



A Rare Case Report of Fungal Synovitis Presenting as Pigmented Villonodular Synovitis

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Synovitis is a disorder that develops when the synovial tissue or joint lining becomes inflamed. The term pigmented villonodular synovitis (PVNS) refers to a form of tenosynovial giant cell neoplasm affecting the tendon and joint soft-tissue lining. We describe the rare case of a 37-year-old male presenting with complaints of swelling in the left knee following trauma. Patients have a limited range of motion. There is a previous history of an accident 1 year back for which he was operated. Recent magnetic resonance imaging (MRI) revealed anterior cruciate ligament and posterior cruciate ligament avulsion fracture. The clinical diagnosis and radiologic findings were in favor of PVNS. Histopathological examination revealed the presence of fungus. It was further confirmed by fungal culture, for which the patient was given antifungals and was further followed up in the orthopedics outpatient clinic. Thus, we diagnosed fungal synovitis in a patient who came with clinical presentation and MRI findings suggestive of PVNS.

Key words: Fungal synovitis, pigmented villonodular synovitis, siderotic synovitis

INTRODUCTION

Synovitis is a disorder that develops when the synovial tissue or joint lining becomes inflamed. The term “pigmented villonodular synovitis (PVNS)” refers to a form of tenosynovial giant cell neoplasm affecting the tendon and joint soft-tissue lining in a diffuse manner. It encompasses villous and/or nodular proliferation of synovium along with accumulation of hemosiderin and fat. Joint discomfort and edema can result from this disorder, which is characteristic of all inflammatory joint disorders.¹

Fungal synovitis typically begins with a protracted, indolent phase lasting several months, which delays diagnosis and necessitates ineffective treatments such as intraarticular and systemic steroid injections.² Bones, joints, and periarticular tissues all exhibit a persistent granulomatous process as skeletal symptoms of fungal synovitis. Knees are often afflicted in 50%–70% of cases, although other weight-bearing joints may also be impacted. There may be 4 weeks to several years between the beginning of symptoms and the diagnosis.³ Only mycologic culture can confirm the disease’s diagnosis;

however, it is strongly suspected when the disease’s distinctive fungal hyphae and spores are observed under a microscope.¹ In addition to the production of noncaseating granulomas containing endospores, histopathological hallmarks include PVNS, or a typical pannus formation. Amphotericin B is the most effective treatment for severe fungal infections with skeletal involvement. In monoarticular disease, local amphotericin B has been found to be effective. The second drug of choice is ketoconazole. Open drainage synovectomy and arthrodesis are performed, all of which, if fail, amputation is considered.³

CASE REPORT

The patient, a 37-year-old male worker at a firm who appeared to be in good condition, complained 1 month ago of discomfort and swelling in the left knee following a small contusion to the afflicted knee. Since then, he has had progressively worsening, sharp shooting pain. Pain

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was aggravated on movement and relieved on rest and pain medications. He also had localized, gradually progressive swelling since then. He denied having fever, chills, rigor, nausea, vomiting, and other signs of systemic illness. The patient stated that he had met with an accident 1 year back for which he had undergone a posterior cruciate ligament (PCL) reconstruction as an arthroscopic surgery. Physical examination showed a normal neurological, cardiovascular, and pulmonary examination. He had diffuse swelling over the left knee with fixed flexion deformity of 5° present. He had no open wound, discharge, sinus, bone deformity, or discolorations. The left knee has a painful and limited range of motion. The laboratory investigations revealed anemia with hemoglobin of 10 g/dL, along with mild lymphocytosis and monocytosis. The left knee's plain radiographs showed soft-tissue swelling but no evident joint problems. Magnetic resonance imaging (MRI) of the left knee revealed an old, displaced avulsion fracture of the tibial eminence at the attachment of the PCL. Comminuted, undisplaced avulsion fracture of the tibial eminence was noted on the anterior aspect of the anterior cruciate ligament (ACL). Mild underlying marrow edema was seen. Grade 2 signal was noted in the posterior horn of the medial meniscus. Diffuse synovial thickening was noted around the lesion with severe knee joint effusion. Thus, the MRI was suggestive of ACL and PCL avulsion fracture [Figure 1a-d]. The clinical diagnosis and radiologic findings were in favor of PVNS. The histopathology sample received was in multiple bits, tan-pink in color, and firm to hard in consistency. Microscopic examination revealed siderotic synovitis with collections of foamy macrophages, siderotic pigmentation, and chronic inflammatory infiltration with many congested and dilated blood vessels. Here, it was advised to rule out traumatic etiology or chronic hemarthrosis before considering the possibility of PVNS. No evidence of acute infection, Koch's, or malignancy was found [Figure 2].

The paraffin blocks were sent for review, which revealed diffuse proliferation of synovial lining along with plenty of mononuclear cells containing hemosiderin pigment. The section also showed a focal area of septate-colored fungal hyphae and spores with surrounding inflammatory reaction, confirming fungal synovitis. Hence, fungal culture was advised for confirmation [Figure 3a and b]. The patient was started on intravenous amphotericin B and was further followed up for treatment regularly in the orthopedic outpatient clinic [Figure 3a and b].

