

Arthroscopic Treatment of Localized and Diffuse Pigmented Villonodular Synovitis of the Knee

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ABSTRACT

Pigmented villonodular synovitis (PVNS) is a rare proliferative synovial disorder of uncertain etiology. Two forms of this disorder, a localized (LPVNS) and diffuse (DPVNS) form, are well differentiated. The therapy of choice for LPVNS is arthroscopic partial synovectomy with excision of the lesion. Total synovectomy, whether done arthroscopically or through an open arthrotomy, is the recommended treatment for DPVNS. During an eight-year period 13 patients, six male and seven female, average age 28 years (range, 16 to 60 years) were treated for PVNS of the knee with arthroscopic synovectomy. Average follow-up was 84 months (range, 28 to 127 months). Four patients were affected by localized PVNS and were subjected to partial arthroscopic synovectomy (two to three portals) with a complete lesion excision. The remaining nine patients presented with the diffuse form of PVNS and all of them underwent total arthroscopic synovectomy (five portals). The diagnosis was confirmed by synovial biopsy. Each patient was evaluated before treatment and at final follow-up. Results were assessed clinically, radiographically and subjectively and were rated as excellent, good, fair, or poor. No complications or recurrences were noted in the LPVNS group, and all four patients were rated as excellent. In the DPVNS group, eight patients were rated as excellent and one patient was rated as fair and it was the patient who suffered the only recurrence in our case series. No relevant complications were encountered. No cases of infection, joint stiffness or neurovascular lesions were seen. Arthroscopy has become the golden standard in treatment of LPVNS, and can undoubtedly give results that are as good as with open synovectomy when treating DPVNS, if performed by an experienced arthroscopic surgeon.

Key words: pigmented villonodular synovitis, localized pigmented villonodular synovitis, diffuse pigmented villonodular synovitis, knee, arthroscopy

Introduction

Pigmented villonodular synovitis (PVNS) is a locally aggressive, proliferative synovial tissue disorder. It affects the synovial lining of joints, tendon sheaths and bursas, and may invade and destroy surrounding soft tissue and bone. It was first described by Chassignac¹ in 1852 as a lesion originating from the synovium of the middle finger flexor tendons. Considering the growth pattern, the capacity to destroy integral tissues of the joint, and a high recurrence rate, this disorder was thought to be of neoplastic nature. In 1941, Jaffe et al.² superseded the neoplastic theory by reviewing the histology of these lesions and presenting evidence to support the inflammatory etiology of PVNS. Various chromosomal abnormalities have been described in the literature, the most commonly reported ones being trisomy of chromosomes VII and V^{3,4} and structural rearrangement of chro-

mosome I^{5,6}. Pathohistological findings of PVNS show hemosiderin deposition, a fibrous stroma with zones of sclerosis, histiocytic infiltrates and multinucleated giant cells. To this day, no unanimous agreement has been reached, as the true etiology of PVNS remains to be proven.

Considering the involvement of the synovial tissue, we can distinguish a localized (LPVNS) and diffuse (DPVNS) form of PVNS, both of which may be intra-articular and extra-articular⁷. The localized form implies a single mass present in the synovium, either a nodule, a small tumefaction or a pedunculated mobile mass, while the diffuse form involves the entire synovium of a joint⁸. These two presentations of PVNS are considered most likely to be two extremes on the spectrum of one disease, instead of two individual entities⁹.

PVNS is typically monoarticular, and most frequently involves the knee, hip, ankle, shoulder and elbow¹⁰. Myers et al.¹¹ found the annual incidence of PVNS to be approximately 1.8 patients *per* 1 000 000 population with no environmental, genetic, ethnic or occupational predilection. Men and women are equally affected with the average age of onset usually in the third and fourth decade. DPVNS is markedly more frequent than LPVNS, and has a higher recurrence rate. PVNS still poses a diagnostic challenge, and the correct diagnosis is often delayed after first symptoms appear, mostly due to its insidious onset and nonspecific presentation, as well as very subtle radiographic findings associated with this disorder.

