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ORIGINAL PAPER

Arthroscopically-assisted mini open partial synovectomy for the treatment of localized pigmented villonodular synovitis of the knee. A retrospective comparative study with long-term follow up

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Abstract
Purpose The purpose of this study was to present the long-term results of treatment of localized pigmented villonodular synovitis (LPVNS) comparing two operative procedures of excision of the lesion—the arthroscopic and the arthroscopically assisted mini-open. We hypothesized that the latter approach allowed for treatment of LPVNS with acceptable recurrence rates, complication rates and functional outcomes.
Methods Between 1990 and 2006, 21 patients with LPVNS were treated with partial synovectomy through an arthroscopically-assisted mini open technique (group A), and 23 patients were treated with an arthroscopic excision of the lesion (group B). All patients were clinically examined at one, three, and 12 months post-operatively and graded by the Lysholm knee score and the Ogilvie-Harris score.
Results The mean Lysholm score was improved from 58.7 ± 9 to 94.2 ± 7 for group A ($p < 0.05$) and from 57.4 ± 9.1 to 95.5 ± 8 for group B ($p < 0.05$). The mean Ogilvie-Harris score was improved from 7.2 ± 2 to 11.2 ± 0.9 for group A and from 7.1 ± 2 to 11.75 ± 0.5 for group B ($p < 0.05$). We encountered two cases of CRPS and one case of recurrence of the lesion in group A and no complications for group B.
Conclusions Arthroscopically-assisted mini open partial synovectomy is a safe alternative treatment, especially for surgeons without extended experience in arthroscopic techniques. The arthroscopic localization of the precise position of the lesion and its subsequent mini-open excision is a safe and effective technique with very low morbidity and recurrence rate and equivalent functional outcome to fully arthroscopic excision due to limited incision.
Level of evidence Retrospective comparative study, Level III.

Keywords Localized PVNS · Arthroscopic synovectomy · Mini-open synovectomy · Knee · Surgical treatment · Recurrence

Introduction

Pigmented villonodular synovitis (PVNS) is a rare pathology that affects the synovial joints, tendon sheaths, and bursa membranes. The first ever report of PVNS was in 1852 by Chassaignac [1] who reported a case of a nodular lesion developing in the flexor tendon sheaths of the middle and index fingers. In 1941, Jaffe et al. [2] coined the term "pigmented villonodular synovitis". Granowitz et al. [3] expanded the terminology by distinguishing between the localized (LPVNS) and the diffuse (DPVNS) forms of these synovial lesions.

The localized form is characterized by focal involvement of the synovium either as nodules, small fumefactions, or pedunculated masses, while the diffuse form affects the entire synovium of the affected joint [4]. PVNS affects people in their third or fourth decades of life [5, 6]. Typically, the disease is mono-articular and involves mainly the knee joint, while the hip, the shoulder and the ankle joint follow in frequency [7–9].

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Complete excision of the mass in the affected knee joint is the treatment of choice in LPVNS [10, 11]. This could be done either arthroscopically or through a mini-open arthrotomy [5]. Recurrence of pathology is the main complication of any operative technique [12]. Arthroscopic excision of the lesion is considered the gold standard [13, 14]. There are no reports in the literature regarding the mini-open arthroscopically-assisted localized synovectomy.

The purpose of this study was to present the long-term results of the treatment of LPVNS comparing two operative procedures of excision of the lesion—the arthroscopic and the arthroscopically assisted mini-open. It was hypothesized that the use of the later approach achieved acceptable recurrence rates, complication rates and functional outcomes. Although a retrospective study, it is to our knowledge the only reported study in the literature that directly compared two different procedures for the treatment of LPVNS.

