CASE REPORT

Arteriovenous malformation (AVM) of the maxilla complicated by a local pathogen infection

Bruno Ella, Aurelie Lanotte, Cyril Sedarat, Jean-Christophe Fricain

Department of Odontology and Buccal Health, Faculty of Odontology, Bordeaux 2 University, Bordeaux, France

Correspondence to
Professor Bruno Ella,
tsir.ella@wanadoo.fr

SUMMARY

Arteriovenous malformations (AVM) are usually observed on the skin, but they can also rarely affect the visceral organs and the maxillofacial area. In the maxillofacial area, these lesions can spread and they are potentially dangerous, as a biopsy or even a simple tooth extraction can lead to a catastrophic haemorrhage that may eventually cause death. There are several indications for the treatment, including age, size and type of vascular malformation. The purpose of this report is to describe an AVM on the maxilla, first diagnosed as an epulis, which has been treated for several years as a chronic local periodontitis. The lesion was finally stabilised after an antibiotic therapy. Improved knowledge of the development of these lesions and collaboration between dentists, radiologists and surgeons are necessary to improve the diagnosis and treatment of vascular malformations.

BACKGROUND

Arteriovenous malformations (AVM) are the most frequently occurring superficial vascular malformations.¹ They are congenital malformations of the blood vessels, and in rare cases, they emerge only in adulthood.² AVM can be subdivided according to the type of vessel involved (capillary, venous and arterial) and can be categorised, according to the haemodynamic features, into high-flow and low-flow lesions.³ The capillary bed is replaced by an extensive vascular 'tortuosity' and by the areas of abnormal direct communication between arteries and veins (shunts). AVM in the maxillofacial region are rare, but they can spread and lead to complications, which are difficult to treat. They are potentially dangerous, as a biopsy or even a simple tooth extraction can cause a major haemorrhage that may cause death. Treatment options depend on the stage and type of lesion and on the resulting deformity.^{4,5}

Haemorrhage is the most common manifestation of these lesions and occurs most frequently around a tooth whose root is buried under the malformation. The resulting gingival bleeding is often the reason for consulting a dentist. The patient described in this case has been seen regularly for check-ups during 8 years, but he presented a refractory focus in the premolar-molar area on the right maxilla.

CASE PRESENTATION

A 44-year-old woman with no particular medical history was treated by her dentist for adult periodontitis and underwent scaling and root planning.

The lesion around the first upper right molar had the aspect of a budding lesion, which was painful and bled on contact.

Her dentist had diagnosed an extensive angiomatous epulis between teeth #15 and #16, and he decided to surgically remove the lesion and the #17 tooth, which was severely affected. Ten days after the surgery, a larger epulis reappeared in the same localisation. Some months later, as the lesion developed further, the other teeth (#15 and #16) were extracted, and then the lesion remained quiescent. Ten years later, the lesion reappeared, and the patient was referred to our department (odontology and buccal health).

INVESTIGATIONS

- ▶ Clinical examination revealed an inflammatory lesion, pulsatile, painful and bleeding on contact (figure 1). Its pulsatile nature suggested a diagnosis of vascular lesion, so further tests were carried out in this direction to characterise the lesion.
- ▶ The panoramic x-ray revealed no periodontal lesions.
- ▶ The echo-Doppler and the colour flow Doppler showed a nodular formation of 10–15 mm. It presented a mosaic flow pattern indicating an anarchic vascularisation, as would be seen in the nidus of an arteriovenous fistula. With the pulsed Doppler, the recorded flow was arterialised (systolic-diastolic modulation) with a collapse of peripheral resistance (presence of flow during diastole). This flow was assumed to be an indirect sign of upstream arteriovenous communication (figure 2).

Figure 1 Clinical aspect of the lesion.

To cite: Ella B, Lanotte A, Sedarat C, et al. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2012-007617

Figure 2 Echo-Doppler and pulsed Doppler revealed the anarchic vascularisation of the lesion.

- Arteriography was performed to reveal the precise angioarchitecture of the lesion. It revealed a capillary-venous malformation with very little vascularisation without communication with the venous network. There was no dilatation of the facial artery⁶ (figure 3A,B).
- The patient's haemogram showed no hemopathy and coagulation was normal.

DIFFERENTIAL DIAGNOSIS

Extended angiomatous epulis.

TREATMENT

The lesion was excised under local anaesthesia and a biopsy was taken around tooth #14. The biopsy revealed a significant inflammatory reaction and a highly vascular lesion.

Figure 3 (A) The arteriography displays the angioarchitecture of the arteriovenous malformations (AVM); (B) schematic diagram of the AVM.

Five months later, a localised gingivorrhagia reappeared around the upper right canine, extending towards the right lateral incisor. We therefore decided on another treatment consisting of a CO₂-laser application. The laser treatment was performed under local anaesthesia, using a mean output of 15 W, continuous wave with 90° angle tip. No excessive bleeding was observed during the procedure. No recurrence of the lesion was observed during an 8-month follow-up. Two sessions of laser treatment were performed during a 16-month period and her lesion disappeared.^{7,8} The advantages of this technique include satisfactory bleeding control and clear visibility during the procedure, as well as reduced postoperative pain and swelling.

- The patient was prescribed half a tablet of propranolol (40 mg), two per day, as β -blockers are used in the treatment of some angiomas. The inflammation had disappeared, but there was still a minor recurrence of inflammation and some localised bleeding.
- Since the result was always unpredictable because of frequent recurrences, we decided to take a sample of the flora at the lesion site using C reactive protein with the Paro-diagnostic test (Gaba laboratories).⁹ That analysis revealed two bacteria: *Tannerella forsythia* and *Treponema denticola*, the 'red complex' associated with periodontics.