Management options for PVNS include open, arthroscopic and radiation-induced synovectomy. The question of whether it is more appropriate to treat cases of DPVNS of the knee with traditional open synovectomy or arthroscopic synovectomy is still open for debate.

The aim of this retrospective case analysis is to critically evaluate the results of arthroscopic synovectomy in the knee affected by PVNS, and to determine the safety and effectiveness of the procedure.

Materials and Methods

Thirteen patients, six males and seven females, were treated for PVNS of the knee by a single surgeon (IB) at our institution from 1999 to 2007. Nine patients had diffuse lesions and four had localized lesions. The average age of the patients was 28 years (range, 16 to 60 years). The right knee was affected in three patients and the left knee in 10. An independent examiner reviewed the patients records, and interviewed and examined the patients at final follow-up.

In patients with LPVNS, symptoms mimicked those provoked by an intra-articular loose body (n=1) or a meniscal tear (n=3), and had been present for an average of nine months at first consult. In patients with DPVNS, presenting symptoms were mainly spontaneous swelling, diffuse non-specific knee pain, and a decreased range of motion, which had begun on average 12 months before the first consult.

In each of the 13 cases, arthroscopy was performed under tourniquet control using standard anterolateral and anteromedial portals. In patients with LPVNS a

thorough examination of the knee joint was carried out to exclude presence of any other lesion and was followed by excision of the pathologic tissue and a partial synovectomy. In two LPVNS cases the lesion was located in the posterolateral compartment, which required use of a posterolateral accessory portal. The diagnosis of LPVNS was confirmed by pathohistological analysis of the excised lesions in all cases. In patients with DPVNS, a first, diagnostic arthroscopy was made and a biopsy specimen was acquired. Following pathohistological confirmation of the diagnosis, a complete arthroscopic synovectomy was performed. Five arthroscopic portals (anterolateral, anteromedial, lateral suprapatellar, posterolateral, and posteromedial) were used in all DPVNS patients. The anterior and posterior compartments were fully visualized and synovectomy was performed using a 5.5 mm full-radius synovial resector in oscillating mode. An intra-articular suction drain was used for 24 hours in the LPVNS group, and for 48 hours in the DPVNS group. After the removal of the drain the patients were subjected to intensive physical therapy consisting of isometric quadriceps exercises and passive range of motion exercises with a continuous passive motion machine. Therapeutic goals were set to achieve maximum range of motion, restore quadriceps muscle strength and decrease swelling, pain and inflammation in the early postoperative period.

Each patient was evaluated before treatment and at the final follow-up visit, following the evaluation criteria proposed by Ogilvie-Harris et al.¹² (Table 1). These criteria comprise four parameters, the first being articular pain, followed by synovitis or joint effusion, compromise to the range of motion, and functional ability. Each of these parameters is graded on a scale of 0 to 3 points. At the final follow-up, the patient's condition was rated as excellent (scores between 10 and 12), good (scores between 7 and 9), fair (scores between 4 and 6), or poor (scores between 0 and 3).

Results

All 13 patients were available for follow-up (Table 2). Mean follow-up was 84 months (range, 28 to 127). Four patients were diagnosed with LPVNS of the knee. Mean preoperative evaluation score according to the Ogilvie-Harris criteria was 8.00, which corresponded to a rating of »good«. In these cases, an arthroscopic excision of the

TABLE 1
CRITERIA FOR ASSESMENT OF THE RESULTS ACCORDING TO OGILVIE-HARRIS ET AL.¹²

	0 Point	1 Point	2 Points	3 Points
Pain	Severe	Moderate	Slight	None
Synovitis, effusion	Severe	Moderate	Slight	None
Range of motion*	>20% loss	10 to 20% loss	0 to 10% loss	No loss
Functional capacity	Minimum activity	Some activity	Most activities	All activities

* The normal range of motion of the knee was considered to be 150 degrees of flexion (that is, a loss of 10 *per* cent represents a combined loss of flexion and extension of 15 degrees)

