

Recurrent Hemarthrosis of the Knee Mimicking Pigmented Villonodular Synovitis

Timoret Keren MD, Mark Shahmurov MD and David Hendel MD

Department of Orthopedic Surgery and Institute of Pathology, Wolfson Medical Center, Holon, Israel
Affiliated to Sackler Faculty of Medicine, Tel Aviv University, Ramat Aviv, Israel

Key words: knee, hemarthrosis, villonodular, synovitis

IMAJ 2005;7:50-51

Although most cases of hemarthrosis are post-traumatic, some patients present with hemarthrosis and no history of trauma. The approach to such a patient varies with age since the spectrum of differential diagnoses is completely different in the adult versus the pediatric age group. A complete medical history should be taken, along with laboratory and imaging studies, including arthrocentesis of the involved joint, which should be sent for complete laboratory evaluation, including blood count, chemistry testing and culture.

A relatively uncommon cause of recurrent hemarthrosis is pigmented villonodular synovitis, a primary benign tumor of the synovial membrane, characterized by extensive proliferation and the appearance of villi and nodules. The common presentation is local pain and tenderness, swelling of the synovium, and a brown or bloody effusion. The definitive diagnosis is reached by synovial biopsy. Treatment consists of arthroscopic or open synovectomy and excision of the nodules. The use of low dose beam radiation or intraarticular radioisotope injections has also been reported as supplemental treatment of pigmented villonodular synovitis [1]. Other possible causes of recurrent atraumatic hemarthrosis include benign and malignant tumors (such as hemangiomas, angiodysplasia, synovial chondromatosis, soft tissue sarcoma and multiple myeloma), coagulation disorders and drugs affecting the coagulation system, septic arthritis, nutritional disorders, and idiopathic causes [2].

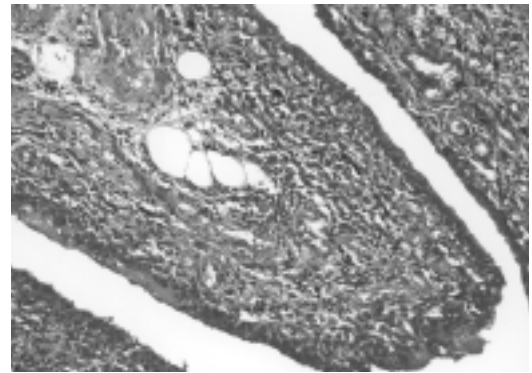
We report a case of an elderly man presenting with recurrent non-traumatic hemarthrosis of the knee treated by primary total knee arthroplasty. This unusual presentation of degenerative joint disease

raises questions about the differential diagnosis of non-traumatic recurrent hemarthrosis of the knee joint.

Patient Description

An 84 year old man was referred to the orthopedic clinic by his family physician. In the previous year he had visited the emergency room four times, each time with the same complaint – a spontaneous swelling of his left knee causing severe and disabling pain, with no history of trauma. The physical examination at all four visits demonstrated a swollen knee with a tense effusion and limited range of motion. A diagnostic arthrocentesis was performed in the emergency room. The patient's history revealed hypertension, prostatic carcinoma for which he underwent a prostatectomy several years previously, and peptic disease. His usual drug regimen consists of aspirin (100 mg/day), a calcium blocker, an alpha blocker and an H2 inhibitor.

Laboratory evaluation revealed a mild microcytic anemia, with no leukocytosis. C-reactive protein and erythrocyte sedimentation rates were normal. Prothrombin time, partial thromboplastin time and fibrinogen levels were within the normal range. A plain X-ray showed moderate joint space reduction, with subchondral sclerosis and osteophytes. He then underwent further investigation with magnetic resonance imaging of the left knee. The MRI scan showed a space-occupying lesion with two cystic components having a heterogeneous texture. Open surgery for complete synovectomy and total knee arthroplasty was performed. At surgery, abundant syno-



Microscopic view of the intraoperative left knee specimen, showing prominent chronic mononuclear inflammatory cell infiltrates rich in plasma cells and lymphocytes, with a tendency to form lymphocyte aggregates (frequently perivascular). There is marked proliferation of the synoviocytes and numerous villi formation, partially organizing fibrin deposits, recent and non-recent hemorrhages and diffuse hemosiderin deposits (stained by Prussian Blue stain).

vial tissue, which was edematous and erythematous with wide areas of brown pigmentation, was found. It penetrated the capsuloligamentous structures within the joint. The articular surfaces had extensive erosions, and the articular cartilage also had a brown pigmentation. Due to the destructive joint changes with subchondral invasion and weakened, loose and eroded capsuloligamentous structures, a constrained rotating hinged prosthesis was implanted.

