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Pigmented Villonodular Synovitis of the Ankle and Subtalar Joint Treated by Surgical Excision and Ligament Reconstructions: A Case Report

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Diffuse pigmented villonodular synovitis (PVNS) involving ankle joint needs complete mass excision and total synovectomy to reduce recurrence rate, while surrounding ligaments can be easily damaged. So the concurrent ligament reconstruction should be considered for post-excisional instability in subtalar joint as well as lateral ankle joint. We describe our experience in the management of a diffuse type PVNS, invades lateral talocrural joint extended to subtalar joint and introduce a new technique of all-in-one reconstruction for anterior talofibular,calcaneofibular and cervical ligament. Our new reconstruction technique applying modified Chrisman and Snook technique is useful in stabilization for deficiencies of the ligament complexafter PVNS excisionat lateral ankle and subtalar joint.

Key Words: Pigmented villonodular synovitis, Subtalar instability, Ligament reconstruction, Cervical ligament

Pigmented villonodular synovitis (PVNS), firstly described by Chassignac in 1852, causes the destruction of the articular cartilage and the instability of surrounding ligaments with the inflammatory response or hyperplasia of the intraarticular synovial cells.[1] The potency to malignant cells is very rare, but PVNS has a high local recurrence rate. So mass excision and total synovectomy is propounded as the therapeutics which lower the recurrence rate and higher the success rate of the complete cure. Specially, two forms of the disease were identified, a localized subtype characterized by a pedunculated lesion and a subtype with diffuse joint involvement. Diffuse type has a high rate of local recurrence, so additional radiation therapy is also considered in the management of refractory disease.[2] PVNS mainly occurs at single joint, such as knee joint or hip joint. However, recently it also frequently occurs at ankle joint.

Ankle joint largely consists of three joint spaces, talocrural joint, subtalar joint and distal tibiofibular syndesmosis. Unlike two other joints, the ankle joint capsule lined with synovial membrane is attached to other surrounding ligaments which keep joint stability. Therefore, it is hard to separate them.[3] Because surrounding ankle ligaments, related to ankle stability, can be damaged during the surgery as mass excision and total synovectomy, initial constructs of insufficient ligaments is strongly emphasized with the possible instability of ankle ligament after the surgery. In reconstruction of lateral ankle ligament, subtalar component also has to be cautious because the lateral talocalcaneal ligament is usually not present or is integrated with the anterior talofibular ligament (ATFL) and calcaneofibular ligament (CFL).[3]

This article presents a case of diffuse type PVNS, invading lateral talocrural joint extending to subtalar joint. We introduce a new technique of all-in-one reconstruction for ATFL, CFL, and cervical ligament.

CASE REPORT

Institutional Review Board approval was obtained from our institution before study onset, and our protocol was also approved.

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Informed consent was obtained from the patient.

A 48-year-old male patient without specific medical history, visited our hospital with severe pain and swelling at left ankle continued for three years. He complained the increasing size of mass and repetitive bouts of ankle sprains. Recently, ankle pain got worse and patient had discomforts even in daily life. At physical examination, there was diffuse swelling of lateral side in his left ankle with mild tenderness. Ankle motion was comparable to the right ankle. American Orthopaedic Foot and Ankle Society (AOFAS) score was 65 points. The simple X-ray only revealed swelling of soft tissue around ankle joint with no specific bony lesion (Fig. 1). T1-weighted sagittal and coronal magnetic resonance imaging showed a predominantly low signal mass extending laterally from the talocrural joint and invading into subtalar joint with focal intermediate-to-high signal area within the proliferative synovial masses on T2-weighted fat suppressed sequence images (Fig. 2).

Figure 1. Preoperative plain anterior-posterior radiograph of left ankle shows diffuse soft tissue swelling, joint capsule distension without bony erosion.

An excisional biopsy was performed under tourniquet control through a curvilinear J-shaped incision on the lateral ankle, and at surgery a 4×5 cm nodular, red, brown and yellow pigmented mass was excised from the left ankle and subtalar joint area (Fig. 3). The gross specimen seemed like a PVNS, and it was confirmed by its histological findings, typical PVNS with the presence of large multinucleated giant cells and hemosiderin deposits (Fig. 4). We put every endeavors to exfoliate the mass from surrounding tissues, with no injury of supporting ligaments. Nevertheless, absence or insufficiency of ATFL, CFL, posterior talofibular ligament (PTFL) of lateral talocrural supporting structures, and cervical, interosseous ligament of subtalar supporting structures were observed. Synovial membrane was completely excised to minimize recurrence. Widening of subtalar joint space in supination-adduction motion and abnormal increased talar motion in anterior draw and adduction of ankle joint, were observed. Therefore we planned to reconstruct ATFL, CFL, and cervical ligament using allogenic tibialis tendon and anchor sutures.