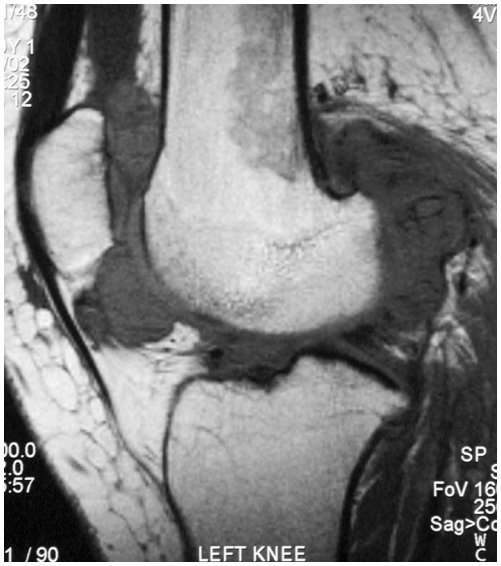

Fig. 1 MRI of a left knee showing a localized pigmented villonodular synovitis (LPVNS) lesion extended from the intercondylar notch to the suprapatellar pouch

Materials and methods

Population

Between 1990 and 2006, 21 patients with LPVNS were treated with partial synovectomy through a mini-open technique (group A) and 23 patients were treated with an arthroscopic excision of the lesion (group B). Inclusion criteria for the study were patients with LPVNS of the knee and patients who completed the entire post-operative examination at final follow-up. Exclusion criteria were extra-articular location of the knee PVNS, diffuse PVNS and cases with recurrence after previous operation.

The average age of the patients was 36 years (range, 14–53 years). The sex ratio was 1.2:1 (15 men, 11 women). The mean duration of symptoms prior to surgical treatment was 6.7 months (range, 2–12 months). The average follow-up was 12 years (range, 5–20 years). Twenty-one lesions (47.7%) were located in the suprapatellar pouch (Fig. 1), while seven (15.9%) lesions were located in the intercondylar notch and the fat-pad (Fig. 2); six lesions (13.6%) originated from the meniscosynovial junction and four (9.1%) lesions from the medial plica, while six (13.6%) lesions were located in the lateral gutter (Table 1).

The clinical symptoms were persistent knee pain (72.7%), effusion of the knee joint (59.1%), locking symptoms (50%), loss of extension (31.8%) and palpable mass (22.7%). Pain, locking symptoms and loss of extension were the predominant symptoms in cases where the lesions were located in the fat pad, the lateral gutter and the medial plica, while effusion and palpable mass were predominant in LPVNS located in the suprapatellar pouch.

Surgical technique

During arthroscopic excision, patients were positioned supine with the affected knee in a leg holder. Tourniquet was applied and arthroscopy of the knee was performed through standard anterolateral and anteromedial portals using a 30° arthroscope. The recognized lesion was dissected from a nodular base using a 4.5- or 5.5-mm shaver and/or a radiofrequency ablator (Fig. 3). When necessary, additional superomedial or superolateral portals were also utilized.

In a mini-open excision, a diagnostic arthroscopy was initially performed in order to recognize and precisely locate the lesion. Then an excision of the nodule and partial

Fig. 2 MRI of the right knee of a 43-year-old female, revealed a localized pigmented villonodular synovitis (LPVNS) lesion located in the fat pad

Table 1 Demographic data

Variables	Group A ($n = 21$)	Group B ($n = 23$)	Statistical significance
Age in years, median (range)	36 (14–54)	38 (17–56)	NS
Gender ratio M:F	11:10	13:10	NS
Duration of symptoms (months)	6.5	6.7	NS
Location of lesion			
1. Suprapatellar pouch	10	11	NS
2. Fat-pad/intertroch. area	3	4	NS
3. Lateral gutter	3	3	NS
4. Medial plica	3	1	NS
5. Meniscosynovial junction	2	4	NS

NS not significant

synovectomy of the surrounding tissues was performed through a small incision (3-4 cm) over the lesion (Fig. 4).

Passive and active rehabilitation exercises were initiated on the first postoperative day and all patients were discharged either the same or the following day.

Methods of assessment via scoring

The primary outcomes collected were post-operative complications and recurrence of the lesion. The following secondary variables were also collected: patient's age and gender, symptoms and duration of symptoms, location of lesion. The systematic diagnostic imaging included an anteroposterior and a lateral view of the knee, and an MRI scan for all patients. MRI scan showed characteristic findings included haemosiderin deposits that resulted in low signal area in both T1- and T2-weighted images. In addition, fat-suppressed sequences showed high signal proliferative masses that also obscure the haemosiderin deposits. Histopathology examination confirmed the diagnosis of PVNS in all cases.

All patients were clinically examined at one, three, and 12 months post-operatively and graded by the Lysholm knee score [15] (a general knee score with primary outcome the knee function and pain, range 0–100) and the Ogilvie-Harris score [16] (a specific score validated for PVNS, provides a scale of clinical severity, range 0–12). Routine follow-up occurs once a year. Excised tissues were routinely sent for histopathology.

