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Uncommon Clinical Manifestation of Pigmented Villonodular Synovitis: A Case Report and Review of Management

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Abstract

Pigmented villonodular synovitis (PVNS) of the knee is a benign but locally aggressive disease of synovial proliferation that occurs in localized nodular and diffuse villous growth patterns. Although inflammatory and neoplastic causes have been hypothesized, etiology remains unknown. Presenting as unilateral knee pain and swelling, PVNS mimics other knee alignments. Radiographs are sometimes unremarkable, whereas magnetic resonance imaging may show characteristic intra-articular masses with signal dropout on T2-weighted sequences. PVNS is surgically treated with open or arthroscopic total or partial synovectomy. High recurrence rates are associated with all treatments of diffuse PVNS.

Keywords: Pigmented Villonodular Synovitis (PVNS), Tenosynovial Giant Cell Tumor (TGCT), Knee Joint, Synovectomy, Magnetic Resonance Imaging (MRI), Recurrence.

Introduction

Pigmented villonodular synovitis (PVNS) is a benign but locally aggressive disease of synovial proliferation that can occur in all joints, tendons, sheaths, and bursae, with a predilection for the knee. The original description of PVNS is credited to Chassaignac in 1852, who described abnormal sarcomatous-like synovial proliferation occurring in the middle and index finger flexor tendon sheaths. The term PVNS was first used by Jaffe et al. in 1941 in their description of 20 synovial lesions occurring in joints, tendon sheaths, and bursa. These lesions had been previously classified based on their predominant histological pattern and location of presentation. Their original names emphasized a perceived neoplastic origin and included terms such as giant cell, fibrohemangioma, benign synovioma, giant cell tumor of the tendon sheath, fibrohemosideric sarcoma, and xanthoma. Jaffe et al. recognized a common histological appearance and attributed variations to differing stages of maturation. They concluded that nodular and diffuse forms of PVNS were histologically similar and part of a spectrum of synovial proliferative disease. They also believed that PVNS was not a neoplastic condition but rather an inflammatory response to an unknown etiologic agent. Their term PVNS was used to emphasize the inflammatory nature of the disease process.

Materials and Methods

A prospective study of a 43-year-old male with a 6-month history of pain and swelling of his left knee, with worsening stiffness of the knee, and a history of a trivial fall, was referred to our orthopedic department for further evaluation from the department of rheumatology after initial rheumatological evaluation. There were no associated symptoms or other joint involvement. Physical examination revealed joint effusion, palpable synovial thickening, and full restriction of the knee range of motion. Radiographic findings showed joint destruction with loss of joint space. Laboratory workup included a complete blood count (E), C-reactive protein (N), erythrocyte sedimentation rate (N), and rheumatological markers (N). A magnetic resonance imaging (MRI) of the right knee revealed diffuse multinodular synovial thickening with multiple foci of gradient blooming and heterogeneous rim

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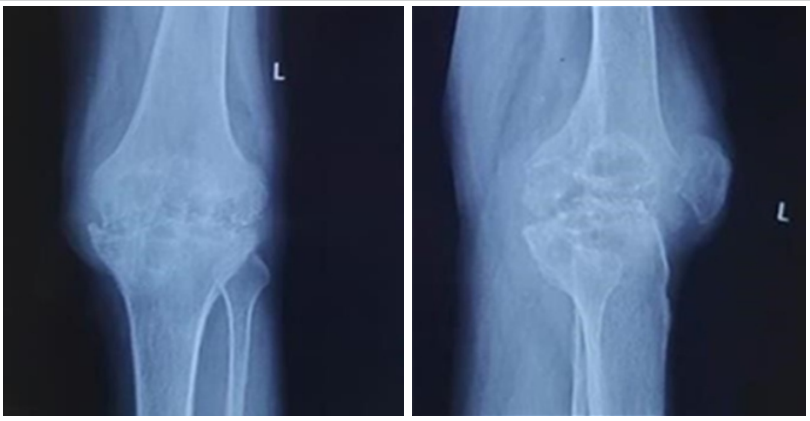

Figure 1:

enhancement.

Management and Results

After initial physiotherapy, the patient was taken to an open arthrotomy with debridement and extensive synovectomy with biopsy. The post-operative clinical course was uneventful.

Physical therapy began on post-operative day 2, once the drain was removed. Suture removal was done on day 12. After confirming the biopsy report as PVNS, the patient was started on radiotherapy and was followed further. At the end of a 1-year follow-up, the patient had improved symptomatically, with improved range of motion, and there was no evidence of recurrence on radiological imaging.

