

Synovial Haemangioma of the Knee: A Rare Paediatric Case Presenting with Knee Pain

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ABSTRACT

Synovial haemangioma is a rare benign vascular neoplasm and is uncommon in children. It occurs usually in the knee and elbow joints and presents as unilateral joint swelling and haemorrhage into the joint causing pain and restriction of movement. Bone is not involved by this tumour. Most patients present with non specific signs and symptoms due to which the diagnosis is delayed and may result in osteoarthritis and cartilage damage. Magnetic Resonance Imaging (MRI) reveals an intraarticular mass. However, histopathology is necessary for a definite diagnosis. Surgical excision is the treatment of choice. Here, the authors present a case report of a 12-year-old male child with left knee pain and movement restriction. MRI showed an ill-defined mass in the suprapatellar region of left knee. The mass was excised; histopathological study revealed proliferation of thin-walled capillaries in lobules below the synovial lining consistent with synovial haemangioma. Close radiological differentials include synovial hamartoma, cystic synovial hyperplasia, tenosynovial Giant Cell Tumour (GCT) and haemorrhagic joint effusions. The histopathological differentials include non specific synovitis, pigmented villonodular synovitis and organising haemorrhage. At one year follow-up, the patient is well and has had no recurrence. To conclude, synovial haemangiomas may be asymptomatic or present with chronic knee pain and movement restriction. Though rare, these benign tumours are usually picked up by MRI and diagnosed on histopathology. Early diagnosis is essential for proper management and to prevent permanent disability.

Keywords: Histopathology, Joint imaging, Magnetic resonance imaging

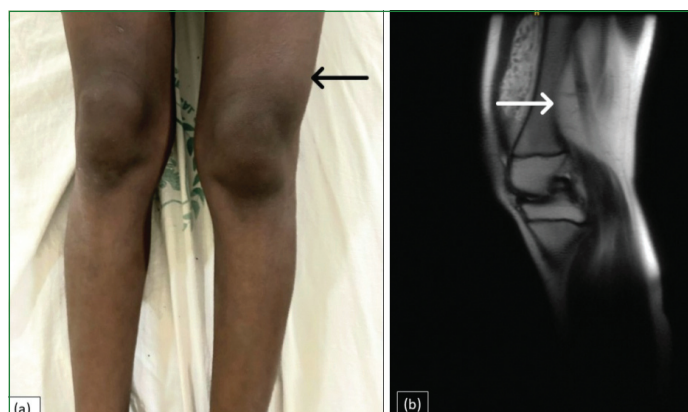
CASE REPORT

A 12-year-old male child presented with complaints of on and off pain and swelling of left knee joint for last two years with restriction of terminal range of motion [Table/Fig-1a]. The pain worsened over time and did not improve with medications. He also had three episodes of fever over the last seven days. He had a history of trauma to left knee two-year-ago. There was no significant family, past medical or surgical history. The child had a normal development and achieved all milestones appropriately. He had previously received local ayurvedic treatment along with over-the-counter painkillers and oral antibiotics with temporary and minimal relief of symptoms. Due to persistent painful swelling of left knee, he walked with a limp but was unable to participate in sports and strenuous physical activities. General physical examination was unremarkable. The Body Mass Index (BMI) was 21 kg/m². Physical examination of the left knee revealed a soft tender swelling measuring 6×5 cm in the left suprapatellar region with local rise of temperature of the skin over the swelling. The range of motion of the left knee was 0-100 degrees with painful terminal flexion. There was no knee effusion or instability of knee joint or signs of meniscus or ligament injury.

Complete blood count details were as follows: Laboratory investigations revealed a Haemoglobin (Hb) level of 13.3 g/dL. The Total Leukocyte Count (TLC) was 12,400/μL. The Differential Leukocyte Count (DLC) showed Neutrophils (N) at 57.4%, Lymphocytes (L) at 29.7%, Monocytes (M) at 3.2%, Eosinophils (E) at 9%, and Basophils (B) at 0.7%. The platelet count was 381×10³/μL. The Absolute Eosinophil Count (AEC) was 1.12×10³/μL, which is elevated compared to the reference range of 0.05-0.45×10³/μL. The Erythrocyte Sedimentation Rate (ESR) was 83 mm in the first hour (reference range: 0-20 mm in the first hour). The quantitative C-reactive Protein (CRP) level was 6.5 mg/dL, which is above the reference range of 0.5-4.9 mg/dL. However, coagulation profile was within normal range. {Prothrombin (PT) 12.4 seconds, (10 to 15 seconds) International Normalised Ratio (INR)-1.12 (0.9 to 1.2)}.

Rheumatoid factor and Serum Antinuclear Antibody (ANA) were negative.

