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Case report

Pigmented villonodular synovitis diagnosed during revision total knee arthroplasty for flexion instability and patellar fracture

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ABSTRACT

Occurring in either a localized or diffuse form, pigmented villonodular synovitis (PVNS) is a disease of unknown etiology that typically presents with insidious onset of pain, swelling, stiffness, or mechanical symptoms as a result of synovial tissue proliferation. PVNS preferentially affects large joints, most commonly the knee. Currently there is no known association with PVNS and total knee arthroplasty (TKA), and to date, there are only a few cases reported in the orthopedic literature in which PVNS was diagnosed after primary TKA. To our knowledge, this is the first case of diffuse PVNS that was discovered at the time of revision TKA for flexion instability and patellar fracture. In this patient, with no known history of PVNS, the diagnosis of diffuse PVNS was made at the time of surgery. She underwent revision TKA, partial patellectomy, and extensive synovectomy.
Level of Evidence: V, Case Report.

1. Introduction

Pigmented villonodular synovitis (PVNS) is a relatively rare disease with an estimated incidence of 1.8 per million people per year. It tends to occur in the third and fourth decades of life and affects both sexes. It can affect any joint, but has a predilection for large joints and is most commonly found in the knee [1]. Although the exact etiology is unknown, some possible theories include recurrent trauma and hemarthrosis, abnormal local metabolic activity, or a neoplastic process.

It is known to be a proliferative disease of the synovium that generally presents in one of two forms, localized or diffuse [2–4]. Localized PVNS classically manifests as a pedunculated, focal expansion of synovium. The diffuse form tends to involve the majority or entirety of the synovium of the involved joint [3,5]. Both forms are characterized as inflammatory processes with hemosiderin deposition within the synovial tissue [5]. Patients present with pain and swelling in the affected joint. Localized PVNS tends to cause less swelling but is more likely to cause mechanical symptoms while diffuse PVNS tends to cause more swelling, pain, and stiffness [2,3,5].

Accurate diagnosis of PVNS can be difficult. Radiographs may reveal bony erosions near articular surfaces and preserved articular cartilage that may degenerate with disease progression; however, the vast majority of radiographs may appear normal [2]. On magnetic resonance imaging, PVNS is generally easier to diagnose as both bony erosions and nodular synovial hypertrophy may be present [6].

Management of PVNS generally consists of surgical synovectomy of the affected tissue which is often accomplished arthroscopically; however, depending on the extent of involvement, diffuse PVNS may require open synovectomy. Other treatment considerations include external beam radiation or intra-articular radiation, especially in recurrent cases that are refractory to synovectomy. Some have also advocated total joint arthroplasty as a viable option in the setting of recalcitrant PVNS [7,8].

To date, there are only four cases reported in the literature in which PVNS was diagnosed following total knee arthroplasty (TKA) in patients who had no known disease at the time of index arthroplasty. These cases include the following: diffuse PVNS associated with recurrent hemarthroses nine years following TKA [9], focal PVNS detected 12 months following TKA [10], diffuse PVNS 18 months following TKA [11], and focal PVNS detected at the time of revision TKA for tibial component loosening five years after the index TKA [12]. We present the first known case of diffuse PVNS associated with flexion instability, polyethylene wear and osteolysis requiring revision.

2. Case report

A 64-year-old woman presented with bilateral knee pain, left greater than right. Nine years earlier she underwent right TKA (Natural Knee, Zimmer Inc., Warsaw, IN) for avascular necrosis related to steroid usage for systemic lupus erythematous. The patient was on warfarin for chronic anticoagulation related to antiphospholipid antibody syndrome. No

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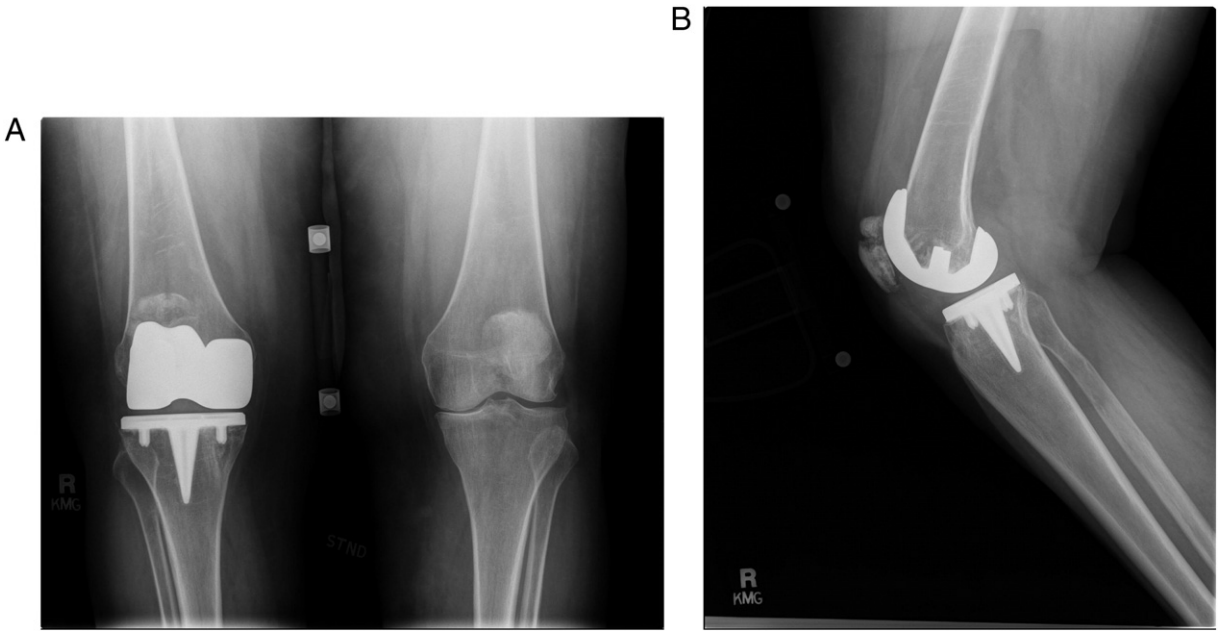

