Acta Orthopaedica et Traumatologica Turcica 53 (2019) 310–312

Contents lists available at ScienceDirect

Acta Orthopaedica et Traumatologica Turcica

journal homepage: https://www.elsevier.com/locate/aott

Intraneural angioleiomyoma of the median nerve presenting as a forearm mass: A case report

H. Utkan Aydın*, Ömer Berköz

Istanbul University, Istanbul Faculty of Medicine, Dept. of Plastic, Reconstructive and Aesthetic Surgery, Hand Surgery Unit, Istanbul, Turkey

ARTICLE INFO

Article history:
Received 4 February 2018
Received in revised form
1 May 2018
Accepted 27 June 2018
Available online 6 July 2018

Keywords:
Angioleiomyoma
Nerve tumors
Forearm mass
Vascular tumor
Peripheral nerve

ABSTRACT

Angioleiomyoma is a solitary form of leiomyoma which is typically encountered in the subcutis. They are mostly seen in lower extremities, and the upper extremity is the second most common location for these lesions. There are only a few reports about the presence of an angioleiomyoma within a peripheral nerve in the upper extremity. Here we report a 56-year-old male patient who was referred to our clinic after an attempt was made for removal of a forearm mass at another institution. The lesion was encased within the median nerve and there was an unusual hypervascularity around the tumor with numerous vessels entering the lesion. Removal of the tumor without apparent damage to nerve fascicles was possible. Histopathological examination of the excision material revealed an intraneural angioleiomyoma. Following surgery, the patient was free of any functional deficits and no evidence of recurrence was observed at one year follow-up. There is no data regarding recurrence in intraneural lesions due to the lack of a large series. It would not be wrong to recommend spare grossly uninvolved fascicles if the nerve in question is not expendable.

Introduction

Angioleiomyoma is a solitary form of leiomyoma which is typically encountered in the subcutis. Angioleiomyomas are differentiated from cutaneous leiomyomas by the presence of numerous thick-walled vessels.[1] These lesions are usually painful and tender; this suggests that like glomus tumors, angioleiomyomas (also called angiomyoma or vascular leiomyoma) may originate from arteriovenous anastomoses.[2] They are mostly seen in lower extremities, and the upper extremity is the second most common location for these lesions.[3] So far, to the best of our knowledge, presence of an angioleiomyoma within a peripheral nerve in the upper extremity has been only reported only a few times in the literature.[4–6] Here we report a patient who presented with an angioleimyoma encased within the median nerve at the forearm level.

Case report

A 56-year-old otherwise healthy male patient was referred to our clinic from a community hospital 6 weeks after an attempt was made to surgically remove a left forearm mass by the local plastic surgeon. The surgeon decided not to remove the mass after the exploration revealed that the lesion was arising from the median nerve and had some macroscopic features which he thought were unusual for a Schwannoma. He took per-operative pictures and closed the skin (Fig. 1). Magnetic resonance images before this removal attempt showed a volar mass located at the distal one third of the forearm measuring 3.3 cm × 1.6 cm × 2.4 cm. The mass was hyperintense on the proton density fat saturated images. It also displaced the flexor tendons volarly and expanded the overlying skin (Fig. 2).

The patient reported that he observed a slight growth after the initial surgery. His physical examination revealed a firm, nontender subcutaneous volar forearm mass. There was no Tinel's sign over the lesion. Neurovascular exam of his hand was unremarkable. The patient was then informed and consented about the possible surgical options including incisional biopsy only, removal of the tumor if it can be easily shelled out from the nerve, or excision of the mass

* Corresponding author. Istanbul University, Istanbul Faculty of Medicine, Dept. of Plastic, Reconstructive and Aesthetic Surgery, Hand Surgery Unit, Çapa, Fatih, Istanbul, 34093, Turkey.
E-mail addresses: utkana@hotmail.com (H.U. Aydın), berkoz@gmail.com (Ö. Berköz).

Peer review under responsibility of Turkish Association of Orthopaedics and Traumatology.

