



Open or Arthroscopic Synovectomy Is the Preferred Management Option in Pigmented Villonodular Synovitis of the Hip Joint Without Evidence of Degeneration: A Systematic Review of 20 Studies

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Purpose: To provide an up-to-date systematic review on the treatment options for pigmented villonodular synovitis (PVNS) of the hip and provide a grade of recommendation using standardized systems. **Methods:** A systematic search of PubMed, Embase, Web of Science, and The Cochrane Library from the date of inception of each database through December 4, 2021, was performed. Studies that described the outcomes of treatment of hip PVNS were identified. These outcomes were discussed and synthesized by three reviewers, and a grade of recommendation was assigned. **Results:** Twenty studies were identified. Seven studies described arthroscopic synovectomy, eight studies described open synovectomy, nine studies described arthroplasty, and one study described osmic acid synoviorthesis. Synovectomy, either open or arthroscopic, had similar rates of disease recurrence. Hip arthroplasty had low rates of disease recurrence compared to synovectomy; however, it was associated with significant risk of aseptic loosening in the longer term. **Conclusion:** Synovectomy, either open or arthroscopic based on surgeon preference, is favored in the treatment of hip PVNS if there is no evidence of joint space narrowing. Arthroplasty should be considered in cases with joint space narrowing or recurrence following joint preservation therapy. There is insufficient evidence to support synoviorthesis either as monotherapy or adjuvant therapy. **Level of Evidence:** IV, systematic review of Level III and IV studies.

Introduction

Pigmented villonodular synovitis (PVNS) is a rare proliferative disorder of the synovium,¹ with a reported incidence of 1.8 per million person years in the

United States of America.² The knee joint is most commonly involved, followed by the hip joint, which accounts for 9 to 15% of cases.^{3,4} PVNS of the hip typically presents in young adults with chronic hip pain, swelling, or recurrent hemarthrosis; however, atypical symptoms, such as locking⁵ or compressive sciatic and femoral neuropathy⁶ have been reported. Previous literature demonstrated increased prevalence in Type 1 diabetics.⁷ Although usually benign and monoarticular, case reports of polyarticular involvement,⁸ as well as lung, abdominal, and vertebral metastases do exist.⁹⁻¹¹

The workup of patients with suspected hip PVNS involves careful history taking and examination, plain film radiography, and magnetic resonance imaging (MRI), and if suspicious, either an open or image-guided synovial biopsy.¹² The gold-standard diagnostic test for PVNS is histological, with hemosiderin-stained multinucleated giant cells and pigmented foam cells seen under high-power microscopy.³ Depending on the extent of synovial involvement, PVNS can be classified

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into diffuse or nodular subtypes, with potential influence on the management options. Even when adequately managed, there is a propensity for recurrence. The recurrence rate is highly variable, ranging from 8% to 60%.¹³

Despite the hip being the second most common location for PVNS, there is little consensus as to its management.¹⁴ The purpose of this study was to provide an up-to-date systematic review on the treatment options for PVNS of the hip and provide a grade of recommendation using standardized systems.^{15,16} We hypothesize that hip preservation techniques should be used when there are minimal signs of arthritis, whereas arthroplasty should be used where there is significant arthritis.

Methods

This systematic review was performed on the basis of the recommendations of Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA).¹⁷ This systematic review protocol was registered with PROSPERO (Registration Number: CRD42022301017).

We performed a systematic search of the literature across multiple databases, including PubMed, Embase, The Cochrane Library, and Web of Science, from the date of inception through December 4, 2021. For studies that described treatment strategies for patients with hip PVNS, both operative and nonoperative were included. The outcomes of interest were disease recurrence, functional scores, and complications. Exclusion criteria included research protocols, epidemiological studies, and studies that described pathologies that were unrelated to hip PVNS, studies describing associations or diagnostic modalities rather than treatment of hip PVNS, systematic or narrative reviews, and case series or studies with fewer than 5 patients with hip PVNS. Studies with less than 5 patients allocated to all treatment modalities were also excluded. Studies were not excluded on the basis of upon their published language or geographic location. Full-text articles not available in English were translated with the help of a translation service.

A literature search using the following search terms and Boolean operators (“pigmented villonodular synovitis” OR “PVNS” OR “giant cell”) AND (“hip”). The title and abstracts were then screened by two reviewers (T.C. and K.W.) independently for relevance and consideration into a provisional list. Further to that, the provisional list would then be assessed independently by the two reviewers after reading the full text for potential inclusion. If there were any discrepancies, the two reviewers would meet with a third reviewer (M.B.) and reach a consensus as to which articles would be included.

Data extraction was performed by the first reviewer and validated by the second reviewer (K.W.). Baseline characteristics of included studies, as well as the

Table 1. Grade of Recommendations

Grade	Description
A (Good)	Level I studies with consistent findings
B (Fair)	Level II or III studies with consistent findings
C (Poor)	Level IV or V studies OR conflicting evidence
	C_f Majority of studies supports the intervention
	C_a Majority of studies against the intervention
	C_i Conflicting studies with no clear majority
I (Insufficient)	Insufficient evidence to make any recommendation

outcome measures, were collected. Analysis of the data was performed based upon three subheadings—joint preservation therapy, arthroplasty, and nonoperative modalities. On the basis of the evidence, a grade of recommendation was assigned on the basis of the recommendations by Wright et al.¹⁶ If a “C” level of evidence was to be assigned, we would then proceed to a subscale, whereby a subscript “f”, “a”, or “c” was applied. These subscripts denoted that the evidence was “for”, “against”, or “conflicted”, respectively.¹⁸ A description of this is shown in Table 1. Subjective synthesis was performed for each outcome of interest, with confidence intervals calculated using the Clopper-Pearson method,¹⁹ with a two-tailed 95% confidence interval utilized if the proportion was greater than 0% and a one-tailed 97.5% confidence interval utilized if the proportion was 0%. The results were then displayed as a Forest plot. Lastly, the methodological quality of studies included was assessed independently by both reviewers using the methodological index for non-randomized studies (MINORS).²⁰

