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Case Study

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The addition of adjuvant radiotherapy in the management of extracranial arterio-venous malformations: a case discussion

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Abstract

Introduction: The management of extracranial arterio-venous malformations (AVMs) is complex and often requires a multidisciplinary approach. Currently, treatment includes surgical resection and embolotherapy.

Methods: We present the case of a foot AVM that was managed with adjuvant radiotherapy after previous surgery and embolotherapy had been attempted, and we discuss the role of radio-therapy in the management of extracranial AVMs.

Results: The malformation was successfully eradicated with complete obliteration of the nidus and no recurrence.

Conclusions: The addition of radiotherapy in the management of extracranial arterio-venous malformations offers promising results using similar doses to those used in brain AVMs.

Introduction

Arterio-venous malformations (AVM) are typically located in the head and neck, pelvis and lower extremity. Occasionally they can be encountered in the foot where they can be painful and interfere with gait.¹ Their only potential cure is the complete eradication of the lesion and its nidus to avoid recurrence, which often requires a multidisciplinary approach.² Surgical resection after embolisation is the most accepted approach, especially in large AVMs.³

Currently, stereotactic body radiation therapy (SBRT) is an established treatment method for AVMs located intracranially, with high obliteration and low complication rates.⁴ The optimal dosage is based on location and volume with typical margin doses ranging from 16 to 25 Gy.⁴ Although the role of SBRT in the management of extracranial AVMs has not been established and the published experience is limited, it has been used in head and neck AVMs safely and effectively using similar doses to those used in brain AVMs.^{5–7}

Hereby, we present the case of an AVM located in the metatarsal area of the foot that was successfully eradicated with the use of adjuvant radiotherapy. Prior to the radiotherapy treatment, the patient had undergone one session of embolotherapy followed by staged surgical excisions with immediate microsurgical reconstruction.

Clinical case

A 33-year-old woman presented with a non-healing ulcer on the internal aspect of her right foot (Figure 1) secondary to an arterio-venous malformation adjacent to the second, third and fourth metatarsal heads that involved the three arterial trunks of the leg (Figure 2A). At the time of diagnosis, she had been offered a Syme's amputation of the foot (above the ankle) at another institution which she had refused, so she inquired about an alternative treatment plan. Her case was discussed with a multidisciplinary team and standard treatment with embolotherapy and surgical resection was decided. She then underwent one session of preoperative embolisation of the tibialis posterior artery with Onyx (Ethylene Vinyl Alcohol Copolymer) followed by two-staged surgical excisions and immediate reconstruction with two latissimus dorsi free flaps, first through a plantar approach and then though a dorsal approach.

One year later, a small residual nidus persisted which was not eligible for surgical treatment due to its entanglement with the pedicle of the second toe. Considering the proven efficacy of radiotherapy in the eradication of intracranial AVMs,⁸ the radiation oncologist proposed applying the same treatment approach. SBRT was selected to minimise the radiation dose to the surgically manipulated surrounding tissue while delivering a high dose of radiation to the nidus. The nidus was the gross tumor volume (GTV), and it was delineated using angio-CT images (4-3 cm/26.0283 cc) and the planning tumor volume (PTV) was generated adding a margin



Figure 1. Aspect of the foot before treatment. Note the ulceration on the internal aspect, secondary to the arterio-venous malformation.

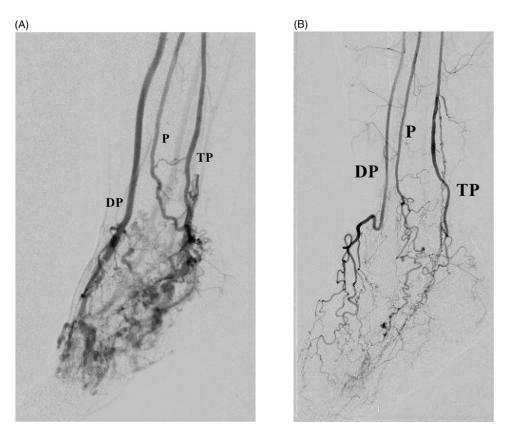


Figure 2. Digital subtraction angiography of the right foot. A. Arteriovenous malformation at the time of diagnosis. Note the involvement of the three arterial trunks of the leg. B. Complete eradication of the arteriovenous malformation 2 years after radiotherapy treatment. (Tibialis Posterior artery (TP), dorsalis Pedis artery (DP), peroneal artery (P)).

of 0.3 cm to the GTV. The prescription dose was 30.94 Gy administered in 3 weekly fractions (one fraction per week for three consecutive weeks) to the 95% of the PTV. Treatment was delivered with volumetric modulated arc therapy technique and photons of 10 Mega-electron volts in flattening filter-free beams. Cone beam computed tomography was the technique used for image-guided radiotherapy, and the acute tolerance of the treatment was excellent.

Two years later, complete eradication of the nidus was evidenced (Figure 2B), and the patient carried out a normal active life with no limitations in gait (Figure 3).



Figure 3. Result 2 years after completion of treatment. Note the two free flaps in place with an adequate aspect of the reconstructed foot and no signs of recurrence.

Discussion

In the management of cerebral AVMs, SBRT is an established treatment that achieves high obliteration with low complication rates.^{4,9,10} Although there is little published experience of using radiation for extracranial AVMs, there are reports of head and neck AVMs that were successfully treated with Cyber Knife,⁶ SBRT⁷ and Gamma Knife⁵ when a small residual nidus persisted after surgery and embolotherapy. In these reports, marginal doses of 22–24 Gy were used and complete obliteration was documented at 3 years post-treatment with no late effects. We describe similar results in our patient with the use of a marginal dose of 30.94 Gy.

We believe that radiotherapy can be safely and effectively applied in extracranial locations for the obliteration of small residual nidus that persist after surgery and embolotherapy. We recommend applying the same principles of dosage, fractionation, and planning as those used for intracranial AVMs.

To the best of our knowledge, this is the first report of an extracranial AVM outside the head and neck area that has been successfully eradicated with adjuvant radiotherapy. The limitations of the current study include that it is a single case with no comparison group. Further studies should be carried out to elaborate on the role of radiotherapy in the management of extracranial AVMs.

Conclusion

The addition of adjuvant radiotherapy in the management of extracranial AVMs can spare morbidity when a small nidus persists after standard treatment and complete surgical resection entails a risk to the surrounding structures. This treatment method offers promising results, using similar doses to those used in brain AVMs.

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Conflicts of Interest. The authors declare none.

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