Simultaneous Anterior and Posterior Synovectomies for Treating Diffuse Pigmented Villonodular Synovitis

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Abstract

Background The current treatment methods for diffuse intraarticular or extraarticular type pigmented villonodular synovitis (PVNS) include arthroscopic synovectomy or staged anterior and posterior open synovectomies. However, it is unclear whether simultaneous anterior and posterior synovectomies achieve local control and recovery of function.

Questions/purposes We therefore determined the recurrence rate and function in patients with diffuse PVNS treated with anterior and posterior synovectomies and adjuvant radiotherapy.

Methods We retrospectively reviewed all 19 patients with diffuse PVNS involving the knee treated with anterior and posterior synovectomies and adjuvant radiotherapy between January 2001 and November 2007. From the records, we determined local recurrence and Tegner-Lysholm scores. The minimum followup was 42 months (median, 98 months; range, 42–143 months).

Results Postoperative MRI revealed residual tumor in five of the 19 patients, although three had no disease progression during followup and had knee scores of 86 to 90. Two patients had recurrences at 6 and 9 months with knee scores of 88 at 42 months and 90 at 68 months. The mean Tegner-Lysholm knee score improved from 59 to 93 points. Mean maximum extension and flexion angles improved from 11° to 2° and from 76° to 127°, respectively.

Conclusions Compared with the literature, simultaneous anterior and posterior synovectomies associated with postoperative radiotherapy provided rates of residual or recurrent tumor and knee function recovery comparable to that with staged synovectomies reported in the literature.

Level of Evidence Level IV, therapeutic study. See Guidelines for Authors for a complete description of levels of evidence.

Each author certifies that he or she has no commercial associations (eg, consultancies, stock ownership, equity interest, patent/licensing arrangements, etc).

All ICMJE Conflict of Interest Forms for authors and *Clinical Orthopaedics and Related Research* editors and board members are on file with the publication and can be viewed on request. Each author certifies that his or her institution approved the human protocol for this investigation, that all investigations were conducted in conformity with ethical principles of research, and that informed consent for participation in the study was obtained. This study was performed at Taipei Veteran General Hospital, Taipei City, Taiwan, ROC.

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Introduction

Pigmented villonodular synovitis (PVNS) is an uncommon disease affecting the synovial lining of joints, bursae, and tendon sheaths. The prevalence has been estimated at 1.8 cases for each million in the population [19]. The most commonly affected individuals are reportedly in their 30s and 40s [6]. Most cases that occur have been monoarticular, and although rare, polyarticular PVNS has been reported [10]. According to one report, the most commonly

involved joint has been the knee, followed by the hip and ankle [18]. Most cases of PVNS are intraarticular whereas extraarticular extension is extremely rare. However, if extraarticular PVNS is present, it will tend to occur in the same joint locations as the intraarticular PVNS [26]. Two types of PVNS were distinguished in 1967 [14]: localized and diffuse. The diffuse type is reportedly 3.1 times more common than the localized type [9, 19].

For patients with localized PVNS, arthroscopic synovectomy reportedly reduces morbidity compared with open synovectomy [12, 29]. However, the best treatment for diffuse PVNS is controversial. Open surgical excision has been the primary method for treating diffuse PVNS [3, 8, 9, 23, 31–33]. Arthroscopic synovectomy reportedly has resulted in smaller incisions and reduced morbidity [29], but owing to reported recurrence rates as much as 46% [6], some have recommended open synovectomy [6, 8, 32]. There are several other disadvantages to treating patients with diffuse PVNS using arthroscopic synovectomy. First, PVNS has the potential to extensively invade adjacent extraarticular structures, such as muscles, tendons, and bones, especially in patients with diffuse PVNS [25]. It is difficult to access PVNS tissue using an arthroscope, especially along the popliteus tendon sheath, beneath the origin of the gastrocnemius muscle, and above the posterior femoral condyle [6]. Second, as patients frequently present with nonspecific symptoms, there is often a long period between the onset of symptoms and accurate diagnosis (average, 16 months to 5 years) [9, 27]. Therefore, there is the potential for local spreading during these long periods. Finally, it theoretically is possible that arthroscopy results in secondary seeding because of limited surgical view and the arthroscopic irrigation system [6].