Results

Search Results

Two thousand one hundred and seventy-nine studies were identified using the initial search strategy, of which 721 duplicates were removed. From the remaining 1,458 studies, a further 1,176 were excluded by screening the titles and abstracts. Full-text analysis was performed for 282 studies, for which 20 studies met our inclusion criteria. The result of our literature search is displayed in Fig 1. A short summary of the key features of study design is shown in Supplementary Table 1. There was significant diversity between studies with poor overall methodology.

Hip Preservation Studies

Fourteen studies described hip preservation (non-arthroplasty) surgical techniques in the management of hip PVNS, of which there were 7 studies that described arthroscopic techniques²¹⁻²⁷ and 8 studies that described open techniques.²⁷⁻³⁴ One study described both open and arthroscopic techniques.²⁷

For studies describing arthroscopic techniques of synovectomy, two studies performed adjuvant

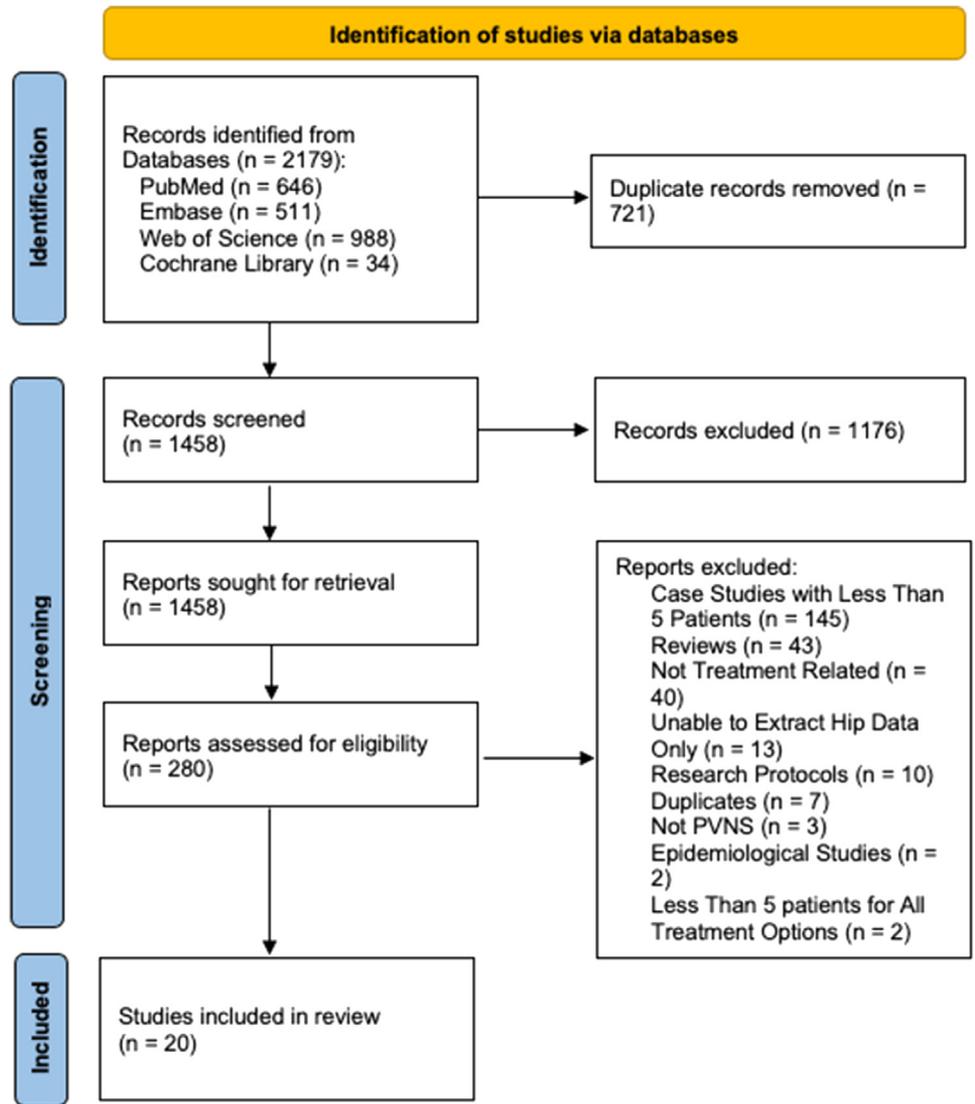


Fig 1. PRISMA flowchart.

