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Accidental diagnosed disease of pigmented villonodular synovitis of the knee joint with multi ligament knee post infection: A case report

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Abstract
Pigmented Villonodular Synovitis (PVNS) is a rare, benign, but potentially locally aggressive and recurrent condition characterized by synovial proliferation and hemosiderin deposition inside the joints, tendon sheaths, and bursae. It usually affects the large joints such as hip, knee, and ankle. We report a case of PVNS of the knee joint in a 45Y old male was treated initially by Arthroscopic Posterior Cruciate Ligament reconstruction, the patient developed knee infection repeated procedures like arthroscopic wash of the knee and at later stage subtotal synovectomy was done and confirmed the disease histologically. At the 7-month follow-up, the patient was pain free and had no signs of disease recurrence.

Keywords: MRI, PVNS, villonodular, synovitis, synovectomy, disease recurrence, case report

Introduction
Pigmented villonodular synovitis (PVNS), coined by Jaffe, *et al*. [1] in 1941 is a rare, benign, but potentially locally aggressive and recurrent condition. It is characterized by synovial proliferation and hemosiderin deposition inside the joints, tendon sheaths, and bursae. It usually affects the large joints, i.e. hip, knee, and ankle, but few cases of PVNS involving small joints have been reported [1]. The most commonly involved joint has been the knee, followed by the hip and the ankle [2]. There are two types of PVNS: localized and diffuse. The diffuse type is reportedly three times more common than the localized type [3].
The etiology of PVNS is not certain but some researchers have debated whether it is inflammatory or neoplastic in origin while others have suggested trauma-induced hemorrhage as an etiology.

Case presentation
A 45-year-old male presented to us with a one year-history of pain, swelling and disability in the left knee joint with twisting injury to knee. The swelling gradually increased over a period of time and was associated with difficulty in walking and standing. The swelling was diffuse and nodular in consistency, measuring 10 cm x 10 cm in size (Figure1). The overlying skin was normal with no signs of inflammation. There was instability of the knee joint on physical examination. Lachmann test-positive, anterior and posterior drawer test is positive.
Range of motion is full.

Clinical image showing the swelling around the knee joint
Radiographs of the left knee were obtained which showed no bony abnormality. A magnetic resonance imaging (MRI) scan revealed a large joint effusion seen predominantly in the suprapatellar recess as well as in the lateral and medial femoral recesses appearing hyperintense on T_2 weighted images. Diffuse synovial thickening was seen which appeared hypointense on T_1 as well as hypo on T_2-weighted and susceptibility images,

- Both ACL and PCL ligaments are torn.
- Both Menisci-Normal
- Lateral Colleteral Ligament-Grade 1 Strain.
- X-ray-uneventful.

Fig 1: Clinical image showing swelling around the left knee joint

MRI of the knee joint-coronal and axial views

After obtaining written informed consent from the patient, she was submitted to surgery

- Surgery-posterior cruciate ligament reconstruction, patient developed infection for with knee aspiration and culture was done. Report showed MRSA infection, patient is put on antibiotics.
- Blood samples showed increased ESR and CRP levels.
- After unsubsided infection patient was subjected to arthroscopioc washout, the infection has not subsided yet.
- Sub-total synovectomy was done using a medial parapatellar approach to the knee joint. The synovium was excised. Intraoperatively, a brownish nodular synovium with grape like tissue was found for which biopsy of the tissue is taken and sent for investigation. Biopsy confirmed of PVNS (Figure 3 & 4).

Fig 2: MRI of the left knee joint-coronal and axial views

Fig 3: Intraoperative image showing excised brownish, nodular synovial tissue

Fig 4: Close-up view of the excised synovial mass

The excised synovium was sent for histopathological examination which showed synovial tissue with hyperplastic synovial lining forming papillary proliferation with oedematous stroma and presence of granulation tissue. The blood vessels were dilated and congested, surrounded by dense inflammatory infiltrate of plasma cells, lymphocytes, and histiocytes. Scattered hemosiderin granules along with hemosiderin-laden macrophages were also seen (Figure 5) all these features confirmed the diagnosis of PVNS.

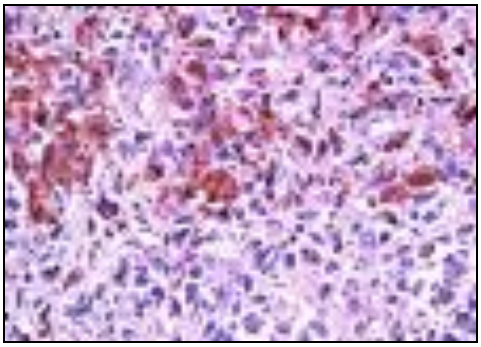

Fig 5: Histopathological slide showing features of Pigmented Villonodular Synovitis (PVNS)

Histopathological slide showing hyperplastic synovial lining with scattered hemosiderin deposits

Postoperatively after subtotal synovectomy, the patient was started on passive knee flexion and extension exercises for 15 days and made to walk after that. Follow-up was taken at regular intervals, and at the 7-month follow-up there were no signs of recurrence both clinically and radiologically, and the patient had full knee range of movement.

Discussion

Patient was treated initially as ligament injury which lead to infection and later confirmed as pigmented villonodular synovitis. Was the disease aggravated after surgery which lead to infection due to blood accumulation and synovial inflammation, or was it a primary infection?

Intra-articular PVNS is an uncommon disease. The prevalence has been estimated to be 1.8 cases per million population [4]. It commonly affects individuals in the fourth and fifth decades [5]. Most cases that occur have been monoarticular but rarely, polyarticular PVNS has been reported [6].

The genesis of PVNS is disputed, most authors regard PVNS as a reactive process, chronic inflammatory process or having a genetic component [9, 10, 11].

The mechanism of bone erosion in PVNS is still unclear. Some believe that pressure within the involved joints increases because of the synovial overgrowth while others believe that the synovium releases a substance that causes bone erosion which in turn results in joint destruction [7].

The reported rate of recurrence is varied. PVNS has been reported to have a high recurrence rate, but it rarely becomes malignant [5]. Surgical excision is the preferred management for both localized and diffuse PVNS with success being dependent on complete resection with clear margins. The best treatment for diffuse PVNS is controversial. Open surgical excision has been the primary method for treating diffuse PVNS [8]. Another method is arthroscopic synovectomy which has the advantage of smaller incisions and reduced morbidity but has reported recurrence rates as high as 46%, so some authors recommend open synovectomy [5]. It is also theoretically possible that arthroscopy results in secondary seeding because of limited surgical view and joint irrigation

system which probably leads to recurrence [5].
In patients with diffuse PVNS, some authors have recommended staged anterior and posterior synovectomies. The recurrence rate associated with such treatment ranges from 14% to 56% [5].
In this patient with anterior medial approach subtotal synovectomy achieved good results.

Conclusions
Infection with PVNS is an unusual condition with a high potential for recurrence and requires excision. PVNS is a rear condition, the clinical presentation of this case the disease was missed initially and later confirmed after open biopsy. Patient with persistent joint swelling associated with pain initially after joint injury for which he was treated with ligament reconstruction which led to increased swelling in the knee joint which subsequently led to infection. We recommend PVNS should be included as a differential diagnosis when evaluating any knee with infection. Patient with PVNS will lead to secondary bleeding in the affected joint with prolong stasis of blood in the joint will lead to chronic inflammation [12, 13].

Conflict of Interest
Not available

Financial Support
Not available

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