TABLE 2
RESULTS OF ARTHROSCOPIC TREATMENT OF OUR PATIENTS WITH PVNS OF THE KNEE EVALUATED BY CRITERIA PROPOSED BY OGILVIE-HARRIS ET AL.¹²

Case	Age (yrs)/ Sex/ Side	Follow up (mo)	Pain		Synovitis		Range of motion		Function		Total Score	
			Pre	Post	Pre	Post	Pre	Post	Pre	Post	Pre	Post
Diffuse PVNS												
1	17/M/L	119	1	3	0	3	1	3	1	3	3	12
2	26/F/L	114	1	3	1	3	2	3	1	3	5	12
3	25/M/L	113	1	3	0	2	1	3	0	2	2	10
4	16/F/R	79	1	3	0	3	2	3	1	3	4	12
5	31/M/L	67	1	3	1	2	1	3	1	2	4	10
6	33/F/L	65	1	3	0	3	2	3	0	3	3	12
7	39/M/L	60	1	3	0	3	2	3	1	2	4	11
8*	16/F/L	55	0	2	0	0	1	1	0	1	1	4
9	22/F/L	28	0	3	0	2	1	3	1	3	2	11
Mean			0.78	2.89	0.22	2.33	1.44	2.78	0.67	2.44	3.11	10.44
Localized PVNS												
1	22/F/R	127	2	3	2	3	3	3	1	3	8	12
2	41/F/L	112	2	3	3	3	2	3	2	3	9	12
3	60/M/L	108	1	2	1	3	2	2	1	3	5	10
4	16/M/R	46	2	3	3	3	3	3	2	3	10	12
Mean			1.75	2.75	2.25	3.00	2.50	2.75	1.50	3.00	8.00	11.50

Note: Pre and post refer to evaluation before surgery and at final follow-up visit

* Patient had a recurrence, open synovectomy performed

lesion was done, followed by resection of the surrounding synovia. The mean postoperative score was 11.50, rated »excellent«. No recurrence was noted in the LPVNS group. In the remaining nine patients affected with DPVNS, total synovectomy was performed arthroscopically, through five arthroscopic portals. The mean score was 3.11 prior to surgery, rated as »poor«, and after the procedure 10.44, rated as »excellent«. There was one recurrence in the DPVNS group, a 16 year old female patient, whose postoperative score was »fair« (4) according to the Ogilvie-Harris evaluation criteria. There were no complications during or after the arthroscopic procedure in this patient. Recurrence was identified clinically, taking into consideration the renewed onset of pain accompanied by swelling, loss of ROM, and was confirmed pathohistologically. The patient underwent an open total synovectomy and no recurrence was noted at final follow up.

Discussion and Conclusion

Diagnosis of DPVNS or LPVNS is rarely clinically evident. According to Flandry et al.¹³, only 17% of patients with PVNS received proper diagnosis before referral. PVNS can often mimic symptoms and signs of various articular disorders, thus posing a great diagnostic challenge for both the surgeon and radiologist. While localized nodular forms often mimic a variety of intra-articular disorders of the knee, such as meniscal tears or loose bodies^{14,15}, diffuse forms are more likely to imitate some

chronic inflammatory states. PVNS should be considered in the differential diagnosis of patients aged 20 to 45 with monoarticular symptoms. Diverse imaging methods are used to narrow down the diagnosis. Plain radiographs are considered a nonspecific and insensitive diagnostic tool¹⁶ but are still widely used due to their availability and can be helpful to a certain degree, especially in longstanding cases in which they show periarticular bone erosions, subchondral cysts and swelling of soft tissue. Aspiration of synovial fluid is a commonly used method and a brown hue in the aspirated fluid is indicative of PVNS. It too lacks specificity and sensitivity, because many other conditions have fluid resembling this. Magnetic resonance imaging (MRI) has become the method of choice for diagnosing PVNS. It is noninvasive, can be highly sensitive and specific using newer sequences and is best used to differentiate between DPVNS and LPVNS, to evaluate the extent of the disease preoperatively, and to monitor patients postoperatively. An important method of diagnosing PVNS is arthroscopy, which enables the physician to make the definitive diagnosis and perform surgery in the same act.