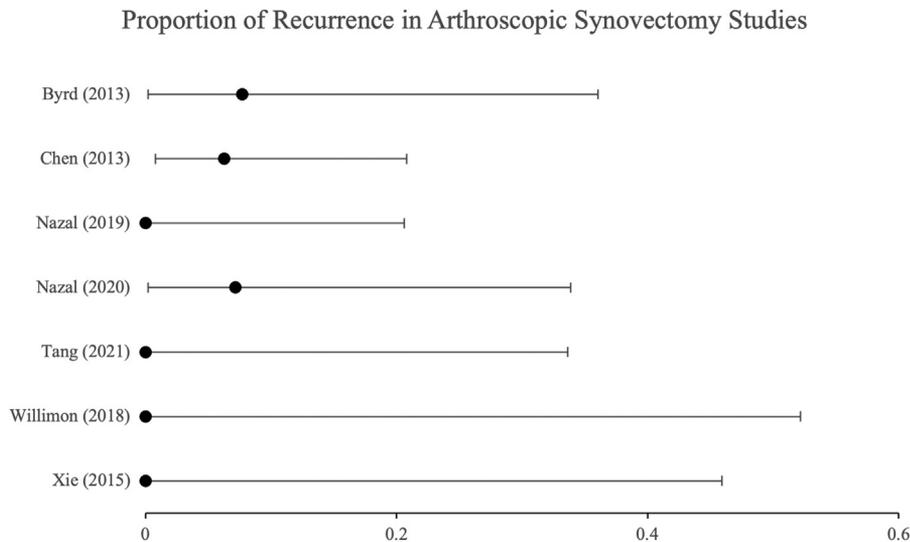


Fig 2. Forest plot for proportion of recurrence in arthroscopic synovectomy studies.

Table 2. Hip Preservation Studies

Author (Year)	Design (Level of Evidence)	Number of Hips	Adjuvant Therapy	Age (Mean, Range)	Gender (M/F)	Type (Diffuse/Nodular)	Final HHS (Mean \pm SD)	Recurrence Rate (%)	Notes	Follow-Up
Arthroscopic Synovectomy										
Byrd et al. (2013)	Case Series (IV)	13	None	26.8 (14 – 46)	9/4	NS	NS	1/13 (7.69%)	Improvement in HHS 27 ± 27	Mean of 17 months (Range: 2 – 60 months)
Chen et al. (2013)	Case Series (IV)	32	Synoviorthesis at 5 weeks postop for 5–6 weeks	41 (18 – 55)	NS	NS	NS	2/32 (6.25%)	One patient who had recurrent disease had repeat therapy, whereas another had arthroplasty	Minimum 24 months
Nazal et al. (2019)	Case Series (IV)	16	None	37 (25–54)	9/7	8/8	78.2 \pm 10.6	0/16 (0%)		Mean of 83 months (Range: 24–123 months)
Nazal et al. (2020)	Case Series (IV)	14	None	32.69	6/8	5/9	74.08 \pm 16.84	1/14 (7.14%)	Patient with recurrence had repeat therapy	Mean of 79.9 months (SD 22.4 months)
Tang et al. (2021)	Case Series (IV)	9	Radiosynoviorthesis (in 5 patients)	24.3 (14 –44)	2/7	5/4	94.6 \pm 4.9	0/9 (0%)		Mean of 55.8 months (range: 24 – 84 months)
Willimon et al. (2018)	Case Series (IV)	5	None	11 (7 – 17)	2/3	1/4	NS	0/5 (0%)	All paediatric patients	Mean of 31.8 months (Range: 12 – 63 months)
Xie et al. (2015)	Retrospective Cohort (III)	6	None	32.07	NS	NS	NS	0/6 (0%)		Median of 108 months

(continued)

Table 2. Continued

Author (Year)	Design (Level of Evidence)	Number of Hips	Adjuvant Therapy	Age (Mean, Range)	Gender (M/F)	Type (Diffuse/Nodular)	Final HHS (Mean \pm SD)	Recurrence Rate (%)	Notes	Follow-Up
Open Synovectomy										
Flipo et al. (1994)	Retrospective Cohort (III)	22	Osmic acid synoviorthesis (in 2 cases)	NS	NS	NS	NS	NS	Bad results in 7 cases 4 patients had arthroplasty, 1 had synoviorthesis, 1 had repeat synovectomy and 1 had observation only	Mean of 34 months (Range 4 – 94 months)
Hufeland et al. (2018)	Case Series (IV)	5	None	19.2 (8 – 28)	1/4	4/1	88.8	1/5 (20%)	Patient with recurrence had arthroplasty	Mean of 96 months (Range 35 – 141 months)
Moroni et al. (1983)	Retrospective Cohort (III)	6	None	23 (13 – 34)	1/4	NS	NS	0/6 (0%)		Mean of 32 months (Range 6 – 96 months)
Ota et al. (2021)	Retrospective Cohort (III)	7	None	NS	NS	NS	NS	0/7 (0%)	Data was part of a larger study	Median of 66 months
Schwartz et al. (1988)	Retrospective Cohort (III)	7	Unclear	36.9 (17 – 59)	NS	NS	NS	0/7 (0%)	Also had patients with arthroplasty	Up to 25 years
Schroder et al. (2012)	Case Series (IV)	5	Radiosynoviorthesis	31 (15 – 65)	NS	4/1	NS	1/5 (20%)	Patient with recurrence had arthroplasty	Mean of 32 months (Range 8 – 56 months)
Vastel et al. (2005)	Retrospective Cohort (III)	8	None	29.75 (23 – 41)	5/3	NS	NS	0/8 (0%)	Also had patients with arthroplasty 4 patients had arthroplasty for progression of arthritis	Mean of 187.5 months
Xie et al. (2015)	Retrospective Cohort (III)	37	None	32.07	NS	NS	NS	3/37 (8.11%)	2 patients with recurrence had repeat therapy and remaining one had arthroplasty	Median of 108 months

F, female; HHS, Harris hip score; M, male; NS, not stated; SD, standard deviation.