Statistical analysis

Statistical analysis using the unpaired Student's t-test and the chi-square test were applied to evaluate significant differences between the two groups (SPSS, version 19.0, SPSS Inc, Chicago, IL, USA). Statistical significance was defined at the 5% ($p < 0.05$) level.

Results

The mean Lysholm score of group A was improved significantly from 58.7 ± 9 pre-operatively to 94.2 ± 7 at last follow-up ($p < 0.05$). For group B, there was also a significant improvement from 57.4 ± 9.1 to 95.5 ± 8 ($p < 0.05$). Mean values at last follow-up were not significantly different between the groups ($p = 0.14$). The mean Ogilvie-Harris score was improved significantly from 7.2 ± 2 to 11.2 ± 0.9 for group A and from 7.1 ± 2 to 11.75 ± 0.5 for group B ($p < 0.05$). There

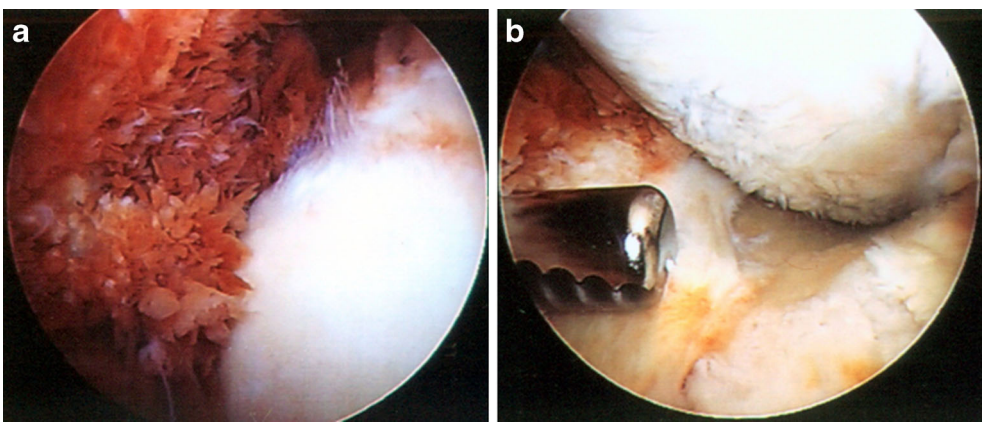

Fig. 3 Arthroscopic view of a pigmented villonodular synovitis (PVNS) lesion located at the meniscosynovial junction (**a**) which was treated with an arthroscopic shaving (**b**)

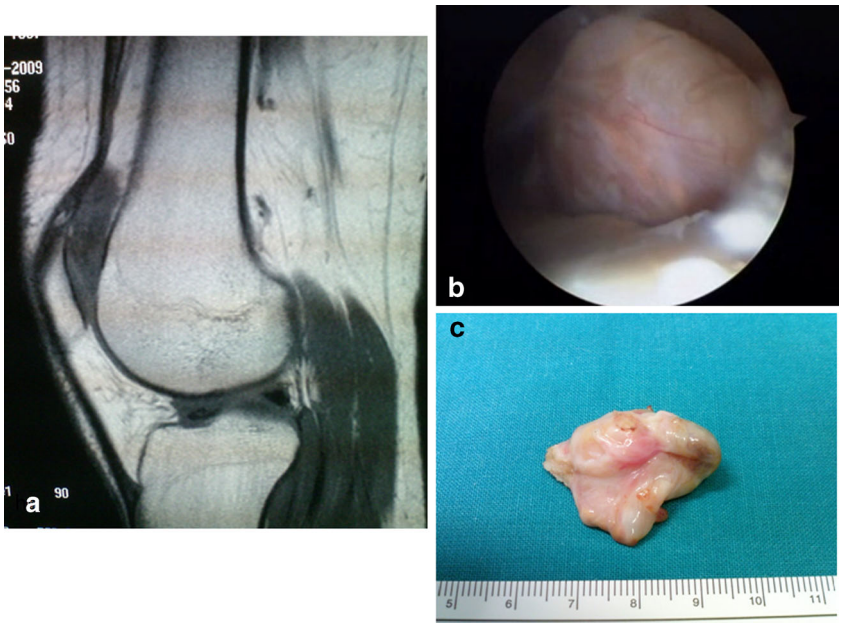

Fig. 4 MRI of the right knee of a 32-year-old male showing a pigmented villonodular synovitis (PVNS) lesion located in the suprapatellar pouch (**a**). The lesion was excised with an arthroscopically-assisted mini-open approach (**b**). A 3-mm LPVNS lesion excised 'en-block' (**c**)

was no statistical difference between the two groups at the last follow-up (p = 0.06) (Table 2).

