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Dental use of cone beam computed tomography in pediatric embolized arteriovenous maxillofacial malformation

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Abstract

Objective: Pediatric facial arteriovenous malformations (AVMs) are rare but can cause potentially fatal hemorrhages during dental procedures and oral surgery. In this article we present a systematic review of the medical open access literature on pediatric facial AVM.

Case report: We illustrate our purpose with clinical dental use of cone beam computed tomography (CBCT) in pediatric embolized facial AVM to define the presence and the position of the right upper impacted canine.

Conclusions: We advocate the use of CBCT as additional imaging tool in the follow-up of pediatric dentomaxillofacial AVM, and for depiction of dentoalveolar structures that are inaccessible by conventional dental radiography.

Keywords: pediatric, arteriovenous malformation, embolization, cone beam computed tomography, CBCT

Introduction

Arteriovenous malformations (AVMs) account for 1.5% of all types of vascular lesions [1], and 60% of them are present in the cervico-facial area [1, 2]. Mortality is in the order of 10-15% of patients due to cataclysmic bleeding [3]. Morbidity occurs to varying degrees in 50% of cases [3].

Pathogenesis

AVMs are congenital malformations of arterial and venous vessels forming connections between vessels of different origins, diameters and resistance [4]. The connections can be direct or pass through a cluster of small dysplastic vessels called the "nidus" [1, 4]. Several factors have been proposed to explain the appearance of this malformation of embryonic origin: an error in the embryonic arterial and venous differentiation [5, 6], a combination of genetic, hormonal, biochemical factors (STAT3 proteins) [6] with the presence of vascular endothelial growth factor [5], a persistence of the arteriovenous ducts of the primary retiform plexus [7], a presence of local ischemia [7].

Characteristics of AVMs

AVMs are present from birth [1, 6]. AVMs are high flow rate malformations [1]. Their expansion is the result of increased blood flow rather than cell proliferation [7]. AVMs have a clear tendency to expand with age causing destruction of soft tissue and adjacent bones [8]. The expansion is also associated with episodes of severe bleeding, loss of function and physical deformities [8]. AVMs can appear as uni-or multifocal type or a diffuse type [8]. Focal AVMs contain a well-defined nidus, the simplest form of which consists of a nourishing artery associated with a venous drainage [8]. Focal type is more easily accessible for surgery and the diagnosis and the treatment can be done in childhood [8]. Diffuse AVMs exceed the limits of topographic units, and are also associated with incomplete prior excision [8]. The nidus can no longer be identified. These type is most common in adulthood [8]. Enlargement of AVMs may also be due to hormonal changes during puberty [1]. It can occur following a stimulus: trauma with hemorrhage, following local infection or tissue destruction [5, 6]. AVMs will not dissapear like hemangioma, but grow slowly or rapidly following a stimulus [6]. The color of the AVM does not change [6].

AVMs were staged by Schobinger in the 1970s [8]. Stage I or quiescence, corresponds to pink or purplish macules with the presence of an arteriovenous shunt detected by Doppler ultrasound; this stage is asymptomatic and corresponds to the patient from birth to puberty; Stage I may be accompanied by erythema and a localized rise in temperature [6-8]. Stage II or expansion, consists of growth and infiltration of deep subcutaneous structures [8]. The lesion is associated with palpable pulsations and audible noise. Tortuous vessels appear at puberty [7]. In stage III or destruction appears dystrophic skin changes with skin ulceration, continuous pain, bleeding, secondary infections and necrosis as well as lytic bone destruction [6-8]. Stage IV is associated with continuous ulcerations and bleeding and heart failure [6-8].

Anatomical locations

The open access articles selected by this review describe the following anatomical locations: the scalp [5, 9], the forehead [10], the ear globe [7], the mandible and maxilla [3], the gingiva, mandible, muscles and skin [11], the nose [12], and the maxillary sinus [13].

Symptoms

Facial AVMs may present with the following symptoms: facial asymmetry, tooth mobility, discoloration of the skin or oral mucosa, palpable pulsation, local increase in temperature [11], spontaneous gingival bleeding, pain associated with thrombosis [11], paresthesia [11, 14-16]. Not all AVMs are pulsatile [6]. These are either very early lesions with a very weak shunt, or lesions where arteriovenous communication is greater on the venous side [6].

Medical imaging

Doppler ultrasound is used as a first-line scan to distinguish between high-flow AVMs and other low-flow vascular (venous, lymphatic) lesions [6]. The gold standard is the angio-CT scan which identifies the vascular support and the selective embolization of the collateral vessels before surgical resection [5, 7].
Magnetic resonance imaging (MRI) can define the extent of the lesion in soft tissues, assess the dynamics of the flow and help differentiate AVMs from vascular tumors [6, 7, 11].
The panoramic radiograph shows a poorly defined radiolucency with "soap bubble" -type boxes, with displacement of the teeth, expansion of the cortices, without destruction of the dental structures [11].

Treatment

Three therapeutic approaches are most commonly cited in the literature to treat AVMs: they are surgical excision, the various types of embolization and the combination of these two techniques [1, 6-9, 11, 14, 15, 17-19].

Surgical excision of AVMs

The objectives of the surgery are the disappearance of symptoms, the preservation of vital functions and the improvement of deformities [6, 18]. The ideal resection of an AVM is a block resection including the surrounding healthy bone, with the ligation of the nutrient vessels if they are recognizable [17]. This is a complex, long dissection surgery with a risk of massive hemorrhage, with the need for immediate reconstruction by flaps, and reserved for specialized centers [6, 8, 15]. Surgical excision with healthy margins is extremely difficult to achieve on the face [6].
Some authors have also proposed curettage of the lesion [11, 15] and stuffing the lesion with bone wax [11, 18], cleaning of the trans-alveolar cavity [11], one [11] or several fenestrations of the vestibular cortex [11], or an injection of hydroxyapatite cement in a mandibular AVM with obtaining complete hemostasis and obliteration of the AVM [11].
Postoperative sequelae are contractile scars, dysphasia, facial asymmetry and malocclusions [15].