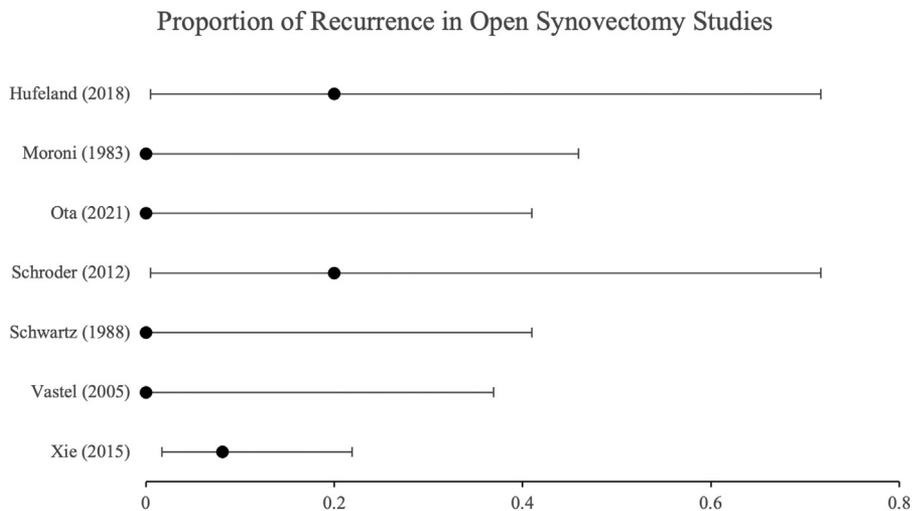


Fig 3. Forest plot for proportion of recurrence in open synovectomy studies.

synoviorthesis.^{22,25} Most of these studies were case series, with a limited number of patients. In total, there were 95 patients managed with arthroscopic synovectomy included in our analysis, with individual studies recruiting between 5 and 32 patients. There was a large variation in the mean age of patients, ranging from 11 to 41 years old. For studies in which the gender of participants was available, there were equal numbers of male and female participants. In terms of the type of PVNS, four of the seven studies displayed this information, with 19 patients having diffuse disease and 25 patients having nodular disease. The largest case series was performed by Chen et al. and had 32 patients. In this series, arthroscopic resection of diseased synovium was performed with synoviorthesis performed at 5 weeks postoperatively.²² Unfortunately, the agent used for synoviorthesis and the frequency of injection were

not clearly described. One of these series performed only arthroscopic synovectomies in pediatric patients.²⁶ The recurrence rate in our included studies ranged between 0 and 7.69% (Fig 2). Four patients had recurrence, of which 2 of these patients had a repeat arthroscopic synovectomy, 1 patient had conversion to arthroplasty, and there was no information on the remaining patient. The follow-up period was variable, with six of the seven studies having a mean follow-up of greater than 24 months. A summary of included studies is displayed in Table 2.

Open synovectomy technique was employed by eight authors, of which one study performed adjuvant radi-synoviorthesis for 6–8 weeks postoperatively.³¹ In our included studies, there was a total of 97 hips and the mean age of study participants ranged from 19.2 to 36.9 years old. The remaining demographical information

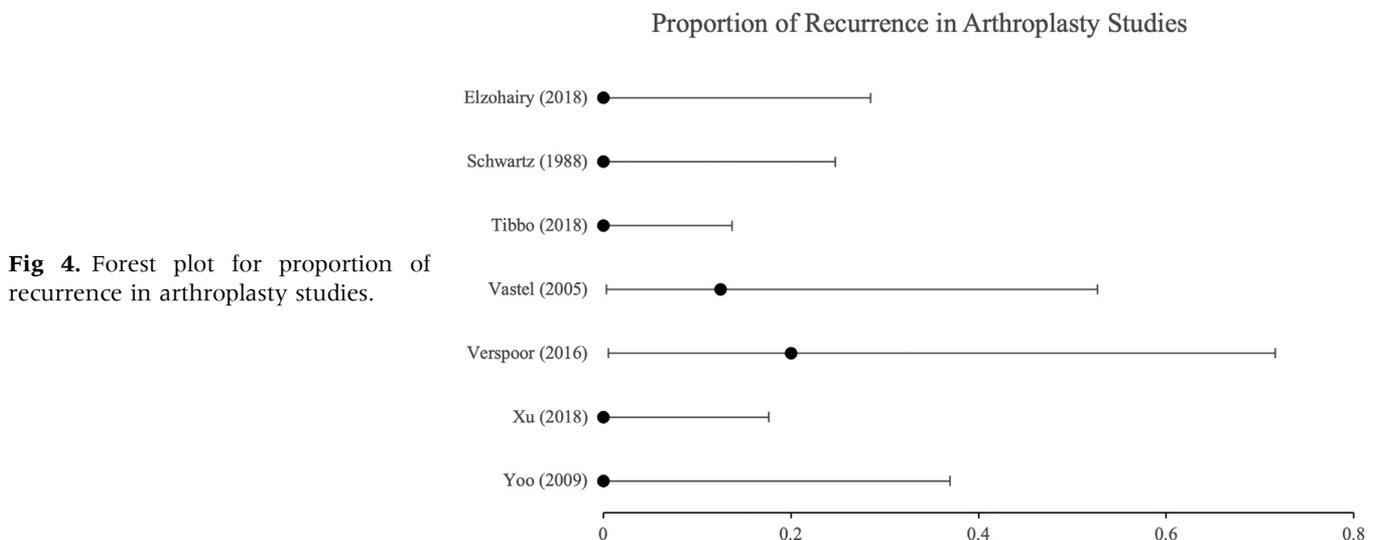


Fig 4. Forest plot for proportion of recurrence in arthroplasty studies.