A microscopic examination of the intraoperative specimen revealed severe degenerative changes with no evidence of tumor, which came as a surprise, contradicting the assumption based on the macroscopic features [Figure]. The patient was referred to a physical therapy clinic for rehabilitation and received no additional treatment. At 1 year follow-up, he was pain-free, with no effusion, and good functional capacity, walking with the aid

of a cane only and a wide range of motion, 0–120 degrees, in the operated knee.

Comment

This case raised several questions concerning the diagnostic and therapeutic approach to recurrent atraumatic hemarthrosis of the knee. The history of this patient, the suspicious MRI and the macroscopic appearance of the disease suggested a working diagnosis of pigmented villonodular synovitis. Due to extensive destructive joint changes, a wide synovial excision and total knee replacement were performed instead of the standard treatment in such cases – namely, an arthroscopic synovectomy, alone or in conjunction with adjuvant treatment modalities (such as cryosurgery, radiation therapy or intraarticular radioactive material injection) [3–5].

Postoperatively, the possibility of adjuvant treatment was contemplated. There

are no reports in the literature about adjuvant treatment for pigmented villonodular synovitis following arthroplasty. This therapeutic enigma was resolved when the results of the histologic examination became known a few days after the operation. Surprisingly, the features were consistent with severe chronic synovitis. No clear morphologic evidence of tumoral nature was seen. It is assumed that the recurrent hemarthrosis in this patient was the result of degenerative joint disease, in the presence of prolonged aspirin treatment and possible recurrent minor trauma. Therefore, the patient received no additional treatment. This case demonstrates an uncommon etiology for recurrent atraumatic hemarthrosis and an unusual clinical presentation of osteoarthritis of the knee.

References

1. Chin KR, Barr JS, Winalski C, Zurakowski D, Brick GW. Treatment of advanced primary and recurrent diffuse pigmented villonodular synovitis of the knee. *J Bone Joint Surg (Am)* 2002;84:2192–202.
2. Cunningham RB, Mariana EM. Spontaneous hemarthrosis 6 years after total knee arthroplasty. *J Arthroplasty* 2001;16(1):133–5.
3. Hamlin BR, Duffy GP, Trousdale RT, Morrey BF. Total knee arthroplasty in patients who have pigmented villonodular synovitis. *J Bone Joint Surg (Am)* 1998;80:76–82.
4. Blanco CE, Leon HO, Guthrie TB. Combined partial arthroscopic synovectomy and radiation therapy for diffuse pigmented villonodular synovitis of the knee. *Arthroscopy* 2001;17(5):527–31.
5. Shabat S, Kollander Y, Merimsky O, et al. The use of surgery and yttrium 90 in the management of extensive and diffuse pigmented villonodular synovitis of large joints. *Rheumatology (Oxford)* 2002;41(10):1113–18.

Correspondence: Dr. T. Keren, Dept. of Orthopedics, Wolfson Medical Center, Holon 58100, Israel.
Phone: (972-3) 502-8383
email: timik@bezeqint.net

Capsule

Reducing plaques in atherosclerosis

Atherosclerosis is the most common cardiovascular disease in Europe and North America. The c-jun-NH2-terminal kinase (Jnk) family is implicated in atherogenesis. Ricci et al. addressed the function of JNK in atherogenesis, using atherosclerosis-prone apolipoprotein E (ApoE)-deficient mice simultaneously lacking either Jnk1 or Jnk2. Jnk2 deletion strikingly reduced plaque formation in ApoE-deficient mice. However, deletion of Jnk1

revealed only a slight effect on atheroma formation. Pharmacologic inhibition of overall Jnk activity substantially suppressed atherosclerosis in ApoE-deficient mice. Specific inhibition of JNK2 activity may thus represent a therapeutic approach to ameliorate atherosclerosis.

Science 2004;306:1558

E. Israeli

Capsule

Emergence of attenuated West Nile virus variants in Texas

In order to understand how West Nile virus (WNV) has evolved since its introduction into North America, Devisa et al. studied the genetic and phenotypic variation among WNV isolates collected in various areas during consecutive transmission seasons. The report describes, for the first time, phenotypic changes occurring in the North American WNV population. Several isolates collected in Texas during 2003 display a small plaque (sp) and temperature-sensitive (ts) phenotype, as well as reduced replication in cell culture, in comparison to isolates collected in 2002 and New York in 1999. Studies of mouse

neuroinvasiveness/neurovirulence also indicate that several of these isolates were attenuated in neuroinvasiveness, but not for neurovirulence. The complete genome and deduced amino acid sequences of several of these isolates have been determined in order to map the mutations responsible for this phenotypic variation. These data indicate microevolution of WNV and the emergence of isolates exhibiting phenotypic variation.

Virology 2004;330:342

E. Israeli