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Case report

An unusual case of pigmented villonodular synovitis after total knee arthroplasty presenting with recurrent hemarthrosis

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ABSTRACT

Pigmented villonodular synovitis (PVNS) is a benign proliferative joint disease, which is a rare finding after total knee arthroplasty (TKA). There is currently no link between PVNS and TKA, and it has been described infrequently in the literature. Its presentation has varied along with the time that it presents postoperatively. We describe a case of a patient who presents with recurrent hemarthrosis 4 years after TKA. The patient had no previous history of PVNS and had an arthroscopy 1 year after the index operation with no evidence of synovitis. We present details of the first case with a review of imaging and pathology and a brief review of the literature on PVNS occurring after TKA.

Introduction

Pigmented villonodular synovitis (PVNS) is a benign synovial joint proliferative disorder with an incidence of 1.8 million cases per year [1]. Two types of PVNS have been described, namely localized and diffuse [2]. The localized form is described as a discrete mass within the synovium, as opposed to diffuse PVNS that involves the entire synovium, whether it be intraarticular or extraarticular [2]. This locally aggressive lesion is poorly understood and usually affects large joints, most often the knee. Patients may present with various clinical findings, but most often, they present with pain and limited range of motion due to recurrent hemarthroses commonly found in the diffuse form [3]. Imaging often demonstrates destructive changes including periarticular cysts, joint space narrowing, and erosions [4,5]. Despite it being a benign process, recurrence rate after initial resection has been noted between 8% and 60% [6]. PVNS after total knee arthroplasty (TKA) is very rare, with limited case reports available [7-11]. Currently there are no published reports describing PVNS 4 years after a primary TKA with recurrent hemarthrosis and no evidence of synovitis 1 year after surgery.

Case history

A 62-year-old female presented with 7-month history of recurrent right knee swelling, pain, and limited range of motion 3 years after primary TKA. At the age of 58 years, the patient's preoperative radiographs of the right knee showed medial and patellofemoral joint space narrowing, subchondral sclerosis, and osteophytes (Fig. 1a and b). Physical examination was consistent symptomatic tricompartmental disease primarily affecting the medial and patellofemoral compartments. The patient failed conservative management including nonsteroidal anti-inflammatory medication, corticosteroid injections, and activity modification. She underwent a cemented right TKA and patelloplasty without complication by an arthroplasty surgeon affiliated with the author's institution. During the procedure, her synovium was examined and not found to be significantly inflamed and was assuredly not notable for any characteristics of PVNS. Six months after her joint replacement, the patient reported a mechanical fall directly onto her right knee. After her fall, the patient complained of persistent anterior and lateral right knee pain. Nine months after her surgery, she had a corticosteroid injection into the lateral aspect of the right femoral condyle for persistent iliotibial band syndrome. The patient had continued pain but full range of motion and no signs of infection on physical examination.

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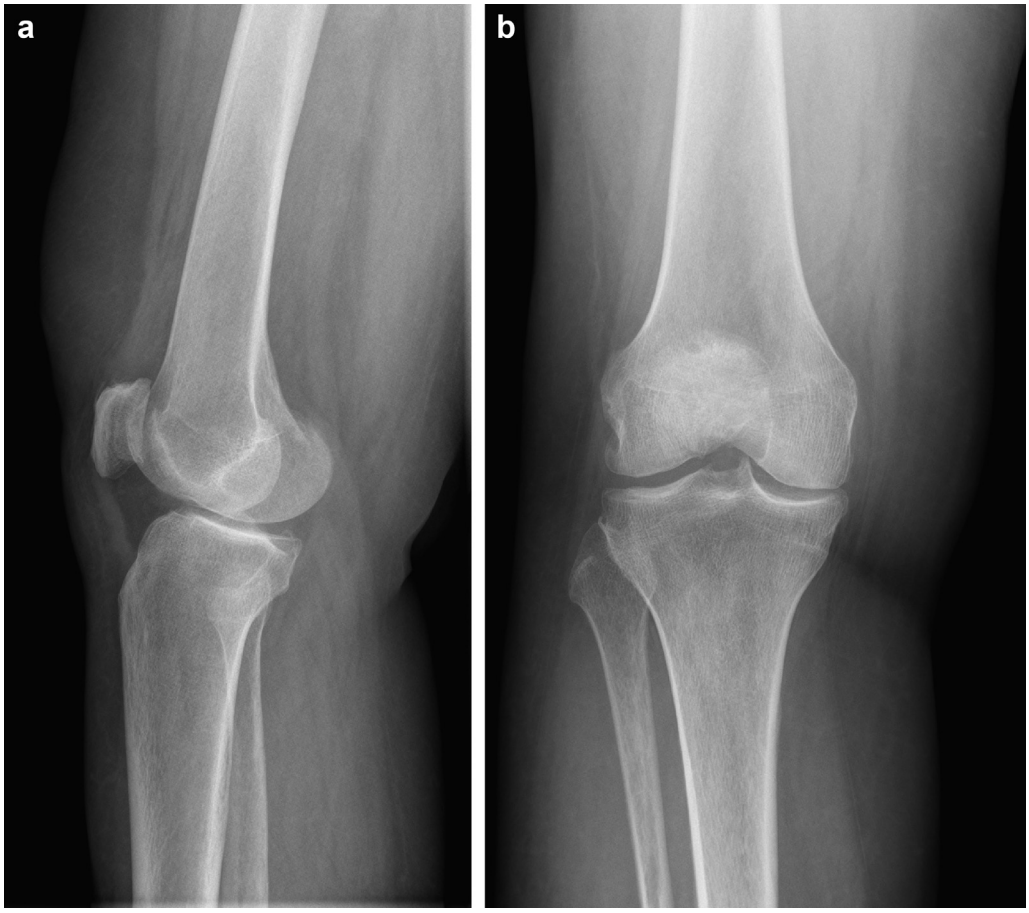