Endovascular embolization of AVMs

The goal of embolization is the occlusion of the AVM nidus [1] to prevent enlargement and bleeding complications [20]. Embolization is more effective than surgical resection alone in terms of preserving functional anatomy especially in pediatric patients during facial growth [6, 15]. Different techniques have been proposed: trans-arterial, trans-venous, direct percutaneous intra-lesional puncture [15, 18].
Different types of substances have been used for embolization such as ethylene-vinyl alcohol co-polymer particles (Onyx®) [6, 8, 18, 19], N-butyl-2-cyanoacrylate [1, 7, 18], the alcohol [7], platinum particles [6], muscles, gel foam, and collagen [1].
Complications from sclerosing agents cause acute or chronic paralysis, recurrence of the lesion, skin ulceration associated with superficial lesions [6, 7].

Combination of surgical excision and endovascular embolization of AVMs

Embolization is performed between 24 hours and 72 hours before surgery to reduce intraoperative blood loss [6, 15, 18, 20].

Prognosis

AVMs are the most aggressive lesions of all types of vascular lesions [1] and are at high risk of recurrence [1]. Diffuse AVMs have a recurrence rate of 93% [16]. AVMs who have bled once are 9 times more likely to bleed within a year [3]. Children with an exacerbation of AVM at a very young age will have a worse prognosis, with more surgeries, greater morbidity and more sequelae than adult patients [6].

We present a unique case of the use of cone beam computed tomography (CBCT) in a pediatric patient with AVM in the upper maxilla for detection and for description of the position of upper right impacted canine.

Materials and methods

For the literature review of AVM we used some aspects of systematic review approach. We used three free databases: PubMed, DOAJ (Directory of open access journals) and Google Scholar. For Pubmed, we carried out the research outside the university servers in order to be able to find the same conditions of access to scientific articles and to CBCT reference images as a private dental practitioner [21]. One observer performed the search for the articles. We have chosen 2 languages: English and French. We also chosen articles with abstracts, and free-access full text articles. The inclusion criteria were: case reports, studies, reviews of pediatric facial unifocal locations (including mandible and maxilla) of AVM.
Exclusion criteria were: adult case reports, locations other than the face, experimental studies, animal studies, vascular tumors, capillary, venous, lymphatic lesions, and conference abstracts. We have also excluded articles without the possibility of accessing the pdf despite the name "open access" provided by the database.
The search equation for Pubmed was as follow (05.04.2021):
(("arteriovenous malformations"[MeSH Terms] OR ("arteriovenous"[All Fields] AND "malformations"[All Fields]) OR "arteriovenous malformations"[All Fields] OR ("arteriovenous"[All Fields] AND "malformation"[All Fields]) OR "arteriovenous malformation"[All Fields]) AND ("paediatrics"[All Fields] OR "pediatrics"[MeSH Terms] OR "pediatrics"[All Fields] OR "paediatric"[All Fields] OR "pediatric"[All Fields])) AND ((ffrft[Filter]) AND (english[Filter] OR french[Filter]))
We found 375 articles, with 16 articles included [1, 3-9, 11, 12, 14-19], and 360 articles excluded.
For the DOAJ database (Directory of open access journals (DOAJ) we used the keywords: "arteriovenous malformations" and "pediatric", and we found (10.04.2021) 80 articles, with 2 articles included [10, 20], and 78 articles excluded.
For the Google Scholar database we used the keywords « pediatric », « facial », « arteriovenous malformations », and we excluded the following terms: « brain »,

« angioma », « cerebral », « pulmonary », « lymphatic », « ileal », « hemangioma », « eye », « cyst », « textbook », « stroke ».

We found (05.04.2021) 193 articles, with 2 articles retained [2, 13], and 191 articles excluded.

Finally we retained 20 articles for the literature review.

Case Report

We present a clinical case of the use of CBCT, which has not yet been described in the open access medical literature, for the management of the consequences of embolized pediatric facial AVMs.

This is a 13-year-old patient sent by the orthodontic department to assess the position of tooth n°13. The patient presented with a right facial AVM for which the transarterial and transvenous embolization with Onyx was performed in 5 sessions at the age of 3-years-old.

Conventional radiography was not helpful because the radiolucency of the embolized nidus was superimposed on the area of interest (Figure 1).

