Late diagnosis of angioleiomyoma of the knee: a case report

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Summary

Angioleiomyoma, also known as angiomma, vascular leiomyoma or dermal angioma is an infrequent benign tumour originating from smooth muscle cells of arterial or venous wall. The peak incidence is between the third and sixth decades of life; a female preponderance has been reported. Minor trauma, venous stasis, hamartomatous changes, arteriovenous malformations and hormonal imbalance especially of oestrogen have been proposed for the etiology of the tumor. Standard treatment includes marginal surgical excision of the tumor. Local recurrence or malignant degeneration after surgical excision is rare. The duration of symptoms prior to diagnosis ranges from weeks to few years. This article presents a patient with a slowly enlarging angioleiomyoma of the knee over a time period of 12 years. The clinicopathological features, diagnosis and treatment are discussed.

KEY WORDS: angioleiomyoma; soft tissue; diagnosis.

Introduction

Angioleiomyoma, also known as angiomma, vascular leiomyoma or dermal angioma is an infrequent benign tumor originating from smooth muscle cells of arterial or venous wall. The peak incidence is between the third and sixth decades of life, with most cases diagnosed in the third and fourth decades of life (1, 2). A female preponderance has been reported (1, 3, 4). The prevalence of this lesion in East Africa has been reported to be 10 times that of the white population living in the same temperate climates (3). Minor trauma, venous stasis, hamartomatous changes, arteriovenous malformations and hormonal imbalance especially of estrogen have been proposed for the etiology of the tumor (4).

Most Authors agree that vascular leiomyoma arises from veins (4). Proliferation of smooth muscle within a haemangioma may produce a vascular leiomyoma that may further proliferate to a simple leiomyoma (1, 4). Other Authors suggested that an appreciable number of angioleiomyomas are not true tumors but vascular malformations. Karyotypic abnormalities including the 6p, 13q and 21q rearrangements and the t(X;10) (q22;q23.2) translocation have also been reported (4).

Case report

A 46-year-old man presented with a 12-year history of a slowly enlarging mass at the medial aspect of his left knee. Pain was sporadic and progressively worse over the last 5 months. Past medical history was unremarkable. The patient denied any history of trauma to the area.

Physical examination showed a slightly mobile tender mass at the medial aspect of his left knee. The skin was intact and there was no erythema or groin lymphadenopathy. Routine laboratory analysis including complete blood cell count, serum chemistries, erythrocyte sedimentation rate, and C-reactive protein were within normal limits. Radiographs of the left knee were normal. Ultrasonography showed an 18.2×10.5 mm, hypoechoic mass with well-defined margins (Figure 1). Color Doppler ultrasonography showed high pressure intratumoural arteries suggesting the presence of muscular arteries. MR imaging showed a soft-tissue mass embedded into the vastus medialis muscle. T1-weighted MR imaging shows a well-defined ovoid mass with signal intensity similar to muscle on T1-weighted sequences and higher signal intensity relative to muscle with a hypointense peripheral rim on T2-weighted sequences (9, 10). The imaging differential diagnosis of angioleiomyoma should include other nodular lesions of the extremities such as lipomas, ganglia, fibromas, schwannomas, haemangiomas, foreign body granulomas, pseudoaneurysms, inclusion cysts, giant cell tumors of the tendon sheath, and glomus tumor. Standard treatment includes marginal surgical excision of the tumor. Local recurrence or malignant degeneration after surgical excision is rare (1, 11). This article presents a patient with a slowly enlarging angioleiomyoma of the knee over a time period of 12 years. The clinicopathological features, diagnosis and treatment are discussed.
imaging showed a low-signal intensity homogenous mass that was isointense to muscle; T2-weighted MR imaging showed high signal intensity heterogeneous areas within the mass (Figure 2A-C).

Through a 5-cm vertical incision, complete excision of the mass was done. The tumor was encapsulated and partially adhesive to the muscle fibers of the vastus medialis muscle. Grossly, the tumor was smooth, elastic soft, white and red colored. Histological sections showed numerous blood vessels of various size and copious fascicles of smooth muscle bundles surrounding the vessels (Figure 3AB). These findings were consistent of angioleiomyoma.

Pain relief was observed immediately postoperatively. However, at 2 years after the operation, new onset of pain and a palpable mass at the area of the previous operation was observed. True-cut biopsy and histological examination of the excised specimen showed local recurrence of the angioleiomyoma; marginal re-excision was done. At the last follow-up examination, 3.5 years after re-excision, local tumor recurrence was not observed.

Discussion

Angioleiomyoma accounts for 5% of all benign soft-tissue tumors (4, 5, 12-15). Three histological types have been reported. The most common solid type is characterized by closely compacted smooth muscle fibers and abundant small, slit-like vascular channels; this type is three times more common in females and typically involves the lower extremities. The venous type is identified by thick muscular walls. The least common cavernous type is characterized by dilated vascular channels with less smooth muscles. The cavernous subtype is four times commoner in males and accounts for the majority of head and upper extremity angioleiomyomas (1).

The typical lesion is a painful, solitary, <2 cm, slow-growing, firm, mobile, subcutaneous nodule (16). Giant angioleiomyomas have also been reported (17-19). Pain is the most common clinical manifestation. Pain originates from smooth muscle contraction, or irritation of the involved nerves. Hypersensitivity to cold is usual (6, 12); this has been attributed
to tumour vessel contraction and ischemia. A variable pain-free interval to diagnosis has been reported ranging from several weeks to a few (4-7) years (1). In the current patient, the tumour was palpable for 12 years with sporadic episodes of slight pain. To the best of our knowledge, a similar time interval has not been previously reported.

MR imaging is the imaging modality of choice to delineate the lesion and define its relationship to adjacent structures allowing better pre-excisional planning (10, 13, 20). Histologically, degeneration and secondary calcification or severe hyalinization in the long-term is typical (18), which, however, was not observed in the present patient. Marginal excision of the tumor is diagnostic and therapeutic, with immediate pain relief, as in the current patient. However, although rare, local recurrence, as in the present case, may occur; malignant degeneration is exceptional (1). In case of local recurrence, the possibility of angioleiomyosarcoma should be evaluated.

In conclusion, angioleiomyoma can grow locally for up to 12 years prior to diagnosis and treatment, and should be included in the differential diagnosis of any lower-extremity soft-tissue mass. Marginal excision is usually sufficient; however, local tumor recurrence may occur.

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References