



Case report

Intra-articular leiomyoma of the knee mimicking a ganglion cyst in a child: A case report and review of literature

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ARTICLE INFO

Keywords:

Knee
Anterior cruciate ligament
Leiomyoma
Ganglion cyst
Arthroscopy
Case report

ABSTRACT

Introduction and importance: Angioleiomyoma is a rare benign soft tissue tumor of smooth muscle getting its origin from the muscular layer of vessel walls. The localized type of leiomyoma occurs on the skin, deep soft tissues, in the genitourinary, gastrointestinal, and respiratory tracts.

Case presentation: We report a case of a 16-year-old patient who presented with an intra-articular leiomyoma developed in the anterior cruciate ligament and treated with arthroscopy.

Clinical discussion: Angioleiomyoma can occur anywhere in the body and is most often seen in the extremities, particularly the lower limbs, but is very rare in the knee joint. Angioleiomyoma should be included in the differential diagnosis of nodular lesions mimicking loose body, such as lipomas, inclusion cysts, ganglion, pigmented villonodular synovitis (PVNS), fibroma, nodular synovitis, hemangioma, synovial sarcoma, myopericytoma, leiomyosarcomas, glomus tumor.

Conclusion: Angioleiomyoma related to the cruciate ligaments of the knee should be considered in the differential diagnosis of a painful knee especially when associated with a decreased of range of motion of knee. The arthroscopic debridement should be considered the treatment of choice in order to reliably restore active ROM.

1. Introduction

Angioleiomyoma is a rare benign soft tissue tumor of smooth muscle origin arising from the muscular layer of vessel walls [1]. The most common presentation is a painful solid subcutaneous mass. The peak incidence is in the fourth to the sixth decades of life, with a female preponderance [2]. Previous literature series showed that this entity can be found anywhere in the body such as the uterus, crania, atrium, hard palate, nasal septum [3,4]. Although angioleiomyoma commonly affects the lower extremities, knee joint is a rare location, especially the intra-articular angioleiomyoma which is presented as loose body. As far as we know, only a minority of individual cases have been reported [5,6].

We present a rare location of an intra-articular angioleiomyoma arising from the anterior cruciate ligament mimicking a ganglionic cyst. This study is reported in line with the SCARE 2020 checklist [7].

2. Case report

A 15-year-old boy was admitted to our orthopedic department for knee pain and gradually movement restrictions in the left knee revealed after a trauma and lasting for one year. The young patient was victim of a fall with landing on the left knee during school accident. The patient had no past medical or surgical history. His family members were healthy. He didn't use any kind of drugs.

On examination, there was no swelling and no inflammation signs. The range of motion was -60° of extension and 80° of flexion. There were negative findings on the Lachman and pivot-shift tests and no swelling or tenderness along the joint line. The range of motion was limited, with loss of extension and flexion. Antero-posterior and lateral radiographs were normal. Magnetic Resonance Imaging (MRI) showed a 33 mm cyst arising from the anterior cruciate ligament (ACL) in its attachment on the intercondylar notch. The cyst is well limited and shows low signal intensity on T1 weighted images and high signal intensity on Fat Saturated Proton Density (Figs. 1, 2). The MRI appearance was compatible

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<https://doi.org/10.1016/j.ijscr.2021.106320>

Received 20 July 2021; Received in revised form 14 August 2021; Accepted 15 August 2021

Available online 18 August 2021

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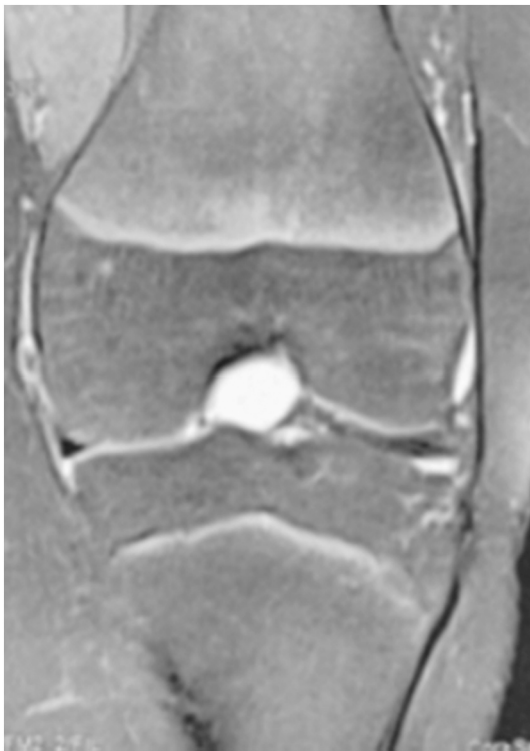


Fig. 1. Frontal MRI showed a 33 cyst on depends of the cruciate ligament.



