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HEMANGIOMA PRESENTING WITH MULTIPLE PHLEBOLITHS: CASE REPORT WITH CBCT FINDINGS

Multiple Flebolit Görülen Hemanjiyom: KIBT Bulguları ile Olgu Sunumu

Hazal KARSLIOGLU, A.Pınar SUMER

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ABSTRACT

Hemangiomas are benign tumors of infancy that display a rapid growth phase with endothelial cell proliferation. Phleboliths are calcified thrombi found in veins, venulae and sinusoidal vessels of hemangiomas. In the head and neck, phleboliths nearly always signal the presence of a hemangioma. Hemangioma with multiple phleboliths is described including its features on panoramic radiography and cone beam computed tomography images.

Keywords: cone beam computed tomography, hemangioma, phlebolith

ÖZ

Hemanjiyomlar, endotel hücrelerinin proliferasyonu ile karakterize bebeklik çağının benign tümörleridir. Flebolitler, hemanjiyomların damar, venül ve sinüzoid damarlarında bulunan kalsifiye trombüslerdir. Baş ve boyunda görülen flebolitler, hemen hemen daima hemanjiyomun varlığını işaret eder. Multiple flebolit bulunan hemanjiyom olgusu, panoramik radyografi ve konik ışını bilgisayarlı tomografi görüntüleri ile sunulmuştur.

Anahtar kelimeler: Flebolit, hemanjiyom, konik ışınlı bilgisayarlı tomografi

INTRODUCTION

Hemangiomas are benign vascular anomalies characterized by an increase in turnover and proliferation of endothelial cells. According to histopathologic features of hemangiomas, capillary, cavernous, mixed types are available.¹ Hemangiomas can not be recognized at birth but arise subsequently during the first 8 weeks of life. They are the most common tumors of infancy, occurring in %5 to %10 of 1 year old children.² The average age at diagnosis is 10 years, with 65% occurring in the first 2 decades of life.³ Hemangiomas occur more frequently in females than in males.^{2,3} The most common location is head and neck.² The precise cause of hemangioma is not known, but, either hormonal changes and traumas have been postulated as being involved.^{4,5} The predominant complaint is presence of a slow-growing palpable mass that can fluctuate. The skin overlying the show hemangioma will often increased vascularity, giving the hemangioma a bluish tint. Clinical manifestations such as swelling, pain, discoloration, pulsation, compressibility, thrills, bruits can be seen.⁶⁻⁸ Phlebolith responds to auscultation in the hemangioma of the cavernous type if it originated from a vein or soft tissue hemangioma.⁹ Increasing venous pressure, changes the size of the hemangioma. Masses in the head and neck hemangiomas enlarge during conditions that increase venous pressure, such as crying, laughing or performing handstands.¹⁰ Changes in blood flow in hemangiomas may induce phlebolith formation. Phleboliths are calcified thrombi found in veins, venulae and sinusoidal vessels of the especially cavernous type hemangiomas. The phlebolith comprise a mixture of calcium carbonate and calcium phosphate salts.³ They are in the idiopathic calcification classification that occurs in normal tissues despite normal serum calcium and phosphate levels. They are usually multiple, varied in size, randomly distributed. Phleboliths do not show any symptoms and they may be found during routine imaging. Although standard radiographs are an important

diagnostic tool for the diagnosis of phleboliths in the mass, computerized tomography (CT), magnetic resonance imaging (MRI) and ultrasonography play an important role in the diagnosis of hemangioma.¹¹ MRI is very useful for the detection of vascular lesions^{10,12}, but the detectability of phleboliths in CT is greater than that of MRI.¹³ Phleboliths usually have a lamellar appearance such as bull's eye but they can also have a homogenous radiopaque image with an oval or round shape, approximately 6 mm in diameter.³

The presence of phleboliths demonstrated on panoramic radiography is rarely described in the literature. The following report presents the first reported case of multiple phelobolits with cone beam CT and panoramic radiography in a 37 years old female patient with head and neck hemangioma.