Fig. 1. Pre-operative radiographs demonstrating AP (a) and lateral (b) views of a right total knee arthroplasty with polyethylene wear, osteolysis, and non-united patella fracture of the right knee.

mention of any abnormal proliferation of the synovial tissue was noted at the time of index arthroplasty. She initially did well following her index procedure until she suffered a right patellar fracture without extensor mechanism disruption approximately one year post-operatively. This was treated conservatively with immobilization and did well for a number of years until she presented to our clinic (nine years following her original TKA). During her initial evaluation at our institution, she was found to have generalized right knee pain for nine months and evidence of flexion instability on examination, as well as end-stage osteoarthritis of the left (contralateral) knee. Radiographs of the right knee revealed asymmetric medial polyethylene wear and tibial osteolysis (Fig. 1). She had no history of recurrent effusions or hemarthrosis in either knee. She underwent primary TKA of her left knee followed by right TKA revision 11 months later. The left primary TKA was uneventful and she went on to a successful outcome.

The revision right TKA was performed for flexion instability, polyethylene wear, osteolysis, and non-united patella fracture (Fig. 1). The knee was not aspirated prior to surgery because the index of suspicion for infection was low and serum inflammatory markers (sedimentation rate: 22 mm/h and C-reactive protein: <3.0 mg/dL) were both normal. The procedure was carried out using her previous incision and a standard medial parapatellar arthrotomy. Upon entering the joint, profuse and extensive synovial hypertrophy was encountered. Histologic examination revealed hyperplastic synovium with a papillary and multinodular proliferation of epithelioid mononuclear cells admixed with focal areas of hemosiderin-laden macrophages and scattered multinucleated giant cells (Fig. 2). There was significant wear of the polyethylene insert medially but minimal backside wear. Following removal of all prosthetic components, an extensive total synovectomy was completed (Fig. 3).

Following the synovectomy, a partial patellectomy with excision of the proximal pole was performed. The remaining patellar fragments were fixed with Dacron tape, imbricating the quadriceps tendon into good approximation with the patella. This was reinforced with non-absorbable interrupted sutures. The patella was resurfaced in the standard revision fashion by removing the prior implant, re-cutting the surface with a saw in a free hand fashion, and drilling for a new cemented implant [13]. The femoral and tibial components were revised to stemmed prostheses (NexGen, Zimmer Inc., Warsaw, IN) utilizing five millimeter distal augments on the femur, and a well-balanced result was obtained with adequate stability and range of motion (Fig. 4).

Eighteen months following revision arthroplasty, the patient had minimal pain, excellent range of motion (0 to 120°) and was very satisfied with her knee. She reported no episodes of swelling in the knee and

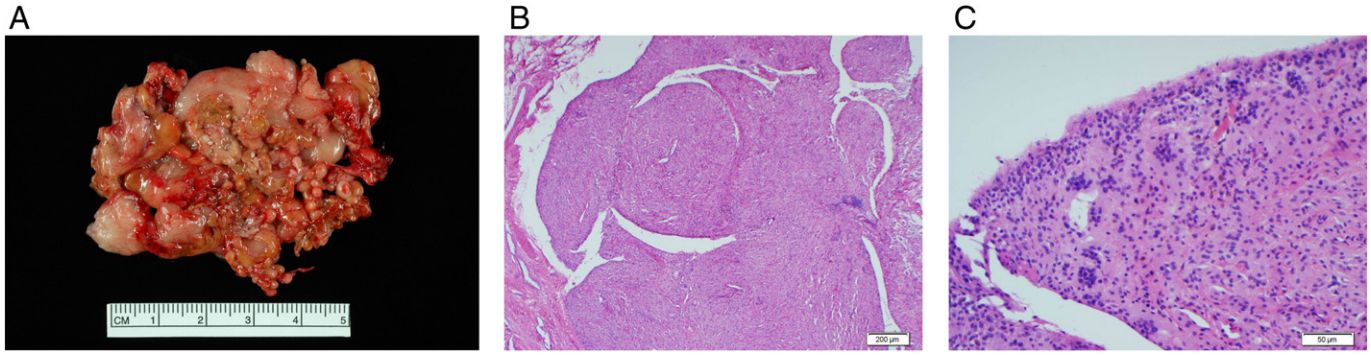

Fig. 2. Initial resected synovial specimen with PVNS. Grossly (a) the lesion consists of tan colored fibrous tissue with a firm, nodular appearance. Microscopically, there is a multinodular proliferation of hyperplastic synovium (b) which is composed of epithelioid mononuclear cells admixed with focal areas of hemosiderin-laden macrophages and scattered multinucleated giant cells (c).