International Journal of Medical and Dental Case Reports (2018), Article ID 140618, 4 Pages

CASE REPORT

Incidental-imaging finding in the practice of oral diagnosis/oral medicine. A case report: Phleboliths in the maxillofacial region with literature review

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Received 12 April 2018;
Accepted: 01 June 2018

doi: 10.15713/ins.ijmdcr.88

How to cite the article:
Carrera JF, Mupparapu M, Bugueno JM. Incidental-imaging finding in the practice of oral diagnosis/oral medicine. A case report: Phleboliths in the maxillofacial region with literature review. Int J Med Dent Case Rep 2018;5:0-0.

Abstract

Aim: This case describes multiple phleboliths found incidentally during a routine dental examination. **Background**: Phleboliths are pathological entities that are often associated with hemangiomas or vascular malformations in the maxillofacial region. **Case Report**: A 56-year-old female presented for comprehensive dental examination. Extraoral examination showed no facial skin abnormalities, lymphadenopathy, or salivary gland enlargement. Intraoral examination disclosed normal oral tissues, partially edentulous jaws, multiple dental restorations, recurrent caries, and marginal periodontal disease. Dental radiographs were obtained including an orthopantomograph, which revealed multiple well-defined, rounded radiopaque masses with laminated appearance, approximately 6–8 mm in diameter, located on the right side, and mainly superimposed on the coronoid process of the mandible. Magnetic resonance imaging with contrast confirmed the presence of low signal intensity foci suggestive of phleboliths, involving the inferior aspect of the right temporalis muscle, and extending anteriorly to the lateral pterygoid muscle within the retro antral fat of the right face, most compatible with a slow flow vascular malformation. Computed tomography angiography of head and neck with contrast verified the presence of multiple phleboliths involving the muscles of mastication on the right face and with no evidence of hemodynamically significant stenosis of the cervical and head blood vessels.**Conclusion**: Phleboliths in the maxillofacial area are mostly asymptomatic, and could represent the presence of a serious vascular anomaly. **Clinical Significance:** Dentists should be aware of these calcified bodies to avoid the risk of hemorrhage during oral and maxillofacial treatments. The clinician must be vigilant when considering, particularly, surgical procedures in the head and neck region.

Keywords: Incidental findings, oral medicine, vascular malformation

Introduction

Panoramic radiographs are valuable diagnostic tools that present a general view of the jaws and contiguous elements and are routinely used in a comprehensive dental examination. These radiographs may also display structures, anatomical variations, or alterations that, in asymptomatic patients, might have an important clinical significance. These are general referred to as incidental findings and may cause clinical problems and affect the individual's oral and general well-being.[1]

Some of the most common of such findings detected during routine radiographic imaging include calcifications of the stylohyoid ligaments as well as carotid arteries, phleboliths, sialolithiasis, and tonsilloliths.[1] Phleboliths are calcified thrombi located in blood vessels, frequently with the occurrence of hemangiomas or vascular malformations. Alterations in blood flow dynamics may originate thrombus formation, which eventually are calcified and produce phleboliths.[1,2] They are generally numerous, variable in size, and produce no subjective symptoms.[2-4]

To the best of our knowledge, phleboliths displaying on panoramic radiography are not often reported in the literature, and possibly no former study has incorporated plain radiography, magnetic resonance imaging (MRI) and computed tomography (CT) angiography of head and neck with phleboliths in the maxillofacial region.[4,5] This case describes the presence of multiple phleboliths involving some muscles of mastication, emphasizing the characteristics apparent in plain radiography, MRI, and CT angiography.

Case Report

A 56-year-old Hispanic female presented to the Department of Diagnostic Sciences and Oral Medicine, College of Dentistry, at the University of Tennessee Health Science Center for comprehensive dental examination. Her chief complaints were caries and oral hygiene. Her medical history included hypertension, hypercholesterolemia, and hypothyroidism. Her medications included Pravastatin, Lisinopril, Levothyroxine, and Amlodipine. The patient reported no allergies. Her family history was non-contributory and she denied current use of tobacco, alcohol, and recreational drugs. The review of systems was within normal limits. Physical examination revealed a well-nourished woman in no apparent distress. Extraoral examination showed no signs of facial skin lesions, lymphadenopathy, or salivary gland enlargement. Gross cranial nerve exam was normal. Intraoral exam revealed normal oral soft tissues, partial edentulous maxillary and mandibular areas, multiple dental restorations, recurrent caries, and generalized periodontal disease. A set of bitewing, periapical, and panoramic radiographs were taken to complement the clinical examination. The panoramic radiograph revealed multiple well-defined, rounded radiopaque masses with laminated appearance, approximate 6–8 mm in diameter, located on the right facial side, mainly superimposed to the coronoid process of the mandible [Figure 1]. An MRI with contrast was indicated for additional evaluation and diagnosis that confirmed the presence of low signal intensity foci suggestive of phleboliths, involving the inferior aspect of the right temporalis muscle, and extending anteriorly to the lateral pterygoid muscle within the retroantral fat of the right face, most compatible with a slow flow vascular malformation [Figures 2 and 3]. Then, A CT angiography of head and neck with contrast was taken verifying the presence of multiple phleboliths involving the muscles of mastication on the right face and with no evidence of hemodynamically significant stenosis of the cervical and head blood vessels [Figure 4]. The patient was instructed to have the indicated dental treatment, which was mostly conservative.

