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Massive bleeding after a tooth extraction: Diagnosis of unknown arteriovenous malformation of the mandible, a case report

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ABSTRACT

INTRODUCTION: Arteriovenous malformations (AVMs) are very rare. Only 5% of them occurs in the jaws but they can manifest with dramatic bleeding and be life-threatening.
PRESENTATION OF CASE: We report the case of a 11-year-old healthy girl who presented a massive hemorrhage after extraction of the right mandibular first primary molar. This patient received a blood transfusion and was hospitalized in pediatric intensive care unit. CT angiography highlighted an AVM of the mandible. Treatment consisted in selective embolization.
DISCUSSION: A review of the literature shows that the majority of AVMs of the jaws are often unknown until severe bleeding occurs during dental surgery. The low specificity of radiological signs on panoramic radiography makes the diagnosis particularly challenging. Their management requires an interdisciplinary approach. Selective embolization has a place of choice in the treatment of these complex pathologies.
CONCLUSION: Although AVMs of the jaws are rare, they are frequently revealed through a massive hemorrhage during tooth extraction. Dentists have to suspect them when young patients present some clinical features, as spontaneous gingival bleeding, unexplained dental mobility, or facial asymmetry.

1. Introduction

Jaw's arteriovenous malformation are very rare pathologies but they can lead to dramatic bleeding and even death [1].

We describe here the fortuitous diagnosis of a mandibular arteriovenous malformation (AVM) after deciduous tooth extraction. We will also discuss the diagnostic pitfalls through a short literature review. This work has been reported in line with the SCARE criteria [2].

2. Presentation of case

Our case concerns a 11-year-old girl without medical past history. She was transported to the emergency room for uncontrolled and recurrent oral bleeding. Anamnesis reported a first episode of massive hemorrhage one month ago, immediately after the extraction of the lower right first primary molar. There was no family history of bleeding disorder as hemophilia. Preoperative dental panoramic showed several lytic bone lesions (Fig. 1). Blood loss was assessed at 1L. Electrocoagulation in operating room and blood transfusion were necessary. This first treatment has been delivered in another health care.

The patient was taken to the emergency room of Mercy Hospital (Metz, France) one month after the first hemorrhagic event. Vital parameters on admission revealed a low blood pressure (97/59 mmHg), tachycardia (121/min) and no breathing frequency anomaly (18/min). Hemoglobin rate was measured at 7,6 g/dL and blood transfusion was given (2 Units of RBC).

Physical examination was unremarkable: no facial asymmetry, skin discoloration or cervical node were noted. Intra-oral examination revealed a pulsatile blood flow concerning extraction socket of 84. Continuous pressure and antifibrinolytic agent as tranexamic acid were not able to stop bleeding. Only a silicone compression tray has controlled hemorrhage.

Monitoring in pediatric intensive care unit was required, so the patient has been transferred to University Hospital (Nancy, France). Her general condition has improved. On admission, blood pressure

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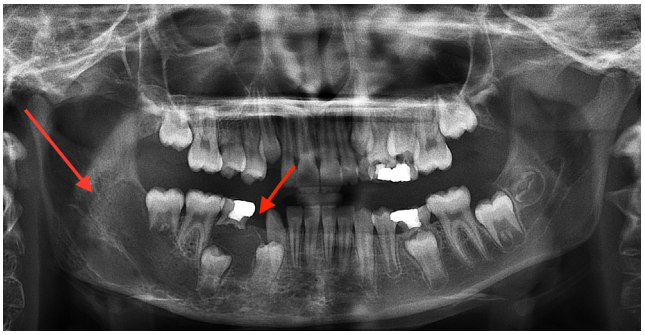

Fig. 1. Dental panoramic radiography with radiolucencies (arrows).

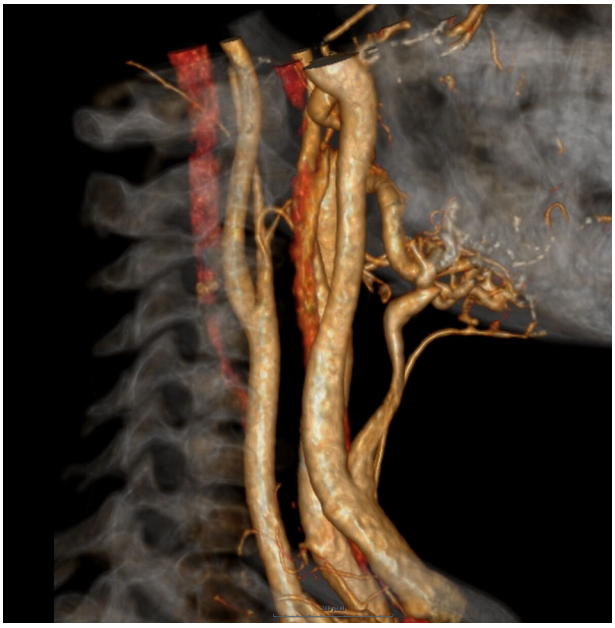

Fig. 2. Three-dimensional arterial reconstruction of CT angiography showing a large AVM of the mandible with arterial supply from right external carotid.

was measured at 109/68 mmHg, heart frequency at 108/min and hemoglobin at 9 g/dL.

Although oral bleeding was controlled, a vascular malformation of the mandible was strongly suspected at this time. CT angiography confirmed the presence of a large AVM fed by a branch of the right external carotid (Fig. 2).