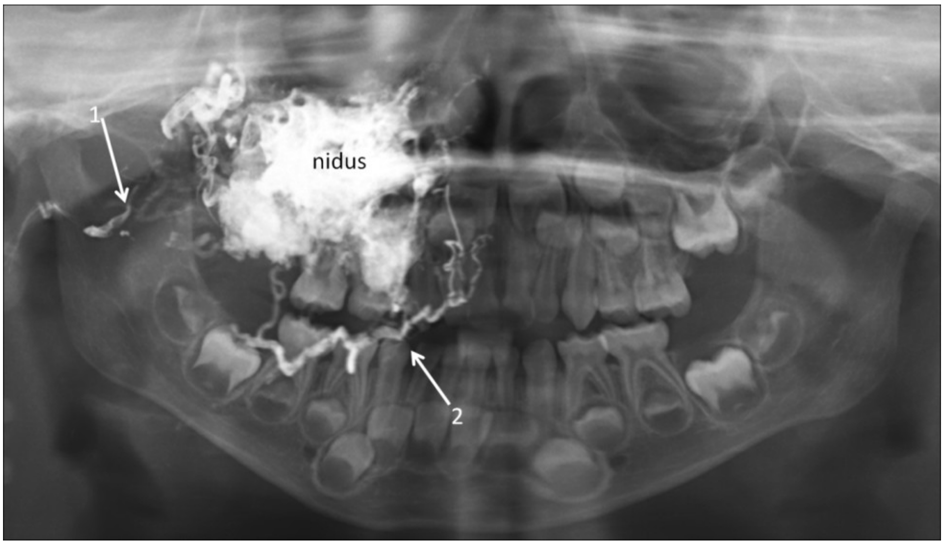

Fig. 1. Panoramic x-ray. Nidus of AVM superimposed on the lateral and posterior region of the right upper maxilla. 1. Embolized right maxillary artery. 2. Embolized right facial artery.

The CT scanner was too irradiating for the dental indication in pediatric patient. We therefore chose to use the cone beam computed tomography (CBCT) with application of the ultra low dose pediatric radiological protocol (field of view of 16/6.2cm with 200µm slice, 90kVp, 4mAs, time scanning of 6.09 seconds, distance area product of 128.8mGy x cm^2).

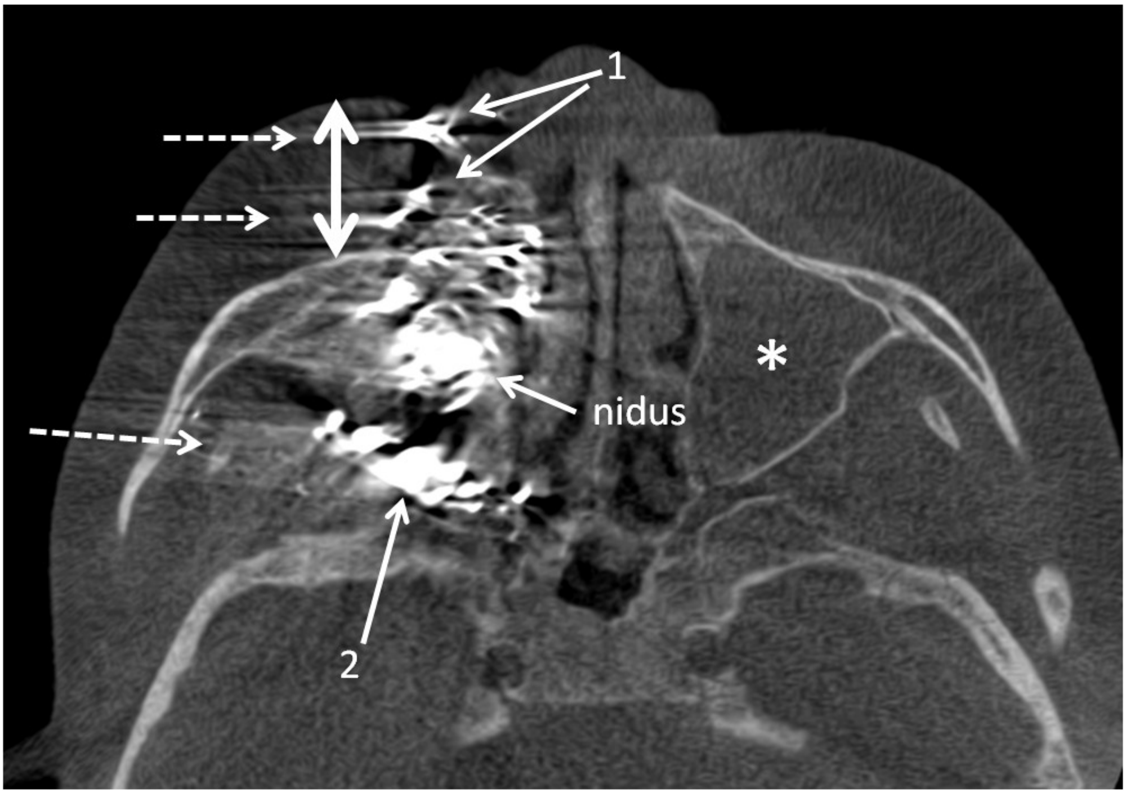

Fig. 2. CBCT axial view. 1. Branches of the right facial artery. 2. Branches of the right internal maxillary artery. * filling of left maxillary sinus. Dashed arrows: metal-type artifact stripes from embolization product projected to the right side of the image. Double arrow: thickening of the soft tissues of the right side in front of the right maxillary sinus.