https://doi.org/10.1016/j.aott.2018.06.009

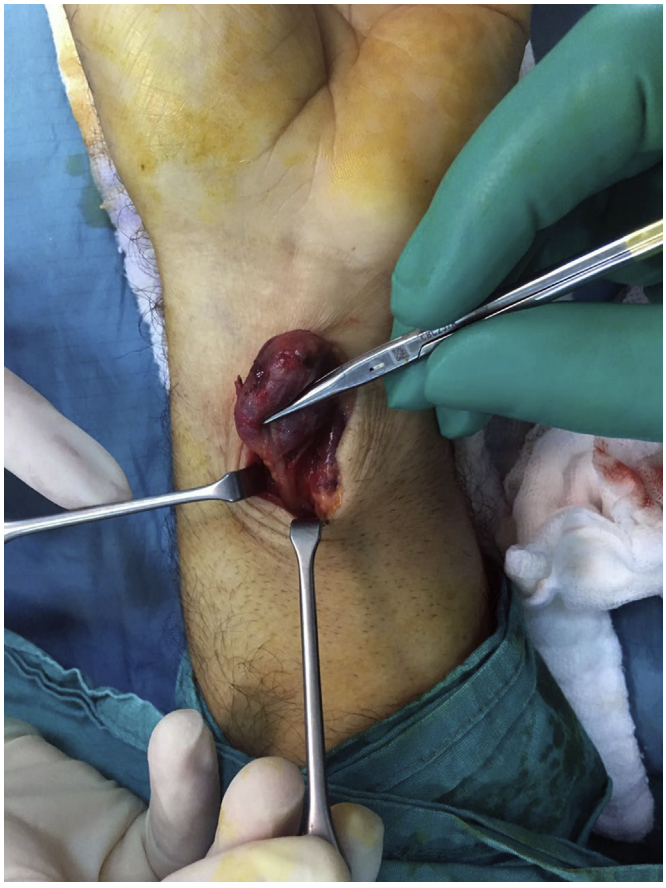

Fig. 1. Photo taken during the first removal attempt. The surgeon decided not to excise the mass since he observed findings unusual for a Schwannoma which was his preoperative diagnosis.

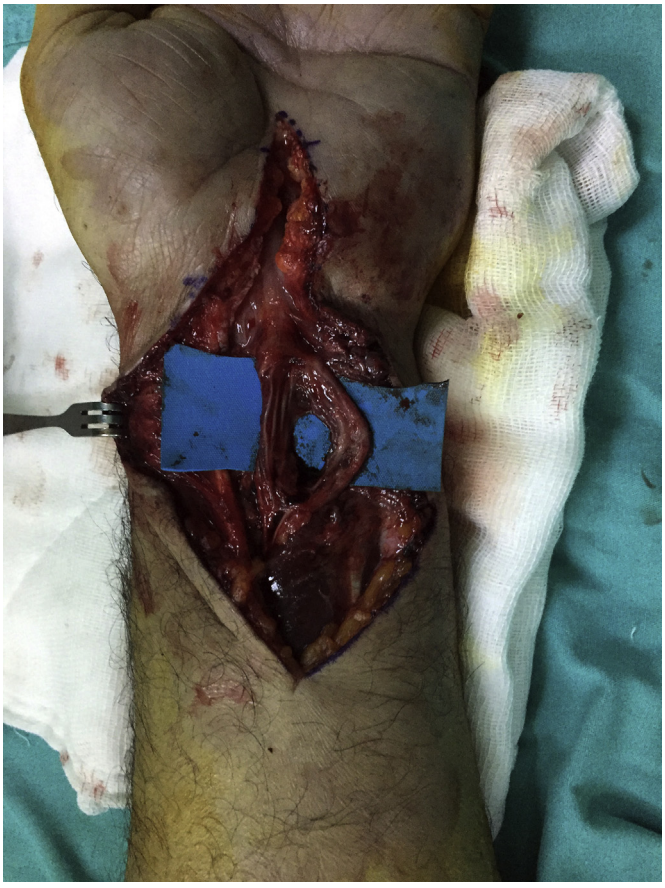

Fig. 3. Intact median nerve fascicles after excision of the lesion.

with the portion of the involved and cable grafting with autologous nerve (Fig. 3).

We used an incision overlying the previous scar to expose the lesion, the incision was extended distally to release the carpal tunnel. We encountered a 3 × 2 cm large, well-circumscribed and elastic mass within the median nerve which was covered by the fascicles of the nerve. Similar to the observations during the previous surgery, we also noted that there was an unusual hypervascularity around the tumor with numerous vessels entering the

