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A rare case of extensive diffuse nonpigmented villonodular synovitis as a cause of total knee arthroplasty failure



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ABSTRACT

INTRODUCTION: Nonpigmented villonodular synovitis (non-PVNS) is a benign proliferative disease involving the synovium. It is a rare condition that is little recognized. Non-PVNS has been reported as a cause of total knee replacement failure.

PRESENTATION OF CASE: We report a case of extensive diffuse non-PVNS in a patient with tibial component loosening after total knee replacement and review the related literature.

DISCUSSION: It is reported that pigmented villonodular synovitis (PVNS) occurs less frequently than non-PVNS after knee replacement. However, there are many more case reports of PVNS than non-PVNS after knee arthroplasty in the English-language literature.

CONCLUSION: Previously, there were no reported cases of extensive diffuse non-PVNS after total knee arthroplasty (TKA). This case study highlights an unusual case of non-PVNS as a cause of TKA failure. We propose that non-PVNS should be considered as a differential diagnosis in patients after TKA who present with recurrent pain and effusion/hemarthrosis of the knee, and that it is one of the causes of implant loosening after TKA.

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1. Introduction

Pigmented villonodular synovitis (PVNS) is a rare, benign proliferative disease of the synovial membrane of joints, tendon sheaths, and extra-articular bursae, which may be locally aggressive.^{1–3} Although Chassignac originally defined this disease in 1852,⁴ the term PVNS was first used by Jaffe et al. in 1941.² The knee joint is most commonly affected during the third or fourth decade.⁵ There are diffuse and focal forms of the disease.^{3,6}

PVNS after replacement arthroplasty is thought to result from a reaction to polyethylene, metal, and cement wear.^{7,8} Histological examination of the lesion is necessary to establish the diagnosis.^{9,10} However, a diagnosis can be made using plain radiographs, computerized tomography (CT), magnetic resonance imaging (MRI),^{9–11} bone scintigraphy, and positron emission tomography.¹² MRI is the preferred imaging technique.^{9–11}

Nonpigmented villonodular synovitis (non-PVNS) is more common than PVNS.⁷ Although it was reported that non-PVNS may occur after replacement knee arthroplasty,^{4,7,8} all villonodular synovitis cases after arthroplasty in the English-language literature are pigmented.^{3,6,13}

2. Presentation of the case

A 53-year-old male who presented with swelling and pain in the right knee for 1 month had undergone cemented right total knee arthroplasty (TKA) 4 years earlier because of osteoarthritis in another clinic. There was no documental information indicating whether any potential signs for PVNS in the synovial membrane were determined at the time of surgery. However, the patient had no history of PVNS after surgery. During physical examination, swelling, effusion, and mediolateral instability of the knee joint were found. The patient had no history of trauma or infection, and had a good range of movement. Blood tests, including for inflammatory and rheumatological markers, were normal, while plain radiography was consistent with suspected loosening that was found as a radiolucent lesion below the tibial component (Fig. 1).

After initial presentation, arthrocentesis of the right knee was performed. The fluid aspirate was hemorrhagic or serosanguinous, with no growth on cultures. Initially, a conservative treatment was

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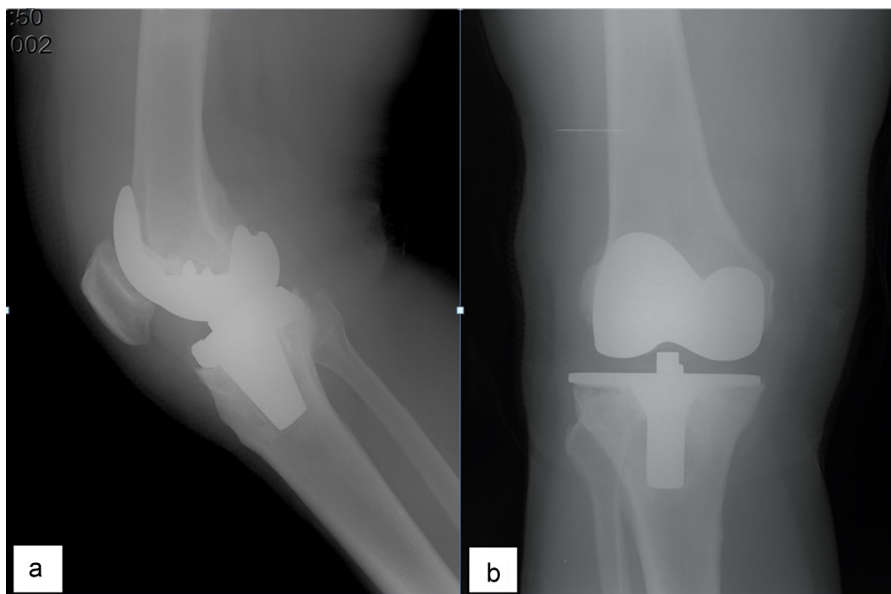


