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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.

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-PNVS 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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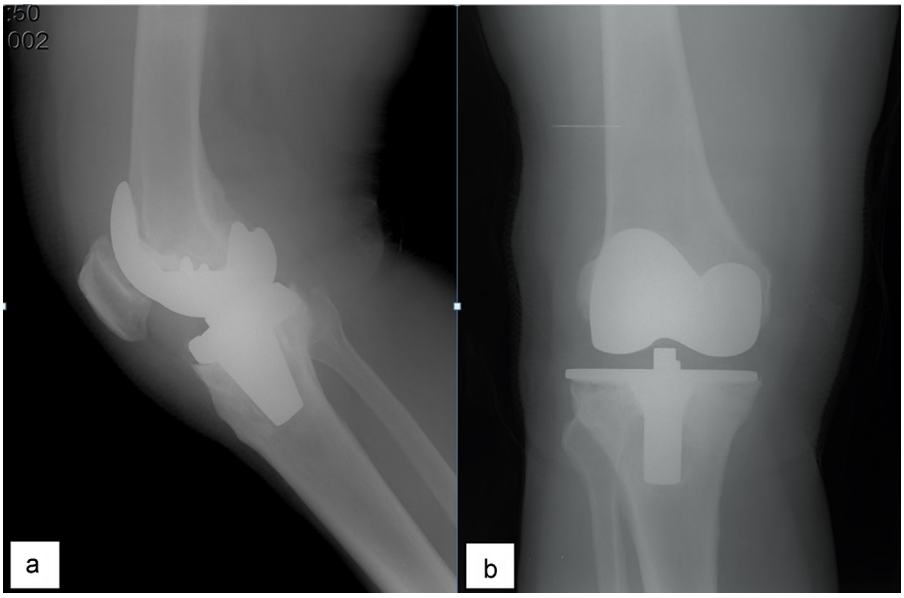

Fig. 1. Pre-operative anteroposterior (a) and lateral (b) radiographs of the knee show loosening and osteolysis under the tibial component in the medial compartment.

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 performed (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

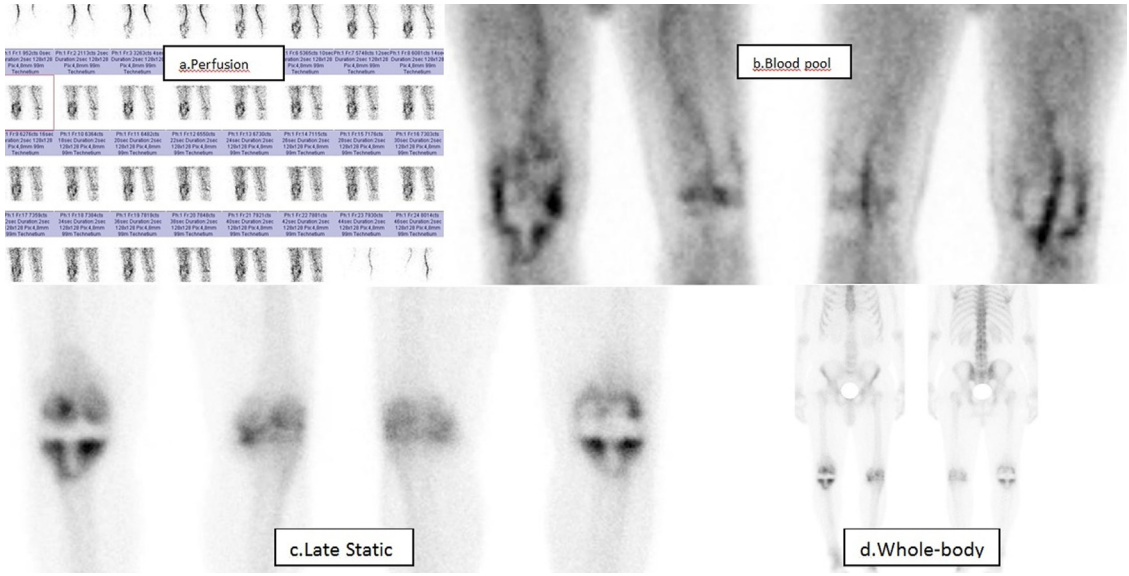

Fig. 2. Perfusion, blood pool, static phase, and anteroposterior whole-body images (three-phase bone imaging using technetium 99 m methylene diphosphonate). Periprosthetic hyperperfusion, hyperemia, and increased periprosthetic activity are seen around the tibial component of a right total knee replacement.