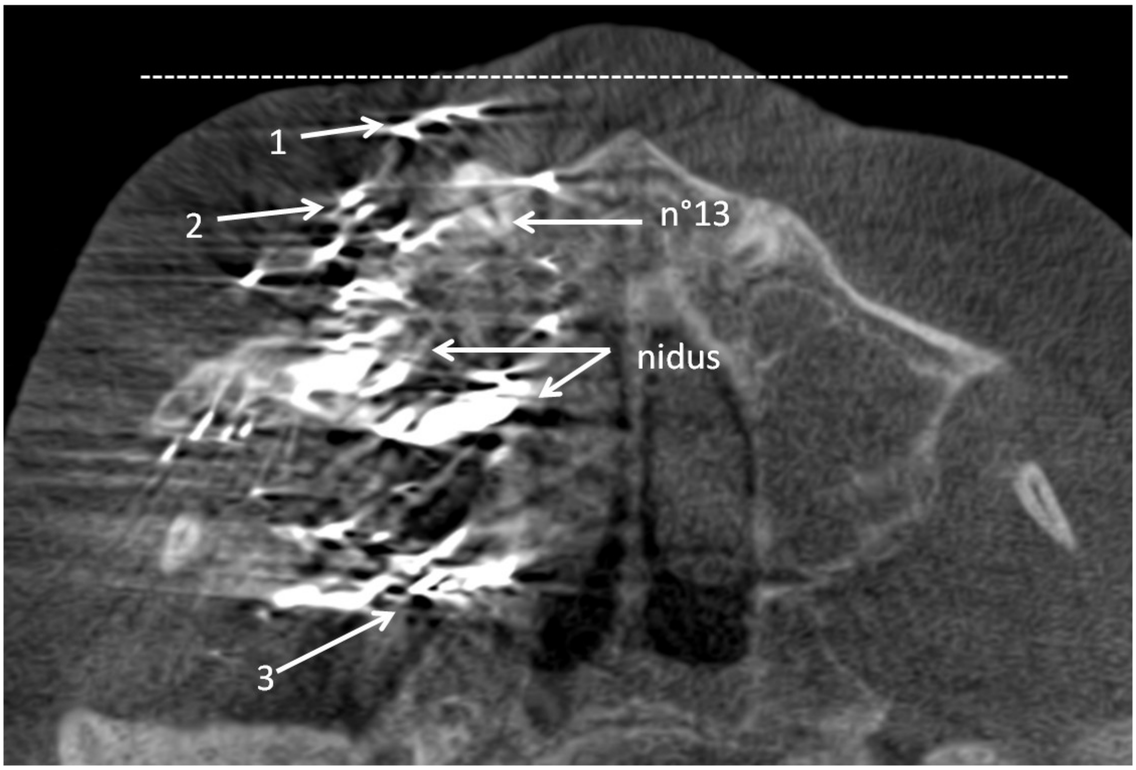

Fig. 3. CBCT axial view. 1. Right lateral nasal artery. 2. Right facial artery. 3. Branches of the right maxillary artery. Dashed line: thickening of the soft tissues of the right side in front of the right maxillary sinus.

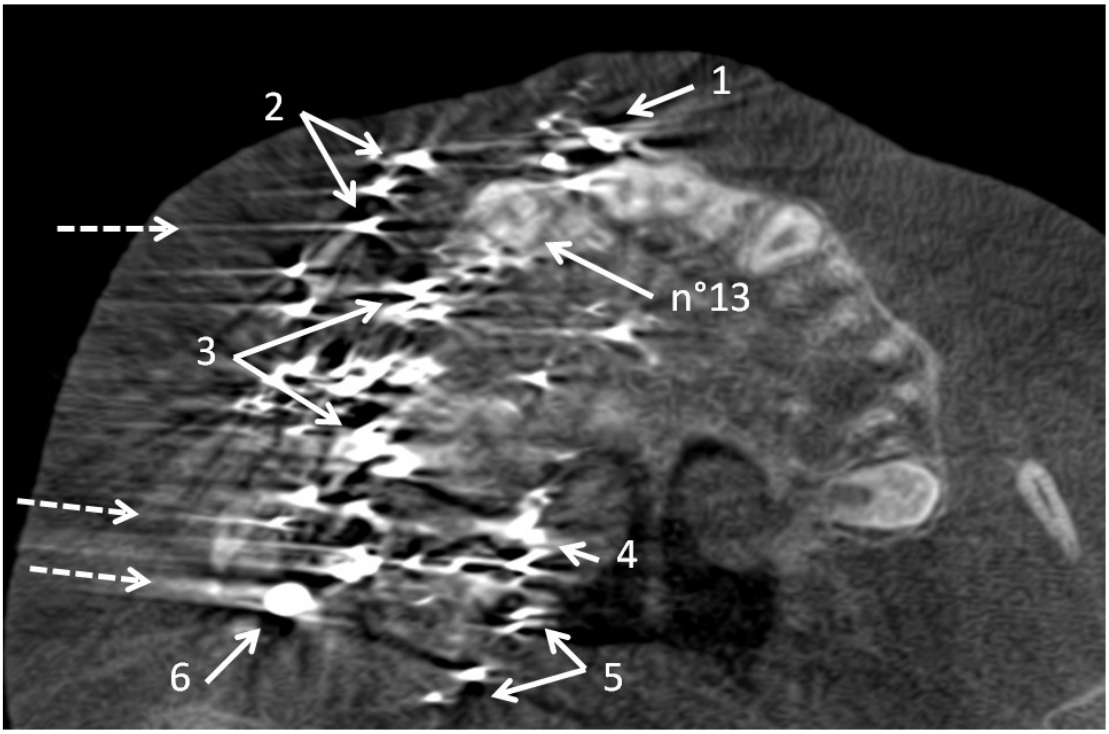

Fig. 4. CBCT axial view. 1. Right superior labial artery.2. Right facial artery. 3. Right buccal artery. 4. Right descending artery. 5. Right internal maxillary artery. 6. Right inferior alveolar artery. Dashed arrows: metal-like artifact streaks from embolized product in arteries.

