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Mucosal necrosis of the palate after embolization for labial arterioveinous malformation

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Disclaimer: the views expressed in the submitted article are our own and not an official position of the institution or funder.

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Abstract

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Objective: Cervico-facial arteriovenous malformations (AVMs) are complex and rare vascular lesions, and present in 0.1% of the population. Of traumatic or congenital origin, they are characterized by variable growth, and their complications can be disfiguring and potentially fatal. The treatment of choice is embolization followed by surgery if necessary. The main complications are recurrence and post-operative bleeding.

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Case report: We report the rare case of a 59-year-old female patient who underwent embolization of a right upper labial and jugal AVM, followed by complete necrosis of the right hemi-palatal mucosa associated with dental mobility and pain. Follow-up at 6 months showed complete reepithelialisation of the palate.

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Conclusions: Soft tissue necrosis after AVM embolization is a rare event and is more commonly described after embolization for epistaxis. The evolution is generally favourable within a few weeks.

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Keywords: arteriovenous malformations, embolization complication, palate necrosis

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Introduction

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Cervico-facial arteriovenous malformations (AVM) are complex and rare vascular lesions, present in 0.1% of the population [1]. Of traumatic or congenital origin, they are characterized by variable growth, and their complications can be disfiguring and potentially fatal. We described a rare case of post-embolization necrosis of the palatal mucosa for a labial and jugal AVM.

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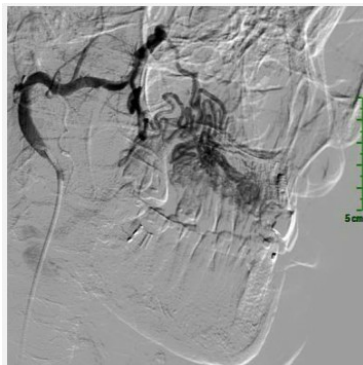
Case report

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A 59 years-old woman, with unremarkable past medical history, presented with a large (> 6 cm) and recurrent right upper lip and jugal AVM (Figure 1).



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Fig. 1. Pre-embolization angiography of the labial and right jugal AVM.

The patient had been treated 20 years ago by sclerotherapy of the lesion. More recently and due to progression of the disease the patient was treated again 3 times by embolization in a period of time of 2 years. The first embolization (Glubran® and Onyx 18®) was uneventful. The second treatment (ethanol) was associated with headache. For the last treatment, Glubran® 25% and ethanol were injected into the pedicle that supplies the nidus. This was followed by rapid complete necrosis of the right palatal mucosa, associated with diffuse teeth ache and mobility of the teeth n°14, 16 and 17. The patient received thalidomide treatment one day before the embolization and for 3 months after embolization to slow down the neoangiogenesis. All the procedures were performed by a highly trained interventional radiologist, under general anesthesia, and by a right femoral approach.

Oral examination three days after embolization showed necrosis of the mucosa overlying the right hard palate, and roots exposure of the teeth n°16 and 17 (Figure 2).



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68 **Fig. 2. Oral aspect of right palatal mucosa three days after**
69 **embolization.**
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71 The bone seemed to be intact clinically. No oroantral nor oronasal fistula was
72 found. The lesion was painful, and speaking and eating were difficult. No other
73 symptoms were present (no cutaneous lesions, no paresis).
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75 The CT scan presented a complete occlusion of the right descending palatine ar-
76 tery (Figure 3).
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79 **Fig. 3. Post-embolization CT scan showing occlusion of the right**
80 **descending palatine artery (red arrow).**
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88 The bone presented no sign of necrosis. Panoramic X-ray showed only the
89 embolization material (Figure 4).
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Fig. 4. Panoramic X-ray with embolization material in the right cheek and right upper lip.