The treatment modalities for LPVNS and DPVNS vary significantly, although the main principle of treatment is very similar. All abnormal synovial tissue must be eradicated, thereby removing the risk of further joint destruction and recurrence. An exception to this principle is made when the disease is recurrent and some of those patients benefit from total joint arthroplasty.

Resection of the PVNS mass along with a rim of surrounding healthy synovium is the most appropriate treatment of LPVNS, giving best results and very low recurrence rates. Arthroscopy is the golden standard in treatment of localized lesions, corroborated by various authors; Ogilvie-Harris et al.¹² reviewed 25 cases of both LPVNS and DPVNS treated arthroscopically. Five LPVNS patients were part of the cohort and were managed with excision followed by partial synovectomy. All five patients improved symptomatically and in terms of function, and there were no recurrences. Kim et al.¹⁷ treated 11 LPVNS patients with arthroscopic partial synovectomy, and detected no recurrences. Dines et al.¹⁸ reviewed 26 LPVNS cases, of which 12 were treated by arthroscopic partial synovectomy, no recurrences were reported. Moskovich and Parisien¹⁹ evaluated arthroscopic partial synovectomies done in nine patients with LPVNS and detected no recurrences. De Ponti et al.²⁰ reviewed four LPVNS patients that were subjected to arthroscopic partial synovectomy, no recurrences were reported. Bojanić et al.²¹ reviewed three cases of LPVNS of the knee managed with arthroscopic excision of the lesions and reported no recurrences. Although recurrence is very rare after arthroscopic treatment of LPVNS, it should be noted that the possibility of inadequate resection of the affected tissue always exists. De Visser et al.²² reported one recurrence (11%) in nine LPVNS cases managed with arthroscopic partial synovectomy.

In contrast, the treatment of DPVNS of the knee is still a matter of debate. A wide array of options exist for treating knee DPVNS; an extensive open anteroposterior approach, an entirely arthroscopic approach, a combination of anterior arthroscopic and open posterior synovectomy, and a subtotal synovectomy followed by adjuvant radiotherapy. Recurrence rates for DPVNS vary from 8% to 46% with all treatment methods^{12,20,23–27}. The decision whether to operate arthroscopically, through classic open arthrotomy, or those two combined is made according to the MRI findings of the extent to which the joint is affected by the disease. Patients with DPVNS confined only to the joint are candidates for arthroscopy by an experienced surgeon. Large popliteal masses or any extra-articular involvement are generally considered to be criteria against arthroscopy and imply the open or the combined anterior arthroscopic and posterior open approach.

Byers et al.²³ reported a 46% recurrence rate in 1968, using open total synovectomy in 24 cases of DPVNS. Flandry et al.²⁴ performed an open total synovectomy using anterior and posterior incisions in 25 cases of knee DPVNS in 23 patients (two bilateral), and noted only two recurrences (8%) at five year follow-up. Johansson et al.²⁵ performed open synovectomies in 24 patients with DPVNS, of which 18 returned for examination. Out of the 18 patients available for follow-up, they noted recurrence in six (33%).

Ogilvie-Harris et al.¹² treated 20 patients for DPVNS of the knee and were the first to do so using arthroscopic synovectomy, total and partial, respectively. The recurrence rate was 9% in patients treated with total syno-

vectomy compared to a 56% recurrence rate following partial arthroscopic synovectomy. De Ponti et al.²⁰ treated 15 patients for DPVNS, eight with partial arthroscopic synovectomy and seven with extended arthroscopic synovectomy. Patients treated with arthroscopic partial synovectomy had worse clinical outcomes and a markedly higher recurrence rate (50%–75%) than those treated with arthroscopic extended synovectomy (20%). Zvijac et al.²⁶ treated 12 patients for DPVNS of the knee with arthroscopic total synovectomy, and reported a 17% overall recurrence rate.

