



Chronic pain of the elbow due to angioleiomyoma: a case report

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Angioleiomyoma is a rare and benign smooth muscle tumor arising from the tunica media of vessels. This tumor can be found throughout the body but occurs most commonly in the lower limbs.⁵ It is typically described as a sole small and slowly growing subcutaneous lesion in majority <2 cm. The typical patient is a woman in her third to sixth decade presenting a painful nodule in which pain is triggered by touch, wind, cold, or any other condition described by the patient.^{7,8}

Both ultrasound and magnetic resonance imaging (MRI) are unspecific.⁸ The relative rarity of this disease, the atypical clinical appearance, and the absence of specific imaging lead to an important medical and surgical wandering impacting the daily lives of those patients. We report the case of a 72-year-old woman presenting a painful angioleiomyoma of her right elbow with a diagnosis delayed by 6 years.

She was informed that the data concerning this case would be used for publication, and she gave her consent.

Case report

A 72-year-old woman, retired, right-handed, nonsmoking, without past medical history, presented to our orthopedic surgery facility for right elbow pain evolving for 6 years.

She consulted different physicians before our consultation. A rheumatologist injected her right elbow with cortisone without any improvement 5 years before. Two orthopedic surgeons with upper limb specialty recused her from surgery. A third one performed a lateral epicondylitis surgery 3 years before our examination without any improvement.

Institutional review board approval was not required for this case report.

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Ultrasounds and MRI were performed before the first surgery, but the report did not mention the tumor.

At the date of our consultation, the pain was described as paroxysmal, triggered by the touch of a precise point just posterior to the lateral epicondyle. The palpation of this point triggered a painful crisis that can last for up to 40 minutes and recur 2 to 3 times per day.

Clinical examination was normal, the flexion and extension of the elbow was normal so was median, radial, and ulnar nerves examination. There was not any motor nor sensitive deficit.

The clinical presentation was consistent over the past 6 years.

First of all, all these elements made us think about a glomeric tumor.

Thereby, an MRI with specific sequences was performed at our institution exhibiting a well-defined mass at the posterior-lateral face of the elbow, close to the distal part of the triceps. The lesion presented a low intensity on T1-weighted sequence, a high intensity on DP-FAT SAT sequence, a homogeneous enhancement after gadolinium injection (Figure 1). The radiologist described the aspect of this mass as atypical and could not rule out a glomeric tumor. Retrospectively, the tumor was viewable on the first MRI, but the first radiologist did not mention its presence.

After a collegial discussion including radiologists and orthopedic surgeons, an excision of the lesion was performed under locoregional anesthesia with the use of a tourniquet. It was decided to carry out a more invasive cancerological surgery only if histologic examination would have found signs of malignancy. The incision was centered on the mass. Subcutaneous dissection was performed. The mass was found deep to the triceps and anconeus aponeurosis, it was not adherent to the muscle and presented a thick capsule (Figure 2). Its removal was uneventful.

The radiohumeral articulation was visualized after excision and was normal.

