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Case Report

Angioleiomyoma in the posterior knee: A case report and literature review

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ABSTRACT

The authors present a case of angioleiomyoma situated in the posterior knee.
A 47-year-old Caucasian woman presented in 2011 with recurrent stabbing pain on the lateral aspect of her right knee. She reported having pain for the last 6 years. She had no history of trauma. In 2008 she was treated with a diagnostic arthroscopy and transposition of the tibial tuberosity, with no benefit to her symptoms. Electromyography of the lower limbs showed asymmetry of the amplitude of sensitive action potential of the superficial fibular nerve. Based on the clinical suspicion of entrapment of the common fibular nerve at its bifurcation, a surgical exploration was performed, but pain persisted. In 2014, ultrasonography localized at the trigger point showed a solid ovular formation of 1 cm in diameter situated on the posterior aspect of the external femoral condyle in proximity to the joint capsule, which was confirmed by magnetic resonance imaging (MRI). Surgical excision of the 1-cm diameter tumor mass relieved the symptoms immediately and permanently. Histology evidenced the presence of a solid-type angioleiomyoma.
The presence of an angioleiomyoma at the knee joint is very rare and few cases are reported in the literature. To the authors' knowledge this is the first time an angioleiomyoma in the posterior knee has been described. In case of unexplained and persistent pain in and around the knee, clinicians should be aware of the atypical locations of this tumor, considering that its surgical excision alone may relieve symptoms permanently.

1. Introduction

Angioleiomyoma is defined as a blood vessel leiomyoma, which is considered to be a benign neoplasm originating from the tunica media of the blood vessel walls [1]. The histological appearance is characterized by smooth muscle and prominent blood vessels [2–5]. The incidence of angioleiomyoma is about 5% amongst all soft tissue tumors, with the lower limbs being most commonly affected [2]. However, this type of neoplasm is rarely located at the knee joint, and few cases have been reported in the literature [6–12]. The tumor is common in middle-aged adult patients, especially females who appear to be affected twice as often [2,8,13–16]. The typical clinical symptom includes localized pain in the subcutaneous region [2,17], which is why it was improperly named in the past as "tuberculum dolorosum" based on the belief that the neoplasm was a cutaneous leiomyoma [18,19]. Diagnosis before surgery tends to be quite uncommon, considering that angioleiomyoma is rarely seen and that orthopedic surgeons have limited awareness in this field [20]. Patients may receive repeated conservative and/or surgical treatment, especially if the neoplasm is situated in proximity to the knee joint [8,21,22]. These rare tumors located in the knee can be misdiagnosed with other common pathologies like meniscus or cartilage disease, which is what occurred in this case-report

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patient. This case report is based on a patient with an angioleiomyoma situated in the posterior knee. It is believed that, to date, there have been no other similar cases reported in literature.

2. Case report

A 47-year-old Caucasian woman was examined at the present hospital in 2011 for recurrent stabbing pain on the lateral side of her right knee. She had no history of trauma, appeared to be in good health and was not on any regular medication. There was no past history of oral medication with hormonal contraceptives. The onset of pain was sudden and intense, with radiation in both proximal and distal directions along the lateral side of the limb that lasted for about one minute, which occurred about once or twice daily on a regular basis over several years. The patient kept a meticulous diary of her disease history, which included specific activities and events that triggered pain, the frequency of the painful events, and the subjective therapeutic success of treatments (Table 1). The patient stated that symptoms began in 2005 without any apparent reason, and continued to date.

She underwent ultrasonography and Doppler-sonography in 2005, which were both normal. She then had a radiograph of her knee, which showed slight narrowing of the medial joint line. Magnetic resonance imaging (MRI) of the knee in 2006 reported moderate trochlear dysplasia with excessive lateral pressure of the patella. In 2008, she underwent diagnostic arthroscopy and transposition of the tibial tuberosity without any symptomatic benefits.

At the first clinical evaluation of her knee in 2011, there were no signs of meniscal pathology or instability. The pain could be triggered by digital pressure just posterior to the fibular head and seemed to be of neuropathic origin, being described by the patient as an electric shock migrating along the limb. A new MRI of the knee was obtained in 2011, which seemed substantially normal. Electromyography of the lower limbs was performed that same year, which showed asymmetry of the amplitude of sensitive action potential of the superficial fibular nerve. An MRI of the spine showed some minor lumbar protrusions in absence of root compression. Based on the clinical suspicion of an entrapment of the common fibular nerve at its bifurcation, a surgical exploration was proposed and accepted by the patient in 2012. However, the symptoms persisted after the surgery. In order to rule out subtle nerve root compression situated in the lumbar spine that could explain the symptoms, an MRI in the orthostatic position was performed in 2013, which confirmed the presence of minor lumbar protrusions in the absence of root compression. The patient tried different kinds of treatment during her illness (oral administration of alpha lipoid acid for six months as well as pregabalin 225 mg/day for two months, physiotherapy, osteopathy, etc.) without any relief of symptoms. Five consecutive sessions of acupuncture in addition to a 15-day treatment with clay packs seemed to decrease the pain intensity and frequency for a short period of time. In 2014, further ultrasonography was performed and, for the first time, the examination localized at the trigger point showed a one centimeter diameter, solid, ovular vascular formation situated on the posterior aspect of the external femoral condyle in proximity to the joint capsule, which was then confirmed by a new MRI of the knee (Figs. 1 and 2). The new MRI was compared with the previous MRI performed in 2011, which retrospectively confirmed that the neoplasm was already present, but was not diagnosed in 2011 by both the radiologist and treating surgeon. The patient underwent surgical excision of the one centimeter tumor mass, which relieved symptoms immediately and permanently (Figs. 3 and 4). The histological assessment confirmed a solid-type angioleiomyoma.

3. Discussion

Subcutaneous soft-tissue masses can be divided in five categories: (1) mesenchymal tumors, most frequently presenting as lipomas, angiomas, peripheral nerve sheath tumors and malignant fibrous histocytomas, the last one representing the most common subcutaneous malignant soft-tissue tumor; (2) skin appendage lesions; (3) metastatic tumors; (4) other tumors and tumor-like lesions rarely encountered in the adult population, such as myxomas, lymphomas and granuloma annulare; (5) inflammatory lesions [23]. The present patient presented a subfascial angioleiomyoma, which is an uncommon mesenchymal neoplasm located in the posterior region of the knee. It is believed that there are few studies about subfascial soft-tissue tumors in the literature; they mainly describe typical and atypical lipomatous tumors or liposarcomas [24].

Leiomyoma of the soft tissues is a benign neoplasm derived from smooth muscle cells. It can be divided into three groups: superficial, vascular and deep leiomyoma; angioleiomyomas are a variant that have a typical presenting onset as a painful

Table 1
Number of pain recurrences related to specific activities, and observed by the patient from 2009 to 2013.

ACTIVITY	NUMBER OF PAIN RECURRENCES
when startled	14 (0,7%)
sneezing	27 (1,4%)
sitting	27 (1,4%)
during fitness	61 (3,2%)
sitting on toilet	86 (4,5%)
standing still	287 (15,0%)
walking	525 (27,3%)
sleeping or while getting up	894 (46,5%)

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