In a very recent study, Sharma and Cheng²⁷ reviewed 49 patients affected with PVNS of the knee, 37 of those affected with DPVNS. The authors divided patients into three groups according to received treatment: the first group consisted of 13 DPVNS patients treated with anterior arthroscopic synovectomy, eight DPVNS patients treated with anterior arthroscopic synovectomy combined with open posterior synovectomy constituted the second group, and 16 DPVNS patients treated with open anterior and posterior synovectomy constituted the third group. Twelve patients out of 13 from the first group, two out of eight from the second group and three out of 16 from the third group suffered a recurrence. The authors concluded that the highest recurrence rate was in the group of patients with DPVNS treated with anterior arthroscopic synovectomy only, while the excision of the posterior disease in DPVNS patients improved the recurrence-free survival in patients.

Radiation therapy has been an alternative mode of treatment in patients with synovitis for many years^{28,29}. Radiation induced synovectomy has produced ambiguous results, and carries with it serious potential complications such as skin reactions, poor wound healing, joint stiffness and radiation-induced sarcomas. Various authors have used radiation, whether intraarticular or external beam radiation, as an adjuvant treatment following surgical excision of DPVNS. Blanco et al.³⁰ reviewed 22 patients treated with arthroscopic partial synovectomy and adjuvant low-dose external beam radiation and reported a recurrence rate of 14%. They also reported a 9% rate of posttreatment joint stiffness in the patient series. Shabat et al.³¹ combined open partial synovectomy with intra-articular yttrium Y-90 administered six weeks after the initial, surgical synovectomy, and reported one recurrence out of 10 patients with DPVNS. Chin et al.³² treated a series of 30 patients using combined open anterior and posterior synovectomy with adjuvant intra-articular radiation, and reported a recurrence rate of 17%. Recently, Nassar et al.³³ treated 12 patients affected with extensive DPVNS of the knee using combined open anterior and posterior synovectomy followed by external beam radiation therapy administered six to eight weeks after the surgery, and reported no recurrences or complications at 20–36 months follow-up. These studies suggest that adjuvant radiation synovectomy should be considered in advanced cases of recurrence and relapsed disease, cases with extensively infiltrating disease and cases

in which posterior compartment access is difficult due to anatomy.

We performed arthroscopic total synovectomy using five arthroscopy portals in all of our DPVNS patients, taking care that all parts of the knee were thoroughly inspected and all abnormal synovial tissue was excised. Arthroscopic synovectomy has many advantages over classic arthrotomy. It is a minimally invasive procedure and carries with it less intraoperative blood loss, quicker functional recovery, and less postoperative complications such as pain and joint stiffness. Additional arthroscopy portals (posterolateral and posteromedial) offer better access to the posterior compartments of the knee. Utilizing all five portals is a technically demanding procedure and should be done by seasoned arthroscopists. Arthroscopy is not, however, an inherently complication-free method. There is a tentative risk of joint seeding and portal contamination^{32,34}. Also, a subcutaneous contamination of an arthroscopy portal was described by Lu³⁵. In our study, no recurrences were found in patients with LPVNS at final follow-up. One patient in the DPVNS group had a recurrence and needed additional surgery. Taking into consideration results and recurrence rates of other authors, we believe we have shown that with a good understanding and knowledge of arthroscopy, it is possible to carry out a precise total synovectomy. We managed to compensate the lack of MRI diagnostics, which was the main limitation of our study, by using long term follow-up (mean 84 months, range 28 to 127). Thorough clinical examinations were performed, according to the clinical assessment scores, radiographs were taken pre- and post-operatively, all the time being fully aware of their restrictions. This clinically oriented follow-up may have caused us to underestimate the exact recurrence rate in our study, but up to this day all of the clinical

markers of recurrence such as swelling, ROM limitations and pain, have remained negative in all but one patient.