In patients with diffuse PVNS, some authors have recommended staged anterior and posterior synovectomies [21, 32]. The recurrence rate associated with such treatment ranges from 14% to 56% [32, 33], although the absence of postoperative MRI studies may have underestimated the number of recurrences. Given the concerns with arthroscopic synovectomies, we presumed open anterior and posterior total or subtotal synovectomies were the best treatment for diffuse PVNS. However, total synovectomy is difficult to perform and may compromise the nerves and vessels adjacent to the PVNS tissue; it sometimes could be difficult to perform total synovectomy. Further, one report suggests total synovectomy may increase the risk of subsequent osteoarthritis [31]. Nonresectable PVNS tissue also may be controlled using adjuvant therapy, such as with intraarticular instillation of radioactive isotopes (ie, yttrium-90 [13, 16, 28] or dysprosium-165 [5]) or low-dose external beam radiation [3, 5, 17]. However, radiotherapy has been associated with side effects. For example, intraarticular injection of yttrium-90

has been associated with radionecrosis of the soft tissue [24] and systemic leakage of the radioactivity with the potential to cause infertility [7, 24]. To reduce concerns regarding side effects, we used low-dose external beam radiation (30 Gy in 10 fractions). However, it is unclear whether simultaneous anterior and posterior synovectomies combined with postoperative radiotherapy improve joint function and prevent local recurrences.

We therefore determined (1) local disease control, (2) the improvement of joint function, (3) the influence of joint function for patients with residual or recurrent disease, and (4) the complications of simultaneous anterior and posterior synovectomies with adjuvant radiotherapy in patients with diffuse extraarticular PVNS of the knee.

Patients and Methods

Between January 2001 and November 2007, we retrospectively reviewed all 19 patients with diffuse extraarticular PVNS involving the knee treated with simultaneous anterior and posterior synovectomies and adjuvant radiotherapy. The indications for simultaneous anterior and posterior synovectomies and adjuvant radiotherapy were (1) limiting knee symptoms including pain and swelling and (2) patient dissatisfaction with knee function. We had no absolute contraindications specifically related to this type of surgery. During this period, we treated all patients meeting the criteria in the same manner. Nine patients were men and 10 were women, and their mean age was 43.4 years (range, 29–59 years). The minimum followup was 42 months (median 98 months; range, 42–143 months). No patients were lost to followup. No patients were recalled specifically for this study; all data were obtained from medical records and imaging. All of the patients provided informed consent for the treatment. We had prior approval of our institutional ethical committee for the study.

The most common signs and symptoms of PVNS were knee pain and joint effusion (Table 1). The mean duration from the onset of symptoms until accurate diagnosis was 12.7 months (range, 4–36 months) (Table 2). We recorded the prior diagnoses of patients referred to our institution (Table 3). Three patients had been misdiagnosed with

Table 1. Patients' symptoms and signs

Symptom/sign	Number of patients
Pain	17
Joint effusion	19
Locking	4
Instability	1
Popliteal mass	3