Discussion

Phleboliths are calcified structures that, usually, present as incidental findings, and by themselves produce no subjective symptoms; therefore, they may be found during routine imaging or during studies of vascular lesions.[2,4,5] Occurrence of multiple calcified thrombi (phleboliths) is considered pathognomonic for hemangiomas and vascular malformations in the oral and maxillofacial region.[6,7] However, they may be the only remaining sign of a childhood vascular lesion encountered in adults and are more distinctive of low-flow vascular abnormalities.[3] Vascular anomalies usually comprise two distinct clinical and histopathological conditions: Hemangioma and vascular malformations.[3] Hemangioma is the most frequent benign soft tissue neoplasia of childhood, usually occurring in the first three decades of life, and evolving as newly developed vessels from hyperplastic endothelial cells through the action of angiogenesis.[3,4] Intramuscular hemangiomas (IMHs) are congenital lesions that represent just 1% of all hemangiomas. 15% of IMHs are located in the head and neck region, being the

Figure 1: Digital panoramic radiography shows multiple phleboliths on the right mandibular side

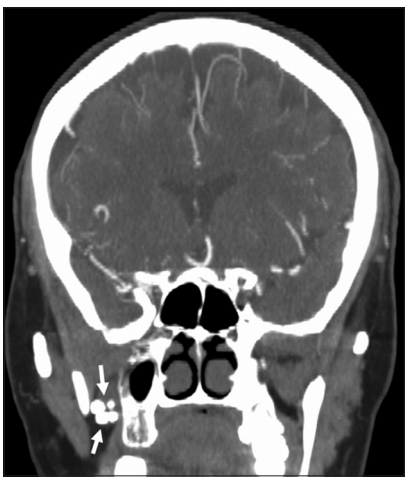

Figure 2: Contrast enhanced T1W coronal images showing the phleboliths in the vicinity of the right lateral pterygoid muscle

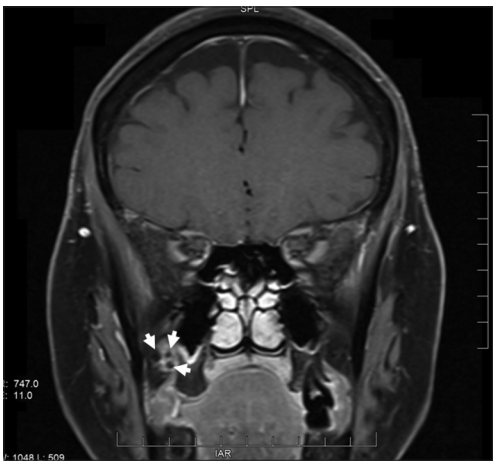

Figure 3: Contrast enhanced T1W coronal magnetic resonance imaging showing the phleboliths within the hemangioma on the right side in the pterygomaxillary fissure

masseter, and/or buccinator muscles the most common sites. Phlebolith formation within IMH occurs in approximately 25% of cases, and the presence of multiple phleboliths might be due to the long-term presence of hemangioma and stagnant blood.[8]

Vascular malformations can be high-flow anomalies of arterial and arteriovenous defects and slow-flow lesions, involving capillary, lymphatic, and venous malformations (VMs). Nearly 40% of VMs are localized in the head and neck region, and these are sometimes related with the presence of phleboliths.[1,3]

The pathogenesis of phlebolith begins when variations in blood flow dynamics, within vascular malformation, cause thrombus development.[9] Thrombi which are fixed to the vascular walls, consecutively undergo a slow calcification process with calcium carbonate and calcium sulfates deposited within, forming the core of the phlebolith. This process of calcification continues and extends to the periphery with concomitant lamellar fibrosis giving rise to histological onion ring-like appearance. The replication of this process conducts to the growth of the phlebolith, which may eventually cause symptoms in cases of constant blood stasis as in vascular malformations.[3,6-8] Differential diagnosis of phleboliths in the head and neck area includes sialoliths, tonsillolihts, atherosclerotic plaques in the carotid artery, healed acne lesions, calcified lymph nodes, cysticercosis, and military skin osteomas.[2-5]