Applying modified Chrisman-Snook technique, we designed allotibialis tendon to replace ATFL and CFL. We used allograft tendon so as to avoid donor site morbidity at the ankle and also to avoid sacrifice of a peroneal tendon which are dynamic lateral stabilizers. The ends of the allotendon were sutured for a length of 1.5 cm with No. 2 vicryl using a whip stitch. A fibular bone tunnel was then created using a 4.0-mm drill beginning above the origin of the ATFL and directed 45° oblique to the mid-fibular

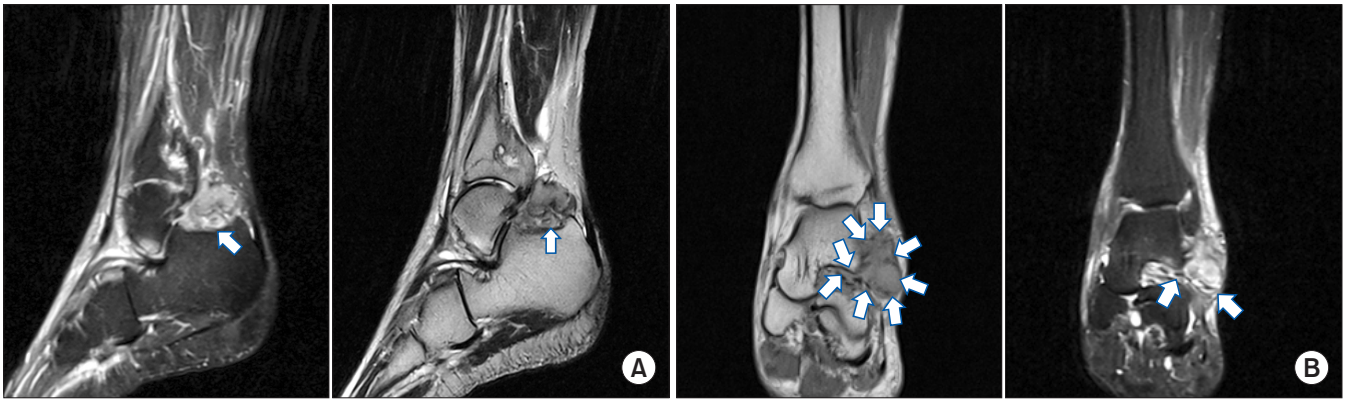

Figure 2. (A) These are magnetic resonance imaging of left ankle. T1- and T2-weighted fat suppressed sagittal images show large sized mass of low signal intensity in posterior aspect of ankle surrounding high signal intensity with focal intermediate signal intensity (arrows). (B) Coronal images show mass (arrows) involved into subtalar joint.

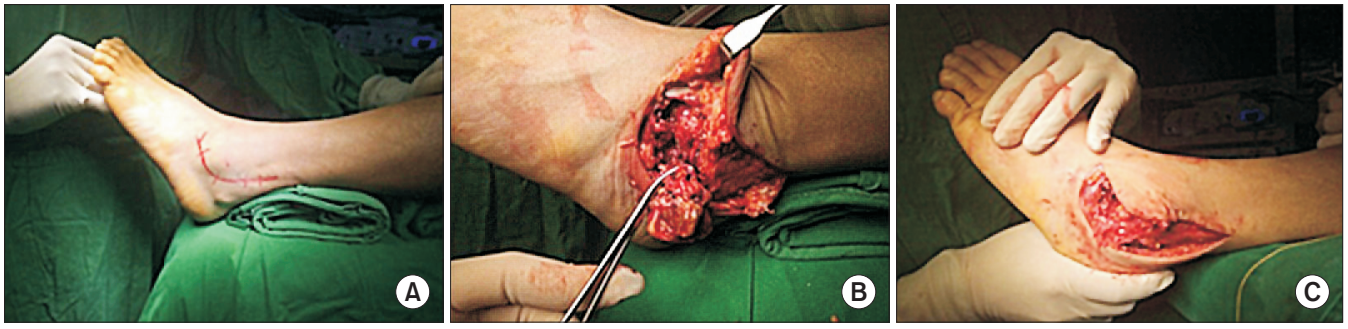

Figure 3. (A) Curvilinear incision was done on lateral side of ankle. (B) Yellowish-brown color mass with localized dark brownish portion involves overall capsular structures in the lateral talocrural joint, and cervical and interosseous ligament into subtalar joint. (C) Widening of subtalar joint space in supination-adduction motion and abnormal increased talar motion in anterior draw and adduction of ankle joint was identified after mass excision.