DISCUSSION

PVNS most commonly affects the knee, typically monoarticular, followed by the hip, ankle, and then the elbow

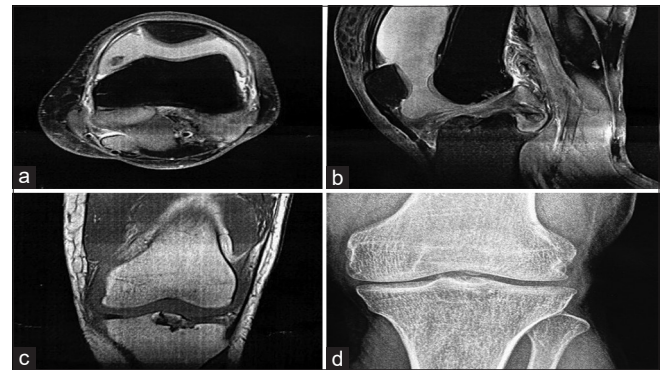


Figure 1: (a-d) Diffuse synovial thickening around the knee joint with severe effusion and also anterior cruciate ligament and posterior cruciate ligament avulsion fracture

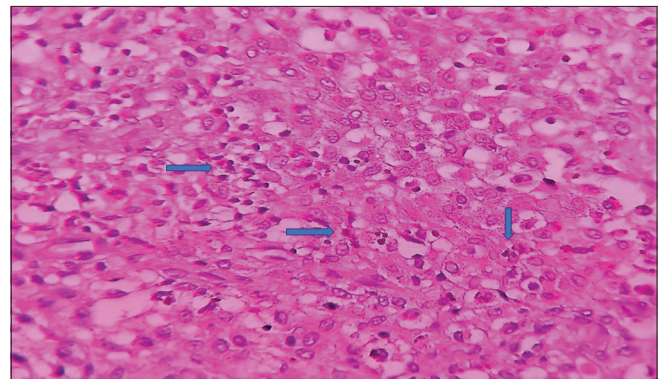


Figure 2: Photomicrograph shows features of siderotic synovitis with collections of foamy macrophages, siderotic pigmentation, and chronic inflammation (Blue arrow) (H and E, ×400)

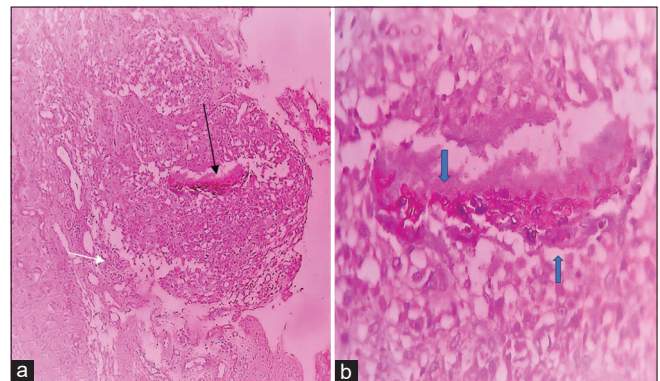


Figure 3: Photomicrograph shows diffuse proliferation of synovial lining along with plenty of mononuclear cells (white arrow) containing hemosiderin pigment a focal area of septate-colored fungal hyphae (black arrow) (a) (H and E, ×200). (b) A focal area of septate-colored fungal hyphae and spores with surrounding inflammatory reaction (blue arrows) (H and E, ×400)

as well. It often exhibits the same symptoms, including discomfort, edema, instability, and restricted range of motion. Although it is a benign lesion, the proposed management is open or arthroscopic synovectomy.⁴

MRI is supposed to be the most diagnostic for PVNS. In relation to benign synovial disorder, studies have mentioned extensive differential diagnoses that must be kept in mind, include osteoarthritis, tuberculous arthritis, synovial hemangioma, hemophilic arthropathy, and synovial chondromatosis.⁵ However, the MRI findings of a prominent low-intensity signal seen with T2-weighting and blooming artifact from the hemosiderin are almost diagnostic for this lesion.⁶ This case report shows that chromogens should be included in the list of diseases to better describe infectious fungal synovitis.⁵

More recently, the study documented the case of a male patient who had knee discomfort, edema, and mechanical symptoms. It was eventually discovered that the patient had primary synovitis caused by fungi in the knee. It highlighted MRI findings that showed material that was widely scattered and had signal intensity characteristics compatible with hypertrophied synovium. The indicated lesion exhibited a high signal on T2-weighted fast spin echo imaging, compared to a medium signal on T1-weighted spin echo imaging.⁵

Fungal prosthetic joint infections, even though they are rare, represent a serious, life-threatening, devastating complication.⁶ Such infections provide a special surgical and medical challenge in updated knee arthroplasties. The research described a rare instance of *Candida parapsilosis* following a revision knee arthroplasty.⁷

In the present case, the clinical examination and X-ray findings concluded it to be a case of PVNS. Further, MRI also showed findings consistent with it, thus focusing the initial plan of treatment to be surgical excision (synovectomy).

CONCLUSION

In our case, we diagnosed a patient who came with clinical presentation and MRI findings suggestive of PVNS to be suffering from fungal synovitis. The patient was treated with antifungal medications and was under close follow-up in the orthopedics department. It was only after the pathological analysis that fungal infection was thought of in the primary differentials.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent form. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that his name

and initials will not be published, and due efforts will be made to conceal his identity, but anonymity cannot be guaranteed.

Acknowledgments

Dr. Mangesh Londhe has diagnosed fungal synovitis in the case. He has played a major role in identifying the fungus seen in the clinically diagnosed case of Pigmented villonodular synovitis. He has a major contribution in the manuscript.

Data availability statement

The data that support the findings of this study are available from the corresponding author, Mallika Agarwal (MA), upon reasonable request.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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