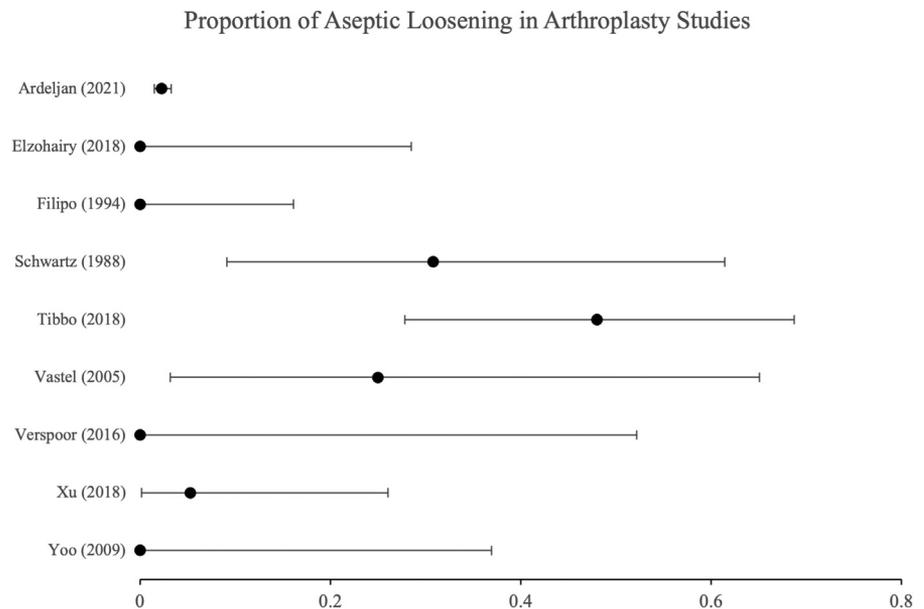


Fig 5. Forest plot for proportion of aseptic loosening in arthroplasty studies.

was unfortunately poorly collated. Only two studies described the type of hip PVNS, in which the majority had diffuse disease. Individual studies included were a mixture of retrospective cohort studies and case series and had between 5 and 37 participants, giving a total of 69 patients. The largest series was performed by Xie et al., who performed a retrospective cohort analysis on 237 patients with PVNS, for which, 37 patients had hip PVNS managed with open synovectomy.²⁷ On the basis of the data we collected, the recurrence rate was between 0 and 20% (Fig 3). Five patients had recurrence, of which 3 of these patients were managed with arthroplasty, and the remaining two had repeat open synovectomy.

Filipo et al. conducted a national survey of the management of hip PVNS in France, whereby open synovectomies were performed in 22 cases. Among these 22 cases, 12 patients received a total synovectomy only, 8 patients received a partial synovectomy only, one patient had a partial synovectomy with osmic acid synoviorthesis, and the remaining patient had a total synovectomy and osmic acid synoviorthesis. In their series, a good outcome was seen in 11 of the 13 total synovectomies, whereas this was reduced to 3 of the 9 partial synovectomies.³³ Concerningly, in one series by Vastel et al., half of the 8 patients who received open synovectomy had conversion to hip arthroplasty at the end of the study period (between 10 and 28 years) due to progression of arthritis.³² Three out of the four of these patients had evidence of early osteoarthritis. The mean follow-up was at least 5 years in almost all of the included studies. A summary of included studies is displayed in Table 2.

In summary, there was poor-quality evidence to support either open or arthroscopic synovectomy, with most demonstrating reasonably low recurrence rates with either hip preservation techniques (Grade C_f). There was insufficient evidence to suggest whether arthroscopic synovectomy should be favored in nodular disease or vice versa (Grade 1). There was also insufficient evidence to support the use of adjuvant synoviorthesis (Grade 1).

Hip Arthroplasty Studies

Nine studies described the use of hip arthroplasty for management of hip PVNS,^{30,32,33,35-40} of which 3 studies also described joint preservation techniques in separate cohorts.^{30,32,33} In terms of study design, there was a mixture of case control, case series, and retrospective cohort studies. The mean age was less heterogeneous between studies, ranging from 34.8 to 41.4 years old. Most studies performed a total synovectomy to remove all diseased tissue. One study only performed this for patients with “active” disease, defined by the presence of proliferative synovial tissue and histologically confirmed PVNS.³⁷

In total, 1,350 joints were investigated, with a large variation in the number of participants in individual studies. Of the seven studies that described the gender of the participants, there was a predilection for males, with 812 males and 504 females. Six of these studies described the indication for arthroplasty, in which most of them was performed for joint space narrowing.^{32,33,36,39,40} One study by Verspoor et al. performed arthroplasty for three of its five included patients due to extensive disease.⁴¹ Six studies

