

Treatment of Pigmented Villonodular Synovitis of the Knee

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Purpose: We aimed to determine the rate of local recurrence, the rate of postoperative complications, and the functional outcome at final follow-up of surgical and nonsurgical treatment approaches for pigmented villonodular synovitis (PVNS) of the knee. **Methods:** Medline, Embase, and the Cochrane Library were systematically searched for studies that reported the results of treatment for any type of PVNS between January 1, 1950, and August 1, 2013. Two authors extracted the data independently using predefined data fields including study quality indicators. **Results:** Sixty studies (1,019 patients) met the inclusion criteria. Thirty-five presented data on the treatment of localized pigmented villonodular synovitis (LPVNS), 40 on diffuse pigmented villonodular synovitis (DPVNS), 1 on extra-articular LPVNS, and 7 on DPVNS with extra-articular involvement. Many therapeutic options were reported. Depending on these options, DPVNS recurred in 8% to 70% of the series and LPVNS recurred in 0% to 8% of the series. For LPVNS, the 2 most-reported options were open localized synovectomy and arthroscopic local synovectomy. Between these 2 courses of treatment, no difference was found in terms of local recurrence (8.7% for open synovectomy and 6.9% for arthroscopic synovectomy) and postoperative complications (<1% for open synovectomy and 0% for arthroscopic synovectomy). For DPVNS, the 2 most-reported options were open total synovectomy and arthroscopic total synovectomy. Between these 2 courses of treatment, no difference was found in terms of local recurrence (22.6% for open synovectomy and 16.1% for arthroscopic synovectomy). However, we found a lower rate of reported complications between open synovectomy (19.3%) and arthroscopic synovectomy (0%). Internal irradiation or external beam radiation as an adjuvant treatment to surgical synovectomy seemed to decrease the rate of local recurrence in DPVNS cases with a high risk of recurrence. Finally, we found a great heterogeneity in the way the functional results were reported, and no valid conclusion could be made based on the data we extracted. **Conclusions:** We found no difference in local recurrence rates after open or arthroscopic surgery for either LPVNS or DPVNS. However, a lower rate of postoperative complications was reported after arthroscopic surgery for DPVNS. **Level of Evidence:** Level IV, systematic review of Level IV therapeutic studies.

Pigmented villonodular synovitis (PVNS) is a rare proliferative process that affects the synovial joints, tendon sheaths, and bursa membranes. In 1852 Chassaignac¹ reported the first case of a nodular lesion developing in the flexor tendon sheaths of the middle and index fingers. In 1864 Simon² described a large pedunculated nodule in the knee joint. In 1909 Moser³

provided a description of diffuse PVNS. In 1941 Jaffe et al.⁴ coined the term “pigmented villonodular synovitis”; then, Granowitz and Mankin⁵ expanded the terminology by distinguishing between the localized and diffuse forms of these synovial lesions. Recently, the World Health Organization indicated that PVNS and tenosynovial giant cell tumor are synonymous.

The estimated incidence of PVNS is around 1.8 per million population.⁶ PVNS is usually a monoarticular process that mainly involves the large joints. The knee joint is the most common location reported, but cases in the hip, ankle, shoulder, and elbow are regularly presented.^{4,6} PVNS is usually diagnosed in patients aged in their thirties or forties, and although it is believed to be more common in male patients, PVNS does not seem to have any gender-based predisposition.⁶⁻⁹ The clinical presentation of PVNS is highly variable, and the diagnosis is often delayed. The most effective screening device to obtain a diagnosis is magnetic resonance imaging.¹⁰ Recently, it was noted that PVNS/tenosynovial giant cell tumor cases commonly have over-expression of colony-stimulating factor 1.

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The aim of treatment of PVNS is to remove all abnormal synovial tissue to relieve pain, lower the risk of joint destruction, and avoid local recurrence. Several options have been proposed for the knee joint—from observation until total knee arthroplasty becomes necessary to external beam radiation, radioactive synovectomy, or surgical synovectomy. Depending on the synovial extension of PVNS, surgical synovectomy can be performed by open or arthroscopic techniques. Finally, some clinicians have suggested combined treatment. Unfortunately, because of the extremely low incidence of PVNS and the long delays before local recurrence, large randomized controlled trials have not been performed and no large studies have clearly shown any superiority of 1 option over another.