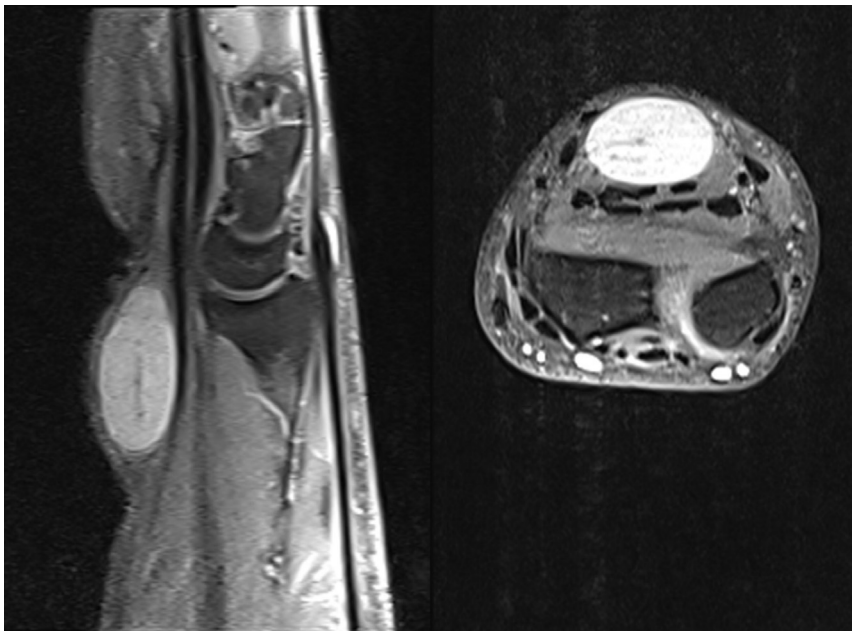

Fig. 2. Axial and sagittal magnetic resonance images of the mass before the first surgery.

lesion. Since we felt that the removal of the lesion was possible without damaging the surrounding nerve fascicles, we proceeded with a tumor excision after carefully cauterizing feeding vessels.

Histopathological examination of the lesion revealed that it was a benign appearing spindle cell tumor containing cavernous vascular structures. The spindle cells were immunoreactive to smooth muscle actin (SMA). Lymphangioma was excluded due to negative staining with D2-40. These findings confirmed a diagnosis of an angioleiomyoma.

We encountered a hematoma at the surgery site on the first postoperative day despite meticulous hemostasis and placement of a drain. We managed this complication with removal of a couple of sutures and draining the hematoma. There was no motor or sensory neurological deficit. The rest of the post-operative course was uneventful and there was no recurrence at 12 months. During the last follow-up visit, the patient had 5 mm static two point discrimination on median innervated fingertips and 5/5 abductor pollicis brevis muscle power according to Medical Research Council Manual Muscle Testing system.

Discussion

When found at other locations in the body, the treatment of angiomyoma is simple excision. Although Duhig and Ayer reported no recurrence after excision in their series, Hachisuga et al, reported two cases with recurrence.[3,6] There is no data regarding recurrence in intraneural lesions due to the lack of a large series. It would be not wrong to recommend spare grossly uninvolved fascicles if the nerve in question is not expendable. Due to vascular character and surrounding feeding vessels, a thorough hemostasis is necessary to avoid further complications. Care must be taken to avoid injury to the nerve while using electrocautery and ligation should be preferred whenever possible.

Unlike cutaneous leiomyomas, these tumors develop later in life, usually between the fourth and sixth decades, as solitary lesions. These lesions are more common in females.[3,7] Interestingly, similar to our present case, both cases of angioleiomyomas involving upper extremity nerves in previous reports also involved median nerve at forearm level.

Per-operative photographs may be helpful if the patient is going to be referred to another facility for a definitive treatment. These photographs will help with the planning of a secondary surgery. However, a fine needle biopsy or open biopsy before definitive surgery is necessary to avoid a diagnostic mistake. Imaging studies and histopathologic examination must be the first priority and the first operation should have been planned as a biopsy procedure with a small incision. If the mass had been a malignant tumor, contamination would have been more severe because of a large incision. The consent form and given information should cover procedures which may be necessary if the mass is identified as a malignant tumor. Although not desirable, it is possible to cancel a tumor removal after exploration, do a biopsy for histopathological diagnosis and refer the patient to a tertiary care unit if the surgeon feels uncomfortable dealing with apparently complex pathology.

References

1. Goldblum JR, Folpe AL, Weiss SW. Benign tumors of smooth muscle. In: Goldblum JR, Folpe AL, Weiss SW, eds. *Enzinger and Weiss's Soft Tissue Tumors*. Philadelphia, PA: Saunders/Elsevier; 2014:524–548.
2. Ekestrom S. A comparison between glomus tumour and angioleiomyom. *Acta Pathol Microbiol Scand*. 1950;27:86–93.
3. Hachisuga T, Hashimoto H, Enjoji M. Angioleiomyoma. A clinicopathologic reappraisal of 562 cases. *Cancer*. 1984;54:126–130.
4. Jing SS, Giesen T. Intraneural angioleiomyoma of the median nerve at the wrist. *J Hand Surg Eur Vol*. 2015;40:639–640.
5. Piers W, Terrono AL, Hayek J, Millender LH. Angiomyoma (vascular leiomyoma) of the median nerve. *J Hand Surg Am*. 1996;21:285–286.
6. Sünram F, Hippe P. Radial nerve paralysis in congenital angioleiomyoma. *Handchirurgie*. 1979;11:27–29.
7. Duhig JT, Ayer JP. Vascular leiomyoma. A study of sixtyone cases. *Arch Pathol*. 1959;68:424–430.