Table 3. Hip Arthroplasty Studies

Author (Year)	Design (Level of Evidence)	Number of Hips	Prosthesis	Adjuvant Therapy	Age (Mean, Range)	Gender (M/F)	Indication	Prior Treatment	Improvement in HHS (Mean)	Recurrence Rate (%)	Aseptic Loosening	Follow-up
Ardeljan et al. (2021)	Case Control (III) with non-PVNS as control	1240	NS	Unclear	NS	779/461	NS	NS	NS	NS	28/1240 (2.26%)	2 years
Elzohairy et al. (2018)	Case Series (IV)	11	Uncemented MOP (8)	Total Synovectomy	38.2 (30 – 50)	6/5	Lytic lesions with joint space narrowing in all patients	0/11	48.2	0/11 (0%)	0/ 11 (0%)	Mean of 7.2 years (5 – 10.5 years)
Flipo et al. (1994)	Retrospective Cohort (III)	21	Cup Arthroplasty (8) Unspecified Total (13)	Total Synovectomy	NS	NS	Joint destruction in total arthroplasty group	0/21	NS	NS	0/21 (0%)	Mean of 34 months (Range 4 – 94 months)
Schwartz et al. (1988)	Case Series (IV)	13	NS	Total Synovectomy	36.9 (17 – 59)	NS	NS	Unclear	NS	0/13 (0%)	4/13 (30.77%)	Up to 25 years
Tibbo et al. (2018)	Case Series (IV)	25	Cemented (7) Uncemented (13) Hybrid Cemented Stem (3) Resurfacing (2)	Synovectomy in 20 patients with active disease	39 (16 – 67)	9/16	NS	15/25	30	0/25 (0%)	12/25 (48%)	Up to 20 years
Vastel et al. (2005)	Retrospective Cohort (III)	8	Cemented (4) Cup Arthroplasty (3) Monopolar (1)	Total Synovectomy	41.4 (24 – 61)	4/4	Joint space narrowing in 6/8 patients	0/8	NS	1/8 (12.5%)	2/8 (25%)	Mean of 17.9 years
Verspoor et al. (2016)	Case Series (IV)	5	NS	NS	33.4 (16 – 49)	3/2	Extensive disease or arthritis	3/5	NS	1/5 (20%)	0/5 (0%)	Mean of 10.3 years
Xu et al. (2018)	Case Control (III) with non-PVNS as control	19	Uncemented COC (19)	Total Synovectomy	35.2 (22 – 58)	7/12	Cystic erosions and joint space narrowing in all patients	5/19	43.9	0/19 (0%)	1/19 (5.26%)	Mean of 8.7 years
Yoo et al. (2009)	Case Series (IV)	8	Uncemented COC (3) Uncemented COP (2)	Total Synovectomy	34.8 (20 – 68)	4/4	Complete obliteration of joint space in 5/8 patients	3/8	47.3	0/8 (0%)	0/8 (0%)	Mean of 8.9 years (Range 4.3 – 13.5 years)

COC, ceramic-on-ceramic; COP, ceramic-on-polyethylene; F, female; HHS, Harris hip score; M, male; MOP, metal-on-polyethylene; NS, not stated.

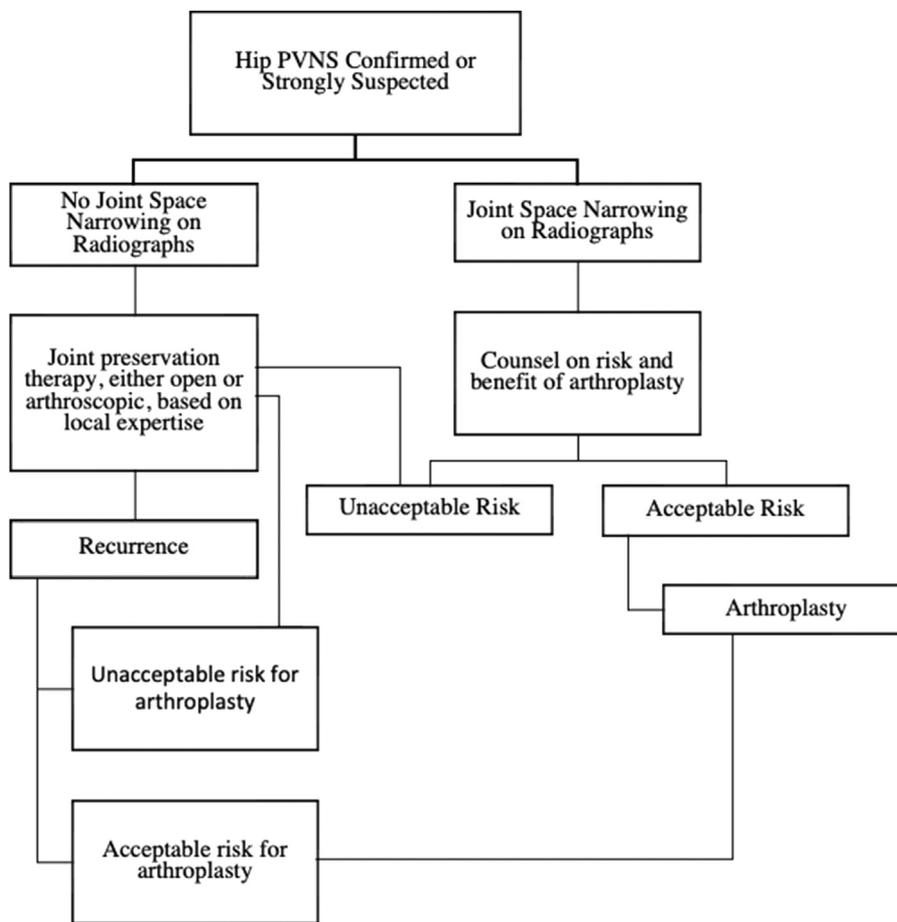


Fig 6. Recommended treatment algorithm.

described if patients had prior treatment for PVNS, for which 26/76 (34.21%) patients had received some form of treatment for hip PVNS prior to attempting arthroplasty. These included 21 cases of synovectomy (7 arthroscopic, 8 open, 6 unspecified), one case of synovectomy followed by resurfacing hemiarthroplasty, 2 cases of acetabular hemiarthroplasties and 2 cases of acetabular bone grafts.

The largest study we have included was a case control study by Ardeljan et al. of 1,240 participants with hip PVNS. The authors compared the outcomes of patients with hip PVNS undergoing arthroplasty with a general cohort. Of note, there was significantly increased odds of implant complications (aseptic loosening, revision,

and prosthetic joint infection) at 2 years. These patients also had a longer length of stay and increased odds of 90-day all-cause readmission.³⁵ Flipo et al. performed total hip arthroplasty in 13 patients and cup arthroplasty in 8 patients, for which the indication for total hip arthroplasty was joint destruction. Interestingly, poor outcomes were observed in half (4/8) of the cup arthroplasty patients, but none in the total hip arthroplasty group.³³ However, another study by Xu et al. used cementless hip arthroplasty with ceramic-on-ceramic bearing surfaces, due to the relatively younger ages of this patient cohort and the need to optimize component survivorship, demonstrated good survivorship in this small case series.³⁹

Table 5. Summary of Recommendations

Recommendations	Grade of Recommendation
Synovectomy (open or arthroscopic) should be performed in the treatment of hip PVNS if joint preservation is desired	Grade C_f
Insufficient evidence to support either open or arthroscopic synovectomy in the treatment of nodular disease versus diffuse disease, and vice versa.	Grade 1
Arthroplasty should be considered in patients with hip PVNS with joint space narrowing	Grade C_f
Arthroplasty should be considered in patients with hip PVNS with failed joint preservation therapy	Grade C_f
Insufficient evidence to support synoviorrhesis either as monotherapy or adjuvant therapy	Grade 1

In terms of overall outcomes, Harris Hip Score was reported in four studies with a mean improvement between 30 and 48.2 points. Recurrence rates were displayed in 7 studies, ranging from 0 to 20%, of which there were two cases of recurrence (Fig 4). Despite lower recurrence, aseptic loosening was reported between 0 and 48% of patients, necessitating revision (Fig 5). Three long-term follow-up studies by Schwartz et al. have demonstrated alarming rates of aseptic loosening of 4/13 (30.7%) at 25 years, 12/25 (48%) at 20 years and 2/8 (25%) at 14 years respectively.^{30,32,37} In most of these studies, the revisions were in patients who received hip arthroplasty with conventional polyethylene, cup arthroplasty, or cemented total hip replacements of the Charnley era. There are insufficient long-term data on uncemented hips with ultra-high molecular weight polyethylene (UHMWPE) as liners, although few studies seem to suggest a better survival.^{32,40} A summary of included studies is shown in Table 3.

In summary, our analysis suggests that arthroplasty should be considered in patients with hip PVNS with evidence of joint space narrowing, or in cases of failed joint preservation therapies. Although there is a lower incidence of disease recurrence compared to hip preservation techniques, there is an increased risk of aseptic loosening. It is possible that with recent advancements in technology, such as the use of UHMWPE liners or ceramic-on-ceramic constructs, the risk of this may be less; however, there is a lack of long-term data.^{36,39,40} Nevertheless, literature seems to support arthroplasty as a treatment modality in patients with evidence of significant hip degeneration, with adequate counselling on the risk and benefit (Grade C₁).

Nonsurgical Techniques

Only one study described nonsurgical techniques as monotherapy. Flipo et al. performed osmic acid synoviorthesis in 12 patients, of which 7 had a good outcome.³³ Four of their patients had a poor outcome, in which total synovectomy was performed in three cases, and repeat therapy was performed in one case. Radiosynoviothsis was described as adjuvant therapy in 2 studies to synovectomy;^{25,31} however, information on outcomes was only available for one study. In that study by Schroder et al., 1 of 5 patients who received open synovectomy with adjuvant radiosynoviothsis had recurrence.³¹ In summary, there is insufficient evidence to recommend synoviorthesis either as monotherapy or adjuvant therapy (Grade 1). This recommendation reflects a lack of studies, rather than a lack of efficacy.

Risk of Bias Analysis

Risk of bias analysis was performed using the MINORS criteria. Of note, almost all studies did not

perform prospective collection of data. However, most studies had a clearly stated aim, included consecutive patients, and had adequate follow-up. More than half of the studies had a less than 5% attrition rate at the conclusion of the study. The results of these studies are shown in Table 4.

Discussion

The findings of our systematic review suggest that patients with hip PVNS should be evaluated for evidence of substantial arthritic changes using plain-film radiographs. If there is minimal to no degeneration of the joint space, we recommend hip preservation surgery with either an arthroscopic or open synovectomy, depending on resources available. If there is substantial destruction of the joint, arthroplasty can be considered. Patients should be counselled on the risk and benefits of arthroplasty, specifically, the lower risk of recurrence in arthroplasty, but also the risk of aseptic loosening of the prosthesis in the longer term. In cases of failed hip preservation surgery, arthroplasty may be considered as an alternative to repeat synovectomy. Our proposed evidence-based decision algorithm is displayed in Fig 6, and a summary of our recommendations is shown in Table 5.

