Hemangioma of the face and neck with phleboliths: Diagnostic imaging with X-ray and CT

Abstract:
Hemangiomas found in the head and neck regions can cause functional and aesthetic complications. The therapeutic approach to this condition presents both challenges and risks, mainly when surgical procedures must be performed due to the risk of hemorrhaging. In this context, the need for tooth extraction can represent a factor of great concern. The present study describes the case of extensive cavernous hemangioma, associated with a large quantity of phleboliths, affecting the orofacial and neck region in an adult woman. Embolization and controls were performed as a means of surgical treatment.

Keywords: Hemangioma; Cone-Beam Computed Tomography; Angiography; Diagnosis.
INTRODUCTION

Hemangiomas are benign vascular lesions of the soft tissue characterized by the abundant proliferation of blood vessels, which is quite common in children but can also affect adults.\(^1\) In the orofacial region, this type of lesion can affect the skin, muscles, and salivary glands.\(^3\) Bone deformity and skeletal hypertrophy can also be associated with hemangioma; however, intraosseous invasion is rare.\(^3\) This condition’s pathogenesis is unknown and commonly appears as a congenital anomaly but can also result from traumas and hormonal alterations.\(^2\)

Changes in the blood flow within hemangiomas often result in thrombi and phleboliths.\(^7\) Hemangioma in the head and neck is rarely associated with a large number of phleboliths; what is common are 2 to 6 and a maximum of 20 phleboliths in a given hemangioma.\(^9\) Thus, the present study sought to illustrate clinical, radiographic, and tomographic characteristics of an extensive hemangioma, associated with a large quantity of phleboliths located in the head and neck.

CASE REPORT

A 21-year-old healthy female patient was referred to the radiology clinic for an imaging study of an intraoral lesion. The patient needed to undergo the extraction of impacted mandibular left third molar; however, the lesion extended into the tongue region of the left mandibular molars. The extraoral exam, upon palpation, showed a firm facial and cervical swelling with a rubbery consistency, which was painless and which confirmed a severe facial asymmetry on the left side. No medical history of a facial or cervical trauma was reported before the appearance of swelling. The skin overlying the swelling presented normal aspects, with no discoloration. Digital palpation revealed no fremitus or pulsations. The intraoral exam showed a clearly defined extensive bluish lesion that extended to the tongue, oral mucosa, lips, pharynx, floor of the mouth, and soft palate, containing characteristic aspects of a hemangioma. No associated lymphadenopathy was identified. The dentition appeared to be in good condition (Fig. 1).

For an initial imaging evaluation of the lesion, a panoramic radiograph, as well as posteroanterior and lateral cephalometric, were taken using a KODAK 8000C Digital Panoramic and Cephalometric System (Eastman Kodak, Rochester, NY) appliance, at 140 kHz, 60-90 kV and 2-15 mA. The panoramic radiograph and the cephalometrics showed an alteration in the curve of the lower border of the mandibular body and in the trajectory of the mandibular canal on the left side, confirming a skeletal mandibular asymmetry in the patient. Many circular and oval bodies, varying in size, randomly distributed, radiopaque in the periphery and radiolucent in the center, with a characteristic aspect of a “target” and suggestive of phleboliths, were observed in the left mandibular and cervical region of the patient, apparently located only the soft tissue (Fig. 2).

It was necessary to perform a Computed Tomography (CT) to determine the exact location of the calcified structures and their relationship with the adjacent anatomical structures, as well as to provide a better evaluation of the extension of the vascular lesion. The CT images were taken using a multislice CT device with 64 detectors (Somatom Sensation 64; Siemens Medical Solutions, Erlangen, Germany). To acquire these images, the following factors were used in the multislice CT: 5s, 120 kV, 150 mAs, collimation of 64 x 0.6 mm, thickness of 0.5 mm, resolution of 0.4 x 0.4 x 0.4 mm\(^3\). The CT images showed an increase in the volume of soft tissues in the left mandibular region, as well as a large quantity of phleboliths, of varying sizes, in the submandibular region, extending to the base of the tongue, as well as to the parotid space on the left side, with a cavernous hemangioma aspect fed mainly by the branches of the lingual artery (Fig. 3). Bone rarefaction was observed adjacent to tooth 38 and a bone defect was found in the lingual cortical of the mandibular body, which were suggestive of an osteolytic lesion resulting from a vascular lesion. Also observed was a bone defect in the vestibular cortex of the mandible, in the left mental foramen, also caused by the lesion.