Figure 1. (a-b) Preoperative lateral and anteroposterior radiographs of the right knee demonstrating medial and patellofemoral joint space narrowing, subchondral sclerosis, and osteophytes.

One year after her initial surgery, she was referred to another surgeon for arthroscopic debridement for suspected traumatic fat pad syndrome. Preoperative physical examination demonstrated a thickened anterior fat pad with no evidence of infection. Figure 2a-c shows intraoperative images with no evidence of synovitis or hemarthrosis. There was also no evidence of synovitis as mentioned in the operative report. Furthermore, normal scarring and no inflammatory synovitis were reported. The patellar component was

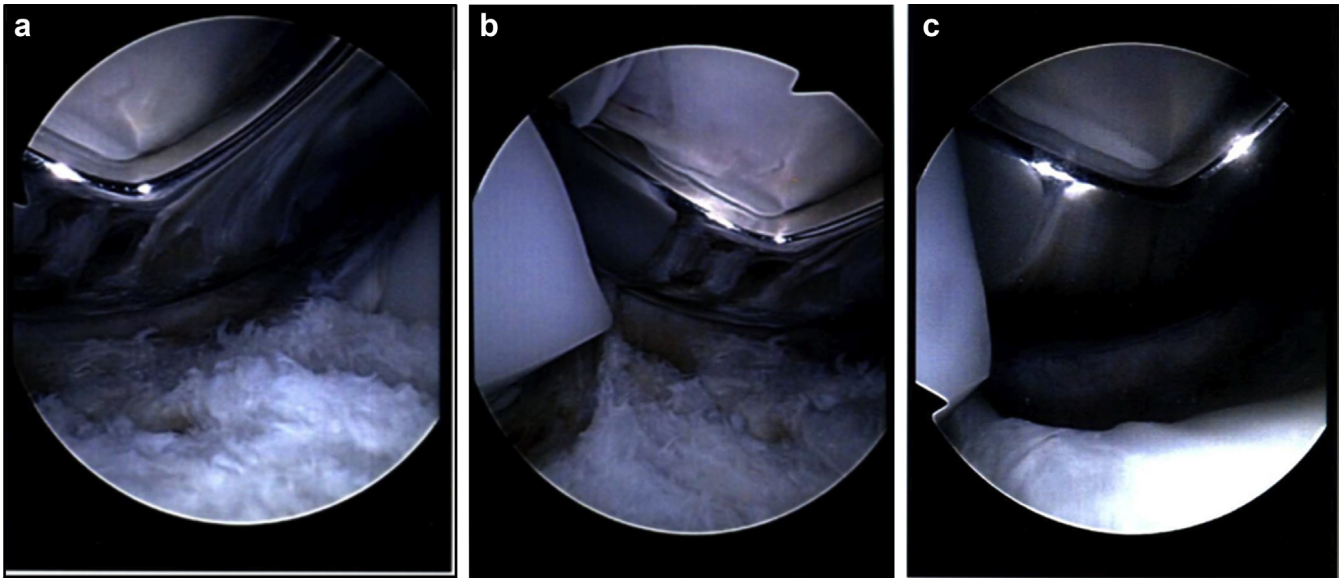

Figure 2. (a-c) Intraoperative arthroscopic images of knee showing no signs of synovitis or hemarthrosis.