Fig. 2. The cyst is well limited and shows high signal intensity on MRI Fat Saturated Proton Density.

with an intrarticular muroid cyst of the anterior cruciate ligament. Surgery was performed under general anesthesia and intubation.

It was carried out by our orthopedic surgery team of Habib Bougatfa Hospital. This procedure didn't need advanced technical or logistical resources and could be attempted by average knee arthroscopists.

We proceeded with arthroscopy in order to excise the mass and to have a biopsy. Arthroscopy revealed a voluminous tumor with homogenous solid aspect (Figs. 3, 4). The anterior and posterior cruciate

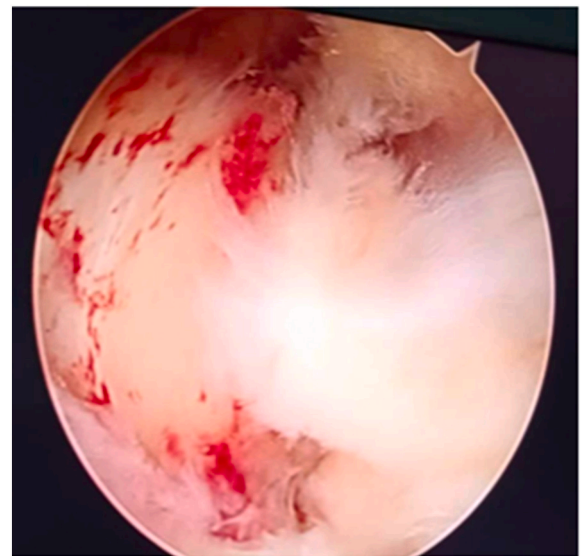


Fig. 3. A vascularized mass developed around the central pivot of the knee.

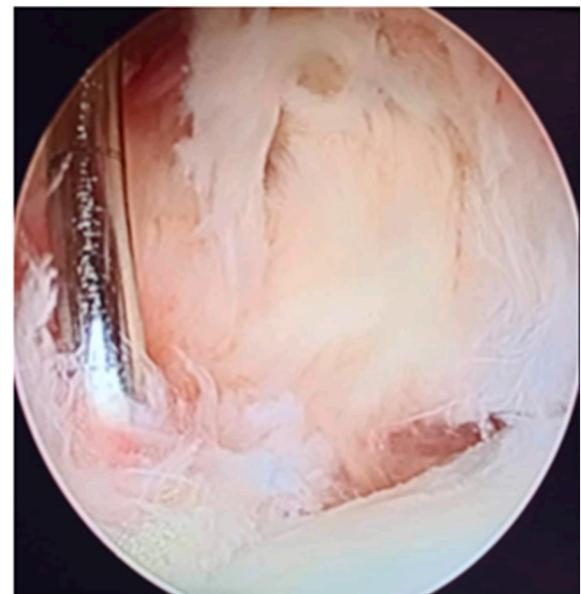


Fig. 4. Arthroscopic intraoperative image of the mass developed around the central pivot of the knee.

ligament was intact.

Pathological examination of the surgical specimen showed a mesenchymatous proliferation of benign appearance made up of spindle-shaped cells, devoid of atypies and mitotic activity, arranged in entangled bundles and encircling multiple vessels of the venous type, with a dilated congestive lumen and sometimes narrowed.

Histological examination of the mass with selective immunohistochemical stains showed positive for caldesmone and negative for desmin, CD34 and P100 (Fig. 5). The diagnosis of a leiomyoma is made.

