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Case Report: A Rare Case of Intra-articular Cavernous Hemangioma of the Knee

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Authors' contributions

This work was carried out in collaboration between both authors. Author TK wrote the manuscript and managed the literature search. Author MD approved the manuscript. Both authors read and approved the final manuscript.

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Case Study

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ABSTRACT

Aims: Intra-articular cavernous hemangioma is a rare benign tumour with fewer than 200 cases described in literature. It is a form of congenital vascular malformation which can be difficult to diagnose in routine clinical practice.
Case: We present case of a 20 years old female with recurrent painful knee swelling, which remained undiagnosed for 9 years. Blood investigations were equivocal.
Discussion: Although rare, such tumours can be frequently disabling with pain, recurrent effusions and restricted range of motion being amongst the common symptoms. Multiple episodes run the risk of causing cartilage damage at an early age. Imaging studies like MRI remain gold standard.
Conclusion: High index of suspicion is required for early diagnosis and optimum management.

Keywords: Adolescent; hemangioma; knee; MRI; cartilage damage.

1. INTRODUCTION

Hemangiomas are tumours characterised by an increased number of normal or abnormal vessels filled with blood. These constitute around 7% of all benign tumours of infancy and childhood; most are present from birth and may initially increase in size, but many eventually regress spontaneously. While the majority of cases are localized, some can occur extensively

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(angiomatosis). Malignant transformation is generally rare. The histological and clinical variants which are described include - capillary hemangiomas, Juvenile hemangioma ("strawberry type"), Cavernous hemangioma and Pyogenic granuloma. Cavernous hemangiomas are large dilated vascular channels which in comparison to capillary hemangiomas are more infiltrative, frequently involve deeper structures and often do not resolve spontaneously. Histological examination reveals an un-encapsulated mass with blood-filled spaces separated by connective tissue stroma. Depending upon the site of origin, they may remain asymptomatic or may lead to significant complications (if located in the brain) [1]. Soft-tissue hemangiomas can be categorized based on the site of origin as cutaneous, subcutaneous, intramuscular, synovial or subsynovial [2].

2. CASE PRESENTATION

A 20-year-old female patient presented to us with a 9-year-old history of recurrent pain with swelling in the left knee with no antecedent history of any injury to the knee. The patient also had an initial restriction in the Knee ROM which had subsided when she presented to us. She had a knee aspiration done 4 years back, but its details were not available with the patient. All previous documents were similarly not available. The affected thigh was slightly atrophic. On local knee examination, a boggy swelling was palpable mainly over the anteromedial aspect of the left knee. The swelling was tender on palpation with vague margins and soft consistency. The knee range was from 0 to 100 degrees without laxity. As expected the X-ray of the affected knee showed abnormal soft tissue shadows but no bony abnormality.

An MRI scan with contrast was carried out which showed the presence of a poorly marginated lesion located within the medial recess of the suprapatellar bursa and insinuating into the medial infrapatellar Hoffa's fat pad, further extending into the medial infrapatellar subcutaneous tissue.

The lesion closely abutted the anterior body and the anterior horn of the medial meniscus.

The lesion measured approximately 5.8 x 2.3 cm in maximum transverse dimension and approximately 5.2 cm in craniocaudal extent.

An Angiography of the lower limb showed that the hemangioma was supplied by feeder vessels arising from the mid-popliteal artery. We consulted the Vascular Interventional Radiology department, which advised to perform sclerotherapy for the alleviation of the lesion. The patient underwent the procedure uneventfully and was subsequently discharged. Post-procedure MRI was not carried out. However, she presented again after 2 months with swelling over the anteromedial aspect of the knee and on-off pain.

In view of the recurrence of symptoms, a decision was made to perform open surgical excision of the lesion. The option of Arthroscopic excision was also considered, but, it was rejected for 2 reasons; firstly, in view of the risk of significant bleeding with mechanical instrumentation of the tumour and secondly because the lesion, in this case, was not a pedunculated tumour which facilitates arthroscopic removal [3,4].

The patient was given Spinal with Epidural anaesthesia. In a supine position, with a tourniquet applied the regular midline knee incision was taken. With meticulous attention to bleeder's, a medial parapatellar approach was used to open the joint. Due to the prior intra-venous sclerotherapy, the size and appearance of the lesion was subdued. The lesion itself was visible as a bluish-purple mass with mulberry like appearance which was friable and prone to bleed on handling. After completing the arthrotomy we attempted to dissect out the entire tumour, however, in view of the extremely friable nature, it was not possible to dissect the entire lesion *en-masse*.

