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Case Report

A rare cause of knee pain in adult – pigmented villonodular synovitis: A case report

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Abstract

A proliferative disease of the synovial membrane of joints, tendon sheaths, or bursae is known as pigmented villonodular synovitis (PVNS). PVNS is a locally destroying fibro histiocytic proliferation that occurs intra-articular and is thought to be a diffuse analog of tendon sheath giant cell tumours. We report a case of PVNS of a knee joint in an adult male patient who had subtotal synovectomy alone, without adjuvant therapy, for chronic knee pain and swelling. After 15 months, the patient showed no symptoms of recurrence and was pain-free.

Keywords: Knee pain, Pigmented villonodular synovitis, Hemosiderin, Tendon sheath giant cell tumour.

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1. Introduction

Pigmented villonodular synovitis (PVNS), term coined by Jaffe, et al. in 1941 to describe idiopathic villous overgrowth of synovial membrane and typical pigmentation usually found in a single joint.[1] The name PVNS signifies the colour change due to deposits of cholesterol and hemosiderin on gross examination. It is most commonly seen in third decade, slightly more often in males.[2]

Although the cause is unknown but clonal abnormalities and the capacity for autonomous growth suggest PVNS is a true neoplasm. The proliferative nodules tend to bleed, and as hemosiderin accumulates in the joint over time, the nodules take on a distinctive coloration known as “crank case oil” appearance of the affected joint fluid. The knee has been the most often affected joint, with the hip and ankle following closely behind.[3] PVNS comes in two forms: diffuse and localized. According to reports, the diffuse kind is three times more prevalent than the confined type.[4]

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 haemorrhage as an etiology. A breakpoint in the colony stimulating factor-1(CSF-1) gene at a 1p13 breakpoint and translocation at the COL6A3 gene at 2q35 are the causes for development of PVNS.

2. Case Report

A 42-year-old male presented to us with an eight-month history of pain and swelling in the left knee joint. The swelling gradually increased over a period and was associated with difficulty in walking and standing. The swelling was diffuse and nodular in consistency, measuring 15 cm x 10 cm in size (**Figure 1**). Upon examination, there were no sinuses, scars, or signs of infection. On physical examination, the knee joint did not appear unstable. There was no involvement of any other joint. The left knee's radiographs revealed no bone abnormalities (**Figure 2A,B**).

A significant joint effusion that was visible in the suprapatellar recess as well as in the lateral and medial femoral recesses and appeared hyperintense on T2-weighted images was discovered during an MRI scan. The synovium was hypertrophied, and diffuse synovial thickening was observed. It appeared hypointense on T1 and T2-weighted susceptibility imaging. There was a multilobulated lesion in the patella-femoral joint space, anteromedial joint space, and

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anterolateral joint space that was continuous with the synovium. Thick enhancement was observed along the synovium on the postcontrast scan. These characteristics were all in line with the pigmented villonodular synovitis diagnosis (**Figure 3 A,B**).

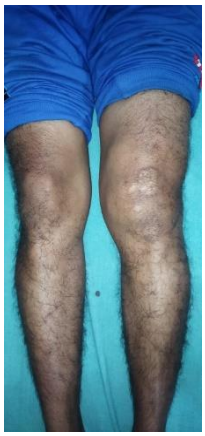

Figure 1: Clinical picture of left knee while presentation

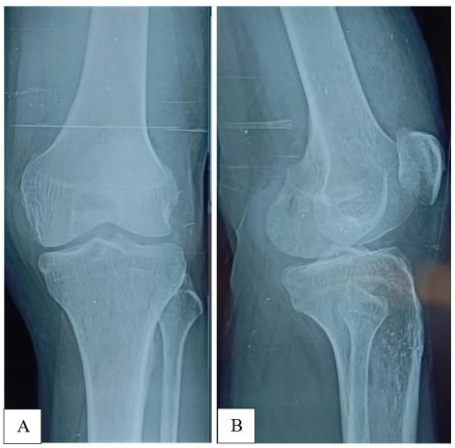

Figure 2: A, B) Preoperative radiograph shows no bony abnormalities)

After taking informed and written consent patient was posted for surgery. A medial parapatellar approach was used to perform a sub-total synovectomy on the left knee joint. A brownish nodular synovium was discovered during surgery, which was also suggestive of PVNS. (**Figure 4 A,B**)

The excised synovium was sent to a histopathological analysis, which revealed the presence of granulation tissue and hyperplastic synovial lining that formed papillary proliferation along with oedematous stroma. The blood vessels had a dense inflammatory infiltrate of lymphocytes, histiocytes, and plasma cells surrounding them, causing them to dilate and become congested. Hemostain-loaded macrophages and dispersed hemosiderin granules were also observed. Each of these characteristics supported the PVNS diagnosis. **(Figure 5)**