CASE REPORT

A 37 year old woman patient referred to dentomaxillofacial radiology clinic, with a complaint of severe pain in the right mandibular posterior area occurred after the first molar tooth extraction. The patient's medical and family history was unremarkable. Extraoral examination revealed hemangioma on the right side that had been presented since 2 years old and gradually increased in size. A diffuse right facial swelling caused facial asymmetry was visible, occupying the buccal soft tissue area. There was a bluish discoloration of the overlying face skin (Figure 1).



Figure 1: The appearance of Hemangioma as extraoral.

Intraoral examination showed discoloration of oral mucosa in the related region (Figure 2).



Figure 2: The appearance of Hemangioma in buccal and palatinal mucosa.

Palpation indicated that the lesion was nonpulsatile. Bruit that is heard with stethoscope, did not reveal on auscultation of head and neck. Lymphadenopathy was not Panoramic presented. radiographic examination revealed radiopacities in variable sizes from 2-8 mm characteristic of phleboliths (Figure 3).



Figure 3: A panoramic radiograph showing multiple round target-like radiopacities varying in size from 2 to 8mm (arrows)

Cone beam CT images showed the existence of multiple phleboliths (Figure 4,5). The patient had no complaint such as pain or aesthetic discomfort about hemangioma. She was only enrolled for treatment of alveolitis.



Figure 5A. The appearances of phleboliths on axial cone beam CT image



Figure 5B. Sagittal cone beam CT image



Figure 4: Cone beam CT images on the coronal plane show multiple calcifications consistent with phlebolith (arrows).



Figure 5C. Cross-sectional cone beam CT image (C) (arrows).

DISCUSSION

Hemangiomas are benign vascular lesions that cause symptoms such as mass, swelling, pain and discoloration.⁶ They occur on both skin and mucosal surfaces frequently in the head and neck region² but are rare in the oral cavity.¹⁴ Presented case was unilaterally seen in the head and neck skin area and in the oral cavity mucosa.

Phleboliths are calcified thrombi found within vascular channels and occur frequently in the presence of hemangiomas or vascular malformations. They arise from injury to a vessel wall or result from stagnation of the flow of blood. Phleboliths generally cause no symptoms.

Radiologically, they are seen either radiolucent or radiopaque. A fibrinous component is attached to the developing phleboliths and becomes calcified. Repetition of the process causes a layering effect and so phlebolith usually has a concentric ring or onion like appearance.¹⁵

Characterization and spread of tumors can only be recognized by CT or MRI. CT with contrast is an excellent imaging technique for revealing phleboliths.⁵ Hemangiomas are usually seen as masses with well-defined phlebolith on the tomography. MRI can produce high signal intensity representing blood, in addition focal heterogenities representing areas of thrombosis, fibrosis or calcifications. Hemangiomas show hyperintensity on T2-weighted images and isointensity on T1-weighted images.10,16 Phleboliths were seen as a nonenhancement nodular structures in contrast-enhanced T1weighted MR scans and hypointense structures in T2-weighted MR scans.¹⁰ Ultrasonography is a cheap and noninvasive method and phleboliths were seen on imaging as multiple hyperechoic areas within the mass.³ In this case report cone beam CT was used for detailed examination of phleboliths with lower radiation dose and cost, significantly higher compared spatial resolution with CT. According to our knowledge, the present case report is a unique one evaluating phleboliths using cone beam CT that can provide useful information about location and extent of the phleboliths.

Head or neck phleboliths must be differentiated from other calcifications, such as sialoliths, calcified lymph nodes, foreign bodies, tonsilloliths, atherosclerotic plaques in the carotid artery, healed acne lesions, cysticercosis, and miliary skin osteomas, that occur in the same area.¹⁵ Sialoliths appear on a single line when seen more than one, whereas distributed. randomly phleboliths are Phleboliths usually coexist with hemangiomas.³

The treatment of the hemangiomas is based on location, accessibility, depth of invasion, age, and cosmetic appearance. Treatment modalities include the use of corticosteroids, interferon alfa 2a or interferon alfa 2b, ligation of the feeding vessels, cryotherapy, embolization and sclerotherapy, fibrous agents and lasers.^{1,5} Periodic observation is also an alternative treatment.¹¹ In the present case, since the patient does not have any aesthetic or functional complaints, she is being followed.

