Localized Pigmented Villonodular Synovitis of the Knee Joint: A Case Report

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Learning Point of the Article:
Localized Pigmented Villonodular Synovitis (LPVNS) is a very rare disease and it is silent and insidious, symptoms are non-specific, nevertheless, orthopedic surgeons should keep this entity in mind.

Abstract

Background: The localized form is characterized by local participation of the synovium as a nodule or pedunculated mass. The incidence rate of PVNS is estimated to be 1.8 per million people—the localized type is just one-quarter of that. The aim of this study is to remind orthopedic surgeons about the unusual properties of LPVNS located in the knee.

Case Report: A 39-year-old man presented to our clinic with pain and discomfort in his right knee. Magnetic resonance imaging showed an intra-articular mass in the infrapatellar region of the knee adjacent to the Hoffa fat pad. The mass was hypointense in the T1 sequence and heterogeneously hyperintense in the T2 sequence, which may be considered a local type of tenosynovial giant cell tumor (LPVNS). Excision was carefully performed without penetrating the tumor. The macroscopic appearance of the tumor was yellow-reddish and brown in color. Histopathologic examination revealed pigmented villonodular synovitis of the local type.

Conclusion: Even though the LPVNS of the knee is an uncommon intra-articular phenomenon, orthopedic surgeons should not overlook this lesion based on imaging findings, and open excision should be regarded as a reliable treatment option.

Keywords: Pigmented Villonodular Synovitis, Synovial Tumor Like Lesions, Knee pain, Intraarticular Mass.

Introduction

Jaffe et al. coined the term “PVNS” for the first time in 1941 [1]. Pigmented villonodular synovitis (PVNS) most frequently occurs in the knee as a proliferative phenomenon that involves the synovial membrane [2, 3]. It is considered a benign lesion of unknown etiology. Some authors postulate that it is probably caused by inflammation, trauma, toxins, allergies, clonal chromosomal abnormalities, or aneuploidy [4, 5]. The disease can be classified into two types: diffuse and localized. The diffuse form (DPVNS), as the name indicates, attacks practically the entire synovial cellular membrane of the affected joint. The localized form (LPVNS) is characterized by local participation of the synovium as a nodule or pedunculated mass. This type is generally a solitary mass of pedunculated or, much less frequently, 2–3 nodules yellowish-brown in color [6, 7, 8, 9, 10]. When LPVNS affects the knee, it is generally located in the anterior compartment [6, 7]. The incidence rate of PVNS is estimated to be 1.8 per million people—the localized type is just one-quarter of that [11]. Approximately 85% of PVNS occurs in the fingers, whereas 12% of the tumors are located in the knee, elbow, hip, and ankle [12]. PVNS can occur at any age, although it is most common between the ages of 30 and 50, with a female preponderance of 2:1 [13]. The aim of this study is to remind orthopedic surgeons about the
unusual properties of LPVNS located in the knee.

Case Report

A 39-year-old man entered our clinic with pain and discomfort in his right knee. The pain had persisted for 2 weeks, after a sprain of the right knee while playing soccer. A physical examination revealed mild pain and tenderness in flexion and extension, while the patellar tendon contracted in extension. Lateral and medial McMurray tests were negative. Routine laboratory tests including ESR, CRP, and serum uric acid levels were normal. Radiographs showed no bony pathology (Fig. 1). Magnetic resonance imaging (MRI) showed an intra-articular mass of 2.7 × 2.1 × 1.3 cm in the infrapatellar region of the knee adjacent to the Hoffa fat pad. The mass was hypointense in the T1 sequence (Fig. 2) and heterogeneously hyperintense in the T2 sequence (Fig. 3), which may be considered a LPVNS. Based on these findings, open surgical excision of the mass was decided. Under spinal anesthesia, the longitudinal midline approach with medial parapatellar arthrotomy was used. The mass was identified in the Hoffa fat pad (Video 1). Excision was carefully performed without penetrating the tumor, and the macroscopic appearance of the tumor was yellowish-red and brown in color (Fig. 4). A histopathologic examination revealed LPVNS (Fig. 5, 6). The patient was pain free 3 weeks after surgery. After a 1 year follow up, no recurrence had been detected.

