

One step open synovectomy without adjuvant therapy for diffuse pigmented villonodular synovitis of the knee in a soccer player

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Summary

Pigmented villonodular synovitis is a proliferative disease involving joints, bursas and tendon sheaths with typical histological changes in the synovial tissue. A local and diffuse form are described. Aetiology is uncertain, MRI is helpful to establish the diagnosis, which is confirmed with by biopsy. Treatment is based on the principles of tumor resection: arthroscopically in the local form, or by a open synovectomy in the diffuse form, often followed by adjuvant radiotherapy. The rate of recurrence is high, but differs in consideration of the treatment chosen. We report a 35 year old soccer player with diffuse pigmented villonodular synovitis with a history of chronic swelling knee. The patient underwent a one step open synovectomy without adjuvant therapy, with the conservation of the heads of gastrocnemius muscle. At five years of follow-up, the patient had no sign of recurrence of the condition. One step open synovectomy in this patient showed an excellent outcome with the return to his previous sport. The short follow-up and the neoplastic characteristic of the disease cannot exclude the risk of recurrence.

Key Words: Diffuse pigmented villonodular synovitis, Open sinovectomy, Soccer.

Introduction

Pigmented villonodular synovitis (PVNS) is a proliferative disease of the synovial joints and usually involves bursas and tendon sheaths, with an annual incidence of 1.8 pa-

tients *per million per year* in the population. Rare in children, it has the highest prevalence between the second and fourth decade. There is a slight predominance of male patients (1,2). The first case was described by Chassaignac in 1852, later by Moser, while Jaffe *et al.* introduced the term of pigmented villonodular synovitis (PVNS) in 1941 (3-5). PVNS was firstly related to inflammation, hemarthrosis and repetitive trauma. Genetic investigations suggest a clonal expression of neoplastic proliferations (6-8). Host factors, such as cellular immunity, might be associated with the genetics factors (9). Histological features are: villous processes; proliferating synovial cells; deposits of hemosiderin and inflammatory infiltrate (1,10). PVNS has been classified as isolated or local and diffuse forms, usually monoarticular. The knee is the most commonly involved joint, followed by the hip, in the diffuse form; the ankle and the shoulder, in the local presentation. Clinical symptoms are not specific, and may vary according to the joint involvement; pain, swelling, joint effusion, limitation of the range of motion, joint locking with stiffness are the main features. The diagnosis is made usually after a joint aspiration, where the characteristic brown synovial fluid (hemosiderin deposits) should be detected. After that, a synovial biopsy is necessary (6). MRI is useful to detect intra-articular synovial nodules, and to exclude the diffuse form of PVNS to perform a preoperative planning. Arthroscopic synovectomy generally is indicated in the local form while open synovectomy, usually in two steps, is performed in the diffused form in order to obtain a complete excision of the PVNS (11-14). PVNS is a benign neoplasia with a higher rate of recurrence, up to 46% , especially in the diffused form, therefore several authors recommend adjuvant therapies after the complete surgical excision as intra-articular radiation, extra-beam radiation, or anti-inflammatory molecules (11,15). Joint arthroplasty is reserved for severe joint involvement cases and multiple recurrences.

Case presentation

A 35 year-old man soccer player presented with a one year chronic swelling and increasing pain of the right knee. At the first clinical examination, the knee was painful, there was joint effusion and palpable masses on the anterior and posterior aspect of the knee, the range of motion was limited to 100° of flexion and 10° of extension, clinical test for ligaments or menisci injuries were negative. Radiographs were reported as normal, MRI showed hypertrophic synovia with joint effusion in the anterior aspect of the joint and in the popliteal fossa (Fig. 1). A posterior knee aspiration was performed and 200 mL of synovial brown effusion was aspirated. (Fig. 2) After that, we proposed to the patient to perform an open anterior and posterior synovectomy in a one step procedure, based on the standard method described by Kingsley *et al.* (13).