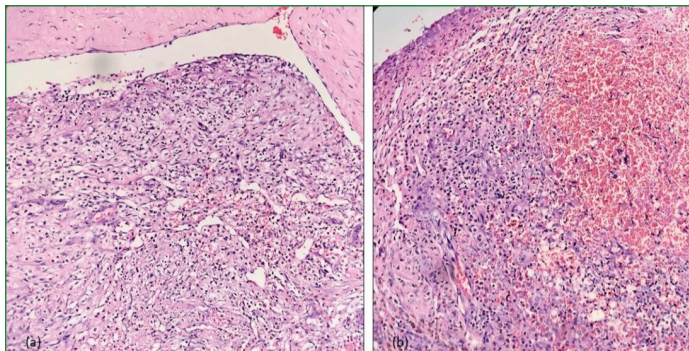
Further evaluation by imaging studies was done. Left knee X-ray radiograph showed no specific abnormality. However, MRI revealed an ill-defined lobulated T1 hypointense, T2 hyperintense lesion showing central hypo-attenuating areas and measuring 5.5×7.0 cm involving the suprapatellar region of left knee [Table/Fig-1b].



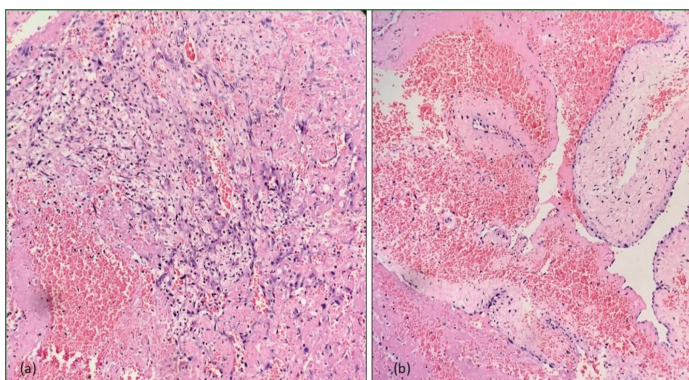
[Table/Fig-1]: a) Swelling in the left suprapatellar region; b) MRI of left knee with an ill-defined lobulated T1 hypointense lesion involving the suprapatellar region.

A provisional clinical differential diagnosis of pigmented villonodular synovitis, haemangioma and bursitis were considered. Complete surgical excision of mass was done and sent for histopathological study. Gross specimen revealed multiple fragments of greyish brown fibrofatty tissue altogether measuring 6.5×5×2 cm which was entirely submitted for sectioning. On microscopic examination, there was presence of numerous small sized capillaries in lobules beneath a normal appearing synovial lining [Table/Fig-2a,b]. Proliferation of numerous variable sized capillaries was seen with wide areas of haemorrhage and clusters of haemosiderin-laden macrophages. Rest of the tissue showed mostly fibro-collagenous

tissue with synovial lining and mild lymphoplasmacytic inflammatory cell infiltration [Table/Fig-3a,b]. A final histopathological diagnosis of synovial haemangioma was made without any evidence of atypia. The histopathological differential diagnoses include diffuse type of tenosynovial GCT (earlier known as pigmented villonodular synovitis), non specific synovitis, and organising haemorrhage.



[Table/Fig-2]: a) Section shows synovial lining epithelium beneath which there is lobular proliferation of small thin-walled capillaries (H&E, 200X); b) Section shows part of synovial epithelial lining with areas of haemorrhage and proliferation of thin walled capillaries along with inflammatory cells (H&E, 200X).



[Table/Fig-3]: a) Section showing wide areas of haemorrhage along with proliferation of lobules of small capillaries (H&E, 400X); b) Section showing synovial lining epithelium and areas of haemorrhage in the fibrocollagenous tissue (H&E, 200X).

Postoperatively, the patient recovered uneventfully and attained complete range of motion in one month with no residual pain or limitation in movement in left knee.

DISCUSSION

Knee pain is unusual in children and warrants immediate attention to exclude underlying disease. Amongst various causes, synovial haemangioma is one rare cause with an incidence of <1% [1]. It was first enumerated by Bouchut in 1856 [2].

Synovial haemangioma is a benign neoplasm and can occur at any site of the body. However, due to its rarity and absence of specific clinical signs and symptoms, it is clinically missed. Proper imaging modalities and histopathological evaluation are necessary to confirm the diagnosis [3]. It requires immediate treatment to prevent complications like haemarthrosis and damage to intra-articular cartilage involving major joints of the body, especially the knee joint [4].

The discussed case was 12 years at presentation, unlike the common age of presentation in adolescents and young adults [5]. Knee joint is a rare site of occurrence of synovial haemangioma; also it usually involves the suprapatellar region rather than the infrapatellar region [6], as in the present case. In the present case, the patient had chronic knee pain for the previous two years; hence there was delay in diagnosis due to non specific signs and symptoms of the disease.

In the present case, elevated inflammatory markers (leukocytosis, ESR, and CRP) along with chronic unilateral knee pain and swelling raised the possibility of several inflammatory and infective differential

diagnoses such as juvenile idiopathic arthritis, septic arthritis, tuberculous synovitis, reactive synovitis, and pigmented villonodular synovitis. Juvenile idiopathic arthritis was considered due to chronic knee involvement; however, absence of morning stiffness, diffuse synovial thickening, and negative rheumatoid factor excluded inflammatory arthritis. Septic arthritis and tuberculous synovitis were considered because of intermittent fever and raised inflammatory markers, but the prolonged clinical course, absence of joint destruction or significant effusion, and lack of granulomatous or suppurative inflammation on histopathology were against infective aetiology. Pigmented villonodular synovitis was an important radiological differential; however, histopathological examination did not reveal multinucleated giant cells or diffuse haemosiderin-rich histiocytic proliferation. Elevation of absolute eosinophil count, may be due to some allergic reaction or due to intake of over-the-counter drugs.