Two patients of group A developed chronic regional pain syndrome (CRPS) two months post-operatively which resolved in time in one of the cases, while the other patient had residual decreased ROM at the latest follow-up. We encountered only one case of recurrence of lesion (4.7%). One patient with a fat-pad nodule who was treated with the mini-open technique developed a recurrence at one year post-operatively. He underwent arthroscopic synovectomy combined with intra-articular radiation therapy with Yttrium-90 without any subsequent complication. No post-operative complications were encountered in group B. There was no statistical difference between the two groups regarding the complication (p = 0.06) and the recurrence rates (p = 0.1).

Discussion

PVNS is a rare condition that affects the synovial joints, tendon sheaths and bursa membranes. There is no consensus about the aetiology of PVNS. There are four theories prevalent throughout the literature, each one focusing on one or more of the cellular elements observed with PVNS [3, 5] with the last two as the most widely accepted: (1) disturbance of lipid metabolism, (2) repetitive injury or trauma, (3) chronic inflammation, (4) benign neoplasia. In a recent study, Richter et al. [17] studied the role of adipocytokines in the pathogenesis of knee joint osteoarthritis. These bioactive proteins were found to cause changes of the synovial membrane, such as hypertrophy, hyperplasia and synovial tissue thickening. Nevertheless, the exact role of these peptides in inducing synovitis and PVNS is unknown. Recently, the World Health Organization indicated that PVNS and tenosynovial giant cell tumor are synonymous [12, 18]. Granowitz et al. [3] distinguished PVNS in two forms, the diffused and the localized. The localized form makes up to 15–21% of PVNS cases and the knee is affected in two-thirds of cases [10]. PVNS has been found in several locations within the knee joint: meniscosynovium junction, suprapatellar pouch, intercondylar notch and Hoffa's fat-pad, lateral and medial gutter, and the posterior compartment [13, 19, 20]. In our study the most common

Table 2 Scoring data

Score		Group A ($n = 21$)	Group B ($n = 23$)	Statistical significance
Lysholm score	Preop	58.7 ± 9	57.4 ± 9.1	NS
	Postop	94.2 ± 7	95.5 ± 8	NS
	Statistical significance	P < 0.05	P < 0.05	
Ogilvie-Harris score	Preop	7.2 ± 2	7.1 ± 2	NS
	Postop	11.2 ± 0.9	11.75 ± 0.5	NS
	Statistical significance	P < 0.05	P < 0.05	

NS not significant

location of the lesion was the suprapatellar pouch, which is in accordance with other studies [6, 14, 21]. Diffuse pain and effusion are the predominant symptoms followed by mechanical locking. Our results showed that effusion is the primary symptom in case of suprapatellar pouch location of lesion whereas locking and loss of extension are present when the lesion was located within the fat-pad, lateral gutter or medial plica [21, 22]. Classically, the localized nodular form consists of an isolated circumscribed or pedunculated lesion of the synovium. Histologically, these lesions are characterized by a proliferation of polyhedral synovial-like cells with multinucleated giant cells responding to haemosiderin deposition in areas of hyalinized collagen tissue, with occasional foam cells [5, 8].

Mean functional scores in the present study were excellent for both groups. There was significant improvement in function with both techniques used. That is consistent with other studies that reported in functional scores [5, 6, 14, 23]. Although the mean value of both scores was slightly superior in the arthroscopy group, there was no significant difference between the two groups.