With careful dissection, we were able to remove the tumour growth from the infra-patellar fat pad, from the close vicinity of Deep Medial Collateral ligament, and the mass from the medial recess of the suprapatellar bursa. The intra-op pictures provided show lesion prominently in the medial recess of the suprapatellar pouch, and within the Hoffa's fat pad. The samples collected and sent for histopathological evaluation (slide pictures are also provided).

The histopathological evaluation confirmed the presence of dilated vascular channels and lakes of blood cells, thus confirming our diagnosis of cavernous hemangioma.

3. DISCUSSION

Synovial hemangiomas arise from any synovium lined surfaces, including along the course of

tendons or from within the joints. If arising in the vicinity of tendons it may closely resemble a tenosynovial giant cell tumour. The intra-articular variety comprises of a discrete mass lined by synovium [5]. Histologically, the tumours are cavernous hemangiomas in which the vessels are separated by edematous, myxoid or focally hyalinized matrix, occasionally containing inflammatory cells and siderophages. The synovium overlying the tumour is sometimes thrown into villous projections, and its cells contain moderate to marked amounts of hemosiderin pigment. Pathological evaluation relies on recognising that the underlying vessels are too numerous and large for the area in question [5]. Synovial hemangiomas are further classified based on the nature of the predominant vessel within the lesion into the following types: capillary (25%), cavernous (50%), mixed (20%) and pure venous type (5%) Many of those in the synovial membrane are of mixed capillary and cavernous types [6]. Another classification system, used primarily by interventional radiologists and orthopaedic surgeons, classifies them by their anatomical relationship to the joint: juxta-articular, intra-articular or intermediate type. Juxta-articular hemangiomas are situated on the outside of the joint capsule, with no intra-articular involvement. However, intra-articular lesions are actually situated within the joint capsule itself, and the last type, intermediate, show features of both former types. Most reported cases have been of juxta-articular and intermediate types [2].

Intra-articular cavernous hemangioma is an extremely rare form of synovial hemangioma with fewer than 200 cases reported in world literature [7,8]. This tumour was first described by Bouchut in the year 1856 [9]. Rather than being a true neoplasm, it is thought to be a form of congenital vascular malformation [9]. The lesions can be diffuse or localised. The knee joint is the most common location, other reported locations include the wrist, elbow and ankle [10].

Most of the affected patients are in the age group of adolescence or early adults. Sometimes the symptoms may date to childhood. The most common presenting complaint is a pain in the joint. Other complaints can be on-off swelling (recurrent effusions), restricted range of movement, and sometimes a palpable mass [11].

A study done by Devaney et al. estimated the common symptoms to be pain and swelling (31%), pain alone (31%), painless mass (31%) and recurrent intra-articular haemorrhage (5%) [12].

Patients may also develop recurrent episodes of locked knee. Any incidental trauma can cause bleeding from the hemangioma with resultant hemarthrosis. Thus, recurrent knee effusions with or without associated pain in the joint of an adolescent/ early adult should alert a clinician to the possibility of intra-articular synovial hemangioma. History of recurrent non-traumatic haemarthrosis in a patient with a normal coagulation profile should raise a suspicion of synovial haemangioma [13]. It is important to note that the recurrent intra-articular haemorrhages can lead to chondral damage and arthropathy similar to that seen in haemophilic patients [14].

A synovial hemangioma may be mistaken for pigmented villonodular synovitis (PVNS) [6]. The best method to differentiate remains histopathological examination. Other differential diagnosis includes synovial sarcoma, other arthropathies (rheumatoid arthritis, juvenile chronic arthritis, hemophilic arthropathy, synovial osteochondromatosis, or lipoma arborescens) usually being distinguished clinically or after MRI interpretation [15].

