Pigmented villonodular synovitis of the knee in a patient on oral anticoagulation therapy: a case report

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Abstract

Introduction: Pigmented villonodular synovitis is a disease which affects the synovial joints and tendon sheaths. Although the exact aetiological factors are not known, we believe that recurrent haemarthrosis has a role in the aetiology of this condition.

Case presentation: A 62-year-old Caucasian man presented with gradually worsening pain and stiffness in his right knee. The patient was on anticoagulation therapy and had been treated for recurrent episodes of spontaneous haemarthrosis of the knee. The International Normalized Ratio on each occasion suggested poor control of the anticoagulation therapy. A diagnosis of pigmented villonodular synovitis was made based on intra-operative findings and was further confirmed by a histopathological examination.

Conclusion: This report is presented to highlight the unusual association of haemarthrosis and pigmented villonodular synovitis.
knee replacement. Intraoperatively, the synovium was found to be hypertrophic and stained reddish orange and the synovial fluid was reddish-orange in colour (Figure 1). These appearances suggested a diagnosis of PVNS. A synovectomy was performed, which was then followed by a total knee replacement. A synovial specimen was sent for histopathological examination. The microscopic features were consistent with a diagnosis of PVNS (Figure 2). The postoperative period was uneventful and the patient was asymptomatic after three years of follow-up treatment.

Discussion

PVNS typically occurs in adults in their third or fourth decade of life, with a male-to-female ratio of 1.9 to 1.3. Involvement is usually monoarticular [2]. The knee joint is the most frequently affected site, followed by the fingers, feet, ankles, hips, wrists and shoulders in a decreasing order of frequency [2,3].

Since the first description of this condition by Jaffe et al. in 1941 [4], the aetiology of this benign tumour involving the synovial membrane has remained unclear. Jaffe proposed that a hypervascular cellular phase occurs after trauma producing hyalinization and fibrosis [4]. Various aetiologies including trauma [2], inflammation [3], haemorrhage [5], neoplasia [6] and genetic factors [7] have been suggested. Chronic recurrent microtrauma and haemarthrosis have also been postulated [2].

It has also been postulated that PVNS in children arises through a different mechanism to that in adults, and it is also possible that not all lesions interpreted as PVNS share the same mechanism [8].

There are very few cases of PVNS reported in patients on anticoagulation therapy [1] and with a bleeding disorder [9]. In our patient, the symptoms in the knee worsened following these repeated episodes of haemarthrosis and the INR on these occasions showed a poor control of his anticoagulation therapy.

Conclusion

This case supports the argument of earlier reports [1,9] that repeated haemarthrosis may have a role in the aetiology of PVNS. We hope that this study will encourage the reporting of similar cases to lead to a better understanding of the aetiology of this condition.

Abbreviations

INR: International Normalized Ratio; PVNS: pigmented villonodular synovitis.
Competing interests
The authors declare that they have no competing interests.

Authors’ contributions
SS made substantial contributions in acquiring data, reviewing the literature and preparing the manuscript. BR performed the knee replacement operation and also contributed in reviewing the literature and drafting the manuscript. SSB gave final approval to the draft to be published. All authors read and approved the final manuscript.

Consent
Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

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