Discussion

In this case presentation, we described the clinical and radiologic features of LPVNS and explained the rationale for treatment using open excision. A comprehensive literature search of localized tenosynovial giant cell tumor or localized pigmented villonodular synovitis of the knee since 1996 was performed, and 20 articles with 42 cases were found [10, 11, 14, 15, 16, 17, 18, 19, 20, 21, 22, 23, 24, 25, 26, 27, 28, 29, 30, 31] (Table 1). Only ten articles with ten cases involved the infrapatellar fat pads, excluding two pediatric patients [10, 14, 15, 17, 18, 19, 20, 21, 23, 28] (Table 2). The ten articles were analyzed to compare the quality of the evidence and the didactic features of the articles.

The majority of articles in the literature included histopathologic photographs, intraoperative photographs, and pre-operative MRI (Table 3). Rarely, authors reported conventional radiographs or macroscopic photographs of the tumor. Intraoperative video images of the tumor with applied exposure were not used in any article. In this article, we would like to add to the literature a visual teaching tool with a prepared video of photographs and a video of the case.

It is still debated whether the treatment of LVPNS should be arthrotomy or arthroscopy. There is no consensus on this issue. Considering the basic principles of tumor surgery, open surgery

Figure 1: Conventional lateral radiograph of the knee demonstrates no bony pathology.

Figure 2: Sagittal T1-weighted image, arrow shows hypointense mass located in Hoffa fat pad.

Figure 3: Sagittal T2-weighted image, the arrow shows heterogeneously hyperintense mass located in Hoffa fat pad.

Figure 4: Gross examination of the tumor mass: The yellowish-red and patchy brown colored macroscopic appearance of the tumor was noted. sizing 5.5 × 3 × 2 cm.

Figure 5: Tumor area consisting of mononuclear cells with histiocytoid appearance, accompanied by xanthoma cell clusters consisting of foamy histiocytes and fewer perivascular lymphocytic inflammatory cells are seen. (H-E Paint, ×100).

Figure 6: Histopathologic appearance of the excised lesion. Note the cell tumor consisting predominantly of mononuclear cells, rarely including multinucleated giant cells. Enlarged view of my previous picture. (H-E Paint, ×200).
seems to be more feasible than arthroscopy. This is because it has the advantage of excision of the tumor from the knee without penetrating and possibly seeding to the joint.

Although the total recurrence rate of PVNS varied from 7% to 44% [32, 33], one study reported a rate of 18% after arthroscopic treatment of LPVN within a 6–9 month follow-up [34]. The

### Table 1: Articles in the literature and the number of patients presented in these articles.

| S. No. | The authors, journal, and year                  | The title                                                                 | Number patients |
|-------|-----------------------------------------------|---------------------------------------------------------------------------|-----------------|
| 1     | Lucas. Arch Pathol Lab Med. 2012              | Tenosynovial giant cell tumor: case report and review.                     | 1               |
| 2     | Beytemur et al. Case Rep Orthop.2014          | Localized giant cell tenosynovial tumor seen in the knee joint.            | 1               |
| 3     | Camillieri et al. Orthopedics. 2012           | Intra-articular tenosynovial giant cell tumor arising from the posterior cruciate ligament. | 1               |
| 4     | Ghnaimat et al. Electronic Physician. 2016    | Giant cell tumor of tendon sheath in the knee.                            | 1               |
| 5     | Atik et al. Eklem Hastalik Cerrahisi. 2017    | Localized pigmented villonodular synovitis in a child knee.               | 1 (Pediatric age) |
| 6     | Kim et al. Clin Orthop Relat Res. 2000        | Arthroscopic treatment for localized pigmented villonodular synovitis of the knee. | 11              |
| 7     | Ajik et al. Arthroscopy. 2001                 | Localized pigmented villonodular synovitis of the knee.                   | 4               |
| 8     | Sun et al. Oncol Lett. 2012                   | Giant cell tumor of the tendon sheath: A rare case in the left knee of a 15-year-old boy. | 1 (Pediatric age) |
| 9     | Flevas et al. J Orthop Case Rep. 2021         | Arthroscopic removal of tenosynovial giant-cell tumors of the cruciate ligaments. Presentation of two cases. | 2               |
| 10    | Godoy FA et al. Rev Bras Ortop. 2015          | Localized pigmented villonodular synovitis: Case report.                  | 1               |
| 11    | Kim et al. Arthroscopy. 2005                  | Clustered localized pigmented villonodular synovitis.                     | 2               |
| 12    | Abdalla et al. Rev Bras Ortop. 2015           | Synovial giant cell tumor of the knee.                                    | 1               |
| 13    | Tu et al. Int J Surg Case Rep. 2022           | Tenosynovial giant cell tumor of cruciate ligament: A case report and review. | 1               |
| 14    | Xu et al. Int J Clin Exp Pathol. 2015         | Tenosynovial giant cell tumor arising from the posterior cruciate ligament: A case report and literature review. | 1               |
| 15    | Sheppard et al. Clin Imaging. 1998            | Giant-cell tumor of the tendon sheath arising from the posterior cruciate ligament of the knee: A case report and review of the literature. | 1               |
| 16    | Otsuka et al. Arthroscopy. 1996               | Tenosynovial giant-cell tumor arising from the anterior cruciate ligament of the knee. | 1               |
| 17    | Gülenç et al. Acta Chir Orthop Traumatol Cech. 2018 | Arthroscopic excision of tendinous giant cell tumors causing locking in the knee joint. | 7               |
| 18    | Lee and Wang. Clin Orthop Surg. 2014          | A tenosynovial giant cell tumor arising from femoral attachment of the anterior cruciate ligament. | 1               |
| 19    | Aksoy et al. Singapore Med J. 2009            | Tenosynovial giant cell tumor of the posterior cruciate ligament and its arthroscopic treatment. | 1               |
| 20    | Agarwala et al. J Clin Orthop Trauma. 2015    | A rare case of giant cell tumor arising from anterior cruciate ligament: its diagnosis and management. | 1               |