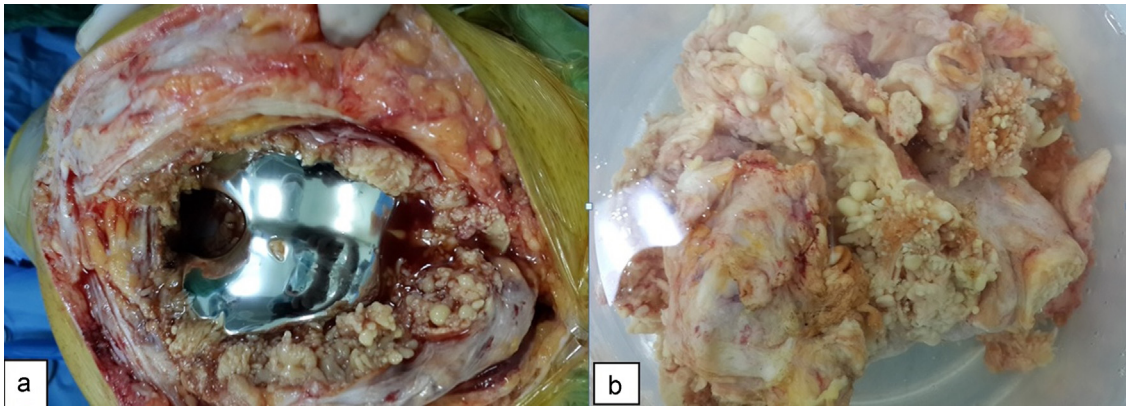

Fig. 3. Before (a) and after (b) synovectomy. Intra-operative photographs of the knee show diffuse papillae and nodules in the proliferating synovium, with yellow-brown areas (typical appearance of villonodular synovitis).

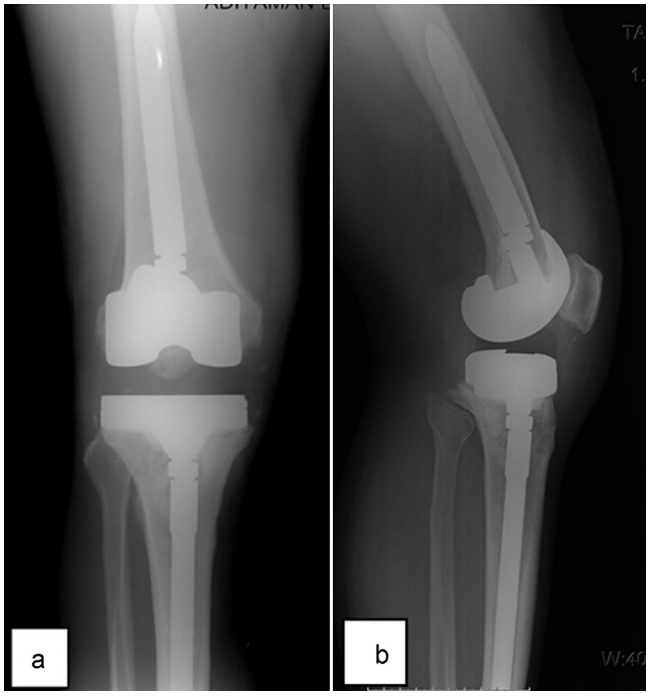

Fig. 4. Post-operative anteroposterior (a) and lateral (b) radiographs of the knee show acceptable alignment and a well-fixed tibial component after revision total knee replacement.

range of motion was normal. The patient could walk without support and was able to negotiate stairs without any assistive device.

3. Discussion

Tenosynovial giant cell tumors (GCTT) are a group of generally benign intra-articular and soft tissue tumors with common histological features.[14] Various terms have been used to describe the different manifestations of synovial-type giant cell tumors, including PVNS (malignant, localized, and diffuse-type), diffuse-type villonodular synovitis, localized nodular synovitis, bursitis, and synovitis. These lesions are often classified as localized or diffuse types, depending on the growth, a distinction believed to have clinical relevance.[14,15] Localized tumors are generally indolent, whereas diffuse tumors are locally aggressive, often with multiple recurrences. The diffuse type widely infiltrates and entraps adjacent soft tissue, and frequently erodes bone, in contrast to the localized type.[14] In reviewing the pathology slides, it was difficult to differentiate between PVNS and non-PVNS involvement in hemosiderin deposition. Diffuse-type villonodular synovitis and PVNS have similar age distributions, locations, and symptoms.[14,16] Although there has been considerable speculation regarding PVNS etiology, its cause remains unknown.[17] While most authors propose that it is an inflammatory process, some suggest that it is a benign neoplasm.[16] Other possible etiologies are metabolic disturbance, trauma, and hemorrhage.[5] PVNS can be confused with other intra-articular pathologies.[18] Bouali et al. suggested that diagnosis of PVNS must be made with a high degree of clinical skepticism and using a synovial biopsy until a more sensitive noninvasive test can be developed for cases in the early developmental stages.[1]