Fig. 1. Pre-operative anteroposterior (a) and lateral (b) radiographs of the knee show loosening and osteolysis under the tibial component in the medial compartment.

performed (aspiration of the knee, ice, splinting, rest, analgesic, and anti-inflammatory drug). Because the effusion and pain were recurrent, the aspirate fluid and blood were analyzed, and all parameters were found to be within the normal ranges. A three-phase bone scan (technetium 99 m methylene diphosphonate) showed increased hyperperfusion and hyperemia around the tibial component of the right TKA both in the perfusion and blood pool phase. Increased focal uptake around the tibial component was shown in the late static phase (Fig. 2). This condition was associated with aseptic loosening of the tibial component, and the patient was hospitalized for revision surgery. During revision surgery, the orthopedic surgeon unexpectedly found excessive diffuse proliferation of the synovial hypertrophy, including papillae and nodules, with yellow and brown areas surrounding the prosthesis (Fig. 3). The tibial components were loose and accompanied by bone loss, while the

polyethylene liner was severely damaged. After complete resection of all the pathological tissues, revision TKA was completed (Fig. 4).

Macroscopically, the specimen surface was covered with nodules and short-long papillae. On histopathological examination, the specimen showed papillary villous structures surrounded by the synovial cell layer, which consisted of different sized and shaped cells (Fig. 5a). There was inflammatory cell infiltration by multinucleated giant cells, histiocytes, and a small number of lymphocytes under the synovial cell structures on the surrounding papillary villous structure (Fig. 5b). Immunohistochemically, there was a diffuse infiltration of CD68-stained histiocytes. Histological analysis demonstrated the typical appearance of non-PVNS (Fig. 5c).

At a 6-month follow-up after surgical resection, no recurrence due to total surgical synovectomy was observed, and the patient reported no knee pain and only mild residual swelling. The knee

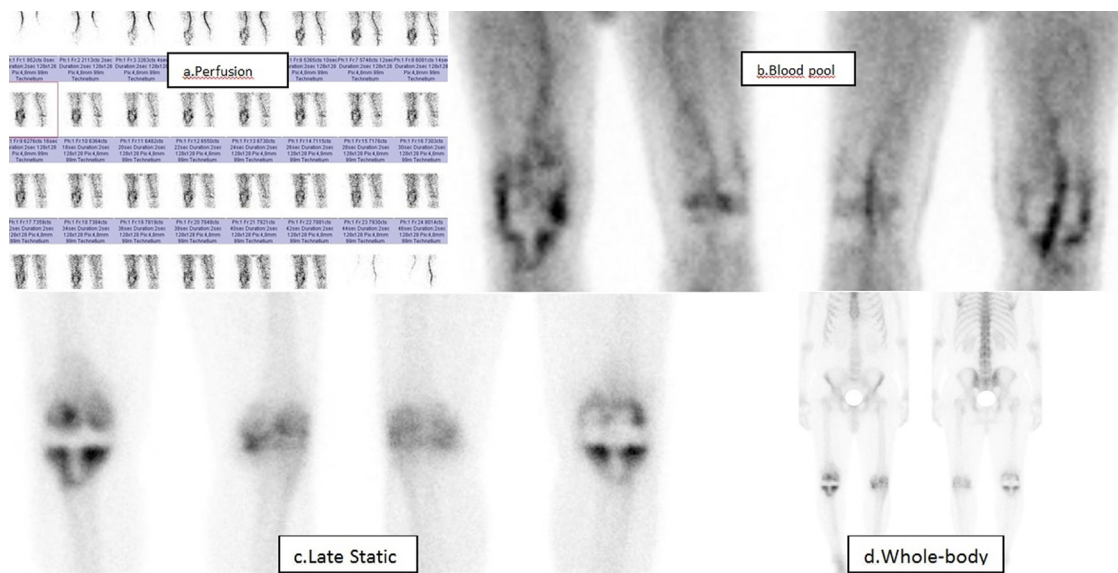


Fig. 2. Perfusion, blood pool, static phase, and anteroposterior whole-body images (three-phase bone imaging using technetium 99 m methylene diphosphonate). Periprosthetic hyperperfusion, hyperemia, and increased periprosthetic activity are seen around the tibial component of a right total knee replacement.

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