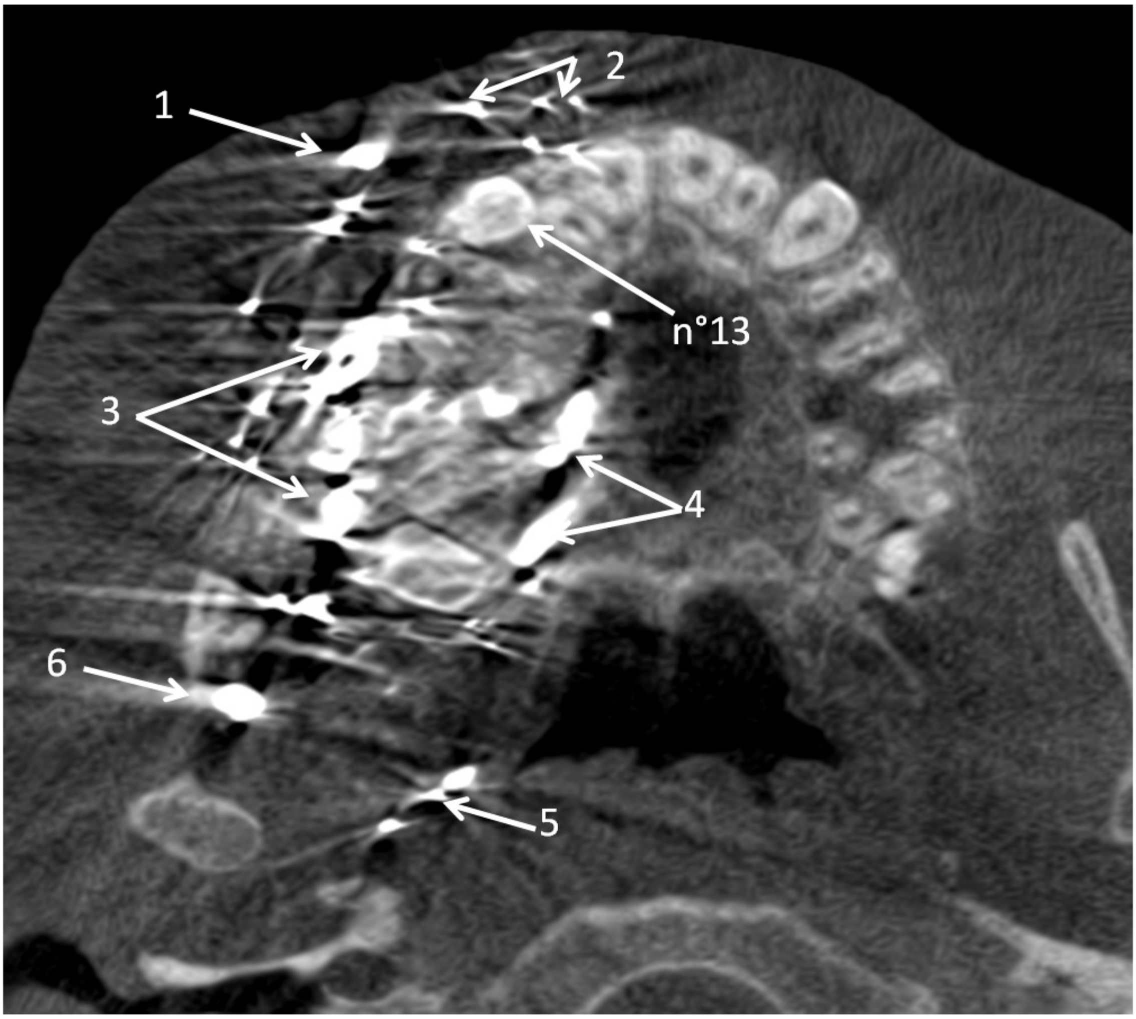

Fig. 5. CBCT axial view. Right facial artery. 2. Right superior labial artery. 3. Right buccal artery. 4. Right palatine artery. 5. Right internal maxillary artery. 6. Right inferior alveolar artery.

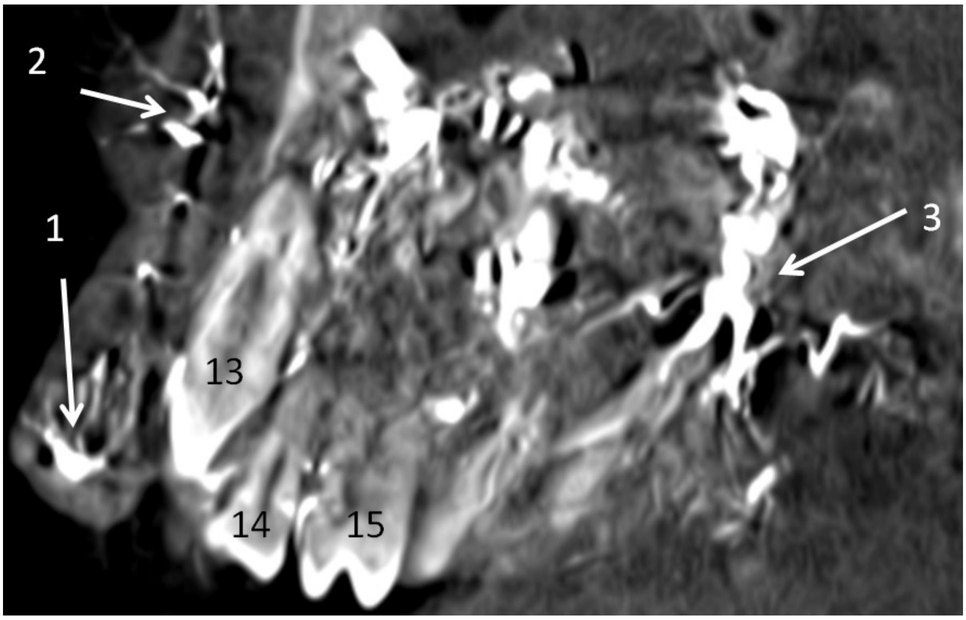

Fig. 6. CBCT MPR reformatted 2D view. 1. Right superior labial artery. 2. Right lateral nasal artery. 3. Right descending palatine artery. Impacted tooth n°13, tooth n°15 with rotation.