Table 2. Patient demographics and outcomes

Patient number	Age (years)	Sex	Duration of symptoms (months)	Previous surgery	Preoperative functional score*	Postoperative functional score*	Preoperative ROM (degrees)	Postoperative ROM (degrees)	Recurrence	Residual tumor	Followup (months)
1	59	Male	20	No	86	97	10-120	0-130	No	No	130
2	50	Female	36	No	86	94	0-110	0-135	No	No	52
3	54	Female	6	No	71	95	10-90	0-125	No	No	80
4	29	Male	4	No	69	97	0-20	0-130	No	No	143
5	36	Male	8	Excision (anterior)	11	87	10-50	5-120	No	Yes	116
6	34	Male	4	ACL reconstruction	88	88	20-80	10-110	Yes	Yes	68
7	56	Female	18	Excision (posterior)	40	86	15-70	5-120	No	Yes	119
8	57	Male	10	No	84	95	10-100	0-130	No	No	55
9	31	Female	6	No	25	95	10-50	0-135	No	No	120
10	56	Male	12	No	54	100	10-50	0-135	No	No	90
11	35	Female	10	No	53	90	10-70	5-120	No	Yes	96
12	32	Female	15	No	52	94	15-90	0-135	No	No	108
13	36	Female	13	No	72	95	5-65	0-135	No	No	84
14	45	Male	14	No	54	90	15-120	0-135	No	No	124
15	40	Male	8	No	52	90	15-50	5-120	No	No	110
16	38	Female	16	No	66	94	15-70	0-120	No	No	60
17	49	Female	13	No	51	90	20-90	10-110	Yes	Yes	42
18	46	Male	14	No	53	99	10-80	0-135	No	No	98
19	42	Female	10	No	58	95	10-75	0-135	No	No	102

ACL = anterior cruciate ligament; * Tegner-Lysholm knee score.

Table 3. Prior diagnoses

Diagnosis	Number of patients
Sarcoma	3
ACL injury	1
Meniscus tear	3
Rheumatoid arthritis	4
No specific diagnosis	8

ACL = anterior cruciate ligament.

sarcoma involving the popliteal area. Eight patients without definitive diagnoses had been treated with NSAIDs. Pathologic analysis of tissue specimens obtained from all of the patients revealed gross and histologic evidence of PVNS. Two patients (Patients 5 and 7) were referred to our institution for treatment of local recurrence of diffuse PVNS at 6 and 10 months postoperatively, respectively. The previous surgeons performed only an anterior approach to excise the anterior lesion but left the posterior lesion. Neither of these two patients received adjuvant radiotherapy in the previous hospitals. The third patient (Patient 6) who underwent prior surgery presented with knee pain and instability. Anterior cruciate ligament reconstruction and partial meniscectomy were performed at another hospital. However, painful swelling of the same knee developed 4 months later, so he came to our institution for treatment. All patients had preoperative MRI to determine the extent of the disease. Localization of the disease was determined by T2-weighted images.

All surgery was performed by one surgeon (WMC). The patient was in the supine position during surgery. After tourniquet inflation, the surgeon performed an anterior midline longitudinal skin incision and standard medial parapatellar arthrotomy. He excised as much PVNS tissue, including the medial gutter, lateral gutter, intercondylar notch, and suprapatellar pouch, as possible. A closed suction drain then was placed and the wound was closed in layers. For the posterior approach, the patient was in the prone position. After tourniquet inflation, a lazy s-shaped incision was made across the popliteal fossa. The medial head of the gastrocnemius muscle was identified and gently retracted laterally along with the neurovascular bundle. The surgeon fixed the knee at approximately 30° to reduce the tension of the neurovascular bundle, then excised the extracapsular PVNS tissue first, and longitudinally incised the posterior joint capsule to facilitate removal of all accessible PVNS tissue located in the posterior knee. Another drain tube was inserted and the wound was closed in layers. The mean tourniquet time was 41 minutes for the anterior procedure and 68 minutes for the posterior procedure.

Ankle pump exercise was encouraged immediately after the surgery to prevent deep vein thrombosis. All patients

started intermittent passive motion of the knee on the second postoperative day. We ensured patients achieved extension to 0° and flexion to at least 110° before they were discharged from the hospital. The mean hospital stay was 8.4 days (range, 7–13 days).