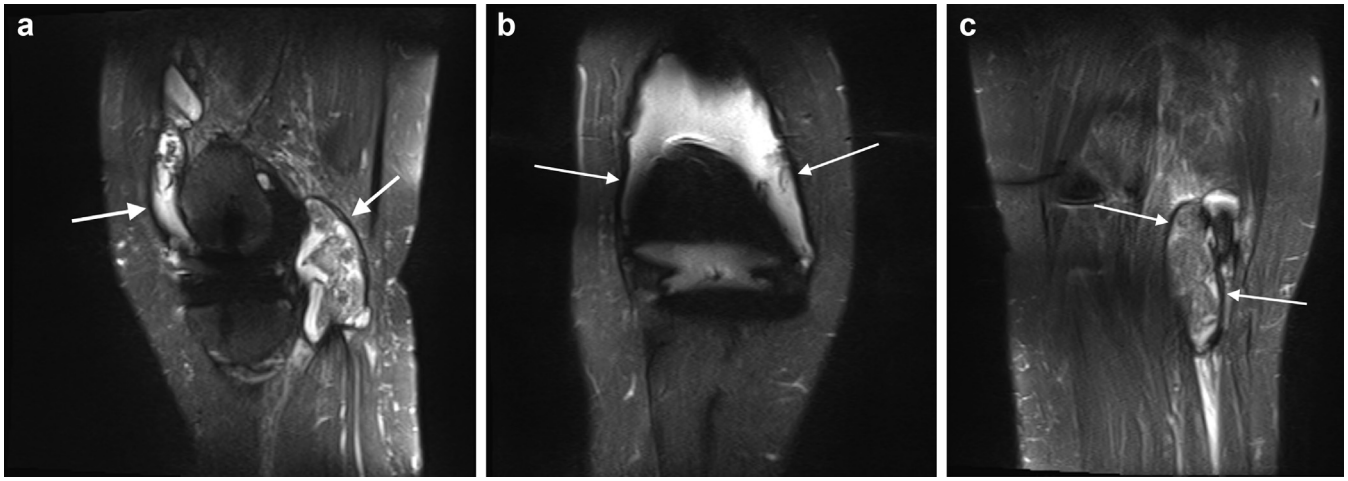

Figure 3. (a-c) T2 weighted MRI images of the sagittal, anterior coronal, and posterior coronal views of the right knee demonstrating a moderate knee effusion (left arrow in panel a, arrows in panel b) with a large popliteal cyst and internal debris posteriorly (right arrow in panel a, arrows in panel c).

observed to track normally, and there was no evidence of either tibial tray loosening or patellar component loosening. The patient reported mild symptomatic improvement after her arthroscopy.

The patient presented to our office 3 and a half years after her initial surgery complaining of 4 months of recurrent swelling and pain in her right knee of atraumatic onset. She stated that she had visited multiple outside emergency room facilities and had 3 aspirations of her right knee in the span of 2 months. On examination, the patient had a moderate effusion and anterior knee pain with palpation. Aspiration during March 2016 from an outside hospital was bloody in appearance, with a total nucleated cell count of 3220. Cell count was unable to be adjusted for hemarthrosis [12] as serum complete blood count was not provided by the outside facility. Cultures from all previous aspirates showed no bacterial growth. Her sedimentation rate and C-reactive protein level were 21 mm/h and 1.28 mg/dL, respectively. Magnetic resonance imaging (MRI) of the right knee demonstrated a moderate knee effusion with a large popliteal cyst, reactive synovitis, and internal debris posteriorly, suggesting the possibility of PVNS [13] (Fig. 3a-c).