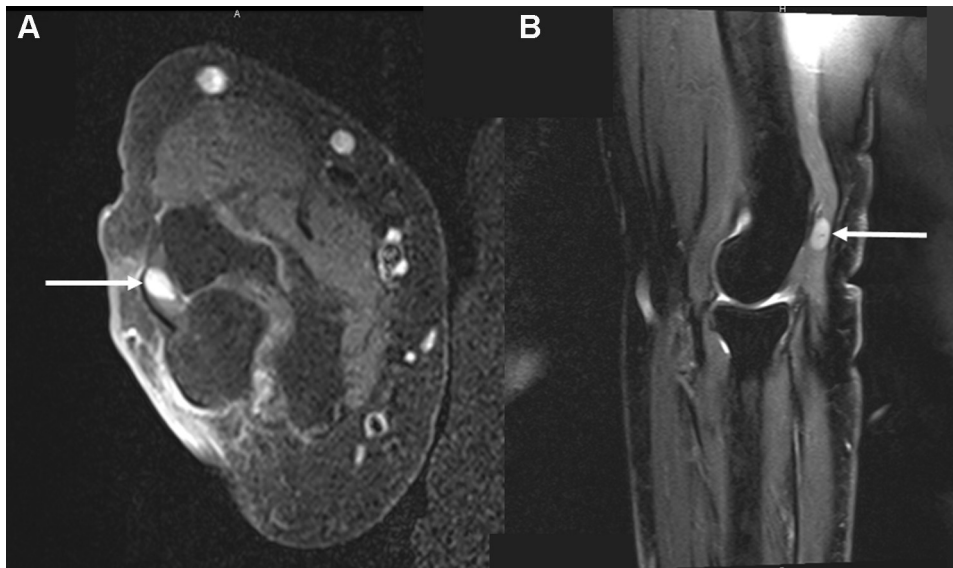


Figure 1 (A) Magnetic resonance imaging (MRI). Axial T1 TSE with gadolinium injection. *White arrow*: Ovoid mass deep at the junction between the anconeus and distal triceps muscle, well defined. The lesion is homogeneously enhanced by gadolinium. (B) MRI. Sagittal DP FS. White arrow pointing the lesion.



Figure 2 Perioperative view of the tumor.



Figure 3 The tumor after excision. Note the well-defined and ovoid shape of the tumor without any sign of malignancy.

The piece of resection was a 1-cm, firm, ovoid and well-defined mass (Figure 3).

Postoperative course was uneventful.

During the follow-up, she described a total disappearance of her painful crisis and was highly satisfied with the surgery.

Histologic examination reported a tumor composed of vascular channels and smooth muscle consistent with an angioleiomyoma, subtype solid (or capillary).

Discussion

Angioleiomyoma is a rare, benign smooth muscle arising from the tunica media of the vessels. This tumor is mostly found in the lower limb. Its development at the elbow is rare, in a cohort of 562 patients, Hachisuga et al⁵ only report 27 cases at the elbow.

Angioleiomyoma occurs preferentially in women in their third to sixth decade; our patient was slightly older.

A majority of these tumors are generally <2 cm, which is consistent with our case.^{5,8}

Classically, the clinical presentation is a history of a subcutaneous mass causing a chronic pain characterized by acute crisis triggered by touch or other conditions described by the patient such as cold.^{3,7,8} In our case, the tumor was deeper under the anconeus aponeurosis, reinforcing its atypia. Hasegawa et al⁶ demonstrated that the pain could be caused by nerve fibers located within the tumor. Dramis et al reported the case of an elbow fixed in flexion due to an angioleiomyoma irritating the anterior capsule.²

Ultrasounds and MRI are described as atypical and cannot rule out a glomus tumor, a ganglion, a neuroma, or another soft-tissue tumor. Although it is nonspecific, those examinations reveal a benign appearing lesion with well-defined margins, a homogeneous structure.⁸ A recent MRI study of ten angioleiomyomas described its MRI aspect as isointense to muscle on T1-weighted spin echo with heterogeneous increased T2W/STIR signal intensity. Its enhancement after IV gadolinium is variable from diffuse to heterogenous. The authors underline a typical aspect of linear or branching hyperintensity on T2W or STIR image.⁴

Thus, the diagnosis is rarely performed preoperatively and the definitive diagnosis is completed with histologic examination.

Under histopathologic examination, 3 subtypes of angioleiomyoma are described: capillary, cavernous, and venous. The proportion of each subtype differs in the different studies. The solid subtype is known to be the most frequent in women; our case is consistent with this. Hachisuga et al⁵ described a positive correlation between the solid subtype and the prevalence of pain which is not found by other authors.⁸

Its nonspecific clinical and imaging presentation is a cause of delayed diagnosis, 6 years in our case, detrimental to the quality of life of the patient.

Initially, our case was thought to be a glomus tumor following the notion of paroxysmal pain triggered by the touch. Even if the MRI was not specific of glomus tumors, they are known to cause this kind of clinical presentation.¹

Surgery offers an immediate relief, and postoperative course are uneventful. This is the treatment of choice especially that morbidity is very low and recurrence is described in few cases.⁵

Conclusion

This case emphasizes that angioleiomyoma as a secondary diagnosis of chronic elbow pain that needs a close clinical examination and appropriate imaging to prevent delay in diagnosis. Its surgical excision is the gold standard as it offers immediate relief and very low recurrence rates.

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