Plain radiographs may show joint effusion, soft tissue masses, phleboliths and bone erosions; or may appear innocuous. Ultrasonographic examinations may differentiate solid lesions from cystic ones and reveal the vascular nature of the lesions. Computed tomography may rarely demonstrate the lesion; however, the findings are non-specific and there is the added risk of radiation [16]. The gold standard investigation is MRI both for detecting the tumour and determining the size and extent. Additionally, it can detect any chondral degeneration if present [6,10,13,14,17]. The lesion appears as an intra-articular lobulated mass which is often not sharply defined with an intermediate signal intensity on T1-weighted images, isointense or slightly hyperintense than surrounding muscles. The lesion appears hyperintense on T2-weighted and fat-suppressed images. Thin, serpentine and low-intensity septa are also detected on T2-weighted and fat-suppressed images. After contrast administration, the lesion shows marked enhancement. The MRI is also used to differentiate the lesion from other pathologies [16]. Angiography not only shows the extent of the lesion and its vascular origin, but also provides the opportunity for therapeutic

embolisation of a major feeder vessel in the same sitting. Angiography may fail to show the hemangioma if the vessels are thrombosed [13].

Treatment recommendations for synovial hemangioma include embolisation, open surgical resection, arthroscopic excision and arthroscopic ablation using a laser [14]. Pre-operative embolization is useful for diffuse lesions with large feeding vessels [14,4]. Arthroscopic excision is preferable for localised pedunculated tumours [18]. It is important to note that mechanical instrumentation during arthroscopy can cause significant bleeding, which may be difficult to control because the electro-cautery device may be ineffective in coagulating the friable hemangiomatous tissue [14]. Open surgical resection with partial or total synovectomy is generally suitable for diffuse lesions. It also helps to reduce rates of recurrence by enabling total resection of the lesion. The role of arthroscopic ablation of an intra-articular hemangioma using holmium:YAG laser was advocated by Shapiro and Fanton [14,19].

Although synovial hemangiomas are notoriously difficult to diagnose in routine out-patient