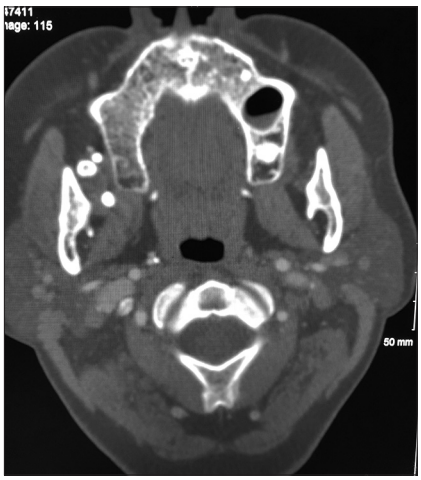

Figure 4: Computed tomography angiography axial image showing multiple round, well-corticated phleboliths situated between the maxilla and mandibular ramus

Studies about hemangiomas and venous abnormalities connecting imaging methods have been reported in the literature intending to improve the diagnosis of these changes and the presence of phleboliths.[1] Previous cases reported in the literature, during the past 15 years, have showed the presence of these calcified bodies in different vascular anomalies in the maxillofacial region [Table 1][2-10] Plain X-rays represent a valuable diagnostic tool in diagnosing phleboliths within hemangiomas or vascular malformations because of the characteristic aspect of the calcified bodies. Radiographically, they appear as spherical and oval laminated forms that have either a radiolucent or a radiopaque center, and repetition of this calcification produces an onion-like appearance or concentric rings.[3,4] Other diagnostic imaging techniques such as CT, MRI, and ultrasound increase the accuracy and have a substantial function in the pre-operative diagnosis of IMHs. Alterations in the masseter muscle can be diagnosed reliably using ultrasound. MRI is considered the most convenient imaging modality for tissue characterization and detection of the extent of vascular lesions, with its excellent spatial resolution in soft tissue assessment and the benefit that no radiation is involved. Overall, vascular malformations and hemangiomas demonstrate hyperintensity on T2-weighted images and isointensity on T1-weighted images due to the enhanced free water present within stagnant blood in the vessels.[4,5] However, phleboliths are not clearly identified in MRI film because of their very subtle signal intensity. They are best recognized on plain radiograph and CT scan.[1] Like in our case, most phleboliths in the VMs of the head and neck region are asymptomatic, and consequently need no treatment. However, surgical removal of phleboliths might be indicated to achieve symptom relief, if they contribute to the aggravation of symptoms of VM such as pain, stasis, inflammation of the nearby tissues, and foreign body sensation.[5,7] Definitely,

Table 1: Previous cases of phleboliths in the literature from 2003 to 2017

Presence of phleboliths	Reference	Gender	Age	Site
VA	Scolozzi *et al*., 2003	F	92	Retromolar trigone, BM
IMH	Kanaya *et al*., 2008	F	14	Cheek
VM	Su *et al*., 2009	F	14	SM
IMH	Mandel *et al*., 2010	F	56	Buccinator-masseter
VA		F	64	Tongue
IMH	Zengin *et al*., 2013	F	21	Masseter
VA	Eivazi *et al*., 2013	20 F 8 M	7-72	Cheek, SM, eyelid, neck
IMH	Kamatani *et al*., 2014	F	51	Tongue
Non-vascular	Nagaraja *et al*., 2016	M	49	Cheek
VM	Uzun *et al*., 2017	F	19	Tongue, masseter, SCM

IMH: Intramuscular hemangioma, VA: Vascular anomaly, VM: Venous malformation, SCM: Sternocleidomastoid, SM: Submandibular, BM: Buccal mucosa

the removal of phleboliths alone does not treat the VM, and, in many cases, a complete removal of the VM is not possible.[5,7] The therapeutic method is based on clinical factors such as age, cosmetic appearance, lesion size, depth of involvement, the vicinity of vital structures, and distinctness of the lesion margin. Many forms of therapy for IMHs or VMs have been suggested including recommendation of low-molecular-weight heparin, sclerotherapy, laser therapy, or surgical volume reduction if appropriate.[4,5,7,8] While a particular procedure can be enough for some smaller, well-contained lesions, for large VM a thorough surgical excision is needed, which has a higher risk, and generally requires a multimodality approach. Even with this approach, local recurrence rates fluctuating from 9% to 28% have been reported.[7] Minimal feeding vessels and residual tumor are accountable for this recurrence rate. In addition, in some cases of IMHs, entire excision of the masseter muscle has been suggested.[4] In our case, the condition and treatment alternatives were discussed with the patient; however, she opted for declining treatment at this time since was completely asymptomatic.

Conclusion

Characteristic clinical features of vascular malformations may not be fully evident in intramuscular or deeply positioned lesions in the maxillofacial region. Phleboliths, in this region, are usually associated to vascular anomalies and might be incidental findings during routine imaging, like in our case.

Clinical Significance

Dentists should be aware of the presence of these calcified bodies, and great care should be taken to prevent the risk of hemorrhage during dental treatment. The clinician must be certain of the kind and the expansion of the vascular lesions when considering surgery.

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