External beam irradiation with a total dose of 30 Gy in 15 fractions during 3 weeks (five times a week) was initiated for all patients 4 weeks postoperatively. We used the preoperative MRI for planning for irradiation, and determined the planning target volume margin to be 5 mm [15].

We followed patients to check clinical status and detect residual disease or recurrences by performing a clinical examination monthly for the first 3 months, and MRI every 6 months during the first 2 years and then annually thereafter. We recorded the preoperative and postoperative maximum angles of knee extension and flexion. We evaluated preoperative and postoperative knee function using the Tegner-Lysholm knee scoring scale [30]. We defined residual tumor as visible tumor on the earliest postoperative MR images (ie, at 3 months), and local recurrence as when there was no residual tumor after surgery, but evidence of tumor recurrence in the surgically treated knee according to subsequent followup MR images (Fig. 1). If residual tumor or local recurrence was suspected on the basis of physical examination or a patient's complaints, such as pain, swelling, joint effusion, or locking sensation, the interval between MRI studies was shortened to every 2 months. An experienced radiologist (H-THW) interpreted all MRI scans for residual of recurrent PVNS. The indications to perform reoperation for recurrent or residual PVNS were worsening of the Tegner-Lysholm knee scores during the two followups and the lesion size or number increased as observed on two followup MRI studies. We used paired t-tests to determine differences in knee scores and ROM before and after surgery, differences in knee scores between patients without and with residual PVNS, and differences in knee scores between patients without and with local recurrence. We performed all calculations using SPSS (Version 13.0; Chicago, IL, USA).

Results

With the first postoperative MRI at 3 months, we discovered residual PVNS in five of 19 patients (Patients 5, 6, 7, 11, and 17). Of these five patients, two (Patients 6 and 17) also had recurrent PVNS detected by postoperative MRI at 3 and 6 months, but at locations different from the residual PVNS observed on the postoperative MR images at 3 months. None of the five patients with residual and/or recurrent disease had progression observed on subsequent followup MRI studies performed at least 42 months after surgery (range, 42–119 months). Therefore, we did not perform repeat surgery during the current followup.

Fig. 1A–C A 34-year-old man (Patient 6) who underwent anterior cruciate ligament reconstruction at another hospital presented with persistent knee pain and instability. (A) A sagittal T2-weighted MR image shows diffuse PVNS of the left knee after anterior cruciate ligament reconstruction, with wearing and loosening of the graft. The black arrow indicates intact bone proximal to the PVNS, and the white arrow indicates PVNS over the posterior aspect of the distal femur. (B) A sagittal T2-weighted MR image was obtained 3 months

after simultaneous anterior and posterior synovectomies. The black and white arrows indicate recurrent and residual PVNS, respectively. (C) This sagittal T2-weighted MR image was taken 9 months after simultaneous anterior and posterior synovectomies. The black and white arrows show recurrent and residual PVNS, respectively. The tumor has decreased in size compared with the tumor seen on the MR image taken 3 months postoperatively.

The mean preoperative Tegner-Lysholm knee score was 59 points (range, 11–88 points) and it improved ($p < 0.001$) to 93 points (range, 86–100 points). At last followup, the Tegner-Lysholm scores of the three patients with residual tumor without progression were 87, 86, and 90 points, respectively. The Tegner-Lysholm scores of the two patients with recurrence were 88 and 90 points at 68 and 42 months, respectively. The mean postoperative knee score for the five patients with residual or recurrent PVNS was lower ($p < 0.001$) than the mean score for the patients without residual PVNS: 88.2 versus 95.0 points, respectively. Because none of the patients with residual or recurrent disease had progressive functional impairment, no surgery had been performed by the time of the last followup. The mean maximum preoperative extension angle was 11.0° (range, 0° – 20°) and improved to 2.1° (range, 0° – 10°) postoperatively. The mean maximum preoperative flexion angle was 76° (range, 20° – 120°) and improved ($p < 0.001$) to 127° (range, 110° – 135°) postoperatively.