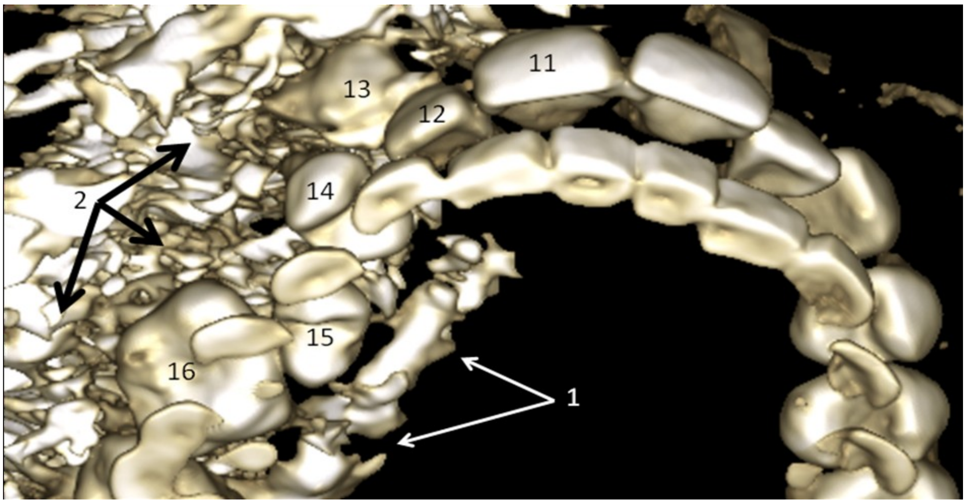

Fig. 7. CBCT 3D reconstruction of upper dental arch. 1. Right palatine artery. 2. 3D reconstruction of massive artifact related with embolization product present in AVM. Tooth n°13 is impacted and in vestibular position. Tooth n°15 is mesially rotated, and situated on the palatine side of the right maxillary alveolar bone.

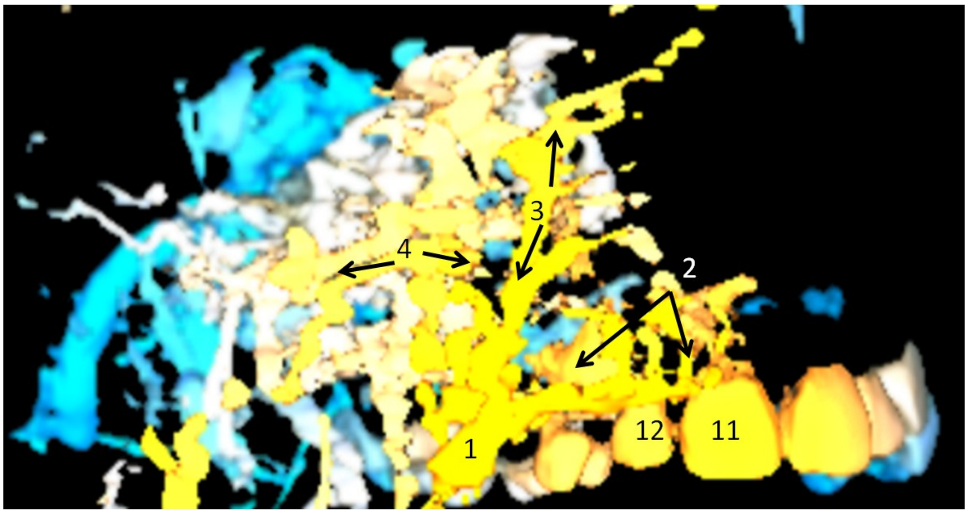

Fig. 8. CBCT 3D reconstruction of the embolized arterial system. 1. Right facial artery. 2. Right superior labial artery. 3. Right lateral nasal artery. 4. Right transverse artery of the face.

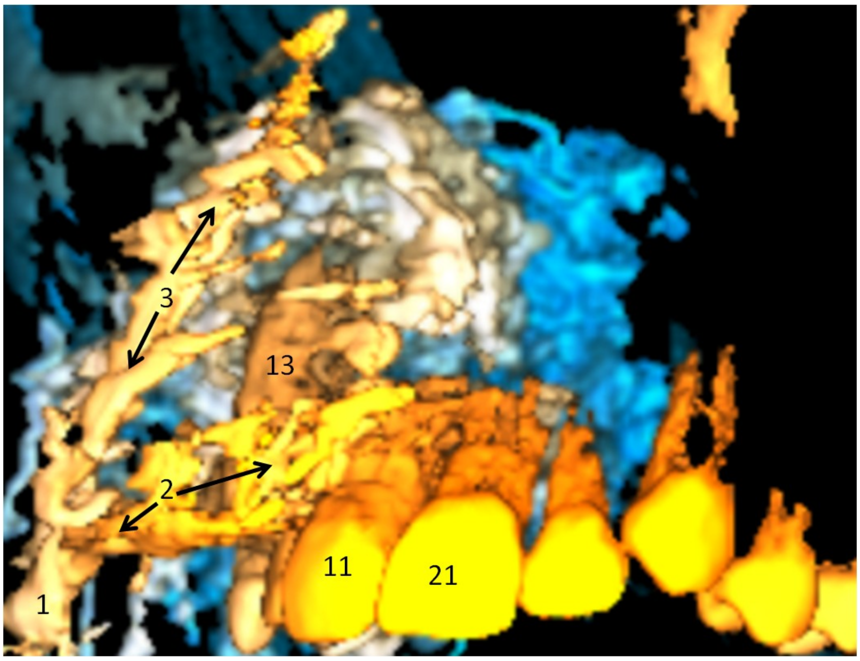

Fig. 9. CBCT 3D reconstruction of the embolized anterior arterial system (facial artery) of the AVM. 1. Right facial artery. 2. Right superior labial artery. 3. Right lateral nasal artery.