On discharge, the patient received analgesic and antiinflammatory agents and physiotherapy. No postoperative complications were noted. The range of motion was 0° of extension and 120° of flexion. Rehabilitation protocol was prescribed for the patient which focuses on addressing knee range of motion (ROM) deficits, initiating knee strength and control, minimizing pain and effusion, and normalizing gait. Since he lives in a rural area, he was unable to do all the rehabilitation sessions.

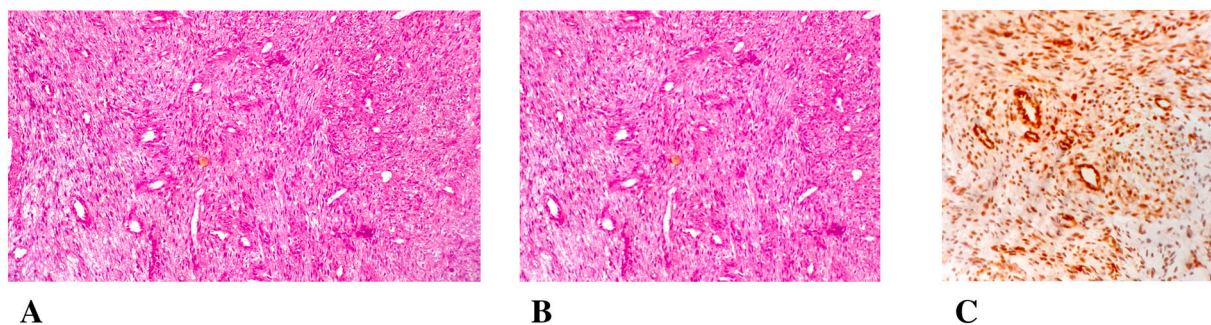


Fig. 5. A: Benign mesenchymal proliferation made up of long, crisscrossing bundles (HE magnification 10×); B: spindle-shaped tumor cells, devoid of atypia and mitotic activity (HE magnification 40×); C: intense and diffuse positivity of tumor cells to the anti-Caldesone antibody (immunohistochemistry (IHC) magnification 40×).

With a one year follow-up, the patient consulted for a restriction of extension with a range of motion was 30° and flexion of 90°. The MRI showed an absence of signs of hemangiomas recurrence and a fibrosis interposing between the PCL and ACL and under the patella and fibrosis was made as a diagnostic for stiffness of the knee (Fig. 6). The fibrosis is probably related to inflammatory changes from the previous surgical procedure and especially localize during arthroscopic exploration in front of the anterior cruciate ligament.

Resection of the fibrosis by arthroscopy was performed by the two classic anteromedial and anterolateral arthroscopic routes. We noted the absence of tumor recurrence intraoperatively. At the six month follow-up, the patient was satisfied with a range of motion was 0° of extension and 120° of flexion and was satisfied with the result.

3. Discussion

The present report describes an unusual case of intra articular tumor with a restriction ROM and pain of knee. The restricted knee motion appeared to be caused by an ACL mass being caught in the notch space upon extension. At microscopic examination, angioleiomyoma shows tortuous vascular channels surrounded by smooth muscle bundles and areas of myxoid change that explain hyperintensity of the tumor on T2. On immunohistochemistry, angioleiomyoma is usually positive for smooth muscle actin, desmin and caldesmon, and negative for myeloid progenitor cell antigen (CD34), with no cellular atypia and/or mitoses. In our case, the immunohistochemical examination of the mass

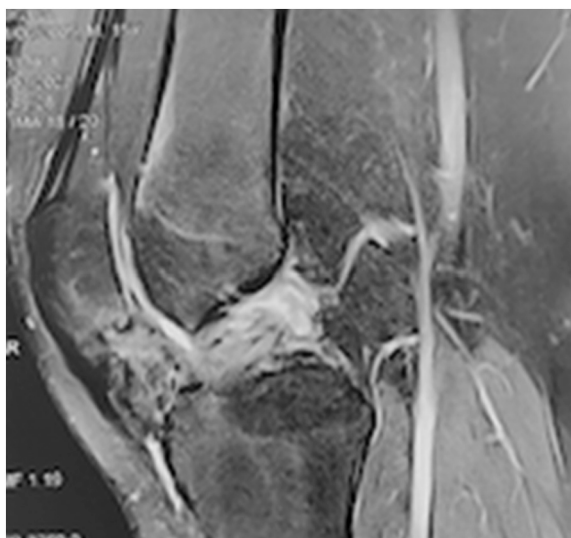


Fig. 6. The MRI showed an absence of signs of hemangiomas recurrence and an intra-articular fibrosis was made diagnosis.

surgical specimen showed positive for caldesone and negative for desmin, CD34 and P100.