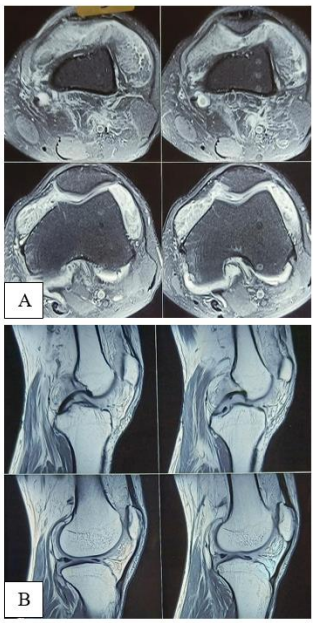

Figure 3: Preoperative MRI (axial and sagittal) of left knee joint – features suggestive of PVNS)

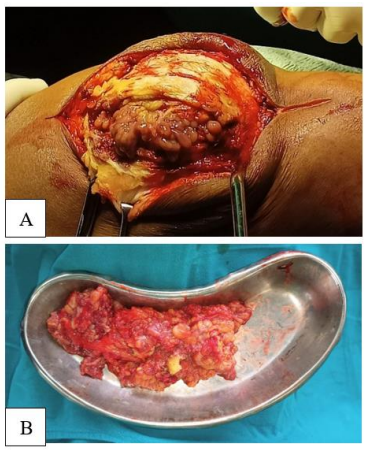

Figure 4: A): Intra operative images of subtotal synovectomy through medial para patellar approach of knee joint; **B):** Resected hyperplastic synovium tissue

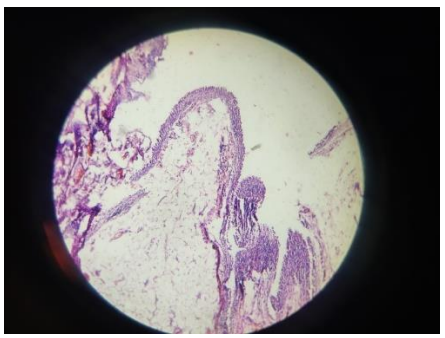

Figure 5: Histopathological image showing papillary hyperplastic synovium with hemosiderin deposition)

Following surgery, the patient was made to walk after 15 days of passive knee flexion and extension exercises. Regular follow-ups were conducted, and at the 15-month, the

patient's knee range of motion was fully restored and there were no clinical or radiological signs of recurrence.

3. Discussion

Pigmented villonodular synovitis is a relatively rare benign tumour that grows slowly and invades the surrounding tissue. It belongs to a group of diseases thought to originate from the bursae, tendons, and synovium of adjacent joints.[5] Large joints, like the knee, are frequently the site of these lesions, but unusual and rare locations of bone and joint involvement have also been documented in the literature. The differential diagnosis consists of septic arthritis, rheumatoid arthritis, haemophilia /hemarthrosis, and synovial chondromatosis. PVNS is distinguished by its ability to infiltrate the subchondral bone and result in erosions and cysts.

An improved functional outcome can be achieved with early surgical excision and prompt care, delayed diagnosis and treatment are associated with poor results. There are, however, few proven prognostic indicators for PVNS recurrence. A big nodule size (>5 cm) and a previous recurrence were found to be associated with recurrence in multicentre research.[6] While arthroscopic excision is the best method for treating localized knee PVNS, open arthrotomy and total synovectomy are associated with a lower risk of recurrence in patients with diffuse PVNS.[7]

The treatment of this disease is challenging. Triamcinolone and pexidartinib, a CSF-1 receptor antagonist, are used as nonsurgical treatments.[8] For symptomatic diffuse type PVNS, synovectomy surgery has shown to be the most successful course of therapy. PVNS is linked to an increased risk of recurrence and rapid degenerative changes in the knee that eventually necessitate arthroplasty.[9] Because external beam radiation can cause uncontrollable complications such joint stiffness, skin necrosis, and the development of secondary sarcoma, it is not a recommended alternative for primary treatment.[10] However, it can be used to manage recurrance. Although mutilating, amputation is the last treatment for extremely serious, incurable diseases.

4. Conclusion

This case report highlights the significance of considering pigmented villonodular synovitis as a crucial differential diagnosis in chronic knee pain, even when there is evidence of degenerative joint disease and a history of trauma. The use of magnetic resonance imaging is essential for precise diagnosis. PVNS has a high recurrence rate and requires excision. It does not need adjuvant radiation if it is easily resectable.

5. Source of Funding

None.

6. Conflict of Interest

None.

7. Consent

Written informed consent was taken from the patient.

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