Median Neuropathy Caused by Intramuscular Venous Malformation in the Brachialis Muscle at the Elbow: A Case Report

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INTRODUCTION

Venous malformations (VMs) are a commonly encountered entity in clinical practice, with an estimated incidence of 1 to 2 in 10,000 births and a prevalence of 1%.¹⁻³⁻⁹ VMs are composed of abnormal collections of veins with a variable luminal size and wall thickness, often multiple.¹⁻²⁻⁷ They are often less well-circumscribed than are vascular tumors, such as infantile hemangiomas, and can be interspersed with adipose tissue or within various kinds of atrophic or degenerative muscle.³⁻⁰ Although all VMs are present at birth, but they are also identified in adolescence and adulthood.¹⁻³⁻⁹ VMs occur at a frequency of 40% in the head and neck region, 40% in the extremities, and 20% in the trunk.² VMs in the extremities often violate surrounding fascial planes and can infiltrate subcutaneous tissue, muscle, bone, joints, neurovascular structures, and even viscera.⁹⁻¹⁰ Intramuscular VMs comprise an uncommon subgroup of VMs.¹⁵ They are often mistaken for tumors, because of a similar presentation and because of improper nomenclature.⁹ Although most intramuscular VMs in the extremities have been reported to present with a growing palpable mass with or without pain,¹⁰ the development of neurological symptoms is rare. The authors report a rare intramuscular VM originating from the brachialis muscle and showing symptoms of median nerve involvement.

CASE REPORT

A 58-year-old, right-handed male patient presented with a gradually growing mass in the distal upper arm and mild weakness in left-hand grasping. The mass occurred 6 months prior to the presentation, and there was no pain when it first occurred. The mass gradually increased: in the morning, it became larger, and in the afternoon, it became smaller (Fig. 1A). A month previously, when he touched the mass, he felt tenderness and tingling in the thenar area of his left hand. Two weeks later, the mass got bigger, and his left-hand grip weakened; when he grabbed an object, he began to drop it.

On examination, there was no objective weakness in forearm pronation, wrist flexion, flexion of the first 3 digits and thumb opposition, or abduction, which are innervated by the left median nerve, nor any objective sensory disturbance in the radial 2/3 of the palm that is innervated by the palmar cutaneous branch of the median nerve. There was no tenderness in the mass itself, but when the medial of the mass was pressed, there was pain along with tingling instantaneously in the thenar side of the left palm. It was not mobile. X-ray examination showed a small calcification in the soft tissue in front of the
Intramuscular Venous Malformation within the Brachialis Muscle

Fig. 1. Presentation of intramuscular venous malformation in the left cubital fossa. (A) Clinical photograph showing a bulging mass in the medial side of the left arm cubital fossa (white circle). (B) On the X-ray, a small round calcification thought to be a phlebolith is seen.

Fig. 2. Magnetic resonance imaging of venous malformation (VM) originating from the brachialis muscle. (A) Axial T2-weighted fast spin-echo image reveals hyperintense signal within the VM (white arrows) with excellent definition of lesion extent. Multiple signal voids (black arrows) indicating vascular channels and fibrous septa are noted within the VM, which is located within the brachialis muscle (asterisk), and the median nerve (white arrowhead) is not involved directly. (B) Axial T1-weighted image revealing isointense VM with lesion extent (white arrows). Multiple signal voids are seen. The brachialis muscle (asterisk) and the median nerve (white arrowhead) are identified. (C) Gadolinium-enhanced fat-saturated T1 image reveals heterogeneously enhanced VM (white arrows). The veins of the antecubital fossa were dilated and showed ectasia. The left median nerve was not directly involved by the lesion, but was displaced by the swollen brachialis muscle (Fig. 2D). With these unique MR findings, the lesion was thought to be a low-flow, VM. Considering the gradual enlargement of the mass and worsening of the pain and neurological symptoms, surgery was planned, and consent was obtained.

