebolitler neredeyse her zaman yumuşak doku hemanjiyomuna işaret eder. Baş-boyun bölgesinde hemanjiyomla birlikte izlenen flebolitler, panoramik radyografilerde belirgin bir görünüme sahiptir. Konik ışınli bilgisayarlı tomografi, flebolitlerin yeri ve dağılımı hakkında yararlı bilgiler sağlar. Bu olgu sunumunda oral hemanjiyom ve ilişkili birçok flebolitin klinik ve radyolojik özellikleri sunulmaktadır.

Anahtar Kelimeler: Hemanjiyom; flebolit;
konik ışınli bilgisayarlı tomografi

Hemangioma is the most common benign vascular tumor of infancy and develops due to endothelial cell proliferation. Hemangioma mostly occurs on head and neck skin and regresses spontaneously within 5-10 years in 50-90% of cases. However, oral lesions show less regression than skin lesions. Hemangiomas are divided into superficial and deep subgroups. Superficial ones are defined as capillaries, deep ones as cavernous and mixed. Blanching of the hemangioma under pressure is characteristic finding.

Hemangiomas are more common in women. The most commonly affected area is the tongue and palatal mucosa, and it is rarely observed in the jaw bones. Hemangiomas are usually asymptomatic but there is a bleeding risk. Diagnosis is based on anamnesis and clinical features.¹

Phleboliths are calcified thrombi found in veins, venules or sinusoidal veins of hemangiomas (especially cavernous type). Head and neck phleboliths are almost always sign of hemangioma.²

Correspondence: Merve Hacer DURAN

Department of Oral and Maxillofacial Radiology, Firat University Faculty of Dentistry, Elazığ, Türkiye
E-mail: mhduran@firat.edu.tr



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RADIOGRAPHIC CHARACTERISTICS OF PHLEBOLITHS

Phleboliths are defined oval/round structures with concentric radiolucent or radiopaque laminations on planar radiographs.² In maxillofacial region, phleboliths are usually observed multiple, randomly distributed radiopaque structures with various shapes and sizes. Conventional radiographs such as panoramic radiography (OPG) are useful for detecting phleboliths but they have some limitations include superpositions and magnifications in structures.³ In cone-beam computed tomography (CBCT), phleboliths are round or oval-shaped, well-circumscribed lesions can increase up to 6 mm in diameter. The internal structure may be homogeneously radiopaque but commonly has layered appearance which giving phlebolith “bull’s-eye” or “target-like” appearance.²

CASE REPORT

59-year-old, healthy female patient applied to our faculty for dental treatment. Extraoral examination revealed a mass located on lower left side of face between lip corner and lower mandibular border, painless on palpation, diffuse and causing malformation. Facial asymmetry and bluish-purple discoloration of skin were present. There was no pulsation and lymphadenopathy. It was learned that lesion was noticed at birth and its size increased with age (Figure 1).

Intraoral examination revealed hemangioma located towards ramus from involved buccal mucosa with diffuse swelling, bluish-purple discoloration. Lesion was moderately-firm and non-pulsative. There was no pain on palpation (Figure 2).



FIGURE 1: Hemangioma causing bluish-purple skin discoloration, malformation and facial asymmetry.

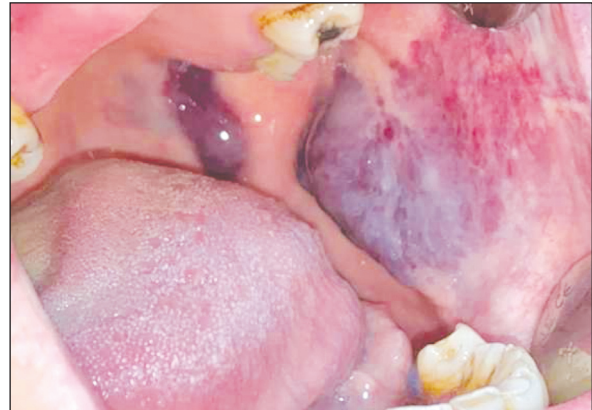


FIGURE 2: Hemangioma with diffuse swelling and bluish purple discoloration from the buccal mucosa to the ramus.

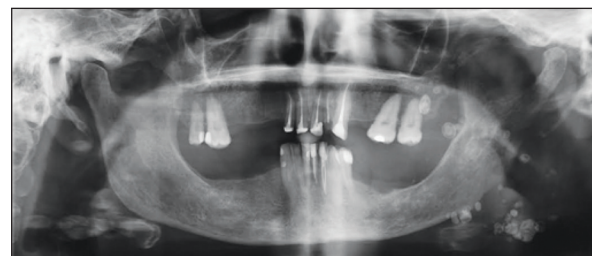


FIGURE 3: Multiple, irregular, target-like radiopaque calcifications in the left mandible and maxilla on panoramic radiography.

OPG showed multiple, target-like radiopaque calcifications in left mandible and maxilla that irregularly located, varying 3-6 mm in diameter (Figure 3). Calcifications were thought to be lymph node calcifications or sialolith, which other calcifications likely seen in same region. Due to presence of oral hemangioma and multiple, irregularly distribution of layered calcifications, preliminary diagnosis of phlebolith and differential diagnosis with other calcifications was made.