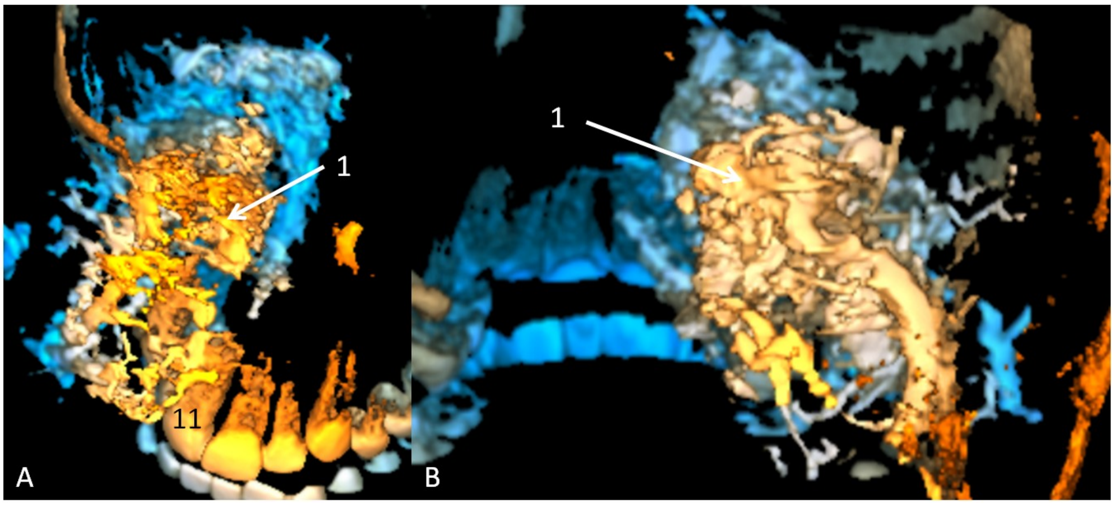

Fig. 10. CBCT 3D reconstruction of the embolized arterial system. A. CBCT 3D reconstruction of the nidus (1). B. CBCT 3D reconstruction of the embolized right posterior (internal maxillary artery) arterial system of the AVM (1).

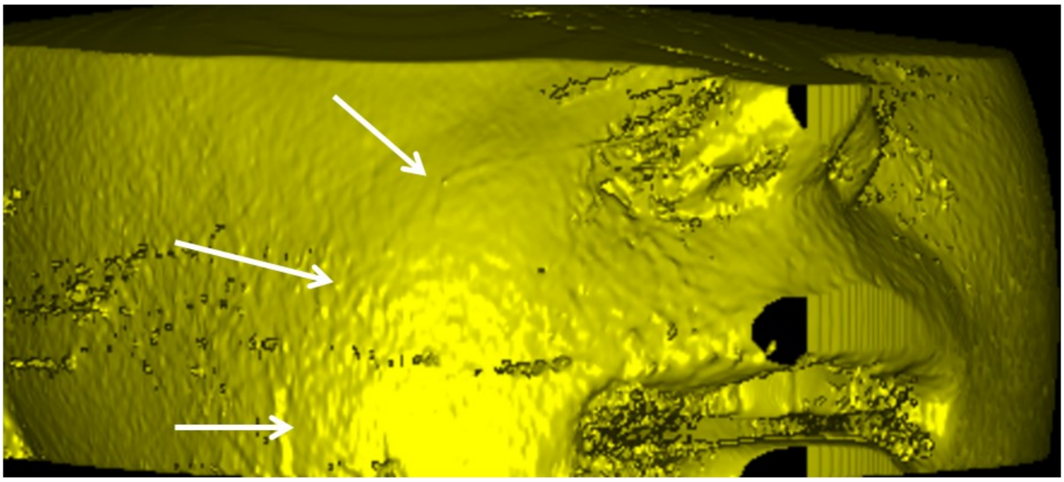

Fig. 11. CBCT 3D reconstruction of the right facial soft tissues (arrows) asymmetry due to AVM.