Review of Literature and Discussion

Diffuse form of PVNS and incomplete tumor resection are two risk factors of recurrence reported in the literature. It has been reported that localized PVNS rarely recurs, with rates of 0–15%. On the other hand, diffuse PVNS has high recurrence rates varying from 9% to 46%. In addition, extra-articular invasion of the lesions has been reported to have a high rate of recurrence. Complete excision of all lesions has been recognized as the critical measure in the prevention of recurrence. For localized PVNS, simple excision of the lesion is the gold standard of surgery, while for diffuse PVNS, adequate synovectomy is very important. The surgical approaches include open surgery, arthroscopic surgery, and a combination of both, followed by radiotherapy. Radiotherapy is the most widely used adjuvant treatment after surgery. It has been shown that radiotherapy reduces recurrence in diffuse PVNS, especially in cases with incomplete synovectomy. Ottaviani et al. recommended intra-articular radiotherapy after partial synovectomy for diffuse PVNS due to the difficulty in completely resecting all lesions. Gouina et al. proposed that adjuvant treatment is necessary after synovectomy for diffuse PVNS. Isotopic synoviorthesis or external radiotherapy may be considered after primary resection, and systemic treatment by targeted therapy or radiation therapy may be an option after synovectomy in cases of recurrence or rapid progression.

In addition, delayed diagnosis is a crucial risk factor for recurrence and poor prognosis, which could be rescued by MRI and post-operative follow-up. Research has shown that two-thirds of local recurrences were diagnosed during the first 2 years, and 90% of all local recurrences occur within this period. Therefore, to detect recurrence in time, regular and timely follow-up is required. For recurrent PVNS of the knee, surgical resection, either open or arthroscopic, is considered the main strategy of treatment. Jobe et al. considered that it is necessary to excise some otherwise normal-appearing fat and areolar tissue with the synovium in recurrent cases. Moreover, external beam radiation is an option as adjuvant therapy.

Conclusion

PVNS is a rare disease that may involve any joint. Standard treatment is surgical resection through open surgery, arthroscopic surgery, or a combination of both. The risk factors

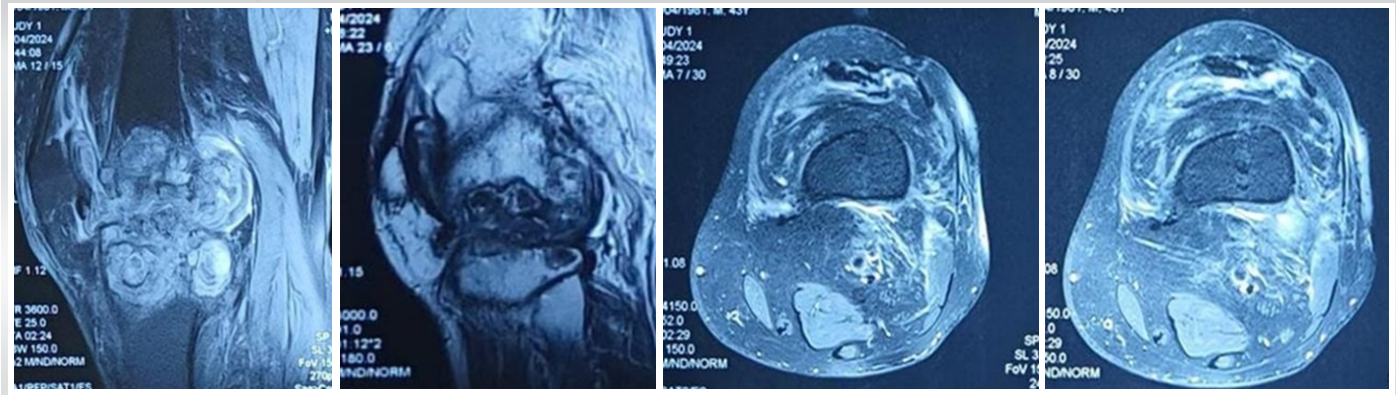

Figure 2: Magnetic resonance imaging showing diffuse multinodular synovial thickening appearing T1 hypointense/T2 heterointense with multiple foci of gradient blooming and heterogeneous rim enhancement on contrast, suggestive of infection/tenosynovial giant cell tumor.

for recurrence include diffuse form of the disease, incomplete resection, location of the lesions, the experience and skills of the surgeon, and adjuvant therapy after surgery. Surgical resection plus adjuvant therapy with close follow-up is recommended for patients with risk factors of recurrence. Early diagnosis and close follow-up determine outcomes.

Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. 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.

Conflict of Interest: NIL; **Source of Support:** NIL

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