Localization, number, shape, distribution, and type of calcification must be accurately evaluated to make accurate diagnosis and determine the treatment requirement. Thereby, three-dimensional CBCT was taken and analyzed in coronal, axial and sagittal sections. On CBCT sections, layered lesions with round-oval shape, 3-6 mm in diameter, regular borders and target-like appearance were determined. Multiple phleboliths were confirmed in CBCT (Figure 4).

The patient had no functional or aesthetic complaints, she was followed-up. Informed consent was obtained that her records could be used in research.

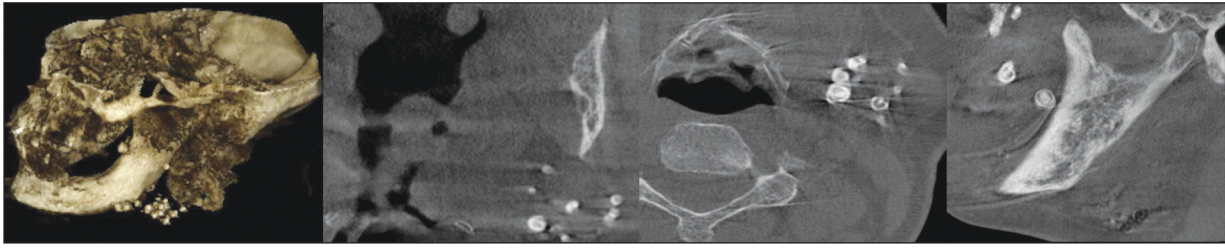


FIGURE 4: Round and oval shaped, 3-6 mm in diameter with regular borders and target-like appearance of phleboliths in coronal, axial, sagittal cone-beam computed tomography sections and three-dimensional reconstruction.

DISCUSSION

Hemangioma is the most common soft tissue tumor of childhood.⁴ Its incidence in head and neck is 60%, also in Caucasian race, women, low birth weight, premature babies are more likely to be seen. Research have shown that its incidence increases in conditions such as advanced maternal age, multiple pregnancy, placental anomalies and preeclampsia.⁵ Hemangiomas cause symptoms include swelling, pain and color change.⁶ Hemangioma shows pigmentation, fluctuation and pulsation. Since hemangioma is vascular lesion, turns white when pressure is applied on.⁷ In our case, there was a hemangioma caused diffuse swelling and bluish-purple discoloration on left cheek and oral mucosa.

Phleboliths are calcifications formed by precipitation of calcium salts after thrombus organization which formed as a result of occlusion in vein/venules.⁷ In our case, hemangioma was observed with multiple phleboliths. Multiple, round/oval, bull's eye or target-like lesions are typical radiographic appearance of phleboliths. Sivrikaya et al. reported a multiple giant phleboliths case and reviewed literature, they stated the phlebolith diameters were between 1-60 mm.⁸ Phleboliths can be seen in magnetic resonance imaging (MRI), ultrasonography (USG), computed tomography (CT) and CBCT.⁷ Contrast-enhanced CT is excellent to reveal phleboliths.⁹ Hemangiomas usually appear as masses with prominent phleboliths on tomography. Hemangiomas show hyperintensity on T2-weighted and isointensity on T1-weighted MRI.^{9,10} Phleboliths appear nodular structures without contrast on T1-weighted MRI with contrast and hyperintense structures on T2-weighted

MRI.¹¹ USG is inexpensive, noninvasive method and phleboliths are seen as multiple hyperechoic areas within the mass.²

Today, CBCT is widely used for maxillofacial imaging due to its lower radiation dose and high resolution compared to CT. Sometimes harmless calcifications can be misdiagnosed, causing unnecessary treatment and costs to patients. The most important criteria in diagnosis of calcifications are: location, distribution, number and shape. Dystrophic calcification in certain tissues may indicate systemic condition and may be indicative of potentially more threatening condition.¹¹ Therefore, phleboliths detected on OPG were examined in more detailed CBCT.

Many treatment modalities have been advised for hemangiomas include cautery, cryotherapy, radiotherapy, sclerosing agents. Sclerosing agents have high response rate, cost-effective, easily obtain. Intralesional injection of 3% sodium tetradecyl sulphate has been used many years for varicose veins, hemorrhoids and hemangioma treatment.¹² Oral propranolol was used to treat heart failure in patients with hemangioma, regression of hemangioma was observed incidentally and led to use of this drug in hemangioma treatment. Subsequently, case reports and trials have been published, it has been preferred as first-line therapy because of its effectiveness.¹³ Reis et al. evaluated the efficacy of topical beta-blockers in patients with superficial hemangioma and stated that topical timolol is promising because its easy to use, systemic side effects are rarely seen and it causes significant regression.¹⁴ Since our patient did not have functional and aesthetic complaints, she was followed-up.

In conclusion, phleboliths observed with hemangioma in head and neck are soft tissue calcifications have distinctive appearance on panoramic radiographs. CBCT provides useful information about location and distribution of phleboliths, as its cost-effective, has lower radiation dose and higher spatial resolution than CT.

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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

All authors contributed equally while this study preparing.

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