A specifically adapted antibiotherapy was prescribed, combining spiramycin (3MUI, three per day) and metronidazole (250 mg, two tablets three times daily).

OUTCOME AND FOLLOW-UP

After 3 weeks of antibiotic therapy, the inflammation was considerably reduced and spontaneous bleeding had stopped. The site was stabilised for over a year, with check-ups every 3 months.

DISCUSSION

We have described here a rare case of facial AVM, complicated by a local infection (*T forsythia* and *T denticola*) and stabilised by antibiotic therapy.

A periodontal AVM is a rare oral lesion that can be misdiagnosed as an epulis. Owing to the pulsatile character of the lesion, this hypothesis can be discarded.

Diagnosis depends on the location and size of the lesion.¹⁰ The evolution of the lesion is unforeseeable and sudden, and any prognosis must be guarded as the patient's life may be at risk.

The fact that the vascular anomaly observed didn't induce any significant change in the upstream arterial network could be explained in two ways: (1) there was no arteriovenous communication or (2) the anomaly had still evolved slowly or was

microscopic, generating only a small local hyper-flow with no repercussions on the existing vascularisation.

In the absence of a nidus or a feeder arteriola, the arteriography neither shows up the vascular malformation nor suggests any embolisation or sclerotherapy.¹¹

Most lesions of this type regress spontaneously or remain stable without any consequences. Some lesions disappear completely without any treatment, if they are diagnosed early after birth (70% of cases), whereas for some lesions with a slow involution phase, further treatment is indicated.

It is important to differentiate manifestations of AVM from all gingivorrhagias that appear harmless, like epulis¹² and other necrotising periodontal diseases.

This clinical case was monitored for more than 10 years, but the outcome was uncertain and unpredictable because of frequent recurrences.

The treatments described in the literature consider the anatomical and functional recovery of the affected area and also the risks involved. Once the disease has been identified and after the treatment, the risks of complications are considerably

reduced. Given that the inflammation and bleeding decreased and remained stable after antibiotic therapy, a bacterial infection could complicate an AVM on the maxilla. Dental treatment must be carried out gently to avoid any bleeding, and regular monitoring is necessary.

Acknowledgements With thanks to Dr Sylvain Catros at Department of odontology and Buccal Health, Faculty of Odontology, Bordeaux University, Bordeaux, France.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 Abouzgia MB, Symington JM. Recurrent arteriovenous malformation of the mandible: a case report. *J Oral Maxillofac Surg* 1992;50:1230–3.
- 2 Bhansali RS, Yeltiwar RK, Agrawal AA. Periodontal management of gingival enlargement associated with Sturge-Weber syndrome. *J Periodontol* 2008;79:549–55.
- 3 Oueis H, Geist JR, Tran MU, *et al.* High-flow arteriovenous malformations of the mandible and the maxilla: report of 2 cases. *Pediatr Dent* 2010;32:451–6.
- 4 Damodar ND, Nandakumar H, Strinath N. Management of arteriovenous malformations in maxillofacial region. *Int J Oral Maxillofac Implants* 2007;36:1037.
- 5 Carneiro S, Batista L, Vasconcelos B, *et al.* Massive oral hemorrhage due to mandibular arteriovenous malformation treated with percutaneous approach: a case report. *J Oral Maxillofac Surg* 2009;67:2525–8.
- 6 Narang S, Gupta R, Narang A, *et al.* Periodontal manifestations and management of a patient with AV malformation. *J Indian Soc Periodontol* 2012;16:266–70.
- 7 Arslan A, Gursoy H, Cologlu S. Treatment of lymphangioma with CO₂ laser in the mandibular alveolar mucosa. *J Contemp Dent Pract* 2011;12:493–6.
- 8 Yoshida H, Yusa H, Ueno E. Use of Doppler color flow imaging for differential diagnosis of vascular malformations: a preliminary case report. *J Oral Maxillofac Surg* 1995;53:369–74.
- 9 Lu Q, Jin L. Human gingiva is another site of C-reactive protein formation. *J Clin Periodontol* 2010;37:789–96.
- 10 Liu DG, Ma XC, Zhao FY, *et al.* A preliminary study of angiographic classification and its correlation to treatment of central arteriovenous malformation in the jaw. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2005;100:473–80.
- 11 Fan X, Zhu L, Zhang C. Treatment of mandibular arteriovenous malformation by transvenous embolisation through the mental foramen. *J Oral Maxillofac Surg* 2008;66:139–43.
- 12 Kuhl SR, Schulze RK, Kreft A, *et al.* Epulis granulomatosa as an oral manifestation of Klippel-Trenaunay syndrome. *J Oral Pathol Med* 2006;35:576–8.
- 13 Chen WL, Ye JT, Xu LF, *et al.* A multidisciplinary approach to treating maxillofacial arteriovenous malformations in children. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2009;108:41–7.

Learning points

- Clinical examination is usually not sufficient to diagnose an arteriovenous malformation.
- Palpation of the lesion is fundamental to guide the diagnosis.
- Complementary tests must be performed for any atypical swelling, atypical lesion or gum bleeding on the maxilla.
- Pulsed Doppler and arteriography are key examinations to explore superficial vascular malformations.
- Bacterial analyses can help to prevent recurrences and to decrease bleeding.
- The diversity of haemangiomas and oral vascular malformations require a multidisciplinary collaboration to obtain an early and accurate diagnosis.¹³

Copyright 2013 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- Submit as many cases as you like
- Enjoy fast sympathetic peer review and rapid publication of accepted articles
- Access all the published articles
- Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow