Clinical outcomes of patients with pigmented villonodular synovitis of the shoulder after arthroscopic synovectomy

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Abstract

Background: Shoulder pigmented villonodular synovitis (PVNS) is a severe clinical condition, while few studies have focused on this situation due to its rarity. This study was to investigate the efficacy of arthroscopic treatment of patients diagnosed with shoulder PVNS.

Methods: From Jan 1st, 2010 to Dec. 31st, 2019, 6 patients (5 females and 1 male) diagnosed with shoulder PVNS underwent arthroscopic synovectomy in our hospital and combined rotator cuff repair was performed in 3 of them. The outcomes of this study include Constant score, Visual Analogue Scale (VAS), University of California, Los Angeles (UCLA) score and American Shoulder and Elbow Surgeons (ASES) score. The data were retrieved from the patients’ medical records.

Results: With a mean follow-up of 52.0 months (range, 28–92 months), the mean difference of Constant, VAS, UCLA and ASES scores were 27.83±21.60, 2.83±2.56, 11.67±10.93 and 17.83±25.35, respectively. Statistically significant improvements were detected in all the patient-reported outcomes except ASES score. One of the patients suffered from recurrence. Two patients suffered from mild complications after the surgeries while both of them achieved satisfactory recovery finally.

Conclusion: Arthroscopic synovectomy in the setting of shoulder PVNS can improve patients’ function. A concurrent rotator cuff repair is recommended if it is needed. The conclusion still needs testifying by further high-quality research with larger sample size.

Keywords: Arthroscopy, Synovectomy, Pigmented villonodular synovitis

Introduction

Pigmented villonodular synovitis (PVNS) is an unusual clinical condition characterized by benign tissue proliferation that involves synovium, tendon sheath, and bursa [1, 2]. The most affected joints are the knee and the hip [3]. The chief complaints include pain, swelling, stiffness, and limited range of motion in the affected joints [4]. Though a rare condition, this benign, highly proliferative neoplasm may result in massive joint destruction [1, 5]. What is more, patients reported by previous studies suggests that shoulder PVNS is often accompanied with massive rotator cuff tear [1, 2]. The poor rotator cuff condition would not only further impair the function of shoulder joint and seriously reduces the patients’ life quality, but also strongly influence the final clinical outcomes.

Shoulder PVNS is known to be extremely rare, accounting for about 2.0% to 8.0% in all PVNS cases...
Accordingly, few studies have focused on this situation, and, as a result, there is no accepted guideline for the treatment of PVNS as far as we know. Currently, the mainstay of treatment for PVNS for the vast majority of patients is arthroscopic and/or open surgery resecting the synovium [4]. Noailles et al [5] conducted a systematic review and suggested that arthroscopic excision of the lesions resulted in a satisfactory clinical outcome without recurrence in shoulder PVNS, but they also stated that it was difficult to recommend open debridement or arthroscopic synovectomy based on current evidence.

This retrospective case series aimed to report the clinical outcomes after arthroscopic synovectomy with/without rotator cuff repair in PVNS of the shoulder. We hypothesized that arthroscopic synovectomy would demonstrate good clinical outcomes in shoulder PVNS.

Materials and methods

Patients
The diagnosis of PVNS was considered when the patient presented the following symptoms before surgery: (1) pain and/or swelling in the affected joint, (2) locking, catching, and instability in the joint, (3) show an increase in the soft tissue density and/or lower bone density around the associated shoulder in plain radiographs, and (4) low signal intensity related to hemosiderin increase in nodule content and high signal intensity related to the increase in fat tissue in magnetic resonance imaging (MRI). The final diagnosis was made based on pathology of the tissue [7]. Patients who underwent open surgery, had bony lesions caused by other diseases or refused surgical treatment were excluded.

Based on symptoms, preoperative MRI and intraoperative pathology of tissue sampling, a total of 544 patients were diagnosed as having PVNS in our hospital from Jan 1st, 2010 to Dec. 31st, 2019. Fourteen (2.6%) of them suffered from shoulder PVNS and 6 underwent arthroscopic synovectomy (Figure 1). A rotator cuff repair was performed when necessary.

Surgical technique
All the operations were performed by one senior surgeon. The patients were positioned in the lateral decubitus position and the affected shoulder was stretched. After general anesthesia, a standard posterior portal and an anterior central portal were established as described in the previous study [8]. An arthroscopic examination was then performed. The mass was removed using a grasper or shaver as well as the peripheral synovitis and the rest of the lesion (Figure 2A&B), and then a thorough synovectomy with debridement was performed with an arthroscopic shaver. Hemostasis was achieved by an ablation device prior to the completion of the procedure. Attempts were made in every surgery to completely resect the pathologic appearing tissue. The effect after removing the lesions and synovium is shown in figure 2C. Tissue samples of synovial tissue were stained

![Flow Diagram](image_url)
with hematoxylin and eosin (HE) stain and examined under a light microscope by a senior pathologist.

After the lesions and hyperplastic synovium were removed, rotator cuff tears were found in 4 cases (case 1, case 3, case 4 and case 6) and the long head of biceps was dislocated and slack in case 1 (Figure 2D). Rotator cuff repair was performed in three patients (case 1,3 and 4). For case 1, after resection of the pathologic tissue and freshness of the insertion site on the greater tuberosity, rotator cuff was repaired by single row with three double loaded 4.5mm anchors (Smith&Nephew, USA). For case 3, The long head of biceps was placed as the Chinese-way described to reconstruct the superior capsular [9]. Then rotator cuff was partially repaired by double row using suture bridge technique by two double loaded 4.5mm anchors and two one footprint ultra PK suture anchors (Smith&Nephew, USA) [10]. For case 4, a limited acromioplasty was performed firstly and then rotator cuff was repaired by two medial suture anchors and one footprint anchor (Smith&Nephew, USA) by suture bridge technique. The proximal ends of the torn tendons retracted back to the glenoid rim in case 6, which was regarded as irreparable rotator cuff tear. Rotator cuffs were intact in other cases.

Postoperative rehabilitation
For patients who underwent both rotator cuff repair and synovectomy, the affected shoulder was immobilized in an arm sling for 2 weeks and the patients could only move their elbows and wrists in this period. After that, passive rehabilitation was applied under the protection of the brace while active exercise started from the 6th week. Resistance exercise was started 12 weeks after the surgery.

For patients who underwent synovectomy only, there was no strict limitation and active range of motion exercise could be performed immediately after the surgery.

Outcomes and statistical analysis
The outcomes of this study include Constant score, Visual Analogue Scale (VAS), University of California, Los Angeles (UCLA) score and American Shoulder and Elbow Surgeons (ASES) score. The data were retrieved from the patients’ medical records. Minimal clinically important difference (MCID) was taken into consideration and the MCID of Constant score, UCLA score, ASES score and VAS were set at 6.3, 3.0, 15.2 and 1.4 respectively according to previous studies [11–13].
The data were analyzed with IBM SPSS Statistics (Version 25.0.0.1). The scores before the surgery and at the final survey were compared and tests for equal variances and normality were performed first. Paired-samples t test was adopted for variables with equal variances. Other way, independent t test was employed. A P value of < 0.05 was considered significant.

Results

Patients

The baseline information of the patients is shown in Table 1. Five of the patients were female and the other one was male. The typical radiological manifestations of PVNS are shown in Figure 3. Their mean age at first surgery was 50.67 years (range, 30-65 years). The mean body mass index (BMI) was 22.29 kg/m² (range, 18.03-28.76 kg/m²). The average follow-up time was 52.0 months (range, 28-92 months). Among them, 4 patients (case 1, case 3, case 4 and case 6) were diagnosed to have rotator cuff tear based on MRI.

Operative findings

All the shoulder joints were filled with brownish synovial fluid and hemosiderin. The reddish-brown synovium was hyperplastic with finger-like villous projections on it (Figure 4A). Typical villonodular lesions were found in 3 cases (case1, 2 and 5). In these 3 patients, more than 5 typical villonodular lesions were

Table 1  Baseline information of patients

| Case No | Age at first surgery, y | Gender | BMI, kg/m² | Side | Size of rotator cuff tear | Type at first diagnosis | Radiotherapy after surgery | Follow-up, mo | Recurrence |
|---------|-------------------------|--------|------------|------|--------------------------|------------------------|---------------------------|---------------|-----------|
| 1       | 45                      | F      | 19.72      | Right | Massive                  | Localized              | No                        | 92            | Yes       |
| 2       | 30                      | F      | 18.03      | Left  | NT                      | Localized              | Yes                       | 62            | No        |
| 3       | 65                      | F      | 28.76      | Right | Massive                  | Localized              | No                        | 44            | No        |
| 4       | 62                      | F      | 23.56      | Right | Massive                  | Localized              | No                        | 40            | No        |
| 5       | 38                      | F      | 21.45      | Right | NT                      | Localized              | No                        | 28            | No        |
| 6       | 64                      | M      | 23.44      | Right | Massive                  | Localized              | No                        | 46            | No        |

BMI: body mass index, M male, F female, NT no tear

* In the subsequent recurrence of the disease, the left shoulder was also affected

* In the subsequent recurrence of the disease, the type changed into diffuse

Fig. 3  Radiological manifestations of shoulder PVNS. A Plain film shows low bone density in the front part of humeral head. B, E MRI shows low signal intensity related to hemosiderin increase in nodule content around shoulder cavity and high signal intensity related to the increase in fat tissue. PVNS, pigmented villonodular synovitis, MRI, magnetic resonance imaging
found within the rotator cuff and bursa mucosa in the articular cavity (case 1), around the tendon of subscapular muscle and long head of biceps (case 2), and in the subacromial space (case 5) (Figure 4B). Most of the nodules were small except the one found in case 2, whose size was about 7cm × 4cm × 2cm.

Pathologic findings
The PVNS diagnosis were confirmed in all patients by final pathologic findings (Figure 5). Histopathologic specimens obtained from the operations had papillary projections of synovial tissue accompanied with hyperplasia. Infiltration of monocytes, plasma cells, lymphocytes were seen. The lesions were composed of matted villi in which thin-walled vascular channels could be seen. The supporting stroma were packed with polyhedral stromal cells, multinucleate giant cells and macrophages.

Functional scores
The functional scores at both first visit and final follow-up were normally distributed. All patients achieved MCID in Constant score and UCLA score improvement except case 6. When all the patients were included in the analysis, statistically significant improvements were detected in all the patient-reported outcomes (PROs) except ASES score. After case 6 was excluded, whose rotator cuff tear was irreparable, significant improvements were found in all PROs including Constant score, VAS, UCLA score and ASES score from pre-surgery to the study endpoint. The clinical outcomes were summarized in Table 2.

Recurrence and complications
Among the 6 included patients, one reported recurrence of shoulder PVNS (case 1). She underwent two more arthroscopic surgeries after the first operation and the disease progressed from localized PVNS to diffuse PVNS at recurrence.

![Fig. 4](image1.jpg)
**Fig. 4** The operative findings during the surgery. **A** The reddish-brown synovium was hyperplastic with finger-like villous projections on it. **B** A lesion (yellow) in the subacromial space and angiogenesis was detected.

![Fig. 5](image2.jpg)
**Fig. 5** Pathological findings of the samples. **A** Photomicrograph (original magnification, × 100; hematoxylin–eosin [H-E] stain) shows villonodular fronds with overlying synovial tissue. **B** Photomicrograph (original magnification, × 400; hematoxylin–eosin [H-E] stain) shows hemosiderin-stained multinucleated giant cells.
Two patients suffered from post-operative complications after the surgeries. Case 1 presented finger numb after the second surgery and the reason may be that brachial plexus was injured during the resection process when we used grasp removed a lesion in axillary capsule region that infiltrated out of axillary capsule. Fortunately, the symptom relieved after approximately 3 months. She stated that the symptom did not significantly disturb her daily life. Case number 2 suffered from rupture of the biceps brachii due to the inevitable injury of long head of biceps (LHB) during the surgery. It was found that the lesion around the tendon of LHB which was described in the operative findings part had invaded into the tendon so that part of the LHB had to be removed(Figure 6). Fortunately, she did not get a Pop-eye sign. The patient was treated with conservative treatment in another hospital and got satisfactory results.

**Discussion**

PVNS is a rare benign tumor characterized by synovial proliferation [5]. Shoulder PVNS is rather rarer [2]. This retrospective case series proved the low ratio of shoulder PVNS in all PVNS cases, which is as low as 2.6%. Furthermore, by reviewing the medical records of these patients,
the clinical outcome of the arthroscopic synovectomy was relatively satisfying based on this study.

Despite the rarity and insignificant metastatic potential, shoulder PVNS can lead to rapid destruction of cartilage and bone and may lead to subluxation and pseudo-paralysis of the affected shoulder [14, 15]. Currently, the mainstream treatments include open and arthroscopic synovectomy [5]. Meanwhile, patients who have shoulder PVNS usually have concomitant rotator cuff tear as well [1]. This is possibly caused by the proliferating nature of PVNS, which can aggravate the degeneration of the tendons. The results of this study demonstrate that arthroscopic synovectomy combined with rotator cuff repair if needed could provide a statistically significant improvement of the patient reported outcomes in patients with shoulder PVNS. What is more, most of the improvements in postoperative scores met or exceeded MCID, which means substantial improvements in patients' quality of life were acquired.

As early as 2001, Mahieu et al. [16] reported 2 cases of shoulder PVNS and concluded that arthroscopic synovectomy was at least as effective for synovectomy compared with open surgery. Gumina et al. [1] conducted a study in which patients with shoulder PVNS combined with massive rotator cuff tear was treated by arthroscopic synovectomy and debridement and they concluded that coexistence of irreparable rotator cuff tear may result in poor functional outcomes. In this study, 4 patients in total had rotator cuff tear and 3 of them received rotator cuff repair. All these 3 patients achieved satisfactory recovery, while the one who received isolated arthroscopic synovectomy got worse PROs after the surgery, implying that the repair is the right option when it could be done.

Using MCID as an assessment tool is one of the strengths of this study. Chiang et al. [17] conducted a retrospective case series in 2009. Like this study, the data of 5 patients with shoulder PVNS were retrospectively collected. They reported that all these patients gained symptomatic and limited functional improvement [17]. However, they did not take MCID into consideration. The study population should be taken into consideration when applying MCID [18, 19]. Since no previous study has decided the MCID in arthroscopic synovectomy of shoulder joint, we used the standard for this study, which is reasonable because of the close association between shoulder PVNS with rotator cuff tear [1]. In this research, the differences of patient-reported outcomes reach MCID in all of the patients except that of ASES score and VAS. The reason for the exception of ASES score may be that the it is a questionnaire focusing on shoulder dislocation and function of shoulder only accounts for 28% of it. The VAS improvement of patient No. 1 did not reach MCID may be due to the repeated surgeries and failed full repair after several times synovectomy so that the remaining tendon quality was not good. In a word, the improvement that arthroscopic synovectomy provides is probably clinically significant based on the results of this study.

Postoperative complications in this study include finger numb (case 1) and rupture of LHB (case 2). The appearance of these complications can be explained by the location of the lesion. Part of the first patient's lesion located around brachial plexus in axillary fossa and the nerve might be injured during the surgery. The lesion of the second patient invaded the LHB and half of the LHB was resected, which weakened the tendon and increased the risk of tendon rupture.

There has been very little literature discussing shoulder PVNS since it is rarely seen and most studies were reported in the form of case report. Compared with open procedures, arthroscopic procedures have certain advantages including less bleeding, less trauma and shorter recovery period. Arthroscopic synovectomy has been proved to be non-inferior to open synovectomy in hip PVNS [20]. Whether it is a viable option in shoulder PVNS is still worth exploring. Pereira et al. [2] reported a case of shoulder PVNS involved subacromial bursa, in which an arthroscopic synovectomy was performed to totally remove the synovium, fluid and loose bodies and the patient got satisfactorily recovered, that is comparable to this study. However, they did not report any specific outcomes such as functional scores of the patient. Serra et al. [21] reported a case of diffuse shoulder PVNS. The patient underwent an arthroscopic near-complete synovectomy and got pain relief and increase of range of motion. However, because of the incomplete synovial resection, the patient received further radiotherapy and presented free of symptoms at 1-month follow-up. Nevertheless, the patient refused further follow-up so that the outcome of a long-term follow-up is unavailable. To our knowledge, although only 5 patients and 8 surgeries were enrolled, this study is still one of the studies with the most cases of shoulder PVNS compared to previous case reports and case series focusing on shoulder PVNS [1, 2, 14, 15, 17, 21–23]. Furthermore, the mean follow-up time of this study is also one of the longest. These advantages have made this research relatively more convincing.

This study does have some limitations. Firstly, it is a retrospective case series with a small sample size from a single institution, and not all the patients underwent the exact same procedure. However, shoulder PVNS is known to be extremely rare and fewer than 40 cases have been reported based on our literature search. As a result, it is quite difficult to collect a larger number
of cases who underwent the exactly same procedure in a single institution. Secondly, this is not a comparative study, so that we could not directly compare the efficacy of arthroscopic and open synovectomy. However, the 3 patients who underwent open surgery went through other concurrent procedures such as humeral head replacement and bone defect filling, making it improper to include them in this study. Further comparative researches with large sample size are still needed. Third, the effect of radiotherapy is not included in this research although it is a safe and effective treatment for PVNS after incomplete resection [24]. The reason is that with only 1 patient went through radiotherapy after the surgery, we cannot draw a firm conclusion. Future studies with more cases treated with radiotherapy will be able to provide more knowledge of it.

Conclusion

Arthroscopic synovectomy in the setting of shoulder PVNS can improve patients’ function. A concurrent rotator cuff repair is recommended if it is needed.

Abbreviations
PVNS: Pigmented villonodular synovitis, MRI: Magnetic resonance imaging, BMI: Body mass index; HE: Hematoxylin and eosin; VAS: Visual Analogue Scale; UCLA: University of California; ASES: American Shoulder and Elbow Surgeons; MCID: Minimal clinically important difference; PRO: Patient-reported outcome; MD: Mean difference; CI: Confidence interval; LHB: Long head of biceps.

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Authors’ contributions
X. T. and J. L. designed the study. L. P. and T. L. analyzed the patient data. L. M. collected the patient data. Y. L. was a major contributor in writing the manuscript and analyzed and interpreted the patient data. All authors read and approved the final manuscript.

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Availability of data and materials
The datasets generated and/or analysed during the current study are publicly available due to privacy restrictions, but are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate
This study was approved by the Institutional Committee on Ethics of West China Hospital, Sichuan University. All authors confirmed that all methods were carried out in accordance with relevant guidelines and regulations. The Ethics committee of West China Hospital, Sichuan University waived the need for patient informed consent as data has been collected from the medical records.

Consent for publications
Not applicable in the declarations section.

Competing interests
The authors declare that they have no competing interests.

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