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Figure 1 - Preoperative sagittal and axial T1 weighted MRI images of the knee showing the diffuse involvement of the synovial tissue with the anterior and posterior joint effusion.

The patient was firstly placed in the prone position and a thigh tourniquet was used. An S-shaped incision was made with the exposure of popliteal space without the release of the heads of gastrocnemius muscle; the neurovascular structures of the popliteal fossa were compressed and displaced laterally by the pathologic tissue. The posterior capsule was dissected, and the intercondylar notch, posterior horns of the menisci and PCL were exposed. The pathologic synovial tissue was identified and excised. Absorbable sutures for the capsule and surrounding tissues were used after the release of the ischemic fascia to control bleeding (Fig. 3).

The patient was then placed supine, and a thigh tourniquet was used. A 16 cm median skin incision and medial peripatellar capsulotomy was performed. The anterior pathological synovial tissue was accurately dissected. Brown nodules of PVNS around the ACL, PCL and posterior aspect of the Hoffa body were removed (Fig. 4). The fascia and subcutaneous tissues were closed with absorbable sutures. No drainage was used, and a compressive bandage was maintained for two weeks. Paracetamol and morphine were used when necessary for post-operative pain control. No adjuvant therapy, intra-articular radiation or extra beam radiation were performed given patient refusal. The patient was discharged after 48 hours. Full weight bearing was allowed from the day after the operation with crutches. Physical therapy after 48 hours and was continued for 8 weeks consisting in isometric reinforcement of periarticular muscles and passive ROM. At 8 weeks, the patient presented full ROM. Home physiotherapy was undertaken. At 10 weeks, the patient returns to run and at 16 weeks to play soccer. The histological exam of the resected pathological tissue (Fig. 5) confirmed the diagnosis of intra and extra articular PVNS.

Physical examination were performed monthly for one year. After that clinical control were delayed every three months in the second year and an MRI was prescribed every year to detect eventual postoperative recurrences.

Discussion

Surgical excision is currently considered the best treatment with the oncological principles, wide resection or marginal resection in conjunction with an adjuvant therapy (13,16). The risk of an extensive open surgical procedures with post-operative complications such as joint stiffness,



Figure 2 - Visible posterior mass in the knee popliteal fossa during the aspiration of a yellow brownish liquid.

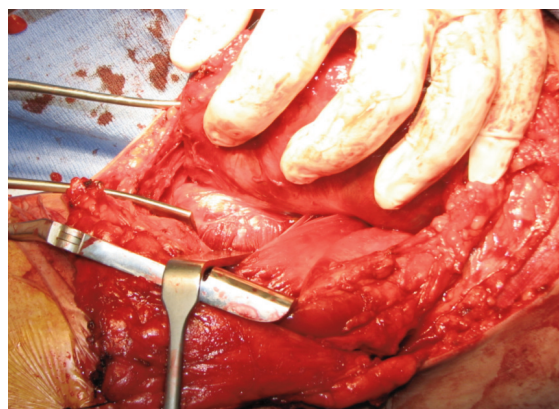


Figure 3 - Exposition of the knee popliteal fossa. Pigmented villonodular tissue is removed without the release of the heads of gastrocnemius muscle.

infection, reduced functional performance has led many authors to try arthroscopic removal with varying success in literature (16). Open synovectomy in two steps, with anterior and posterior approaches, usually with the release of the heads of the gastrocnemius muscle to allow a wide

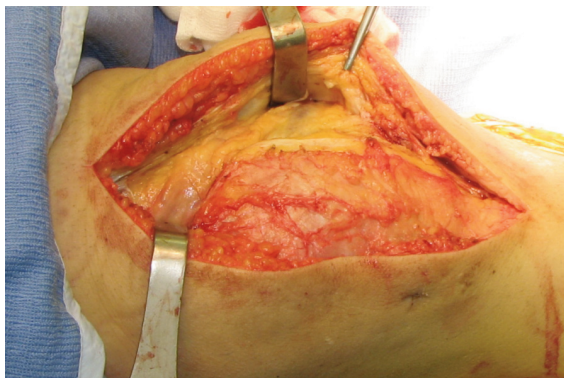


Figure 4 - Median skin approach and peripatellar capsulectomy in the way to remove widely the anterior pathological knee synovia.