Our experience in arthroscopic surgical treatment of PVNS of the knee confirms that arthroscopic surgery is an excellent option for treating LPVNS and a powerful tool in DPVNS treatment when used by a surgeon experienced in arthroscopy.

Although the diagnostic options are getting more and more sophisticated each day, and surgeons are adopting new techniques to treat this disease, no evidence-based algorithm of treatment exist yet. Recently, Kramer et al.³⁶ suggested a well thought out plan of approach to various types of knee PVNS cases. Today, MRI should be regularly used to determine the extent of preoperative disease and the decision whether to operate on a patient arthroscopically or using open arthrotomy should be made on the basis of an extensive MRI study. MRI is also particularly useful for postoperative follow-up of patients for recurrence. Patients with DPVNS confined solely to the joint should be treated with arthroscopic total synovectomy, since it causes much less postoperative complications than the open approach. Patients with extra-articular involvement, large popliteal masses and recurrent disease should be considered for open total synovectomy alone, or combined arthroscopic anterior and open posterior synovectomy. In this study we have shown that it is possible to treat even extensive cases of DPVNS exclusively by arthroscopy, as long as it is done by a surgeon experienced in arthroscopy. It is very important for surgeon to improve and upgrade their skills in arthroscopic surgery to be able to use it to the fullest, and thus contribute to better treatment results, less postoperative complications and lower recurrence rates.

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ARTROSKOPSKO LIJEČENJE PIGMENTIRANOG VILONODULARNOG SINOVIITISA KOLJENA

SAŽETAK

Pigmentirani vilonodularni sinovitis (PVNS) rijedak je proliferativni poremećaj sinovije čija etiologija nije u cijelosti poznata. Razlikuju se dva oblika ove bolesti, lokalizirani (LPVNS) te difuzni (DPVNS). Liječenje izbora za LPVNS je artroskopska djelomična sinoviektomija s ekscizijom lezije. Potpuna sinoviektomija, učinjena bilo otvorenom artrotomijom bilo artroskopski, preporučena je terapija za DPVNS. Tijekom osmogodišnjeg razdoblja trinaestoro bolesnika, šest muškaraca i sedam žena, prosječne dobi 28 godina (raspon, 16 do 60 godina) liječeno je artroskopski u našoj ustanovi zbog PVNS koljena. Prosjek praćenja bolesnika bio je 84 mjeseca (raspon, 28 do 127 mjeseca). Četvoro bolesnika liječenih radi LPVNS podvrgnuto je djelomičnoj sinoviektomiji (koristeći dva do tri ulaza ili portala) te eksciziji lezije in toto. Preostalih devetero bolesnika prezentiralo se s difuznim oblikom PVNS, a u njih je učinjena potpuna artroskopska sinoviektomija. Svakog se bolesnika evaluiralo prije liječenja te prilikom posljednjeg kontrolnog pregleda. Rezultate se procijenjivalo klinički, radiografski i subjektivno te je svakom bolesniku pridodana ocjena odličan, dobar, zadovoljavajuć ili loš. Nije zabilježen niti jedan slučaj povratka bolesti u grupi bolesnika s LPVNS, a svih četvoro bolesnika ocijenjeno je odličnom ocjenom. U grupi bolesnika oboljelih od DPVNS, osmero ih je bilo ocijenjeno odličnom ocjenom, dok je jedna bolesnica ocijenjena kao zadovoljavajuća, to je upravo bolesnica kod koje je zabilježen povratak bolesti. Nije zabilježena niti jedna značajna komplikacija, niti jedan slučaj poslijeoperacijske infekcije, pojave ukočenosti zgloba ili pak neurovaskularnog oštećenja. Artroskopsko liječenje LPVNS u današnje je vrijeme zlatni standard. Također, potpuna artroskopska sinoviektomija može proizvesti rezultate koji su jednako kvalitetni kao i oni otvorene potpune sinoviektomije, pod uvjetom da ju učini kirurg s dovoljno iskustva u artroskopiji.