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Research Article

Evaluation of patients with diffuse pigmented villonodular synovitis of knee with functional scoring systems after surgery and radiosynovectomy

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Abstract

Aim: To evaluate the symptoms and functional status of patients with pigmented villonodular synovitis (PVNS) of the knee using the modified Marshall scoring system and musculoskeletal tumor society (MSTS) rating scale before therapy and six months after radiosynovectomy (RS).

Materials and methods: Evaluation was made of 29 knee joints of 29 patients with DPVNS. Arthroscopic synovectomy was applied to all patients and additional posterior open synovectomy to five. Yittrium-90 (Y-90) citrate colloid was used in the RS procedure. RS was performed 11.8 ± 13.3 weeks after the operation. In the six months after RS, patients were evaluated with the modified Marshall scoring system and MSTS rating scale.

Results: The patients were 17 females and 12 males with a mean age of 31.8 ± 12.9 years (range: 13-54 years). The preoperative modified Marshall score was poor in 24 patients and fair in five patients. In the follow-up period, the scores were excellent in 17 patients, good in 10 patients and fair in two patients. The mean modified Marshall scores increased after the operation and RS from 12.93 ± 2.93 to 25.52 ± 2.90 ($p<0.001$).The mean MSTS scores increased from 17.52 ± 4.82 to 26.86 ± 2.46 ($p<0.001$). In one of four patients with a score of 22 and three patients with a score of 24, maximum scores were obtained in the follow-up period with the effect of surgery and RS.

Conclusion: RS with Y-90 citrate colloid after surgical excision in the treatment of PVNS is a reliable and efficient treatment method for the increase of functional capacity and decrease of symptoms.

Introduction

Pigmented villonodular synovitis (PVNS) is a rare proliferative disease of the synovium with locally aggressive behaviour and high relapse rates after surgical removal [1]. The etiology of PVNS is unknown but the possible mechanism is thought to be associated with reactive process, repetitive microtraumatic or inflammatory events with synovial hemosiderin deposition [2]. Histology reveals hypertrophic synovium with villous, nodular and villonodular proliferation and pigmentation from hemosiderin [3]. PVNS is a benign and destructive disease which results in major symptoms and loss of function leading to amputation [4]. The clinical symptoms are mostly chronic, and years may pass from the first symptoms to medical presentation. In the advanced stages, a synovial mass may invade the chondro-osseous junction with joint destruction [5]. PVNS of the knee has been reported to have a higher recurrence rate when compared with PVNS of other joints [6]. Surgical excision is the treatment of choice with wide excision in localised disease and total synovectomy in the diffuse form [7].

Radiotherapy or intra-articular isotope injection, known as radiosynovectomy may be necessary after tumor resection. Radionuclide emits β-particles which are injected into the articular cavity. Synovial lining cells phagocytise the colloidal particles. Active irradiation with β-particles of the surrounding synovial tissues causes fibrosis and sclerosis of the synovial membrane [8]. Radionuclide choice depends on the synovial thickness in the treated joint and the penetration rate of the β-particles. Y-90 is used in the knee joint due to the high mean tissue penetration of 3.6 mm [9]. Yittrium-90 emits β-particles of 2.27 MeV with a physical half life of 64 hours [10].

In this study, the symptoms and functional status were evaluated of patients with PVNS of the knee using the modified Marshall scoring system and musculoskeletal tumor society (MSTS) rating scale before therapy and six months after RS.

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Materials and methods

Evaluaton was made of 29 knee joints of 29 patients with a diagnosis of histopathological diffuse PVNS. The patients were 17 females and 12 males with a mean age of 31.8 ± 12.9 years (range 13-54 years). All patients underwent arthroscopic surgery and posterior open synovectomy for posterior extra-articular masses was also applied to five patients. RS was performed at 11.8 ± 13.3 weeks postoperatively. Written informed consent was obtained from all patients or the parents of those aged under 18 years.

Procedure of arthroscopic synovectomy

Pneumatic tourniquet with 300 mm Hg was used to achieve bloodless field for arthroscopic synovectomy. Anteromedial, anterolateral, suprapatellar medial and lateral portals, posteromedial, posterolateral and transpatellar tendon portal were used for near-complete arthroscopic synovectomy. Cryo-cuff was used to decrease pain and inflammation and suction drain was kept in the joint for 48 hours. Home exercises were advised for strengthening quadricaps and range of motion.