The patient was taken back to the operating room for anterior synovectomy and polyethylene exchange. A large complex hematoma was found along with an inflamed, diffusely thickened erythematous synovium (Fig. 4a and b). The excised lesion also had some extension into the quadriceps tendon. On microscopy, the

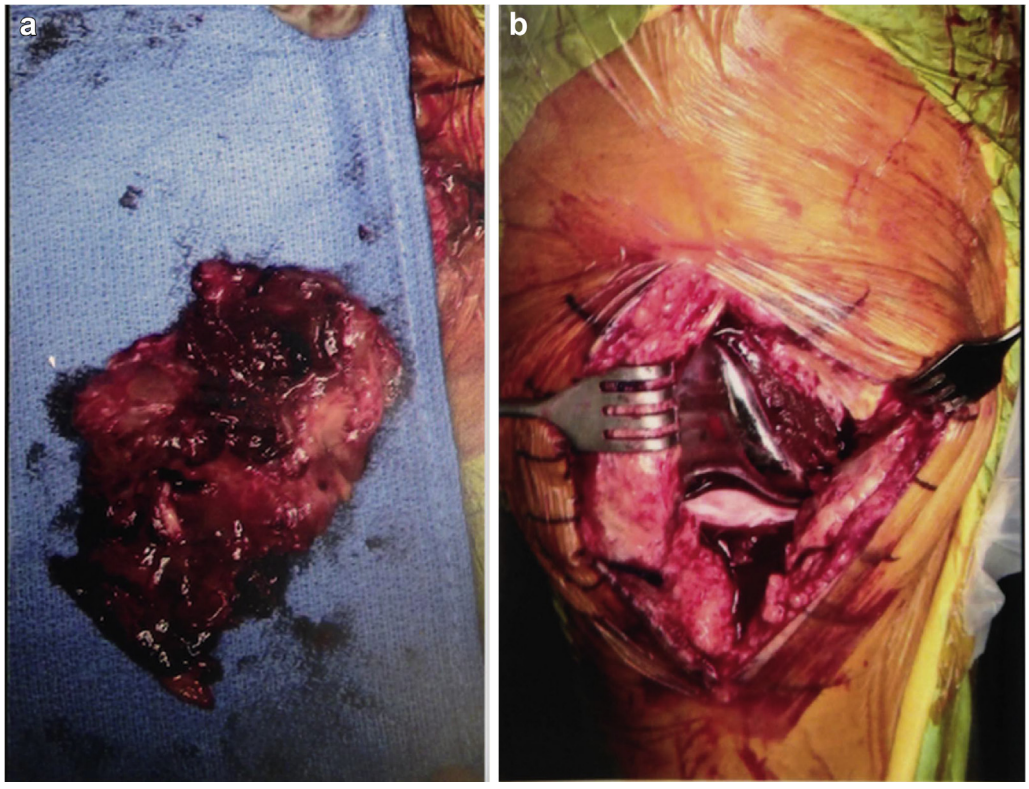

Figure 4. (a) Large complex hematoma removed from the knee joint along with (b) inflamed, diffusely thickened erythematous synovium.

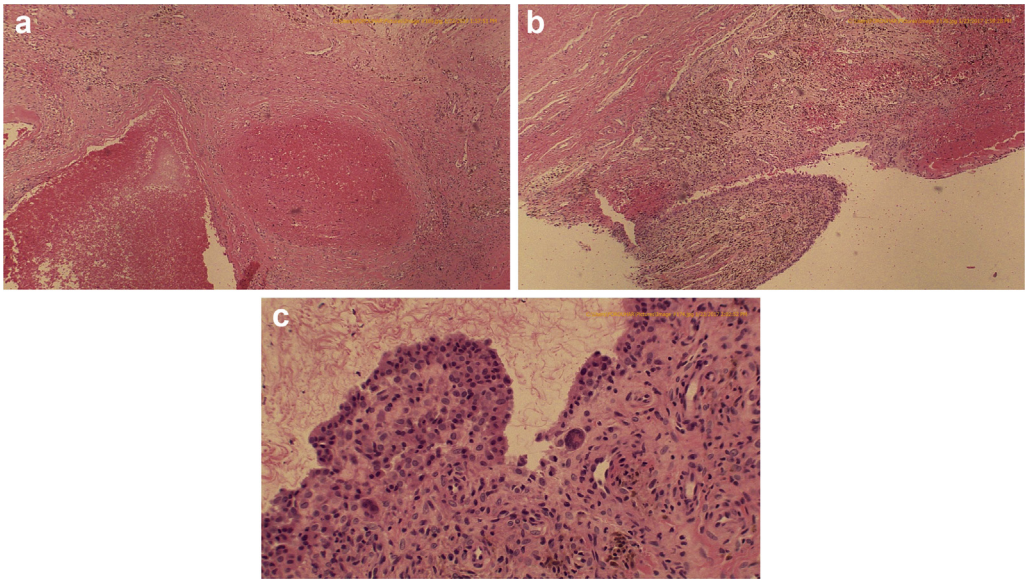

Figure 5. (a) Pathology slides demonstrating fresh and old hemosiderin deposition in the synovial membrane and joint collagenous capsule. (b) Intraparenchymal synovial lining cell groups with (c) occasional giant cell formation.