The purpose of this systematic review was to respond to the following questions: (1) What are the available treatment options for each type of PVNS? (2) What is the rate of recurrence of each treatment? (3) Is there one option with a lower recurrence rate, a lower rate of complications, and better functional outcomes?

Methods

A review protocol was created before the beginning of the study. Electronic databases including Medline (PubMed), Embase, and the Cochrane Library were searched using the terms “pigmented villonodular synovitis,” “knee,” and “treatment.” Our specific search strategy was as follows: [Synovitis, Pigmented Villonodular] AND [Knee Joint] AND [Therapeutics]. The final search was run on August 1, 2013. In addition, references for the relevant citations of selected articles were extracted. The inclusion criteria were studies in English, reporting the results of the treatment of localized pigmented villonodular synovitis (LPVNS), diffuse pigmented villonodular synovitis (DPVNS), or extra-articular PVNS of the knee joint confirmed by a pathologic examination, regardless of the study design. Only studies published after 1950 were examined because, before this period, the distinction between PVNS and other proliferative diseases had not been confirmed and treatment options were significantly different. The exclusion criteria were studies with fewer than 3 patients, studies with only patients aged younger than 15 years, and studies with mean follow-up of less than 12 months. There were no restrictions on the types of treatment.

Two authors (J-C.A. and S.K.) reviewed the titles independently. When the title was relevant, these authors scrutinized the abstract. When the abstract was relevant, they extracted the article and scrutinized the text in detail independently. At each step, any disagreement between the 2 authors (J-C.A. and S.K.) was examined and resolved by consensus of all 6 authors. A data extraction sheet was used for data collection; it was pilot tested for the first 10 studies included in

the review before final use. The extracted data from each selected study included the following: (1) characteristics of the study (design, time frame of inclusion); (2) characteristics of the study participants (age, gender, type of PVNS, number of previous recurrences, last follow-up); (3) type of treatment, such as observation, arthroplasty, external beam radiation, radioactive synovectomy, or surgical synovectomy (open localized synovectomy, open total synovectomy, arthroscopic localized synovectomy, arthroscopic total synovectomy, combined open and arthroscopic synovectomy), as well as any adjuvant treatment; and (4) type of outcome measurement (local recurrence, postoperative complications, knee function at last follow-up).

Because of the expected high rate of descriptive retrospective studies, the risk of bias was evaluated using the STROBE (Strengthening the Reporting of Observational Studies in Epidemiology) checklist to provide a score of 0 to 31 for each included study.¹¹ Although the STROBE criteria do not assess the quality of research, they provide a perspective on the quality of reporting that can be useful for the critical appraisal of published studies.¹¹ No study was excluded based on this score.

The primary outcome measure was the estimated rate of local recurrence for each type of treatment (total number of local recurrences at last follow-up/total number of patients treated). The secondary criteria were the rate of postoperative complications based on the same calculation and functional scores when available.

Results

Studies

A flow diagram for the selection of the studies is shown in Fig 1. Sixty studies were identified for inclusion in the review. No unpublished study was considered for inclusion.

Study Characteristics

Methods. Among the 60 studies included in the final analysis, 1 had a prospective design and 59 had a retrospective design (54 case series, 4 case reports with ≥ 4 patients, and 1 pattern-of-care study) (Appendix Table 1). Of the studies, 2 were published between 1950 and 1959, 2 between 1960 and 1969, 0 between 1970 and 1979, 6 between 1980 and 1989, 10 between 1990 and 1999, 29 between 2000 and 2010, and 11 after 2010. The mean follow-up period was 4.47 years (range, 0.5 to 13.5 years).

Participants. The included studies involved 1,019 patients. All patients had PVNS of the knee joint confirmed on pathologic examination: 323 had LPVNS, 562 had DPVNS, and 82 had PVNS with extra-articular involvement. The type of PVNS was unknown in 52 cases. One hundred twenty-two cases were local

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