Surgical removal of intramuscular arteriovenous hemangioma of the upper left forearm compressing radial nerve branches

Case report

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✓ The authors report on the case of a 32-year-old woman with an intramuscular arteriovenous hemangioma (AVH) of the left forearm with burning pain and paresthesias diffused to the radial nerve–related territories. The patient underwent coil embolization of the AVH and surgical removal of the remnant and regrown AVH. This case demonstrates the safety and efficacy of surgery when interventional radiology fails to achieve complete occlusion. En bloc removal of the lesion was performed through a left elbow cleft incision, and intraoperative electrophysiological monitoring and angiography with indocyanine green (ICG) were performed. The pathological diagnosis was intramuscular AVH. Postoperative follow-up examinations demonstrated the permanent disappearance of the subcutaneous mass and of the patient’s sensory disturbances. Complete excision of the AVH was confirmed on postoperative magnetic resonance angiography, and no surgery-related complications or new neurological symptoms were detected.

Intramuscular AVHs are rare lesions that can be successfully treated with both coil endovascular embolization and surgery; the latter is indicated when endovascular procedures fail to occlude the AVH completely. Intraoperative angiography with ICG can be helpful in confirming the success of the procedure. (DOI: 10.3171/JNS/2008/108/4/0808)

KEY WORDS • indocyanine green angiography • intramuscular arteriovenous hemangioma • radial nerve • vascular hamartoma

Intramuscular AVHs are very rare vascular malformations; the corresponding cutaneous form is more frequent. Both lesions can be defined as vascular hamartomas with a benign course and variable growth pattern. Some of these lesions are present at birth and slowly grow with time, and others spontaneously evolve. Intramuscular AVHs are found mainly in the limbs and scalp and are much more common in women and children than in men. In some cases AVHs are associated with genetic disorders, but they should not be confused with other types of vascular malformations capable of severely altering the pattern of limb growth in infants, such as those that arise in well-defined syndromes such as Parkes Weber syndrome or Klippel–Trenaunay syndrome.

We present the case of the surgical removal of an intramuscular AVH that had been partially embolized 9 months before admission to our institution.

Case Report

History and Embolization. This 32-year-old woman first noticed a nonpainful subcutaneous mass in the anterior surface of the superolateral portion of her left forearm in February 2006. After a few weeks, she began to experience burning pains and paresthesias in the area innervated by the radial nerve. She underwent echography at an outside institution, and a spindle-shaped hypoechocic mass (40 × 36 × 12.8 mm) was revealed. The lesion was located anterior to the proximal third of the radius and appeared hypervascularized on the Doppler study. The vascular structures found inside the lesion had an arteriovenous pattern. The patient then underwent transfemoral angiography, which confirmed the presence of an arteriovenous malformation at the level of the upper left forearm with a complex pattern

Abbreviations used in this paper: AVH = arteriovenous hemangioma; ICG = indocyanine green; MR = magnetic resonance.
of feeding vessels (mostly from the brachial artery) and an early venous drainage (Fig. 1 left). During the procedure, coil embolization of the lesion was performed and achieved ~80% deafferentation (Fig. 1 right).

An echographic control study performed 1 month later showed the persistence of the lesion with reduction in size (25 × 15 × 12 mm). An MR image of the left forearm performed 3 months after embolization demonstrated an increase in lesion size, suggesting partial regrowth (Fig. 2). The examination further demonstrated that the lesion was situated close to the proximal third of the diaphysis of the radius, the round pronator and brachioradialis muscles, the tendon of the biceps brachialis, the median and radial nerves, and the ulnar artery. Electromyography of the upper limbs performed in the same period did not show any relevant alterations in nerve conduction.

Surgical Excision. The patient presented to our institution in November 2006. A Z-shaped skin incision was made, centered on the left elbow cleft. The muscular fascia and layers at this level were opened, and the radial nerve and its branches were identified. At this point a tortuous vascular bunch located anterior to the round pronator was found. It injected into an abnormally round, pale, cherry-colored lesion that was located inferiorly and surrounded by a thin layer of muscular tissue. The lesion was dissected from the surrounding tissues, and the feeding and drainage vessels were isolated and sectioned. En bloc removal of the mass was achieved. The intervention was performed using intraoperative electrophysiological monitoring of all nerve branches close to the hemangioma and ICG fluoroangiography, which demonstrated the vascular nature of the lesion and confirmed its complete removal (Fig. 3). For the latter procedure a microscope-integrated light source containing infrared excitation light (OPMI Pentero surgical mi-
croscope, Carl Zeiss) was used to illuminate the operating field after 25 mg of ICG dye was injected intravenously. Intravascular fluorescence from within the blood vessels was imaged using a video camera attached to the microscope. The angiographic images were observed on the video screen in real time and recorded with a video camera.

**Results**

The histopathological diagnosis was intramuscular AVH. The patient confirmed immediate pain relief and reduction in paresthesias after the intervention, and there were no neurological or vascular complications.

The patient attended 2 follow-up examinations at 1 and 3 months postoperatively. At both visits, the persistent disappearance of the subcutaneous mass on inspection and of the patient’s sensory disturbances was confirmed. Control radiological examination with MR angiography confirmed total excision of the intramuscular AVH (Fig. 4). No surgery-related complications or new neurological symptoms were reported during the follow-up period.

**Discussion**

To our knowledge, this is the first case in the literature in which intraoperative ICG fluoroangiography was used to confirm the location of afferent and efferent vessels and the total removal of a lesion. Furthermore, this case demonstrates that when a complex pattern of feeding and draining vessels is present, a preoperative angiographic examination and intraoperative ICG fluoroangiography can be helpful for surgical planning. Because of the good outcome at follow-up and the absence of recurrence after surgical intervention (as shown on radiological examinations), the latter

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**Fig. 3.** A: Intraoperative photograph of the surgical exposure of the lesion (AVH) with radial nerve (R) branch located ventrally to it. Draining veins (DV) are also shown.  B: Intraoperative ICG angiogram. Lesion appears as a highly vascularized round mass with early draining veins to the left.  C: Intraoperative photograph of the surgical field at the end of the procedure. The veins (sectioned and coagulated) on the left are ventral to the round pronator muscle; the previously positioned coil (C) is at the bottom of the figure.  D: Final photograph of the surgical field before the wound is closed up.

**Fig. 4.** Postoperative images of the left upper limb obtained at the 3-month follow-up examination; T1-weighted, Gd-enhanced MR image (left) and angiographic sequence (right), demonstrating complete removal of the lesion without regrowth.
treatment seems to be indicated in cases of rapid regrowth after coil embolization.

Conclusions

Arteriovenous hemangiomas are rare lesions that are defined as vascular hamartomas with arteriovenous shunts. They can be located in the skin (cutaneous AVHs) and, less commonly, inside the skeletal muscles (intramuscular AVHs). These lesions are most frequently found in women and young adults and can be associated with genetic abnormalities of the connective tissues. In some cases, the contiguity of these lesions to nervous structures suggests that they are tumors of nervous origin. These lesions can be treated successfully with coil embolization, surgery, or both. The results of the present study show that in patients in whom interventional radiological procedures do not prevent a regrowth of the lesion, surgery can allow its total and safe removal. Intraoperative ICG angiography can be helpful in confirming the success of the procedure.

References


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