The CT study conducted by means of an intravenous injection of non-ionic iodinated contrast, computed angiotomography, revealed a large hemangioma on the...
left side, extending from the submandibular region to the left curve of the mandible, containing many phleboliths of varying sizes (Fig. 4). A patent aortic arch, of normal course and caliber, was also observed, as were its main branches.

The mental foramen and the mandibular canal showed an increase in their dimensions on the left side. No exuberant vascularization was identified in the alveolar bone tissue adjacent to tooth 38, which had been impacted. An exacerbation (increase) in the vascularization of the gingival tissue adjacent to tooth 38 was observed, especially in the tongue region.

The CT also presented hypertrophy of the masticatory muscles on the left side as compared to the right side, a lack of definition of the oropharyngeal tissues and an increase in the dimensions of the soft tissues in this region, with the lesion extending into the parapharyngeal region. Its clear prominence brought about by the contrast showed that the hemangioma extended in the direction of the skull, occupying the entire masticatory space and extending into the pterygoid fossa. No presence of arteriovenous fistulas were found within the hemangioma.

**DISCUSSION**

Although they are benign, hemangiomas can cause both functional and aesthetic complications and even pose a threat to life if not properly treated during childhood.  

Hemangiomas are classified as capillary, cavernous, and mixed, based on the size of the blood vessel and the histological characteristics.  

Cavernous hemangioma contains large blood vessels (> 140 µm) and aligned endothelial vascular canals. Capillary hemangioma is characterized by the proliferation of capillaries and contains smaller blood vessels (< 140 µm).
Cavernous hemangioma is less common than the capillary hemangioma and tends to be chronic with slow growth.\textsuperscript{11} The majority of hemangiomas (68\%) found in the head and neck regions are capillary.\textsuperscript{12} In this clinical case, through the clinical and radiographic data, the patient was diagnosed as presenting an intramuscular cavernous hemangioma.

The presence of phleboliths suggests the diagnosis of hemangioma\textsuperscript{2} Changes in the blood flow within the hemangioma result in thrombi and phleboliths.\textsuperscript{7,8} Phleboliths consist of calcified nodular structures and can be seen in x-rays.\textsuperscript{2} Phleboliths are uncommon in the oral region,\textsuperscript{13} and, as they are asymptomatic, are normally found accidentally in routine imaging exams or during the study of vascular lesions. However, once identified, it is necessary to conduct a differential diagnosis with other calcifications of the head and neck region, such as: sialoliths, tonsilloliths, calcified lymph nodes, atheroma, healed acne lesions, cysticercosis, and miliary skin osteomas.\textsuperscript{7}

In most cases, no spontaneous regression of the hemangioma occurs, but an individualized therapeutic approach is necessary. The treatment of the hemangioma is based on its location, growth pattern, access, and extension of the lesion, as well as the impact on aesthetics and the patient’s age.\textsuperscript{9} Some treatment alternatives consist of the use of corticosteroids, cryotherapy, ligation of blood vessels, intravascular embolization, the application of lasers, period observation, and when indicated, surgical excision.\textsuperscript{2,9,13} In the clinical case presented in this study, the patient was referred for medical care. The extraction of tooth 38 and the excision of the lesion were not recommended due to the risk of hemorrhaging and due to the mutilating effect of the procedure. Intravascular embolization and periodic observation of the hemangioma and of the poor position of tooth 38 were recommended.

In conclusion, the use of only one modality of imaging exams is insufficient to diagnose a hemangioma. A clinical exam, associated with a CT study with an intravenous injection of contrast, allows for a more comprehensive view of the extension and degree of the impact of the lesion. The dentist should be aware of the clinical signs and x-rays of this pathological entity to provide a correct diagnosis and recommend the most appropriate medical treatment to the patient.

**ACKNOWLEDGEMENTS**

This project was supported by the Coordenação de Aperfeiçoamento de Pessoal de Nível Superior (CAPES).

**REFERENCES**