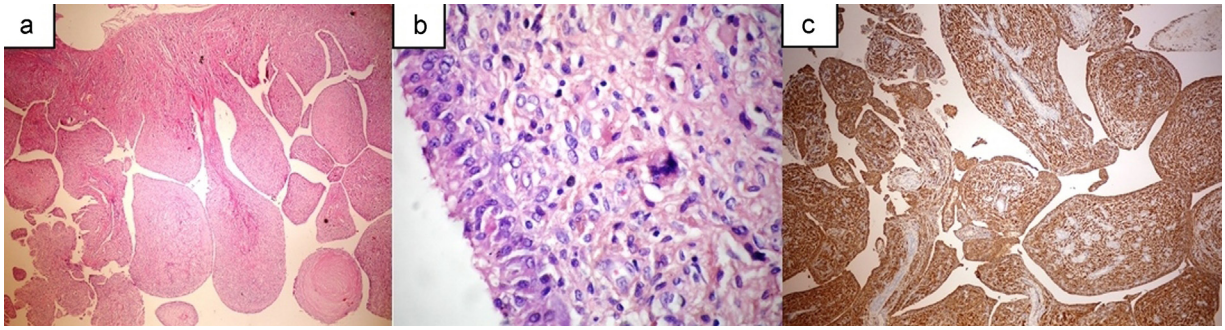

Fig. 5. (a) Photomicrograph of the specimen shows the synovial cell layer which is formed from different sized and shaped cellular areas surrounding the papillary villous structures (hematoxylin and eosin (H&E) staining, original magnification 40×). (b) Photomicrograph showing synovial proliferation with multinucleated giant cells, histiocytes, and a small number of lymphocytes under synovial cell structures surrounding the papillary villous (H&E, original magnification 40×). (c) Immunohistochemically there is diffuse infiltration of CD68-positive histiocytes (original magnification 20×).

PVNS is normally monoarticular, with the most commonly affected joint being the knee.[5,19] It most frequently affects adults in their third or fourth decade without significant gender predisposition and with an deceptive range of symptoms.[5] PVNS may adversely affect the quality of life,[20] because patients often present with symptoms of pain, swelling, stiffness and occasional locking, or signs of a palpable mass.[19] Joint aspiration typically yields hemorrhagic or xanthochromic/serosanguinous (brown, murky) fluid.[1] In the present case, an arthrocentesis was also performed. The aspirate fluid was serosanguinous and no growth was found on cultures.

Macroscopically, the localized form involves only a portion of the synovial surface and there appears to be less villous proliferation than in the diffuse form. In cases of localized PVNS, where the hemosiderin deposition is minimal, the MRI may appear normal.[1] The diffuse form is more common, accounting for almost 75% of all cases. It affects either all of the synovium or extensive areas.[21] Macroscopically, the synovium is prolific with coarse villi, finer fronds, and diffuse nodularity. The synovium is often heavily pigmented, ranging from dark yellow to chocolate brown.[1] In the present case, there was excessive diffuse proliferation of the synovial hypertrophy, including papillae and nodules, with yellow and brown areas surrounding the prosthesis, but without hemosiderin deposition. The tibial components were loose and displayed bone loss, and the polyethylene liner was severely damaged.

Microscopic findings of PVNS include abundant collagen production, proliferation of mononuclear synovial cells and fibroblasts, hemosiderin and lipid deposits, foamy histiocytes, plasma cells, giant multinucleated cells, and xanthoma cells.[1,2,21] The hypertrophic synovium in this disease is typically yellow-brown papillary, villous, nodular, or villonodular, and contains variable amounts of hemosiderin and lipid.[2,5] Hemosiderin-laden macrophages are common in most GCTT. However, there is no involvement of hemosiderin deposition in non-PVNS, whereas prominent, abundant hemosiderin deposition appears in diffuse PVNS.[15,16]

In the present case, the specimen surface was covered with nodules and papillae. On histopathological examination, cellular infiltration consisting of multinucleated giant cells, histiocytes, and a small number of lymphocytes was found under the synovial cell structures surrounding the papillary villous structure. A diffuse infiltration of histiocytes was shown using CD68 staining. However, there were no hemosiderin deposits or hemosiderin-laden macrophages. Histological analysis demonstrated the typical appearance of non-PVNS.