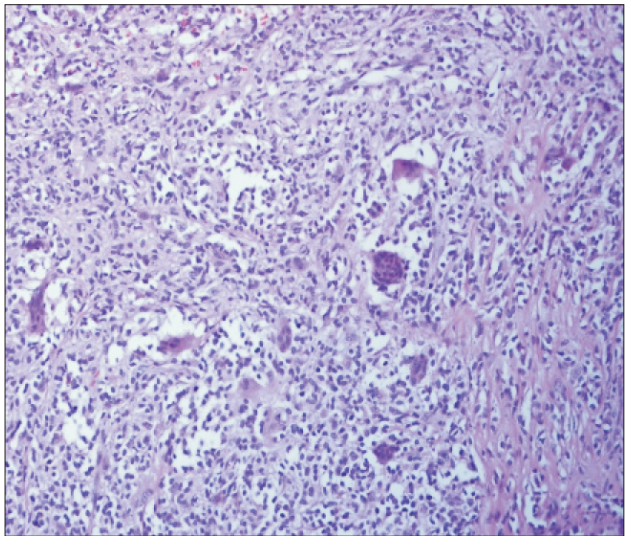

Figure 4. Microscopic finding shows proliferation of fibrous matrix, mononuclear histiocytes, multinucleated giant cells, hemosiderin-laden cells (H&E stain, ×100).

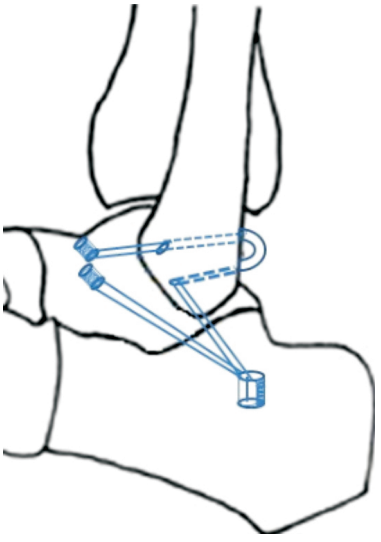

Figure 5. This drawing shows anterior talofibular, calcaneofibular, and cervical ligament reconstruction technique.

axis in a proximal direction. Another fibular bone tunnel was then created to pass from posterior of fibular to apex of anterior fibular after verifying the attachment site of CFL. The one side of looped graft tendon was routed through an oblique osseous tunnel in the talar neck, the other side through an oblique osseous tunnel in the distal fibula, and lastly, through a vertical osseous tunnel in the talus. This orientation of the looped tendon closely duplicated the anatomic ATFL and CFL. With use of an anchor suture, one end of graft tendon was fixed to reconstruct ATFL. A calcaneal bone tunnel was prepared distal to the CFL insertion in a lateral to medial direction. Looped tendon of the other side was fixed into calcaneus with a biodegradable interference screw. After passing the tendon through the calcaneal tunnel, the extra terminal graft tendon was reused to reconstruct cervical ligament (Fig. 5). The surgical wound was then closed and a compressive dressing was applied with a splint to maintain the ankle in neutral extension and slight eversion position. At 1 week postoperatively, a short leg cast with slight hindfoot eversion was applied and the patients were instructed to walk with partial weightbearing. At 6 weeks postoperatively, the cast was removed and an ankle strap brace was applied for another 6 weeks. After cast removal, the patient started ankle stretching exercise, the range of operated ankle movement which recovered fully at postoperative 12 weeks. At postoperative 2-year follow-up, AOFAS score was improved to 92 points, instability of the ankle and subtalar joint and visible sign of reoccurrence were not observed.

DISCUSSION

PVNS rarely occurs at ankle joint, below 10%, compared

to other joints and no standard treatment has been defined.[1] Whether done arthroscopically or through an open arthrotomy, the treatment of choice is synovectomy with excision of the lesion for localized PVNS, and total synovectomy for diffuse PVNS. Because of its high recurrence rate, several trials has been introduced including surgical resection with adjuvant focused external beam radiotherapy,[4] adjuvant intra-articular injections of radioactive colloid (synoviortheses).[5]

Total synovectomy and complete mass excision are recommended with the minimum of soft-tissue disruption in order to prevent joint erosions and degenerative changes. However, because of the anatomical characteristics of ankle joint, integration of capsular structure and surrounding ligaments, it's technically difficult to exfoliate massive tissue without disturbing integrated ligaments. So we removed all affected ligaments during total synovectomy. Therefore, probability of joint instability after surgery is emphasizing the necessity of ligament reconstruction.