Total: 42

### Table 2: The articles according to anatomical location of localized pigmented villonodular synovitis of the knee.

| S. No. | The Authors, Journal And Year                  | Adjacent To The Pcl | Adjacent To The Ac | Retropatellar Area (Suprapatellar Pouch) | Lateral Gutter | Infra Patellar Fat Pad | Anteromedial Synovium Near The Anterior Horn Of The Medial Meniscus, And The Anterior Horn Of The Lateral Meniscus | Posteromedial Compartment, Medial Gutter | Posterior Medial Compartment |
|-------|-----------------------------------------------|---------------------|-------------------|------------------------------------------|----------------|------------------------|----------------------------------------------------------------------------------------------------------------------------------|--------------------------------------------|----------------------------|
| 1     | Agarwala et al. J Clin Orthop Trauma. 2015    | *                   | *                 | *                                        | *              | *                      | *                                                                                                                                 | *                                          | *                          |
| 2     | Lee and Wang. Clin Orthop Surg. 2014          | *                   | *                 | *                                        | *              | *                      | *                                                                                                                                 | *                                          | *                          |
| 3     | Gülenç et al. Acta Chir Orthop Traumatol Cech. 2018 | *                   | *                 | *                                        | *              | *                      | *                                                                                                                                 | *                                          | *                          |
| 4     | Otsuka et al. Arthroscopy. 1996               | *                   | *                 | *                                        | *              | *                      | *                                                                                                                                 | *                                          | *                          |
| 5     | Sheppard et al. Clin Imaging. 1998            | *                   | *                 | *                                        | *              | *                      | *                                                                                                                                 | *                                          | *                          |
| 6     | Xu et al. Int J Clin Exp Pathol. 2015         | *                   | *                 | *                                        | *              | *                      | *                                                                                                                                 | *                                          | *                          |
| 7     | Tu et al. Int J Surg Case Rep. 2022           | *                   | *                 | *                                        | *              | *                      | *                                                                                                                                 | *                                          | *                          |
| 8     | Kim et al. Clin Orthop Relat Res. 2000        | *                   | *                 | *                                        | *              | *                      | *                                                                                                                                 | *                                          | *                          |
| 9     | Camillieri et al. Orthopedics. 2012           | *                   | *                 | *                                        | *              | *                      | *                                                                                                                                 | *                                          | *                          |
| 10    | Godoy et al. Rev Bras Ortop. 2015             | *                   | *                 | *                                        | *              | *                      | *                                                                                                                                 | *                                          | *                          |
| 11    | Ajik et al. Arthroscopy. 2001                 | *                   | *                 | *                                        | *              | *                      | *                                                                                                                                 | *                                          | *                          |
| 12    | Sun et al. Oncol Lett 2012                    | *                   | *                 | *                                        | *              | *                      | *                                                                                                                                 | *                                          | *                          |
| 13    | Flevas et al. J Orthop Case Rep. 2021         | *                   | *                 | *                                        | *              | *                      | *                                                                                                                                 | *                                          | *                          |
| 14    | Atik et al. Eklem Hastalik Cerrahisi. 2017    | *                   | *                 | *                                        | *              | *                      | *                                                                                                                                 | *                                          | *                          |
| 15    | Kim et al. Arthroscopy. 2005                  | *                   | *                 | *                                        | *              | *                      | *                                                                                                                                 | *                                          | *                          |
| 16    | Abdalla et al. Rev Bras Ortop. 2015           | *                   | *                 | *                                        | *              | *                      | *                                                                                                                                 | *                                          | *                          |
| 17    | Aksoy et al. Singapore Med J. 2009            | *                   | *                 | *                                        | *              | *                      | *                                                                                                                                 | *                                          | *                          |
| 18    | Ghnaimat et al. Electronic Physician. 2016   | *                   | *                 | *                                        | *              | *                      | *                                                                                                                                 | *                                          | *                          |
| 19    | Lucas. Arch Pathol Lab Med. 2012              | *                   | *                 | *                                        | *              | *                      | *                                                                                                                                 | *                                          | *                          |
| 20    | Beytemur et al. Case Rep Orthop. 2014         | *                   | *                 | *                                        | *              | *                      | *                                                                                                                                 | *                                          | *                          |

