The Journal of Foot & Ankle Surgery 54 (2015) 160–163

Contents lists available at ScienceDirect

The Journal of Foot & Ankle Surgery

journal homepage: www.jfas.org

Surgical Outcomes After Excision of Pigmented Villonodular Synovitis Localized to the Ankle and Hindfoot Without Adjuvant Therapy

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ARTICLE INFO

Level of Clinical Evidence: 4

Keywords:
ankle
AOFAS score
hindfoot joint
subtalar
synovium

ABSTRACT

Although a benign disorder, pigmented villonodular synovitis (PVNS) has a high rate of recurrence. Because of the high incidence of recurrence and concern about destruction of the affected joint, several adjuvant therapies have been promoted without a clear standard treatment strategy. We reviewed cases of PVNS affecting the ankle and hindfoot joints (ankle and/or subtalar joints) treated with surgical resection without adjuvant therapy in an effort to identify the incidence of PVNS recurrence after excision without adjuvant therapy. Of the 10 cases with a mean follow-up duration of 33.2 ± 19.8 months, 4 (40%) developed a recurrence, with a mean interval of 6 (range 3 to 14) months. At the final follow-up visit, the mean American Orthopaedic Foot and Ankle Society ankle-hindfoot score was 86.6 ± 12. The clinical outcomes of PVNS affecting the ankle and hindfoot joints are associated with a relatively high incidence of recurrence, and additional clinical investigation comparing the incidence of recurrence in patients undergoing excision versus excision with adjuvant therapy is needed for us to better understand this condition and provide more informed recommendations to our patients.

Pigmented villonodular synovitis (PVNS) is a relatively rare, benign, locally aggressive disorder of the synovium of the joints, bursae, and tendon sheaths (1). PVNS has been classified into localized and diffuse forms, which are thought to represent a spectrum of the same pathologic process. The localized form manifests as a pedunculated nodule within the joint, and the diffuse type insidiously infiltrates the synovial lining, leading to osseous erosion (1,2). The most prevalent sites of PVNS involvement include the knee, hand joints, and hip, followed by the ankle and shoulder (1,3–5). PVNS of the ankle joint constitutes approximately 2.5% of all cases (4), with presentations in the subtalar joint, tarsometatarsal bones, and toes relatively rare. A small number of studies have considered PVNS affecting the foot and ankle (6–10); however, these studies did not focus on the foot and ankle exclusively.

For localized forms of PVNS, complete localized synovectomy has been suggested owing to the low rate of relapse (11–15). For diffuse forms, surgical resection with total synovectomy has been classically proposed (12,14). Isotopic synoviorthesis or radiotherapy could be useful, especially for those patients who undergo incomplete synovectomy and for those with recurrence. However, serious complications have been associated with adjuvant therapy, including full-thickness skin necrosis after injection of yttrium-90 in the ankle after subtotal synovectomy (16). Moreover, local complications, such as postradiation fibrosis, swelling, and nonhealing wounds, and malignant transformation have been encountered after radiotherapy. As such, careful considering should be given to identifying those patients who might benefit the most from the use of adjuvant therapies, for whom the benefits of reducing the risk of recurrence will outweigh the risk of complications. Thus, we undertook a retrospective study of a series of patients who had undergone surgical excision of ankle or hindfoot PVNS without adjuvant therapy to determine the incidence of PVNS recurrence in this group of patients.

Patients and Methods

A review was performed of the data from all patients who had undergone surgery (by K.S.S.) for PVNS involving ankle and/or hindfoot joints at our medical center (Samsung Medical Center, Seoul, Republic of Korea) from March 2004 to January 2013. We searched the entire surgical schedule for the study period on our electrical medical information system with the keywords of excision or removal of soft tissue tumors of foot or ankle (by K.S.S. and K.R.K.). We found 23 cases of PVNS around the foot and ankle. Of these, the 10 cases of PVNS involving ankle or hindfoot joints were included in the present study. Their medical records, including the outpatient medical record, admission note, progress notes, operative reports, and radiographs and other diagnostic imaging studies (including ultrasonography [US] and magnetic resonance imaging [MRI]), and the patients' responses to questionnaires were used to analyze the clinical

Financial Disclosure: None reported.
Conflict of Interest: None reported.
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http://dx.doi.org/10.1053/j.jfas.2014.11.008

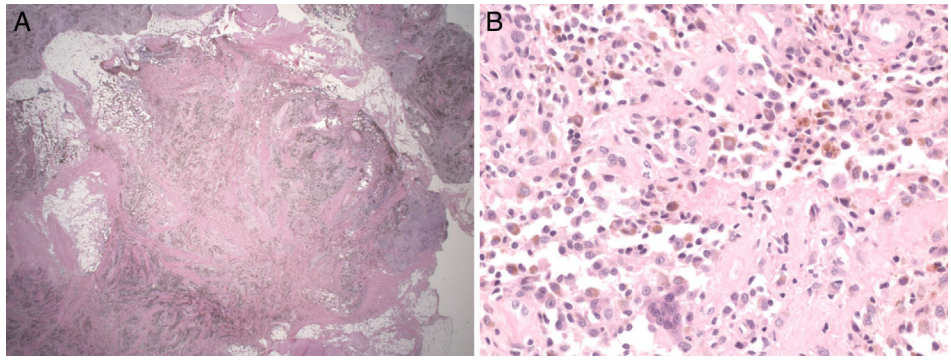

Fig. Histologic appearance of pigmented villonodular synovitis from patient 10 in our case series. *(A)* Low-power view (hematoxylin and eosin, original magnification ×10). *(B)* High-power view (hematoxylin and eosin, original magnification ×400). The mass appeared as nodules and/or villi on the low-power view, with an abundant number of pigmented hemosiderin-laden macrophages on the high-power view.

outcomes (by K.R.K.). In each case, a pathologic diagnosis of PVNS confirmed the diagnosis (Fig). The type of PVNS (i.e., localized or diffuse), was determined mainly using the MRI studies (by K.S.S.). All of the patients were followed up in the outpatient clinic.