The goal of synovectomy is to ensure that all diseased tissue is removed to minimize the risk of recurrence. Although it could be argued that in cases of diffuse PVNS, synovectomy is best performed as an open procedure, the studies that we have included have not been able to substantiate this claim. Additionally, there is insufficient evidence to support adjuvant synoviorthesis. In cases of arthroscopy, the recommendation is to perform a T-capsular arthrotomy to increase the mobility of arthroscopic instruments, hence, allowing adequate resection of diseased tissue.⁴² Radiosynoviothsis or chemosynoviothsis has yet to be adequately investigated in the context of hip PVNS. There is evidence, however, to support the use of adjuvant radiosynoviothsis in patients with knee PVNS.^{43,44} Theoretically, in diffuse cases, there may be a role for adjuvant radiosynoviothsis or chemosynoviothsis as safe surgical dislocation may risk inadvertent damage to the medial femoral circumflex artery and, hence, leading to avascular necrosis.⁴⁵⁻⁴⁷ Ganz et al. have previously demonstrated that anterior dislocation with trochanteric flip osteotomy mitigated the risk of femoral head osteonecrosis.⁴⁸ This technique was used by Vastel et al.³² and Hufeland et al.,²⁸ with 8 and 5 patients respectively. Of these 13 patients, none developed femoral head osteonecrosis, with only one patient experiencing recurrence. There were limited studies on nonoperative measures, such as radiosynoviothsis, external beam radiotherapy, and systemic therapies for the management of hip PVNS. This is despite good evidence supporting the use of CSF-1 receptor inhibitors,

such as nilotinib and pexidartinib for PVNS that is not amenable to surgical resection.^{49,50}

Degenerative disease of the affected joint is not uncommon in patients with hip PVNS. Although the exact mechanism is unknown, evidence of bony lesions on radiographs in patients presenting with hip PVNS has shown to be significantly higher, at 89%, when compared to PVNS of other joints.⁵¹ The median time of diagnosis from the onset of disease has been shown to be 18 months.²⁷ As PVNS expands, pain and swelling lead to range of motion limitations.⁵² Histological analysis demonstrates mononuclear cells, macrophages with extensive hemosiderin stores, and multinucleated osteoclast-type giant cells. As repeated bleeding into the joint occurs, hemoglobin breaks down and deposits in the surrounding tissues.⁵³ The resulting hemosiderin-laden parenchyma leads to further joint destruction in a slow manner—as in hemophilia.^{54,55} In addition, PVNS has been shown to have neoplastic components. Translocations of chromosome 1p13 are present in the majority of PVNS cases with the endpoint effect of overexpressing colony-stimulating factor-1 (CSF-1). As CSF-1 becomes overexpressed, clusters of aberrant cells form to create focal areas of soft tissue hyperplasia in the synovial cells lining joints.⁵⁶ However, the proportion of patients with PVNS who have destructive disease requiring treatment is unknown.

In patients with advanced joint destruction and PVNS, it may be better if arthroplasty is performed. The series by Vastel et al. had demonstrated progression of arthritis in half of the synovectomy group, necessitating arthroplasty, despite no evidence of disease recurrence. Most of these patients had a degree of joint degeneration prior to synovectomy.³² In terms of prosthesis, there is insufficient evidence at present to support either cemented or uncemented implants, or specific bearing surfaces. The increased rate of aseptic loosening was previously thought to be due to the features of older-generation implants. It is hoped that newer uncemented implants would allow bony ingrowth, alongside improved bearing surfaces, such as ceramic or highly cross-linked polyethylene would generate less debris and, hence, reduce the risk of loosening.

Limitations

There are several limitations in our study. First, all of the included studies were retrospective case series, case control, or cohort studies. Such studies of PVNS of the hip are relatively uncommon. However, these studies lack random allocation to study groups, which, in turn, increases the risk of selection bias. The diverse nature of included studies, in combination with poor research methodologies, precluded a formal meta-analysis. Instead, we have displayed our results as Forest plots with summary estimates removed, and we have avoided pooling of outcomes in our discussion, in line with

recommended practice.⁵⁷ With such small cohorts, it is likely that surgeon experience or preference may have played a role in the allocation of one intervention over the other. Second, there may have been publication bias. To reduce the risk of publication bias, we extended our search to wider databases, such as the Web of Science. Third, some of our included studies had a poor display of demographic data, which diminished our ability to perform more detailed analysis. A few older studies utilized cup arthroplasty, as opposed to conventional hip arthroplasty, which may have led to increased rates of adverse outcomes. Lastly, a major limitation was that there was a single large study by Ardeljan et al.,³⁵ containing 86% of the patients in this review, which had the potential to outweigh other studies. This speaks to the rarity of the condition being investigated.

Conclusions

Synovectomy, either open or arthroscopic based on surgeon preference, is favored in the treatment of hip PVNS if there is no evidence of joint space narrowing. Arthroplasty should be considered in cases with joint space narrowing or recurrence following joint preservation therapy. There is insufficient evidence to support synoviorthesis either as monotherapy or adjuvant therapy.

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