Figure 5 - Characteristic macroscopic samples of the pigmented villonodular tissues removed from the anterior and posterior knee joint.

exposure of the joint with adjuvant radiotherapy, seems to be the gold standard (13). In this patient, we decided to undertake a wide resection with an open synovectomy in one step without the release of the heads of gastrocnemius muscles. This surgical procedure has avoided to compromise the functional request of this athlete, with no limitations in the exposure of both compartment of the knee, as we were able to access the whole of the popliteal fos-

sa and intercondylar notch removing the neoplastic tissue. Adjuvant radiotherapy was not performed due the patient refusal. Considering the high rate of recurrence, varying from 8 % of Flandry and Hughston to 46 % of Byers *et al.* (11,15), radical excision of the abnormal tissue must be performed. Schwartz showed that incomplete surgical synovectomy were related significantly to a higher recurrence rate (17). Radiotherapy has been used to prevent the risk of recurrence of the disease after surgical excision, with low dose of radiation or intra-articular injection of radioactive material immediately after the operation or in the case of a recurrence. O' Sullivan *et al.* (18) in 41 patients and Kotwal *et al.* (19) in 48 patients did not observe severe radiotherapy-related complications, even though several cases of periarticular fibrosis and radiation induced sarcoma have been reported in literature. De Ponti *et al.* showed, in 15 patients affected by the diffuse form of PVNS, better clinical results and lower rate of the recurrence in the 8 patients treated with extended synovectomy (20). Recently Chin and Brick reevaluated a group of 38 consecutive patients with persistent extraarticular diffuse pigmented villonodular synovitis of the knee after arthroscopic synovectomy. The authors concluded that, although arthroscopic synovectomy offered a short-term relief, the long term outcome is poor (21).

Early and intense rehabilitation program permitted a good functional outcome with the patient able to return to his previous sport level. At a five year follow up, the MRI (Fig. 6) shows no recurrence, and the patient is still playing soccer without complaining any knee stiffness, swelling or pain. There are reports of excellent results in patient treated surgically with no adjuvant radiotherapy. Radiotherapy seems unnecessary when total and wide synovectomy is performed, and it is reserved to patients with recurrences or those who had partial synovectomy (15). Our open synovectomy approach permitted a wide resection of the neoplastic tissue, and an excellent functional result. We advocate a multicentre randomized study which may be able to clarify whether adjuvant radiotherapy is the best post-surgical treatment for local and diffuse PVNS.

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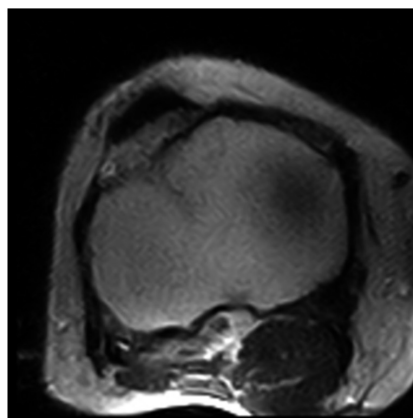
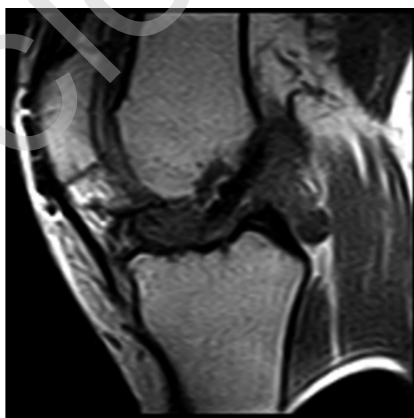


Figure 6 - Sagittal and axial T1 weighted MRI images of the knee after five years of follow-up showing no signs of recurrence.

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