The surgical treatment of lateral joint instability can be classified into anatomic repair and nonanatomical reconstruction. For nonanatomic reconstruction, Elmslie[6] introduced talofibular ligament and CFL reconstruction using tensor fascia at 1934, Chrisman and Snook[7] introduced reconstruction using tendon of fibular instead of tensor fascia at 1969, and many reconstruction techniques have been introduced.

Our case especially had a mass extensively invading into not only ankle joint, but also subtalar joint with multiple ligament insufficiency. Considering the anatomical traits of each ligament, we performed ligament reconstruction for ankle and subtalar joint concurrently applying modified Chrisman-Snook surgery. Because we did total synovectomy and removed all affected ligaments only few ligaments were left. Therefore, we couldn't repair the ligaments anatomically, and performed modified Chrisman-Snook surgery.

Untreated subtalar joint instability results in pain on landing after jump or giving way symptoms, and leads to potentially degenerative arthritis.[8] It is known that stability of subtalar joint is maintained by several ligamentous structures included CFL, ATFL, PTFL, anterior of lateral talofibular ligament, and interosseous ligament, etc. They are agglutinated with lateral synovial membrane and function together. To treat the subtalar instability, several techniques have been suggested, technique using peroneus brevis muscle by Aynardi et al.,[9] reinforcing technique by suturing of the inferior extensor retinaculum by Gould et al.,[10] peroneal tendon transfer technique by Chrisman and Snook,[7] and talocalcaneal ligament reconstruction techniques also have been introduced. However they were technically difficult and did not show the good prognosis.[2] This study introduced a simple reconstruction technique for ATFL, CFL, and cervical ligament using only one tendon allograft.

In summary, diffuse PVNS involving ankle joint needs complete mass excision and total synovectomy to reduce recurrence rate, while surrounding ligaments can be easily damaged. So the concurrent ligament reconstruction should be considered. Our new reconstruction technique applying modified Chrisman and Snook technique is useful in stabilization for deficiencies of the ligament complex of lateral ankle and subtalar joint.

REFERENCES

1. **Stevenson JD, Jaiswal A, Gregory JJ, Mangham DC, Cribb G, Cool P.** *Diffuse pigmented villonodular synovitis (diffuse-type giant cell tumour) of the foot and ankle. Bone Joint J. 2013;95:384-90.*
2. **Frassica FJ, Bhimani MA, McCarthy EF, Wenz J.** *Pigmented villonodular synovitis of the hip and knee. Am Fam Physician. 1999;60:1404-10.*
3. **Hertel J.** *Functional anatomy, pathomechanics, and pathophysiology of lateral ankle instability. J Athl Train. 2002;37:364-75.*
4. **Schnirring-Judge M, Lin B.** *Pigmented villonodular synovitis of the ankle-radiation therapy as a primary treatment to reduce recurrence: a case report with 8-year follow-up. J Foot Ankle Surg. 2011;50:108-16.*
5. **Ward WG Sr, Boles CA, Ball JD, Cline MT.** *Diffuse pigmented villonodular synovitis: preliminary results with intralesional resection and p32 synoviorthesis. Clin Orthop Relat Res. 2007;454:186-91.*
6. **Elmslie RC.** *Recurrent subluxation of the ankle-joint. Ann Surg. 1934;100:364-7.*
7. **Chrisman OD, Snook GA.** *Reconstruction of lateral ligament tears of the ankle. An experimental study and clinical evaluation of seven patients treated by a new modification of the Elmslie procedure. J Bone Joint Surg Am. 1969;51:904-12.*
8. **Karlsson J, Eriksson BI, Renström P.** *Subtalar instability of the foot. A review and results after surgical treatment. Scand J Med Sci Sports. 1998;8:191-7.*
9. **Aynardi M, Pedowitz DI, Raikin SM.** *Subtalar instability. Foot Ankle Clin. 2015;20:243-52.*
10. **Gould N, Seligson D, Gassman J.** *Early and late repair of lateral ligament of the ankle. Foot Ankle. 1980;1:84-9.*