No patient experienced complications, such as neurovascular injury, infection, or wound dehiscence. We identified no complications after low-dose radiotherapy.

Discussion

The best surgical method for diffuse PVNS has been controversial. Arthroscopic synovectomy reportedly resulted in a smaller wound and reduced morbidity [29]; however, owing to reported recurrence rates as much as 56% [20], some have recommended open synovectomy

[5, 6, 11, 21, 32]. Furthermore, some authors have recommended staged anterior and posterior synovectomies for patients with extraarticular diffuse PVNS [21, 32]. However, until now, there were no reported studies in the English literature regarding results of simultaneous anterior and posterior synovectomies combined with postoperative radiotherapy. The purpose of our study was to determine (1) local disease control, (2) the improvement of joint function, (3) the influence of joint function for patients with residual or recurrent disease, and (4) whether there were any complications after performing simultaneous anterior and posterior synovectomies with adjuvant radiotherapy in patients with diffuse PVNS of the knee.

Readers should be aware of the limitations of our study. First, owing to infrequency of diffuse type PVNS, the number of patients in this study was limited. Second, the disease courses among the 19 patients differed. Three had received surgical treatment before our surgery, and this might have affected the postoperative outcome. Third, we had no control group and therefore are unable to conclude if our technique was superior to others; however, we found simultaneous anterior and posterior synovectomies were associated with a low recurrence rate and reliable functional recovery. Fourth, the functional outcome assessment was based on a subjective questionnaire; however, Briggs et al. showed the Tegner-Lysholm knee scores are reliable and valid for determining return to function in patients after ACL surgery [4].

Ohnuma et al. [21] reported five patients with diffuse PVNS of the knee who underwent anterior synovectomy and posterior excision of the bursal tissue using two

Table 4. Comparison of literature reporting results for diffuse type pigmented villonodular synovitis

Study	Number of patients	Incision for open synovectomy	Staged surgery	Postoperative radiation	Local recurrence	Residual tumor	Knee function	Knee ROM	Complication	Followup
Ohnuma et al. [21]	5	Single anterior incision; two posterior oblique incisions	Two stage	No	1 (20%)	1 (20%)	NA	Three (60%) full ROM; 1 (20%) full ROM after manual mobilization; 1 (20%) from 10° to 110°	One (20%) infection	Average, 6.9 years (range, 3.3–8.1 years)
Wu et al. [32]	9	Single anterior incision; single s-shaped posterior incision (staged surgery)	Two stage	Yes*	1 (11%)	NA	Mean knee rating improved from 65.2 to 93.7; functional rating improved from 70 to 96.6	Maximal knee flexion ranged from 90° to 130°; 3 (33.3%) had flexion contractures of 5° develop; 1 (11.1%) from 5° to 112° after manual mobilization	None	Mean, 67 months (range, 37–103 months)
Flandry et al. [11]	23	Single anterior incision; single straight posteromedial incision	Two stage	No	2 (8%)*	NA	28% excellent; 64% good; 8% poor	NA	NA	Average, 58 months
Current study	19	Single anterior incision; single s-shaped posterior incision (one-stage surgery)	One stage	Yes	2 (11%)	5 (26%)	Mean Tegner-Lysholm knee score improved from 59 to 93	Mean maximum extension angles improved from 11° to 2°; mean maximum flexion angles improved from 76° to 127°	None	Median, 98 months

* Dose and frequency were not reported; **diagnosed by physical examination and plain film only; NA = not available.