Clinical and Radiologic Evaluations

Evaluation of the postoperative US and/or MRI scans and the clinical examination findings were used to identify PVNS recurrence. A patient was considered to have developed recurrence if these postoperative imaging studies detected the appearance of a new mass. Routine US was performed at 3 months postoperatively, with MRI studies performed on an as-needed basis. Clinical symptoms such as pain and swelling were also monitored to determine the potential for PVNS recurrence. A recurrence-free period was defined as the interval from the date of surgical resection to the date that follow-up imaging studies identified the presence of recurrent tumor. Any other postoperative complications (in addition to, or instead of, recurrence) were recorded. The American Orthopaedic Foot and Ankle Society (AOFAS) ankle-hindfoot scale (17,18) was used to assess the patients' pain, function, and alignment at the last follow-up visit.

Results

We identified 12 consecutive patients with PVNS localized to the foot or ankle. Of these 12 patients, 10 (83.3%) had undergone surgical resection, consisting of 9 open excisions and 1 arthroscopic excision, without adjuvant therapy. A statistical description of these 10 patients is listed in the Table. Of the 10 patients, 7 (70%) were male and 3 (30%) female. Their mean age was 37.7 ± 11.4 (range 18 to 54) years. The mean follow-up duration was 33.2 ± 19.8 (range 6 to 66) months. The right lower extremity was involved in 7 patients (70%) and the left in 3 (30%). Diffuse PVNS was present in 7 patients (70%) and 3 (30%) had localized PVNS. The PVNS was localized to the ankle in 6 patients (50%), the subtalar joints in 2 (20%), and the ankle and subtalar joints in 2 (20%). The most common initial presenting symptom was a painful, swollen ankle, present in 5 patients (50%). A nonpainful but palpable mass was present in 4 patients (40%) and a painful mass in 1 (10%). The mean AOFAS score was 86.6 ± 12 (range 63 to 100) for the overall group and 85.25 ± 18.1 (range 63 to 100) for the 4 patients (40%) who developed recurrent PVNS.

Surgical resections were performed using an open approach in 9 patients (90%) and arthroscopically in 1 (10%). For the single case with an arthroscopic approach, the debridement was performed using an arthroscopic shaver (patient 2). For the localized forms (patients 8 and 10), the lesion was excised using complete localized synovectomy through an open extensile exposure over the involved joint. For diffuse forms (patients 1, 3, 4, 5, 6, 7, and 9), the PVNS lesions were resected as much as possible using open synovectomy. The PVNS lesion in patient 4 involved both the ankle and the subtalar joints, and this was the only case in our series for which the senior author (K.S.S.) did not perform the initial operation. During the first operation (performed by another surgeon who was not involved with our investigation), according to the previous operation record, complete resection was impossible owing to extensive infiltration involving the distal tibiofibular syndesmosis, ankle, and subtalar joints. After recurrence development and referral to the senior author's (K.S.S.) practice, 4 months after the first attempt at excision, 2 additional operations were undertaken. At the latest follow-up examination, yet another operation was planned.

Our review identified 4 patients (40%) with recurrent PVNS. The mean recurrence-free interval after excision of the lesion was 6 (range 3 to 14) months. Of the 4 patients with recurrence, 2 (patients 1 and 2) had no discomfort or symptoms and had normal AOFAS scores of 100 points, despite recurrence confirmed on US (patient 1 at 16 months postoperatively) and MRI (patient 2 at 3 months postoperatively) studies. Thus, these 2 patients were monitored by periodic outpatient observation. In 1 patient (10%) with recurrence (patient 4), complete resection was not possible because of extensive PVNS infiltration into the distal tibiofibular syndesmosis, ankle, and subtalar joints at the initial operation (not performed at our institution), and 2 subsequent operations were necessary to treat the recurrence. At the latest follow-up examination, a fourth operation was scheduled, and the patient's latest AOFAS score was 63 points. In the remaining patient with recurrence (patient 9), a routine outpatient surveillance US study at 3 months postoperatively detected recurrence, but the patient was asymptomatic. At postoperative month 6, the patient was experiencing discomfort in the medial ankle and great toe, and MRI at 14 months postoperatively identified 2 recurrent masses, 1 along the flexor hallucis longus tendon sheath in the great toe and another superficial to the calcaneus at the midfoot level. However, the patient refused additional surgical intervention and chose to maintain conservative management with intermittent administration of nonsteroidal anti-inflammatory drugs. His last follow-up AOFAS score was 78 points. Two patients (20%) (patients 4 and 7) had secondary arthritic changes of the ankle evident on MRI scans, indicative of stage II osteochondral defect and bony erosion, respectively.

Discussion

A few studies have focused on PVNS affecting the foot and ankle (6–10), but none of these focused only on intra-articular involvement in the ankle and/or hindfoot. The most prevalent presentation in our investigation involved a male patient in his third decade (age 37.7 ± 11.4 years) with diffuse PVNS causing ankle pain and swelling. One previous report (19) also observed PVNS to be more prevalent in males, although the condition has more often than not been observed more commonly in females (6–8,10). The mean age of our patients was 37.7 ± 11.4 (range 18 to 54) years, similar to the mean age