Total sum 10 6 7 7 12 6 3

Total: 42
recurrence rate of 18% in a short period also causes us to favor open excision. In addition, the size of these tumors is usually not suitable for arthroscopic excision. In our case, we preferred open resection, because we were concerned that the tumor would not come out in one piece through the arthroscopic portal because of its size, and moreover, if the surgeon preferred a portion-by-portion excision, it would seed and recur.

**Conclusion**

Even though LPVNS of the knee is an uncommon intra-articular phenomenon, orthopedic surgeons should not overlook this lesion based on imaging findings, and open excision should be regarded as a reliable treatment option.

**Clinical Message**

LPVNS is a rare tumor that rarely affects the knee. However, when it does affect the knee, it is usually localized in the anterior compartment. Awareness and clinical sense are important in diagnosis. Orthopedic surgeons should prefer open excision if the tumor does not appear to come out of the arthroscopic portal in one piece.

### Table 3: The documentation details of the articles

| S. No. | The authors, journal and year | The title                                                                 | Conventional radiograph | MRI | Intra-operative photo or video | Macroscopy | Pre-operative clinical photo | Histopatology |
|--------|--------------------------------|---------------------------------------------------------------------------|--------------------------|-----|-------------------------------|------------|------------------------------|---------------|
| 1      | Lucas. Arch Pathol Lab Med. 2012 | Tenosynovial giant cell tumor: case report and review                      | --                       | *   | *                             | *          |                              |               |
| 2      | Beytemür et al. Case Rep Orthop. 2014 | Localized giant cell tenosynovial tumor seen in the knee joint.            | --                       | *   | *                             |            |                              |               |
| 3      | Ghnaimat et al. Electronic Physician. 2016 | Giant Cell Tumor of Tendon Sheath in the Knee.                            | *                        |     |                               |            |                              |               |
| 4      | Kim et al. Clin Orthop Relat Res. 2000 | Arthroscopic treatment for localized pigmented villonodular synovitis of the knee | --                       | *   | *                             |            |                              |               |
| 5      | Aşik et al. Arthroscopy, 2001 | Localized pigmented villonodular synovitis of the knee.                   | *                        | *   | *                             | *          |                              |               |
| 6      | Sun et al. Oncol Lett. 2012 | Giant cell tumor of the tendon sheath: A rare case in the left knee of a 15-year-old boy | *                        | *   |                               | *          |                              |               |
| 7      | Kim et al. Arthroscopy, 2005 | Clustered localized pigmented villonodular synovitis.                    | *                        | *   |                               |            |                              |               |
| 8      | Abdalla et al. Rev Bras Ortop. 2015 | Synovial giant cell tumor of the knee.1                                   | *                        | *   |                               | *          |                              |               |
| 9      | Gülenç et al. Acta Chir Orthop Traumatol Cech. 2018 | Arthroscopic excision of tendinous giant cell tumors causing locking in the knee joint | *                        | *   |                               | *          |                              |               |
| 10     | Atik et al. Eklem Hastalik Cerrahisi. 2017 | Tendiendous giant cell tumors causing locking in the knee joint.         | *                        | *   | *                             | *          | *                            |               |
| 11     | Current case | Tendiendous giant cell tumors causing locking in the knee joint.         | *                        | *   | *                             | *          | *                            |               |

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