Differential diagnoses in the current case may have included an intra-articular ganglion cyst, giant cell tumor or synovial chondromatosis arising from the cruciate ligament. Those conditions may also cause pain, swelling, tenderness and loss of joint function. The intra-articular ganglion cysts and synovial chondromatosis can cause a limitation of range of motion [8–10].

In the current case, although MRI was very useful in locating the lesion, the findings did not allow us to identify the type of lesion. We initially considered a tumorous condition such as a ganglionic cyst or giant cell tumor, given that an angioleiomyoma is a rare entity and has no distinguishing MRI features.

Angioleiomyoma can occur anywhere in the body and is most often seen in the extremities, particularly the lower limbs [11], but very rare in the knee joint. Al-Jabri and Murty reported a case of an intra articular angioleiomyoma presented with recurrent pain and a soft tissue swelling of knee [12,13]. We reported two cases of angioleiomyomas presenting loose bodies in the knee joint, and the case of angioleiomyoma with calcification in the knee should be the first report in the literatures. Intra-articular tumors are very unusual and physicians are apt to misdiagnose conditions like meniscus tear or arthritis [14]. To avoid making a misdiagnosis, a high index of suspicion is required. Mazen Hamoui et al. suggested that angioleiomyoma should be included in the differential diagnosis of nodular lesions mimicking loose body, such as lipomas, inclusion cysts, ganglion, pigmented villonodular synovitis (PVNS), fibroma, nodular synovitis, hemangioma, synovial sarcoma, myo-pericytoma, leiomyosarcomas, glomus tumor [15]. Usually an angioleiomyoma presents as a painful, solitary, small, slow-growing and mobile lesion, the majority are oval and less than 2 cm in size [16].

There are no specific imaging techniques for vascular leiomyomas. Radiograph plain film has little value in diagnosis. MRI is not specific for diagnosis. The diagnosis of angioleiomyoma must be evoked in front of a well-demarcated mass with isointense signal to muscle on T1, heterogeneous high signal intensity on T2, heterogeneous strong enhancement, and an adjacent tortuous vascular structure is seen in the extremities [17].

It is recommended that the patient obtain full passive and active knee extension range of motion within the first 2 weeks. Failure to achieve full knee extension ROM can have a significant long-term impact on pain, gait and function, with arthrofibrosis as a potential complication. Arthrofibrosis is an inflammatory response which results in joint fibrosis and restricted knee motion [18], which can occur following knee surgery, trauma or immobilization [19]. Prevention of knee joint stiffness and potential arthrofibrosis should be a primary goal early post-operatively [20]. Patient education is crucial to carryover the home exercises designed to address knee extension ROM, with the frequency of physical therapy sessions established to maximize early return of full knee extension ROM. It is important to recognize that even a loss of less

than 5 degrees of knee extension ROM can lead to long-term patellofemoral pain issues, quadriceps strength deficits and a bent-knee gait abnormality [21,22]. In our case the recurrence of the stiffness of the knee is related to the nonproximity of the place of rehabilitation and the lack of education of his parents

4. Conclusion

Although extremely rare, Angioleiomyoma related to the cruciate ligaments of the knee should be considered in the differential diagnosis of a painful knee especially when associated with a decreased ROM and no evidence of osteoarthritis on radiographs. Other pathology can often be present, which may affect the overall outcome, but arthroscopic debridement of Angioleiomyoma should be considered the treatment of choice in order to reliably restore active ROM.

Sources of funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Ethical approval

The study type is exempt from ethical approval.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor in Chief of this journal on request.

Author contribution

Writing the paper, Study Concept: Khezami karim.
Data collection, Study Concept: Bennour Mohamed Amine.
Supervision: Mohamed Amine Bennour.

Registration of research studies

Not applicable.

Guarantor

Dr. Khezami Karim.

Provenance and peer review

Not commissioned, externally peer reviewed.

Declaration of competing interest

This article has no conflict of interest with any parties.

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