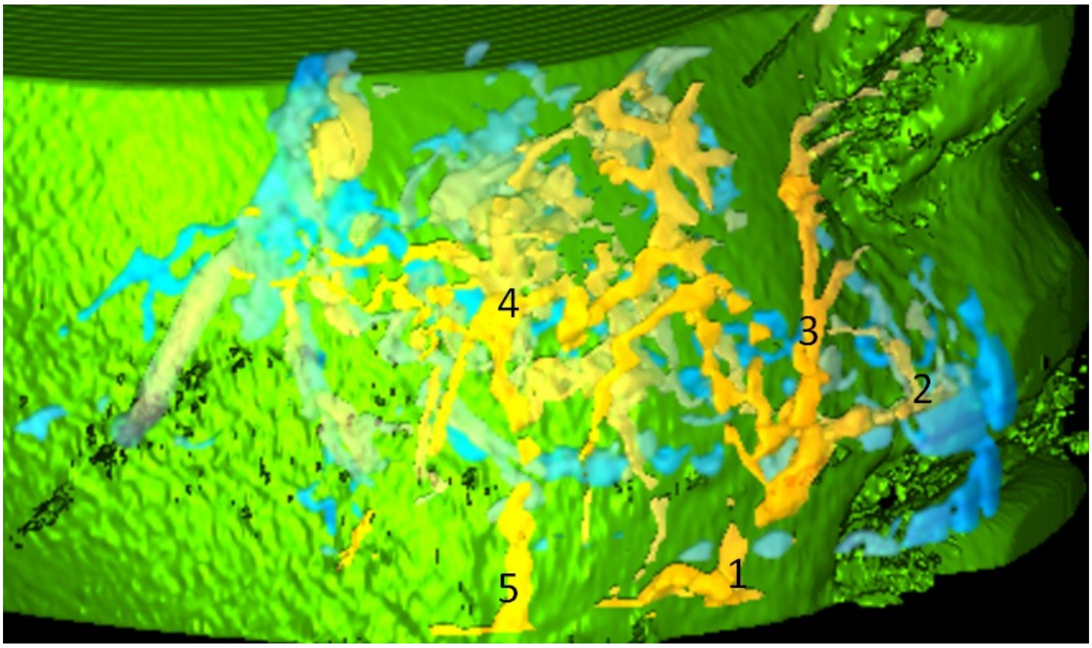

Fig. 12. CBCT 3D reconstruction of the right facial soft tissues and superimposition of the arterial and venous embolized system of AVM.
1. Right facial artery. 2. Right superior labial artery. 3. Right lateral nasal artery. 4. Right transverse artery of the face. 5. Right facial vein.

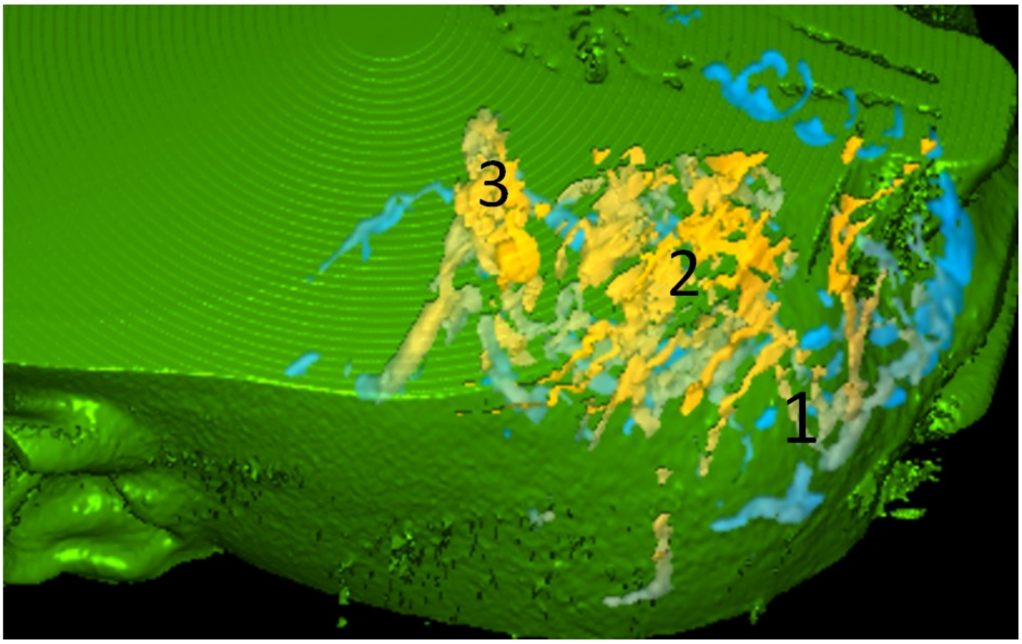

Fig. 13. CBCT 3D reconstruction of the right facial soft tissues and superimposition of the arterial and venous embolized system of AVM. 1. Embolized area of AVM related to the right facial artery. 2. Nidus embolized area of AVM. 3. Embolized area of AVM related to the right internal maxillary artery.

Discussion

Onyx embolization solution is known to present beam-hardening artifact on CT scan [22, 23]. Beam-hardening artifact related to Onyx solution is also present on CBCT (Figures 2-6). In our clinical case the AVM was situated on the right side of the face and the artifact was projected laterally toward the right side of the patient. This situation allows sufficient visualisation of dental arches and of the position of the right upper canine (Figures 3-7, 9).

CBCT allowed global three-dimensional visualisation of the AVM arterial and venous network fixed by Onyx embolization process (Figures 8-10). Anatomical extension of the embolized lesion can also be better understood (Figures 11-13). However, three-dimensional CBCT reconstruction alone (Figure 7) was insufficient to depict the arteriovenous embolized network. We needed to use more advanced CBCT software visualization tools with superimposition of soft and hard tissues to visualize the extension of the lesion (Figures 8-13). Structures close to the observer were presented in yellow and structures far from observer were presented in blue to enhance the perspective (Figures 8-10, 12,13).

In this clinical case the beam-hardening artifact was projected to the right from the right-side embolized lesion (Figures 2-5, 7). It allowed to free the sight on the right dentoalveolar process. However, we don't know yet if a beam-hardening artifact from left-side embolized lesion will be projected to the left or also to the right side of the patient.

Finally, we advocate the use of CBCT as additional imaging tool in the follow-up of pediatric dentomaxillofacial AVM, and for depiction of right dentoalveolar structures that are inaccessible by conventional dental radiography.

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- **Ethical approval**: there was no need for the ethical approval for this case report
- **Informed consent**: we obtained the written informed consent from the mother of the patient, and all the images were anonymized and no private data were provided allowing the patient's identification.

Authors contribution:

Author	Contributor role
Olszewski Raphael	Conceptualization, Investigation, Writing original draft preparation, writing review and editing
Theys Stéphanie	Data curation, Writing original draft p reparation, writing review and editing

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