The treatment of the necrosis consisted of the association amoxicillin/clavulanic acid 875 mg three times per day for 3 weeks, and mouth washes (chlorhexidine 2% alternated with H₂O₂ 3%). Once a week, and for six weeks, local debridement of the necrotic mucosal tissues until bleeding was performed under local anesthesia. Progressive reepithelialization was noted. Mobility of tooth n°14 decreased, but persisted on teeth n°16 and n°17. Local evolution is shown in figures: after 3 weeks (Figure 5), after 2 months (Figure 6), after 6 months (Figure 7), and after 11 months (Figure 8). The superior labial lesion was still present.



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Fig. 5. Oral aspect of the right palatal mucosa after 3 weeks.



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Fig. 6. Oral aspect of the right palatal mucosa after 2 months.



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Fig. 7. Oral aspect of the right palatal mucosa after 6 months. Complete reepithelialization of the palate.



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Fig. 8. Oral aspect of the right palatal mucosa after 11 months.
Stability of the clinical aspect.

CBCT at 3 months showed no bone necrosis. Re-epithelialization of the palatal mucosa was completed at 3 months. The wound healed by secondary intention. The residual problem was the mobility and the pain on teeth n°16 and 17, for which the patient was referred to a parodontologist.

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Discussion

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Typical extracranial locations of AVMs are the face, the oral cavity, and the extremities. The treatment remains a major challenge because the growth pattern is often compared to that of a malignant tumor. Recently, Kansy et al [1], described an algorithm for the management of AVMs: the "Heidelberg's Algorithm". Surgical resection (with or without reconstruction) at 24-48h post-embolization shows a very satisfactory results compared to monotherapy. Incomplete resections or embolization can induce aggressive growth of the residual nidus, and the risk of recurrence can be as high as 50% in the first 5 years [1].

Different materials are used for embolisation such as: ethanol, Onyx 18®, Glubran® [2, 3]. Onyx 18® is a non-adhesive liquid embolic agent made from ethylene vinyl alcohol copolymer and from dimethyl sulphoxide, suspended in tantalum powder [2, 3]. Glubran® is a surgical glue composed of a cyanoacrylate co-monomer [4].

The main complications of embolization are the recurrence and the postoperative hemorrhage. Soft tissue necrosis is less frequently reported, probably due to the rich blood supply to the region [5, 6]. The mechanism is the reflux of particles into proximal vessels [7], when the injection of the material is forceful, or when the particles are very small and could flow back easily [5]. This was not the case here. Rare cases of palatal necrosis have been described following embolization of refractory epistaxis [3, 8], with very similar results of re-epithelialization of the

145 mucosa at 3 and at 6 months. Embolization for tumoral indication [6] or for trauma
146]9] had also led to necrosis.

147 In this case, the descending palatal artery (branch of the internal maxillary artery)
148 was involved while it represents the major blood supply of the region. The multiple
149 vascular anastomoses in this region have allowed the healing of the lesion.

150 Treatment consisted of mouth washes (chlorhexidine 0,2%, H2O2 3%), antibiotics
151 (amoxicillin /clavulanic acid), and local debridement under local anesthesia. Local
152 or distant flap may be necessary in the absence of the healing.

153 Patients need to be informed about this rare major transient complication.

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155 In summary, necrosis of the oral soft tissue after embolization is a rare event. We
156 present the only case of palatal necrosis after embolization of a labial and jugal
157 AVM described in the literature, in a patient with no risk factor: no trauma, no
158 tobacco, no medication use, and no radiotherapy.

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- **Acknowledgements:** none
- **Funding sources statement:** this study does not received any funding
- **Competing interests:** Dr M. Magremanne is member of the Editorial board of NEMESIS. Dr A. Arnal declares no conflict of interest.
- **Ethical approval:** there was no need for the ethical approval for this case report
- **Informed consent:** there was no need for the informed consent for this case report as all the images were anonymized and no private data were provided allowing the patient's identification.

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Authors contribution:

Author	Contributor role
Arnal Etienne Andrea	Conceptualization, Investigation, Data curation, Writing original draft preparation, writing review and editing
Magremanne Michele	Writing original draft preparation, writing review and editing, Supervision

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