Histological examination of the lesion is necessary to establish a diagnosis.[9,10] The history, physical examination, and radiographic studies may assist in making a diagnosis.[1] Plain radiography is nonspecific and classically shows soft-tissue swelling or mass, bone erosion, and subchondral cysts in the joint.[22] MRI is useful in early diagnosis, demonstrating the extent of the PVNS and thus improving the early management of this pathology.[11] Bone scintigraphy is sensitive, with a high negative predictive value, and is thus a useful screening test. Many patients are referred to nuclear medicine specifically to differentiate aseptic loosening from infection. Some investigators reported that increased focal and diffuse activity around components of the prosthesis was associated with loosening. However, increased periprosthetic activity can persist for an extended period during the first year after implantation.[12] CT can help to define extrinsic bone erosion and excavation.[9] In our patient, plain radiographs were consistent with suspected loosening that was found as a radiolucent lesion below the tibial component. In bone scintigraphy, we observed periprosthetic hyperperfusion, hyperemia, and increased periprosthetic activity around the tibial component of the right total knee replacement.

The optimal treatment for PVNS is to complete surgical resection of all the affected synovial tissue.[7,10,11,16] Treatment options include surgical, arthroscopic, and radiation synovectomy, and combined procedures.[10,18] Diffuse PVNS is more difficult to eradicate and is optimally treated using near total or total synovectomy.[10] The treatment of extensive diffuse PVNS of large joints using isolated surgical resection is unsatisfactory because of the high rates of local recurrence. Post-synovectomy adjuvant treatment with radiosynovectomy for extensive diffuse PVNS of the knee joint is a reliable treatment method with good results in regard to the incidence of local recurrence and an acceptable functional outcome.[7,10] In our patient, there was no recurrence 6 months after surgery, although this was a relatively short follow-up period.

Non-PVNS may be seen after TKA and it is thought to occur as the result of a reaction to polyethylene, metal, and cement wear.[7,8] The causal relationship between TKA and PVNS remains unknown. In addition, it has not been explained how failure and loosening after TKA is caused by PVNS.[13] An idiosyncratic reaction in certain patients to fine wear debris was assumed in the single case described so far.[8] Eventually, the common end point of PVNS of the knee leads to destruction of the joint and joint replacement.[23] PVNS should be considered a differential diagnosis of patients who present with recurrent painful swelling of the knee and as one of the causes of implant loosening after TKA. PVNS after TKA is rare, and several cases have been reported after primary knee arthroplasty, appearing a mean of 4.4 years after arthroplasty was performed.[3,6,13] The mechanism of joint arthroplasty failure is not clearly understood. Histological studies on patients with failed metal-on-metal implants have shown a diffuse collection of perivascular lymphocytes and plasma cells in patients with metal hypersensitivity. In contrast, in patients with a metal-on-polyethylene implant the predominant reaction was a histiocytic response. Lymphohistiocytic infiltrates surrounding an implant may indicate intolerance reactions.[24]

Chung and Park proposed that PVNS should be considered in the differential diagnosis of patients who present with painful swelling of the knee and as one of the causes of implant loosening after TKA. Although they could not identify the cause of the loosening in their case study, they suggested that it was more likely to arise from the direct involvement of PVNS than be the result of a reaction to wear debris.[13]

It was reported that PVNS is observed less frequently than non-PVNS after knee replacement.[3,6–8,13] However, there are many more case reports of PVNS than of non-PVNS after knee arthroplasty.[3,6,13]

4. Conclusion

Non-PVNS is histopathologically and clinically similar to PVNS, except for hemosiderin deposition. To date, there are no reported cases of extensive diffuse non-PVNS after TKA. It is interesting that loosening of the tibial rather than the femoral component occurs in patients with villonodular synovitis after TKA. Further research is required to elucidate why the tibial component loosens in patients with PVNS. This study highlights an unusual case of non-PVNS as a cause of TKA failure. We recommend total surgical resection in the treatment of diffuse non-PVNS. If it recurs, radiosynovectomy should be included after surgical excision. In conclusion, 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 following TKA.

Conflict of interest

None.

Funding

None.

Ethical approval

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Author contributions

Hacı Bayram Tosun performed the operation, writing and study design of this paper. Abuzer Uludağ involved in research article and study design. Sancar Serbest involved in research article and editing pictures. Seyitali Gümüştaş involved in research article and writing and İbrahim Halil Erdoğdu contributed in Histopathological examination.

Key learning points

- Non-PVNS (nonpigmented villonodular synovitis) is histopathologically and clinically similar to PVNS, except for hemosiderin deposition.
- Non-PVNS is a rare condition that is little recognized.
- 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.
- It is interesting that loosening of the tibial rather than the femoral component occurs in patients with villonodular synovitis after total knee arthroplasty.
- We recommend total surgical resection in the treatment of diffuse non-PVNS.

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