After we made a lazy S-shaped vertical incision along the medial antecubital area, we carried dissection down to the antibrachial fascia. After securing the left median nerve, we dissected the swollen brachial muscle under microscopic vision.
Table 1. Classification of vascular anomalies according to vascular dynamics

<table>
<thead>
<tr>
<th>Category</th>
<th>Description</th>
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<tbody>
<tr>
<td>I</td>
<td>Hemangioma</td>
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<tr>
<td>II</td>
<td>Vascular malformations</td>
</tr>
<tr>
<td>A</td>
<td>Low-flow (venous malformation)</td>
</tr>
<tr>
<td>B</td>
<td>High-flow (arteriovenous malformation)</td>
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<tr>
<td>III</td>
<td>Lymphangioma</td>
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DISCUSSION

1. Intramuscular VM

Identification and classification of vascular anomalies were hampered historically by the use of confusing nomenclature\(^7\). Early classification suggested by Virchow\(^20\) and Wegener\(^21\) classified vascular lesions according to the pathologic appearance of the vessel\(^7\). Vascular growths were divided into angionomas and lymphangiomas without consideration of the biologic behavior and natural history of the vascular lesions\(^7\). Consequently, there was a tendency to identify any vascular anomaly as a hemangioma\(^5\,\(^7\,\(^3\,\(^3\,\(^3\)). A variety of terms including “venous angionoma”, “cavernous angionoma”, “cavernous hemangioma”, and “phlebangioma” have been used in the medical literature to describe these anomalies\(^5\,\(^7\). These terms have led to confusion with the more common proliferating or true hemangiomas of infancy\(^5\,\(^7\). For example, capillary hemangioma, nevus flammeus, and port-wine stain have all been used in the literature to describe a capillary malformation of the skin\(^10\).

In 1982, Mulliken and Glowacki\(^10\) proposed a modern classification of vascular anomalies according to the lesion’s biologic and pathologic differences; all vascular anomalies were assigned to 1 of 2 broad categories: hemangiomas and vascular malformations. The former category was later expanded to include vascular tumors; the suffix “-oma” was to be reserved for only those lesions exhibiting increased cellular turnover, the classic example within this category being the infantile hemangioma\(^13\,\(^3\). The term “vascular malformation” was applied to those lesions present at birth growing commen-surately or pari passu with the child\(^13\). The vascular malformations were composed of normal “mature” flat endothelial-lined vascular spaces with a normal rate of cell turnover and were further subdivided into capillary malformations, VMs, arterial (arteriovenous) malformations, and lymphatic malformations\(^5\,\(^3\). In 1993, Jackson et al.\(^9\) classified vascular malformations according to flow patterns instead of the former anatomicopathologic classification for ease of investigation and treatment (Table 1). They simplified flow patterns within vascular malformations as either low-flow (VMs) or high-flow (arteriovenous malformations), keeping separate categories for lymphangiomomas and hemangiomas, with the purpose of creating “a system directly related to investigation and treatment”\(^9\).

2. Clinical Manifestation and Diagnosis

The diagnosis of VM and differentiation from other vascular malformations can be usually made by clinical history and
3. Treatment of Intramuscular VM

Treatment is generally indicated if the lesion causes pain, functional impairment, or aesthetic problems, as in craniofacial lesions. With the exception of some superficial VMs where laser therapy is effective, VMs are generally treated with direct surgery and sclerotherapy. Surgical resection is considered preferable if the lesion could be completely removed so as to avoid recurrence. This includes patients with local well-defined VMs that are thrombosed, confined to a single or specialized muscle group, or causing a neurological or compression syndrome, and patients where there is a good possibility of anatomical and functional restoration. Many lesions are infiltrative, however, and involve multiple muscle groups or fascial planes where surgical resection results in an unacceptably high functional and cosmetic deficit. Sclerotherapy has been increasingly incorporated in surgical regimens and now has been considered as an adjunct to surgery or the stand-alone therapy of choice for most VMs. We chose surgical resection in the current case because the lesion was well localized within the brachialis muscle and caused local neurological symptoms by mass effect. The treatment resulted in complete relief of the symptoms of VM, with no recurrence up to 2 years after surgery.

CONCLUSION

Here, we report on a rare intramuscular VM originating from the brachialis muscle in the left arm, which caused the local mass effect, showing the symptoms associated with irritation of the median nerve. The lesion was diagnosed through a typical MR imaging finding. Because the lesion was well-localized within the brachialis muscle, surgical resection resulted in complete symptomatic relief without recurrence.

CONFLICTS OF INTEREST

No potential conflict of interest relevant to this article was reported.

REFERENCES

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