Surgical resection, either through open or arthroscopic technique, is considered the mainstay of treatment for PVNS around the knee [7, 10, 18, 23, 24]. Especially for patients with LPVNS, arthroscopic resection of the lesion is considered the golden standard [13, 14]. The main benefit of arthroscopic synovectomy is a very low post-operative morbidity and a low recurrence rate. Our results are consistent with previous published reports that documented a 0% of post-operative complications [5, 6, 14, 21, 22, 24, 25] and a 0% recurrence rate for LPVNS lesions removed arthroscopically [5, 11, 22, 24, 26]. However, there are several studies that reported a higher complication rate. Loriat et al. [14] reported four complications (20%) on a study of 20 patients with LPVNS treated arthroscopically, while Rhee et al. [6] and Kubat et al. [25] reported 18.2% (2 cases) and 7.7% (1 case) complication rate in case series of 11 and 13 patients, respectively. Open synovectomy is historically associated with longer hospitalization and rehabilitation and can be associated with post-operative stiffness or wound healing complications [18, 27]. One study reported a complication of deep vein thrombosis in one case after treatment of LPVNS with open synovectomy [28].

In regards to the recurrence rates, Byers et al. [29] reported two recurrences on 13 cases of LPVNS treated with open synovectomy, while Schwartz et al. [28] and Sharma and Cheng [30] reported two cases of recurrence each on a 12- and a seven-patient case series, respectively. Treatment of relapsed and recurrent PVNS with synovectomy and the use of adjuvant radiotherapy or intra-articular injection of Y-90 was associated with excellent results [26, 31]. In a recent systematic review [12] the overall recurrence rate was 6.9% (10 cases) and complication rate was 0% in a total number of 144 patients treated with arthroscopic localized synovectomy for LPVNS in 22 included studies. On the other hand, the overall recurrence and complication rate was 8.7% (10 cases) and 0.88% (1 case), respectively, in a total of 115 patients treated with open localized synovectomy for LPVNS, in 17 included studies. Van Heijden et al. [9] reported, in a recent crowd-sourcing study, higher recurrence rates of 58% (69/118), 36% (35/97) and 50% (5/10) for arthroscopic, open and combined arthroscopic and mini-open synovectomy, respectively. We attributed those high rates of recurrence to the study profile as it was a crowd-sourcing study via social media, amenable to the limitations the authors acknowledged.

The reasons for recurrence after the arthroscopic synovectomy was not explicated in any of the studies. We attributed the recurrence to two reasons: (1) the experience of the surgeon in advanced arthroscopic techniques needed to excise the lesion entirely with the arthroscopic shaver, (2) the ability to localize the margins of the PVNS nodules which are less distinct in bigger nodules with coexisting inflammation of the surrounding synovial tissue. Therefore, we have empirically implemented in our practice the following algorithm:

1. Fully arthroscopic synovectomy has been used by surgeons experienced in arthroscopic techniques and/or for LPVNS with small nodules (<3 cm) .

2. Arthroscopically-assisted mini-open synovectomy has been used by less experienced surgeons and/or for larger nodules (>3 cm).

We acknowledge several limitations in the present study. This is a retrospective study which comprised all the potential bias of a retrospectively designed study. A relatively small number of patients were included in the study and the sample size could not lead to safe statistical conclusions. However, the disease is rare and RCTs are difficult to conduct. On the other hand, the two groups were similar and comparable with respect to demographic and disease characteristics. The sample size of both groups are quite sufficient in comparison with other studies and in relation to the rarity of the disease. The advantage that strengthened this study was also the long follow-up (5–20 years), which decreased the possibility of underestimating late recurrence. To our knowledge, this study is the only Level III study that directly compared two different techniques for the treatment of LPVNS. It is one of the largest series of LPVNS of the knee to date and the only study that reported on the mini-open arthroscopically-assisted synovectomy for the treatment of LPVNS.

Conclusion

Although arthroscopic resection of localized PVNS is considered the gold standard treatment, arthroscopically-assisted mini open partial synovectomy is a safe alternative treatment, especially for surgeons without extended experience in

arthroscopic techniques. The arthroscopic localization of the precise position of the lesion and its subsequent mini-open excision is a safe and effective technique with very low morbidity and recurrence rate and equivalent functional outcome to fully arthroscopic excision due to limited incision.

Compliance with ethical standards

Conflict of interest All authors have no competing interests related to the manuscript.

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Ethical approval This article does not contain any studies with human participants or animals performed by any of the authors.

Informed consent As a retrospective study, informed consent was not obtained from the participants included in the study.

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