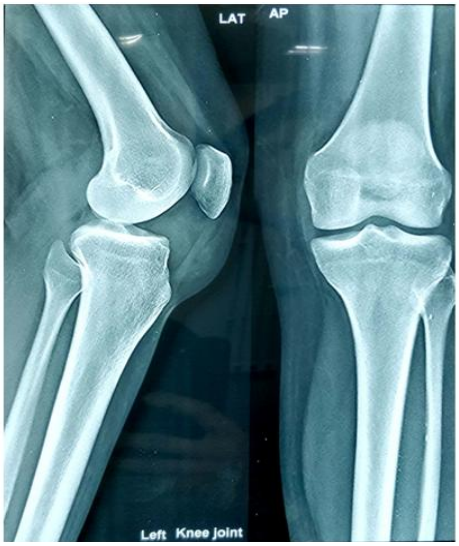

Fig. 1. X-ray Knee joint

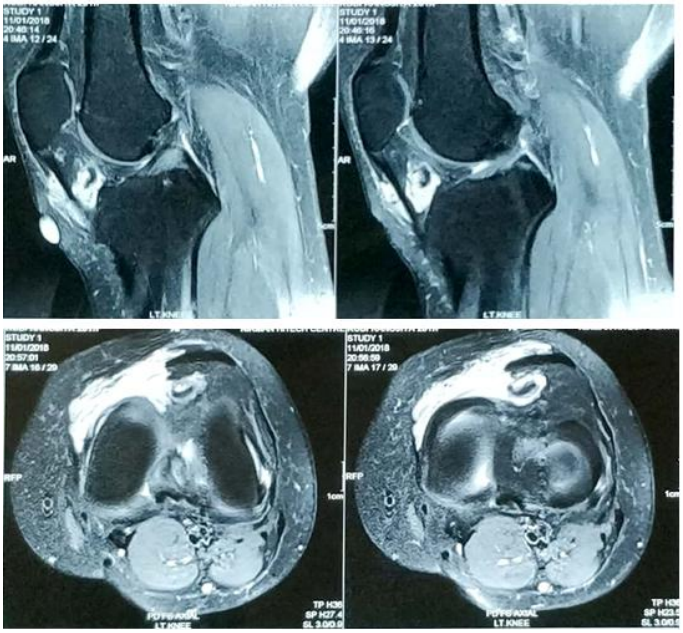

Fig. 2. Sagittal and Axial MRI Images showing the hemangioma

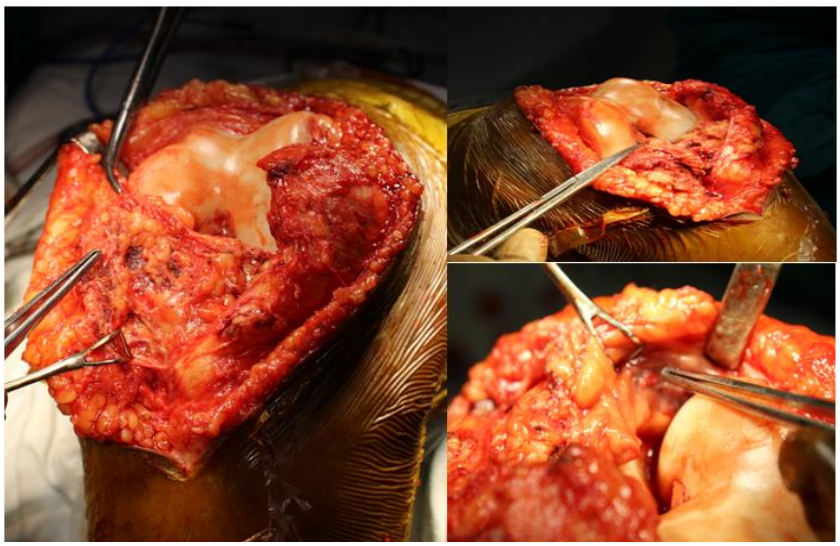

Fig. 3. Intra-operative images demonstrating the lesion

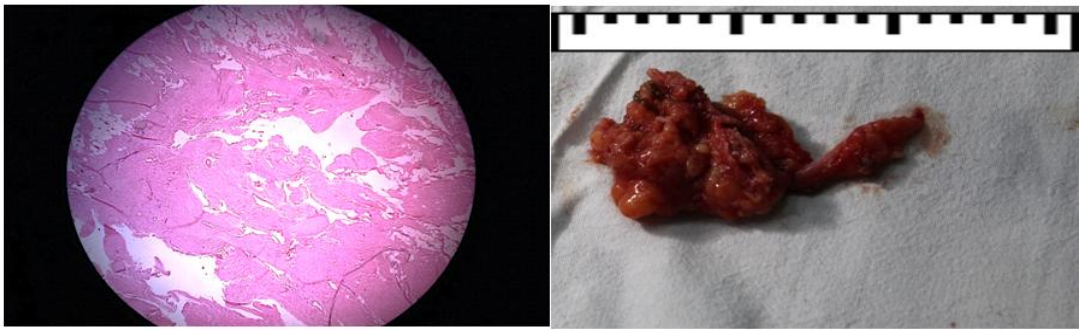

Fig. 4. Image of sample sent for Histo-pathological evaluation with the Slide picture showing blood-filled lakes highly characteristic of Cavernous Hemangioma

department setup, there are cases on record, where patients with a long-standing intra-articular hemangioma have developed a massive swelling infiltrating into the adjacent soft tissues [6,4]. Additionally, there is a risk of arthropathy due to recurrent episodes of intra-articular haemorrhage and mechanical irritation by the tumour [4]. There is also one case in published literature where a long-term lack of treatment led to dysplasia of the femoral condyle [20]. Furthermore, the data regarding the recurrence rate following surgery is also somewhat ambiguous with certain studies asserting that local recurrence should be limited with wide excision [9]; however, there are also other studies which emphasise that recurrence may be actually related to the histopathology of the tumour [3].

Moreover, the majority of literature available on this rare intra-articular pathology is either in the form of individual case reports or small case series'. Hence, additional projects, especially ones with a longer duration of follow-up would be useful to elucidate its management protocol and risk of recurrence.

4. CONCLUSION

Intra-articular synovial hemangioma is a rare benign tumour. Most of the patients are adolescents or young adults, who commonly present with joint pain, with episodes of recurrent effusions. MRI is useful for early diagnosis. A Histopathological evaluation may be needed to rule out Pigmented Villonodular synovitis (PVNS). Starting treatment as early as possible is important to prevent secondary joint degeneration.

CONSENT

As per the International standard, written informed consent was obtained from the patient for the surgery and publication of this case report and the accompanying images. The copy has been preserved with the authors.

ETHICAL APPROVAL

The case was discussed in detail with other senior faculty members, which included a senior Arthroscopy surgeon; before, deciding to go ahead with an open excision of the tumour mass instead of a minimally invasive procedure. The reasons for the same have been mentioned in the text. As per International standards, all requisite ethical permissions were obtained, including those from the Departmental Review Board and the Institute Ethics Committee.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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