Evaluation of chronic knee pain with varying functional disability is difficult as the clinical features are non specific and overlapping. Entities such as Patellofemoral Pain Syndrome (PFPS), chondromalacia patellae, Osgood-Schlatter disease, Plica syndrome, patellar tendonitis and knee bursitis are considered in such cases [7]. Synovial tumours are unusual around the knee and differentials include Pigmented Villonodular Synovitis (PVNS), GCT, lipoma arborescens, synovial chondromatosis, synovial haemangioma, etc., [7].

Synovial haemangiomas are rare vascular tumours accounting for less than 3% of all haemangiomas [2]. X-ray radiography is non diagnostic and can show mild erosions in case of complications. MRI, the primary diagnostic modality, shows a well-defined intra-articular lesion which is hypointense in T1-weighted image and hyperintense on T2-weighted images [7].

The MRI gives an early clue to diagnosis and further management by excisional biopsy is the treatment of choice in most of the cases. Histopathology is the confirmatory diagnostic modality and reveals proliferation of thin-walled capillaries in lobules below the synovial lining along with extensive areas of haemorrhage [5]. Synovial haemangiomas must be differentiated from close radiological mimickers like synovial hamartoma, cystic synovial hyperplasia, tenosynovial GCT and haemorrhagic joint effusions [3]. In cystic synovial hyperplasia, there is proliferation of synovial lining cells and abundant capillaries, fibrosis and cystically dilated spaces (representing areas of degeneration) in the underlying stroma with mild to moderate inflammatory cells infiltration. The gold standard for diagnosis is histopathology. The histopathological differential diagnoses include diffuse type of tenosynovial GCT (earlier known as pigmented villonodular synovitis), non specific synovitis, and organising haemorrhage [8]. Localised type of tenosynovial GCT is a common tumour affecting digits and fingers while diffuse type of tenosynovial GCT is intra-articular and affects larger joints like knee, hip and ankle and are locally aggressive and recur. Histopathologically, these tumours show heterogeneous population comprising of haemosiderin-laden large epithelioid cells with vesicular nucleus, macrophages with pale cytoplasm and reniform nucleus and multinucleate giant cells. Organising haemorrhage shows large areas of haemorrhage, fibrin, few blood vessels and fibroblasts. Non specific synovitis shows synovial hyperplasia, few congested blood vessels and mild to moderate chronic inflammatory cell infiltrate. None of these features were present in the present case.

Synovial haemangioma usually has the following histopathological subtypes- venous, arteriovenous, capillary, cavernous or mixed subtypes. These are classified as such based on the dilatation of vascular channels and presence or absence of vascular smooth muscles. Capillary haemangiomas usually contain small lobules of thin-walled single layer of endothelium lined vascular channels, while cavernous haemangioma consists of dilated vascular channels of varying size which are lined by endothelial cells and vascular smooth

muscle. In the present case, histopathological examination revealed thinned out synovial lining epithelium with underlying stroma showing lobules of capillaries lined by flattened endothelial cells and moderate mixed inflammatory cell infiltration. A final diagnosis of capillary haemangioma was made.

Similar to the cases reported by Shyam K et al., and Hernández-Hermoso JA et al., the present patient presented in the paediatric age group with chronic unilateral knee pain, suprapatellar swelling, delayed diagnosis, and characteristic MRI findings showing T1 hypointense and T2 hyperintense intra-articular lesions [4,5]. Unlike the case described by Suphakitchanusan W et al., where recurrent haemarthrosis and infrapatellar involvement were the major presenting features, the present patient had suprapatellar involvement without recurrent haemarthrosis [1]. Similar to the observations made by Sonobe T et al., MRI played a significant role in narrowing the differential diagnosis preoperatively and demonstrated characteristic signal intensity changes suggestive of a vascular synovial lesion [4]. Histopathological findings in the present study were also concordant with the findings reported by Singh D et al., and Saxena P et al., demonstrating proliferation of thin-walled capillary vascular channels beneath synovial lining epithelium with areas of haemorrhage and haemosiderin deposition [7,8].

In the present case, the young boy regained complete range of movement in the left knee joint after complete surgical excision of the mass was done. The patient is well, has had no further complications and at a year follow-up, there has been no recurrence. Early diagnosis and management prevents various complications like damage to articular cartilage, massive haemarthrosis, synovial degeneration which could otherwise lead to permanent disabilities.

CONCLUSION(S)

Synovial haemangioma is a rare vascular neoplasm and is difficult to diagnose due to its non specific clinical presentation. Imaging modalities and histopathological analysis are helpful in identifying this tumour and differentiating it from other non neoplastic conditions. Complete surgical excision is the treatment of choice in most of the cases and should be done immediately to prevent further complications.

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