In conclusion, panoramic radiography can show presence of phelobolits and cone beam CT can provide useful information about the location and extention of the lesion.

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REFERENCES

1. Allen PW, Enzinger FM. Hemangioma of skeletal muscle: An analysis of 89 cases. Cancer. 1972;29:8-22.

 Neville BW, Damm DD, Allen CM, Bouquot JE. Oral and Maxillofacial pathology.
 3rd ed. St. Louis: Elsevier; 2009.

3. White SC, Pharoach MJ. Oral Radiology Principles and Interpretation. 4th ed. Philadelphia: Mosby Inc; 2000. **4.** Rossiter JL, Hendrix RA, Tom LW, Potsic WP. Intramuscular hemangioma of the head and neck. Otolaryngology Head Neck Surg. 1993;108:18-26.

5. Wolf GT, Daniel F, Krause CJ, Kaufman RS. Intramuscular hemangioma of the head and neck. Laryngoscope. 1985;95:210-3.

6. Aynalı G, Unal F, Yarıktaş M, Yasan H, Ciriş M, Yılmaz О. Submandibular hemangioma with multiple phleboliths mimicking sialolithiasis: the first pediatric case. Kulak Burun Bogaz Ihtis Derg. 2014;24:168-71.

7. Addante RR, Donovan MG. Right facial mass. J Oral Maxillofac Surg. 1994;52:1061-5.
8. Tsang WM, Tong AC, Wu PC. Cavernous hemangioma of the masseter muscle. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 1998;85:629-30.

9. Yıldırım D, Bilgir E. Baş Boyun
Bölgesindeki Yumuşak Doku Kalsifikasyon
Ve Ossifikasyonlari J Dent Fac Atatürk Uni.
2015;13: 82- 90.

10.Zengin AZ, Çelenk P, Sumer AP. Intramuscular hemangioma presenting with multiple phleboliths: a case report. Oral Surg Oral Med Oral Pathol Oral Radiol. 2013; 115:e32-6.

11.Altuğ HA, Büyüksoy V, Okçu KM, Doğan N. Hemangiomas of the head and neck with phleboliths: clinical features, diagnostic imaging, and treatment of 3 cases. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2007;103:e60-4

12.Gold L, Nazarian LN, Johar AS, Rao VM. Characterization of maxillofacial soft tissue vascular anomalies by ultrasound and color Doppler imaging: an adjuvant to computed tomography and magnetic resonance imaging. J Oral Maxillofac Surg. 2003; 61:19-31.

13.Kakimoto N, Tanimoto K, Nishiyama H, Murakami S, Furukawa S, Kreiborg S. CT and MR imaging features of oral and maxillofacial hemangioma and vascular malformation. European J Radiol. 2005;55:108-12.

14.Kripal K, Rajan S, Ropak B, Jayanti I. Cavernous hemangioma of the tongue. Case Rep Dent. 2013; doi: 10.1155/2013/898692.

15.Mandel L, Perrino MA. Phleboliths and the vascular maxillofacial lesion. J Oral Maxillofac Surg. 2010;68:1973-6.

16.Yonetsu K, Nakayama E, Yuasa K, Kanda S, Ozeki S, Shinohara M. Imaging findings of some buccomasseteric masses. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 1998;86:755-9.

Corresponding Author

Hazal Karslioglu, DDS

Ondokuz Mayis University

Faculty of Dentistry

Department of Dentomaxillofacial Surgery

55139 Kurupelit, Samsun TURKEY

Tel : +90 (362) 3121919-8150

Fax : +90 (362) 4576032

E-mail : <u>hazallllk@hotmail.com</u>