Procedure of radiosynovectomy

RS was performed with 185 mBq (5 mCi) Y-90 citrate colloid (CIS Bio International, France) under aseptic conditions. In cases of excess effusion in the joint, fluid was drained before the Y-90 citrate colloid injection. After injection of Y-90 citrate colloid, another syringe with prilocaine (Citanest, AstraZeneca, London, UK) was injected into the knee joint to provide analgesia and to prevent extra-articular leakage of radionuclide. After two minutes of manual pressure to the puncture site, passive flexion-extension of the knee was applied to achieve homogenous distribution of the radionuclide in the joint space. At four and 48-72 hours after RS, whole body and static imaging using Bremsstrahlung settings with General Electric (GE) Millennium MG (Germany) was taken to assess the distribution and leakage of radionuclide in the knee joint. The joint was immobilized for 48-72 hours with a splint to minimize extra-articular leakage. Patients were warned to avoid excessive physical activity for two weeks.

Preoperative and follow-up evaluation

Preoperatively and at six months after RS, the functional results were assessed with the modified Marshall scoring system with a maximum score of 30 points. Below 16 points was evaluated as a poor functional result, 16-20 points as fair, 21-25 points as good and 26-30 points as excellent [11,12] (Table 1). The MSTS rating scale was also applied. This scale has six categories of pain, function, emotional acceptance, support, walking ability and gait with each category rated on a scale of 0-5, where 0 represents poor function and 5 normal functions. The maximum possible score is 30 points [13].

Statistical analysis

Data obtained from the application of the modified Marshall scoring system and MSTS rating scale were analysed using Statistical Package for Social Sciences (SPSS for Windows 16.0). The scores obtained preoperatively and at six months after RS in the Modified Marshall and MSTS scales were compared with the paired sample *t* test.

Results

Bremsstrahlung imaging revealed an even distribution of radionuclide in all the knee joints and no evidence of extra-articular leakage of radioactivity (Figure 1).

Table 1. Modified Marshall knee scoring scale.

Criteria	Points
Subjective	
Pain	0=yes, 1=no
Swelling	0, 1
stair difficulty	0, 1
Clicking, numbness	0, 1
Giving way	0-4
Normal	4
With Athletic Activity Only	2
With Stress Upon ADL	1
Regularly Upon ADL	0
Return to sports or work	0-3
Return, no limitations	3
Return, some limitations	2
Change in occupation	1
Cannot work	0
Objective	
Functional tests	
Duck Walk	0, 1, 2
Run in place	0, 1
Jump one leg	0, 1,, 2
Half squat	0, 1,
Full squat	0, 1,
Specific knee examination	
Tenderness	0, 1
Joint effusion	0, 1
Swelling (soft tissue)	0, 1
Crepitation	0, 1
Muscle power	0-3
Normal (5)	3
Mild weakness (4+, 4)	2
Moderate weakness (4-, 3+)	1
Severe weakness (3 or less)	0
Thigh sizes	0-2
Equal	2
1-2 cm difference	1
>2 cm difference	0
Range of motion	0-3
Normal	3
5 degree extension lossand/or 10 degree flexion loss	2
10 degree extension loss and/or 20 degree flexion loss	1
>10 degree extension loss and/or >20 degree flexion loss	0
Total points	30

ADL: Activities of daily living, 2 = performance without discomfort, 1= performance with discomfort, 0 = cannot perform

The patients were 17 females and 12 males with a mean age of 31.8 ± 12.9 years (range: 13-54 years). The preoperative modified Marshall score was determined as poor in 24 patients and fair in five patients. No patient had a preoperative score greater than 19. In the follow-up period, the modified Marshall score was evaluated as excellent in 17 patients, good in 10 patients and fair in two patients. No patient had a functional score of less than 16 at six months after RS. Of the 24 patients with a poor preoperative functional score, two were evaluated as fair, nine as good and 13 as excellent in the follow-up period. The modified Marshall scores were 12.93 ± 2.93 (range: 6-19) preoperatively and 25.52 ± 2.90 (range: 16-29) at six months after RS. The increase in the modified Marshall scores were statistically significant ($p<0.001$) (Table 2).

MSTS scores were 17.52 ± 4.82 (range: 8-24) preoperatively and 26.86 ± 2.46 (range: 20-30) at six months after RS The increase in the MSTS scores were statistically significant ($p<0.001$). In one of four patients with a preoperative score of 22 and three patients with a score

