Case Report

Management of recurrent bleeding from facial arteriovenous malformations in end-stage liver disease

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Introduction

We report the management by arterial embolisation of recurrent significant bleeding from a facial arteriovenous malformation (AVM) in a patient with end-stage liver disease. Given their propensity to re-bleed, AVMs are often refractory to other forms of treatment, including surgery. Evidence from the management of facial AVMs and our case suggests that embolisation may offer a better initial management strategy in patients with end-stage liver disease related facial AVMs.

Case report

A 49 year-old man, awaiting liver transplantation for Child-Pugh grade C alcohol-related cirrhosis, was admitted to our hospital with diuretic-induced renal dysfunction. He had first presented 6 years previously and had subsequent hospital admissions for recurrent tense ascites, spontaneous bacterial perotonitis, hepatic encephalopathy, oesophageal variceal bleeding and symptomatic hydrothorax, managed with pleurodesis. He ceased drinking alcohol 1 year prior to the present admission and was listed for liver transplantation 9 months later.

This hospital admission was complicated by a sudden onset, brisk, large volume (700mL) apparent fresh haematemesis, leading to tachycardia, hypotension, and necessitating urgent fluid resuscitation. Emergency upper gastrointestinal (GI) endoscopy revealed well-epithelialised oesophageal

varices with no evidence of recent bleeding. Following further active bleeding, it became apparent that the patient had actually bled from an arteriovenous malformation (AVM) on the right side of his upper lip (Figure 1a), sufficient to drop his haemoglobin by 1.2g/dL. This lesion had developed over the last 3 years, gradually increasing in size. There was no pre-existing cutaneous birthmark or history of trauma at this site. The patient also had several spider naevi on his upper body. An arteriovenous Doppler velocity profile at the site of the lesion was consistent with a localised high-flow AVM. The upper lip AVM bled subsequently on several occasions, requiring further transfusions of blood products and necessitated urgent intervention. In view of his decompensated chronic liver disease, surgical intervention was considered unsafe and cryotherapy under local anaesthetic was technically impossible. Therefore, arterial embolisation therapy of the AVM via a right femoral artery puncture was undertaken. Right external carotid and facial artery angiograms during the procedure confirmed a focal high-flow AVM (Figure 1c). Embolisation of the feeding superior labial vessels was achieved using polyvinyl alcohol, via a coaxial 3 French catheter. Post-embolisation angiograms demonstrated obliteration of arteriovenous shunting (Figure 1d). Left external carotid arteriography after embolisation did not demonstrate any supply to the malformation across the midline. There were no adverse clinical events resulting from the procedure and there was no further bleeding. The patient subsequently underwent liver transplantation several weeks later.

Discussion

The case we describe is unusual in that the lesion bled so much that it was mistaken for an oesophageal variceal bleed by the acute medical team managing his case. However, systemic AVMs are a well-recognised cutaneous manifestation of chronic liver disease[1]. They have a high predisposition towards bleeding and are often refractory to surgical treatment alone, owing to revascularisation of the AVM nidus[2,3] Cases in the literature illustrate that treatment with laser, steroids, irradiation or cryotherapy may not be effective in controlling high-flow AVMs[4,5] Arterial embolisation has been widely used in AVM management[6] and is the treatment of choice for facial AVMs, including those involving the upper lip.²⁻⁴ Although AVMs may resolve following liver transplantation[1] there is little information on the best form of management of problematic cutaneous AVMs in the pretransplant setting, where there may be life-threatening coagulopathy. However, the usefulness of embolisation techniques for other complications of end-stage liver disease including the treatment of irresectable hepatocellular carcinomas, oesophageal varices and hypersplenism is well recognised.

Given the propensity to bleed in hepatic decompensation, our case supports the use of embolisation in the management of high-flow cutaneous AVMs in distal sites, such as the face, in such patients. With advances in interventional radiology, it has become possible to cannulate and selectively embolise lesions supplied by smaller, less easily accessible feeding

vessels without adverse effects[4] This offers a better initial management strategy than other interventions for AVMs that are at high risk of bleeding.

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References

- 1 Alcolado R, Bowry J, Winwood P J, Loehry C A. Systemic arteriovenous malformations: a feature of advanced liver disease. *Gut* 1994; 35: 1145–1147.
- 2 Hassard AD, Byrne BD. Arteriovenous malformations and vascular anatomy of the upper lip and soft palate. *Laryngoscope* 1985; 95: 829-32.
- 3 Tan KT, Simons ME, Rajan DK, Terbrugge K. Peripheral High-Flow Arteriovenous vascular malformations: A single-center experience. J Vasc Interv Radiol 2004; 15:1071-1080.
- 4 Bradley JP, Zide BM, Berenstein A, Longaker MT. Large arteriovenous malformations of the face: aesthetic results with recurrence control. Plast Reconstr Surg. 1999;103: 351-61.
- Kohout MP, Hansen M, Pribaz JJ, Mulliken J. Arteriovenous malformations of the head and neck: Natural History and Management. Plast Reconstr Surg. 1998; 102: 643-654.
- 6 Simons ME. Peripheral vascular malformations: diagnosis and percutaneous management. Can Assoc Radiol J 2001; 52: 242-51

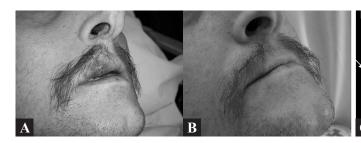




Figure 1: Appearances of AVM and angiograms

(A) AVM before embolisation. (B) AVM after embolisation. (C) Lateral projection from selective right facial artery angiogram demonstrates focal vascular abnormality involving upper lip (see arrow) through which there is rapid arteriovenous shunting. (D) Angiogram after embolisation demonstrates that arteriovenous shunting has been obliterated