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Clinical case

Extensive arteriovenous malformation in the face of a pediatric patient – Case report



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ABSTRACT

Vascular anomalies can be academically divided into two groups: vascular tumors and vascular malformations. Among vascular malformations, we can highlight the arteriovenous malformations, a subtype of anomaly characterized by presenting a high blood flow. The diagnosis is beyond doubt a great challenge for the multi professional team involved in treating these anomalies. Imaging tests such as the ultrasonography, magnetic resonance, CT scan with contrast and angiography are extremely important for the orientation about the characteristics and hemodynamic properties of the blood vessels, for delivering predictable results and for risk reducing of complications. In this paper, we describe a rare case of extensive arteriovenous malformation in the face of a pediatric patient, its clinical characteristics and imaging manifestations as well as the satisfactory clinical results obtained with a combination of sclerotherapy and surgical excision.

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Extensa malformação arteriovenosa em face de paciente pediátrico – Relato de caso

RESUMO

As anomalias vasculares podem ser didaticamente divididas em dois grupos: os tumores vasculares e as malformações vasculares. Dentre as malformações vasculares, podemos destacar as malformações arteriovenosas, um subtipo de anomalia caracterizada por apresentar um alto fluxo sanguíneo. O diagnóstico é sem dúvida um grande desafio para a equipe multiprofissional envolvida no tratamento destas anomalias. Exames imaginológicos como a ultrassonografia, ressonância magnética, tomografia computadorizada com contraste e a angiografia são fundamentais para orientação quanto as características e propriedades

Palavras-chave:

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hemodinâmicas dos vasos sanguíneos, proporcionando resultados previsíveis e menor risco de complicações. Descrevemos um caso raro de extensa malformação arteriovenosa de face em paciente pediátrico, suas características clínicas e imaginológicas bem como os bons resultados clínicos obtidos com a escleroterapia e exérese cirúrgica.

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Introduction

Vascular anomalies are a set of changes of blood vessels that could be associated, or not, to endothelial cells proliferation that results in a vascular hamartomatous growth.¹

There are many discussions about the classification of these anomalies and for this reason, the correct diagnosis is important to establish the most appropriate therapeutic treatment. Although most vascular abnormalities can be clinically diagnosed, it is common knowledge that severe conditions require complementary tests for the differential diagnosis between soft tissue or bone neoplasms.

In 1996, the International Society for the Study of Vascular Anomalies (ISSVA) adopted and revised the classification system created by Mulliken & Glowacki in 1982,² being considered since then the official classification system of vascular anomalies. They proposed the division of these anomalies in two great groups: tumors (infantile hemangioma, congenital hemangioma, tufted angioma, kaposi hemangioendothelioma) and malformations. The vascular malformations can be subdivided into low-flow (capillary, venous or lymphatic) and fast flow malformations (arterial malformation and arteriovenous fistula).³

Vascular malformations (VMs) are disorders of benign origin, unusual to the maxillofacial region. They represent about 6.4% of lesions found in the oral cavity. However, studies on prevalence of these lesions are still scarce. The upper and lower lips along with the oral mucosa have the highest incidence.⁴

Although the VMs in the head and neck region cause significant esthetic defects, nasal obstruction and interference

on speech and dentition, the main concern of patients is the possibility of severe and recurrent hemorrhages.⁵

Complementary methods such as computed tomography, Doppler ultrasonography and magnetic resonance imaging, in association with clinical findings can provide information regarding blood flow characteristics and lesions extent. Generally, low-flow vascular malformations are percutaneously treated with sclerosing agents injection, while in high flow lesions the approach is endovascular, with permanent liquid or solid embolization agents.⁶

The aim of this rare case report of extensive arteriovenous malformation in a pediatric patient's face is to show the importance of a precise diagnosis obtained by imaging exams that assisted the choice of treatment, proving an excellent long-term clinical result.

Case report

A female child, 9 years old, was referred to the oral and maxillofacial surgery service at Baleia Hospital – Belo Horizonte – Brazil, complaining of painless swelling in the chin region. The mother reported that soon after the child's birth a small purplish spot appeared in the chin. Evolution was slow, progressive and associated with recurrent bleeding episodes, sometimes spontaneous. Facial asymmetry and concern about the possibility of bleeding limited the patient's social life and practice of physical activity.

The extra oral clinical examination showed an increased volume of firm consistency, with well-defined margins and blue-purplish coloration on mental region. Blood vessels superficialization was clinically suggested (Fig. 1). It was possible to observe, during intraoral clinical examination, that the lesion extended to the right side of the rear region of the mouth. The anterior lower teeth were crowded and lingualized (Fig. 2).

The intravascular puncture aspiration by an intrabuccal access, revealed the presence of pulsatile blood content. Initially, an ultrasonography was requested to evaluate the characteristics of the soft tissue volume, the distribution of vascular structures and the intensity of blood flow present in the lesion. The ultrasound examination showed an intense arteriovenous flow suggesting the hypothesis of hemangioma.

There was a suspicion of bone tissue involvement due to dental misalignment. Then, we decided not to use resonance imaging exam due to its limitation in analyzing the hard tissues, if compared to the benefits of CT scan with contrast. The CT scan identified a small calcification in the soft tissue, with no bone continuity circumscribed by an intense vascularization area suggesting a phlebolith (Fig. 3).



Fig. 1 – Initial extraoral photography (facial deformity).

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