posterior oblique incisions (three total incisions). However, they did not mention whether these surgeries were simultaneous or staged. After the followup period, one patient had a residual tumor and one underwent reoperation because of recurrence. Additionally, two patients experienced joint stiffness and received manual mobilization after release of the intraarticular adhesion. Wu et al. [32] reported on nine patients with diffuse PVNS treated with anterior and posterior open synovectomies. Unlike us, they used two-stage surgeries, performing the anterior synovectomy first and then the posterior synovectomy 6 to 8 weeks later. After the posterior procedure, they immobilized the knee in 15° flexion to relieve the wound tension. However, three patients experienced limited knee flexion postoperatively (90° in two patients and 100° in one patient). Thus, higher mobility, such as knee function impairment, may be a major concern after open synovectomy. Local recurrence rates after surgical treatment of patients with diffuse PVNS involving the knee have been reported. Flandry et al. [11] reported a local recurrence rate of 8% in patients treated with total synovectomy; however, they used plain radiographs and clinical examination for the postoperative followup owing to unavailability of MRI. Therefore, the true local recurrence rate may have been underestimated. In our protocol, we routinely used MRI for followup, and two of 19 patients experienced local recurrence. This was similar to recurrence rates of 11% to 18% described in previous studies of diffuse PVNS of the knee treated with open synovectomy and radiotherapy [5, 32] (Table 4).

Few studies have addressed the issue of residual tumor after surgery. However, it is difficult to remove all PVNS tissue, even using open synovectomy, in patients with advanced diffuse PVNS. In our study, five patients had residual PVNS tissue after simultaneous anterior and posterior synovectomies, including the two patients with subsequent local recurrence. Compared with patients without known residual tumor, these patients had less satisfactory postoperative knee function scores. As the patients' symptoms were stable and MRI revealed no evidence of disease progression during followup, none of these patients received additional surgery. This suggests radiotherapy plays an important role in controlling the residual tumor. It might also suggest the confounding presence of surgical changes in the knee since MRI appears to be sensitive, but not specific for detecting residual PVNS tissue postoperatively [5]; that is, the MRI findings may represent postoperative change instead of a true residual PVNS tissue. However, longer clinical followup and MRI are needed to determine which of these two explains the MRI findings.

The average Tegner-Lysholm score improved from 59 to 93 after treatment. The mean maximum postoperative extension and flexion angles for our patients were 2.1° and 127°, respectively. Earlier passive motion of the knee and

active quadriceps muscle training started the second postoperative day without the need for knee immobilization, which differed from previous studies [32] and may have contributed to the better functional outcome of our series. Furthermore, the simultaneous procedure could have shortened the hospital stay and reduced the cost of the entire treatment program.

The adjuvant therapy, such as low-dose radiotherapy, could be started earlier. Postoperative radiotherapy was suggested as adjuvant therapy for diffuse-type PVNS, especially for recurrent or retractable PVNS [2, 22, 32]. O'Sullivan et al. [22] reported 13 patients with PVNS treated with radiotherapy (35 to 50 Gy). They concluded radiotherapy could salvage subsequent recurrent PVNS which may compromise the function [22]. Kotwal et al. [16] reported no recurrence of PVNS after surgery followed by postoperative radiotherapy and 6% recurrence after surgery alone. With the simultaneous procedure, we were able to initiate low-dose radiotherapy during 4 weeks after surgery. If we had performed a two-stage surgery (separate anterior and posterior synovectomies), the radiotherapy would have been delayed by several weeks. Concerns regarding the simultaneous procedure are surgical time and wound healing [1]. However, our mean tourniquet time was 41 minutes for the anterior procedure and 68 minutes for the posterior. Additionally, with use of the anterior longitudinal and posterior lazy-s incision, all of the wounds healed well with no superficial or deep infections.

Our data suggest simultaneous anterior and posterior synovectomies with adjuvant radiotherapy are a reasonable option for patients with diffuse PVNS of the knee. Compared with arthroscopic synovectomy, this treatment offers better access to PVNS tissue and seemingly lower potential for disease spread and recurrence. Compared with staged anterior and posterior synovectomies, simultaneous anterior and posterior synovectomies allow earlier initiation of adjuvant radiotherapy and physical therapy. This may result in lower recurrence rates and superior knee ROM and function.

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