After discussion between maxillofacial surgeons and interventional radiologists, angiography to precise cartography of the AVM has been planned (Fig. 3). A partial embolization of branches from the facial artery was first realized to prevent early recurrences. The complexity of the angio-architecture and the multiplicity of feeding vessels have made this first embolization insufficient.

As surgical resection of AVM presents high risks of sequelae and high morbidity, especially when AVM occur before adolescence, further selective embolization sessions were performed [3]. Two sessions of artery embolization and one session of veinous embolization have reduced the lesion size. Six months later, persistence of arteriovenous shunts on angio-MRI dictates a complement of treatment, either intravenously or by direct puncture.

3. Discussion

AVMs are rare anomalies, which can be congenital or acquired [1]. Congenital malformations, as in the present case, result from errors in vascular morphogenesis. Although 50% of AVMs occur in

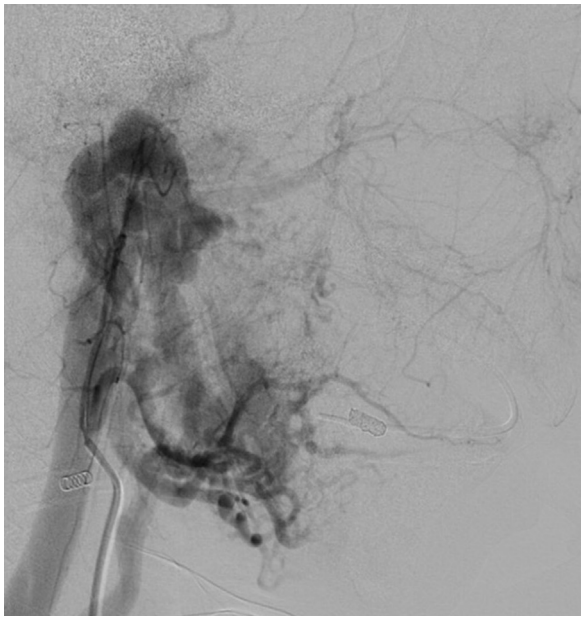

Fig. 3. Angiography of the AVM.

the head and neck region, only 5% concern the jaws. They affect preferentially the mandible than the upper jaw [4–6]. They are present at birth but may not be clinically evident before the second decade. These pathologies are complex, hard to cure, and potentially life threatening [7].

Gelfland described AVM on dental panoramic radiography as multilocular radiolucency with honeycomb or bubble soap appearance [8], whereas Stafne thought they could be like any lytic bone lesions [9]. Differential diagnosis is then possible with several jaws lesions, as odontogenic cyst, ameloblastoma, keratocyst, odontogenic myxoma, central giant cell granuloma, fibrous dysplasia, malignant tumors or metastasis [1].

In the current case, preoperative orthopantomogram revealed two lytic lesions (Fig. 1, arrows). The first one could have been an odontogenic cyst due to 84. The second one could evoke an odontogenic keratocyst or an ameloblastoma.

Further examinations can precise AVMs diagnosis: color Doppler, CT angiography and angio-MRI. The examination of choice is angiography, but is not performed at first-line because of their invasiveness. Moreover, it is usually associated with a therapeutic procedure [5,10,11].

In our case, the absence of suggestive clinical signs made the diagnosis very difficult before the tooth extraction. However, as the first massive bleeding occurred, color Doppler or CT angiography should have been realized.

A review of the literature concerning AVMs of the jaws showed that various symptoms can be present: facial asymmetry, dental mobility or malposition, discolorations of skin or intra-oral mucosa, palpable thrill, spontaneous gingival bleeding, pain, paresthesia [4,12]. Another review of fatal cases showed that AVMs are in majority revealed by a massive hemorrhage during tooth extraction by a dental surgeon [13]. Some authors prescribe a fine needle biopsy; a negative response does not eliminate totally the hypothesis of an AVM, but a positive response confirm the hypothesis and hemorrhage is more easily controlled than after an invasive surgery [10,14].

We recommend further examinations as cone beam computed tomography (CBCT) when doubtful radiolucency is observed on plain radiography. If there is the slightest doubt for a non odontogenic lesion, the practitioner has to investigate the existence of a potential AVM with non-ionizing imaging modalities (color

Doppler, MRI), even if there are no typical clinical signs. If this kind of hemorrhagic complication occurs, the best way to control the bleeding is to replace the tooth into the socket and compress.

4. Conclusion

The absence of pathognomonic radiographic features dictate to include AVMs in differential diagnosis of lytic jaws bone lesions in atypical context. Dentists have to prescribe further examinations before tooth extraction when the radiolucency concern young patients (<20 years) with spontaneous gingival bleeding, unexplained dental mobility, or facial asymmetry.

Consent

All authors assure that alterations to protect anonymity do not distort scientific meaning of the manuscript.

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent has been made available for review to the Editor-in-Chief of this journal.

Author contribution

Nasr Hasnaoui : data collection, patient follow up, paper redaction.

Etienne Simon : patient follow up, paper correction.

Eric Gérard : data study, study design.

Julie Guillet : paper redaction.

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Ethical approval

No ethical approval is requested because we submit a case report and not a research study.

No ethical approval from any committee is submitted.

Guarantor

Francesca Ceci, MD, PhD.

Conflicts of interest

All authors declare that there are no financial and personal relationships with other people or organisations that could inappropriately influence their work.

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