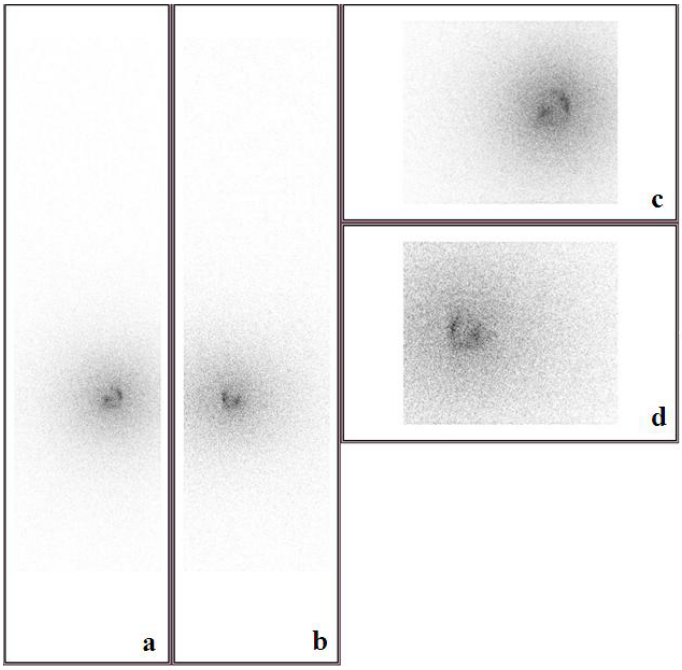

Figure 1. Distribution of radionuclide in all the knee joints.

Table 2. Distribution of preoperative and follow-up functional scores (p<0.001).

	Preoperative	Follow-up
Poor score	24	0
Fair score	5	2
Good score	0	10
Excellent score	0	17

of 24, maximum scores were obtained in the follow-up period with the effect of surgery and RS.

Discussion

PVNS typically occurs before the age of 40 years, presents with pain, swelling and dysfunction in the joints and the knee is the most involved joint [14]. Recurrence rates of PVNS increase with time and as time passes, it becomes increasingly difficult to cure. The quality of life decreases for PVNS patients when compared with the general population [15]. Surgery may not remove all the tissue and RS should be discussed to remove the residue of the synovial membrane in 6-8 weeks after the operation [16]. The use of RS has been shown to be effective in some diseases that involve joints. Improvement rates after RS have been reported as 66.7% in rheumatoid arthritis, 56% in osteoarthritis, and 77.3% in PVNS. Reduction in joint bleeding and factor usage has been shown to be evident in 91% of cases of haemophilia and Willebrand's disease. Treatment failure is especially seen in deformed and unstable joints [17].

Wu *et al.* applied anterior and posterior open synovectomies with external radiation therapy. The mean knee rating and functional rating improved from 65.2 and 70 preoperatively to 93.7 and 96.6 postoperatively. Of nine patients in the series, eight were disease free and one had local recurrence [18]. Nassar *et al.* used external beam radiotherapy after debulking surgery and reported no disease recurrence or progression of bone or articular destruction in any patient, nor any complication related to the surgical procedure or radiotherapy [19].

Oztemur *et al.* applied Y-90 RSV to 17 knees of 17 patients for the treatment of resistant chronic non-specific synovitis and evaluated the efficacy of RSV with the Lysholm score. The combination of RSV with surgical synovectomy was reported to be an effective and reliable treatment method in repetitive, chronic, non-specific synovitis [20]. De Carvalho Jr *et al.* applied external beam radiotherapy after subtotal arthroscopic and open synovectomy and had a recurrence rate of 12.5% at 8.6 years of follow-up for the treatment of diffuse PVNS of the knee joint [21]. Heyd *et al.* suggested the use of radiation therapy in the postoperative setting after incomplete resection and for the treatment of recurrences [5].

Ozturk *et al.* applied the combination of debulking surgery with Y-90 RSV to a small group of seven patients. Synovial thickness disappeared and joint effusion decreased [22]. Zook *et al.* administered 11 injections of P-32 to nine patients, eight of whom had PVNS in the knee and one with involvement of the hip with an overall control rate of 70% [14]. Franssen *et al.* performed Y-90 RSV to diffuse PVNS and recommended Y-90 as an alternative treatment without previous surgical synovectomy or for cases with failure of previous surgical synovectomy [23]. Koca *et al.* applied Y-90 citrate to 15 knee joints and the MRI examination showed no progression, as two cases were seen to be stable, regression was seen in nine and total cure in four cases. The Lysholm score increased from 39 before surgical excision to 83 at the final follow-up examination (24). Shabat *et al.* combined Y-90 RSV and traditional open synovectomy in 10 patients with six knees, three ankles and one hip joint. In one patient, disease was stable with no further joint damage and nine patients had no local recurrence and no progressive bone or joint destruction [25].

Jahangier *et al.* applied second RSV at a double dose when the initial response was inadequate, but the second application with double dose did not contribute to a better result. It was reported that persistent synovitis can be treated with a success rate of 75% in the first month, but prolonged remission was only achieved in 29% of joints [26]. Kamaleshwaran *et al.* used 370 mBq (10 mCi) Y-90 as a primary treatment in the knee joint with diffuse PVNS and obtained pain relief, a decrease in joint swelling and an increase in joint mobility [27].

In the current study, the modified Marshall and MSTS scores increased with the therapeutic effect of the surgery and RS in patients with diffuse PVNS. RS should be applied for an increase in functional capacity and a decrease in symptoms.

Conflict of interest

The authors state that there is no conflict of interest in this paper.

Acknowledgement

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