Conservative management of a palatal ischaemic area following embolization for a cranio-facial arteriovenous malformation (AVM) in a patient with Wyburn-Mason Syndrome

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We read with interest the paper from Santos *et al.* that was published recently (1). Here we present a similar patient with a necrotic mucosal area in the midline of the maxilla, following embolization, in a patient with Wyburn-Mason Syndrome. Wyburn-Mason Syndrome is an exceedingly rare, non-hereditary congenital neurocutaneous disorder leading to arteriovenous malformations (AVMs) (2). It is also known as *Congenital Unilateral Retinocephalic Vascular Malformation Syndrome* and is characterised by arteries that directly connect to veins without intervening capillaries and leads to a fragile mass of abnormal blood vessels found in the midbrain, eyes, orbit and rarely cutaneous nevi.

Our patient was a 58-old female patient with a slow flow vascular malformation lying in the suprasellar/hypothalamic region and extending into the right orbit, face and nose. The AVM consisted of a small and relatively stable intracranial component and an orbital/facial component, supplied by the bilateral ophthalmic arteries, bilateral external carotid system (internal maxillary and facial) and the mandibulovidian branch of the right internal carotid artery (ICA).

The magnetic resonance angiography (MRA) demonstrated multiple small aneurysms in the facial component of the AVM, particularly in the right nasal cavity; this area has shown clinical progression, resulting in the patient suffering several significant episodes of epistaxis. Our patient underwent radiologically guided embolisation for management of the epistaxis. Specifically, Sceptre Mini advanced into left IMA and PHIL 25 was injected until no further devascularisation of the arterial pedicles could be performed with excellent penetration of the arterial pedicles supplying the AVM. Further embolisation was then performed via the right ophthalmic artery via the Sceptre Mini using a mixture of PHIL 25 and 30 with again very good devascularisation of the arterial pedicles supplying the AVM. Shortly after this, she presented to the maxillofacial clinic with a necrotic area of mucosa in the midline of her palate (Figure 1). A CT scan showed no bone involvement. In the absence of infective signs and symptoms she was managed conservatively with analgesia and an antimicrobial mouthwash. At review the necrotic area had spontaneously healed, and no further interventions were deemed necessary (Figure 2).

In general, embolization can potentially cause stroke, ischaemia, skin necrosis, bleeding, blindness, adverse haemodynamic changes and pulmonary embolism (3). We recommend, in specific cases of necrosis in the maxilla following embolization, adopting a cautious initial management in order to allow necrosis to self-demarcate and heal.

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Figure 1 Palatal necrosis at presentation.



Figure 2 Palatal epithelialization following conservative management.

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Footnote

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