lesion was characterized by marked fresh and old hemosiderin deposition in the synovial membrane and joint collagenous capsule. Focally, there was synovial lining cell hyperplasia with formation of villous structures. Moreover, there were intraparenchymal synovial lining cell groups with occasional giant cell formation (Fig. 5a-c). The polyethylene was examined and found to be near pristine, with no evidence of wear. The diagnosis made was PVNS. The patient had dramatic improvement in her anterior knee pain after open excisional debridement of her anterior synovium. However, she continued to have posterior knee pain. The surgeon considered a posterior approach to the knee to perform a repeat synovectomy focused on the posterior capsule. However, on repeat MRI, the synovitis had completely resolved. The patient subsequently began to respond to physical therapy. At 1-year follow-up, she had near resolution of her knee pain. At that time, the patient gave her consent to be the subject of this case report.

Discussion

PVNS is a locally aggressive proliferative disorder of the synovium that is a rare complication after TKA [7-11]. The etiology of PVNS is unclear, although recurrent hemorrhage, neoplasm, and trauma have been reported as possible causes [14]. Polyethylene wears off after TKA is a reported cause of nonpigmented synovitis [14]. Although these present similar to PVNS with proliferate synovitis with giant cell formation, they differ in their lack of hemosiderin deposition [11,14]. We present a unique case of a patient who presented with recurrent pain and hemarthrosis secondary to PVNS 4 years after primary TKA. Our study also provides no arthroscopic evidence of PVNS 1 year after index surgery.

Ballard [7] first described PVNS after TKA in a patient 9 years after arthroplasty. Their case report described a patient with previous gonarthrosis who had an uncomplicated right TKA with 9 years of being asymptomatic. The patient then returned complaining of spontaneous hemarthrosis and swelling with negative cultures on multiple aspirations. They postulated that the wear of the polyethylene along with microtrauma of daily movement caused episodes of bleeding to form pigmented synovitis [7]. Oni and Cavallo described a case of diffuse PVNS (DPVNS) in a patient's knee, occurring within 2 years after primary TKA, similar to our patient [9]. They concluded that their patient's presentation may not have been secondary to the surgery, given its quick development, but rather suggested the condition as a spontaneous occurrence [9]. Tosun et al. [15] reported a case of spontaneous diffuse non-PVNS, which presented in similar timeline to our patient roughly 4 years after surgery. Their patient had no evidence of recurrence in symptoms 6 months after revision surgery and synovectomy [16]. Diagnosis of PVNS after total joint arthroplasty is often difficult to make. Imaging can help guide the diagnosis, including MRI. A gradient recalled sequence on MRI may be helpful in establishing the diagnosis. Friedman et al. [13] showed that PVNS demonstrates characteristic blooming with low-to-intermediate signal on T1-weighted imaging and low intensity on T2 imaging due to iron in the deposits.

Treatments described include complete synovectomy and revision of prosthesis if there is evidence of loosening or failure [6-9]. Given the rarity of spontaneous PVNS after a TKA, there are no current data about recurrence after synovectomy. Houdek et al. [17] reported a recurrence rate of 12% at 14-year follow-up for those who had a previous diagnosis of PVNS before TKA.

Although the features of PVNS were present only focally, the presence of villous configuration of synovial membrane, synovial lining cell hyperplasia, and hemosiderin deposition indicate progression of a chronically hemorrhagic condition to an early form of PVNS. Given the time course and manifestation of PVNS in this patient, it suggests that the etiology of the disease was likely traumatic in nature rather than from reaction to the polyethylene.

Summary

We present an unusual case of PVNS 4 years after TKA, presenting with recurrent hemarthrosis but no evidence of synovitis 1 year after surgery. The etiology of this patient's disease was likely traumatic. Her condition dramatically improved after open excisional debridement of the synovium with evacuation of her hematoma.

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