CASE REPORT

# Pigmented Villonodular Synovitis Arising from Calcaneocuboid Joint in an Army Staff: A Case Report

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## Abstract

Pigmented villonodular synovitis (PVNS) is a locally destructive benign lesion usually affecting the synovial tissue of weight-bearing joints. Herein we reported a 20-year-old male patient who was an army staff with a foot lesion which was painful in army boots. In the beginning, the lesion was considered a ganglion cyst. Further investigations revealed cortical erosion of cuboid bone on the radiograph. Pigmented villonodular synovitis was considered as a probable diagnosis after observation of low signal lesion around the calcaneocuboid joint on both T1 and T2 images of Magnetic Resonance Imaging. The treatment included excision of the lesion and synovectomy of the calcaneocuboid joint. The diagnosis was confirmed with histological studies. At more than one year follow-up, the patient was completely asymptomatic, and there was no evidence of recurrence. This study aimed to raise the awareness of clinicians about the diagnosis of this rare soft tissue neoplasm which might be misdiagnosed as a ganglion or synovial cysts in the hindfoot zone.

**Level of evidence:** V

**Keywords:** Pigmented villonodular, Soft tissue neoplasms, Synovitis, Tarsal joints

## Introduction

Pigmented villonodular synovitis (PVNS) is a rare locally destructive benign lesion usually affecting the synovial tissue of weight-bearing joints (1). This lesion is the result of the synovial proliferation of unknown etiology (2). Traumatic events can be accelerating factors; however, fewer than %50 of patients had a history of previous traumatic events (2). Some authors believe it is a reactive proliferation of synovial tissue, while others consider it as a locally aggressive benign neoplasm (3). In contrast to its unknown etiology, Fletcher et al. found an association between PVNS and the seventh chromosome trisomy in a genetic study (4).

The incidence rate of PVNS is about 1.8 per a million people in the United States of America (5). The PVNS in tarsal joints and toes is uncommon, while PVNS of the foot and ankle zone is reported in 2.5-5% of all cases with joint involvement (1). Moreover, most of the studies on foot and ankle involvements are published as case reports (1–3). Mankin et al. considered the foot and ankle region as the second common site for PVNS involvement (6). Most patients are between 30 and 40 years with similar distribution in both genders (7,8); nevertheless, some authors have reported that it is more prevalent among females (3).

The PVNS of the foot usually presents with firm soft tissue lesion with relative motion which can be mistaken with other common soft tissue masses in this area, like a synovial cyst or ganglion cyst. Most of the lesions are painless and asymptomatic. Some cases have some problems, such as irritation or even ulceration of the lesion in footwear (9).

Diagnosis of PVNS, like other lesions, is primarily based on history and physical examination while further tests include radiology and magnetic resonance imaging (MRI). The cortical erosion and soft tissue lesion shadow are common findings in radiographic tests (9). The MRI can confirm the diagnosis of PVNS by typical features. The PVNS lesion typically is dark on both T1 and T2

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weighted images of MRI (10). In symptomatic localized lesions, surgical treatment is indicated and usually includes complete excision of the lesion with involved synovium and curettage of bony destruction area (9).

This study aimed to raise the awareness of clinicians about the diagnosis of this rare soft tissue neoplasm which might be misdiagnosed as a ganglion or synovial cysts in the hindfoot zone.

## Case presentation

A 20-year-old male patient who was an army staff was referred to the Orthopedics Clinic with a three-year history of a swelling on the dorsolateral side of the left foot. In the beginning, the lesion was considered a ganglion cyst. The lesion size had progressively increased from six months before our initial assessments. The patient complained of foot pain while wearing army boots. Physical examination revealed a tense and cystic lesion with approximately three cm diameter and low mobility and the overlying skin was intact and mobile [Figure 1]. Range of motion in ankle, subtalar, and midfoot joints was normal and pain-free.

According to his medical history and physical examination, the patient was scheduled for further investigation. Radiographic tests revealed a soft tissue density on the lateral side of the foot and a well-defined cortical erosion in the inferolateral side of cuboid bone in the vicinity of the calcaneocuboid joint [Figure 2]. The MRI revealed a T1/T2 low signal mass measured 39×20 mm with mild enhancement at the lateral aspect of calcaneocuboid joint and deep to peroneal tendons. These findings were mostly compatible with pigmented villonodular synovitis [Figures 3-6].

The surgical procedure was performed under spinal anesthesia. The patient was placed in the lazy lateral position. After prepping and draping, the thigh tourniquet was inflated, skin incision was made, and a dark brown mass with areas of yellow discoloration appeared [Figure 7]. It had arisen from the calcaneocuboid joint deep to the extensor digitorum brevis muscle. The lesion was dissected from the adhesions and completely excised with the capsule of the joint [Figure 8].

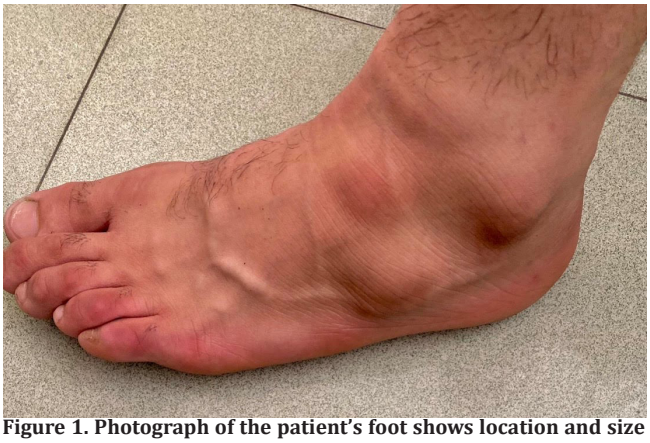

**Figure 1. Photograph of the patient's foot shows location and size of the lesion without skin involvement.**

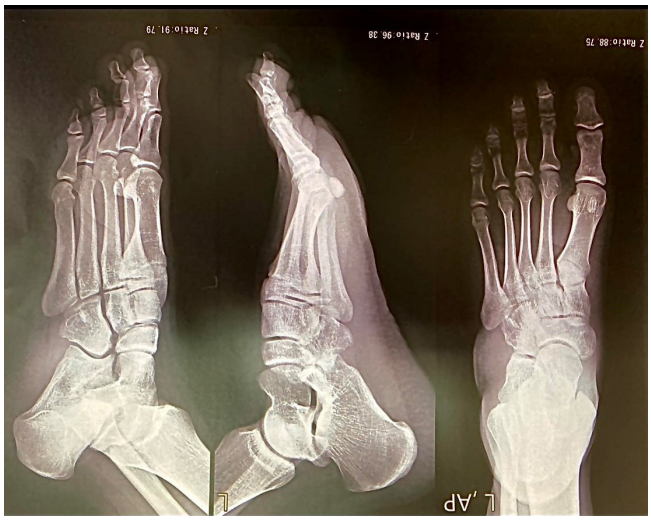

**Figure 2. Radiograph of the patient's foot in three views which demonstrates soft tissue mass shadow in lateral side of the foot and cortical erosion in the cuboid side of the calcaneocuboid joint.**

Afterward, the remains of the synovial tissue and the lesion site were curetted and cauterized circumferentially. The tourniquet was deflated and bleeding points were cauterized. After irrigation of the surgical wound, the skin closure was performed via simple, separated sutures. Histological findings of the specimen revealed large amounts of mononuclear cells with hemosiderin

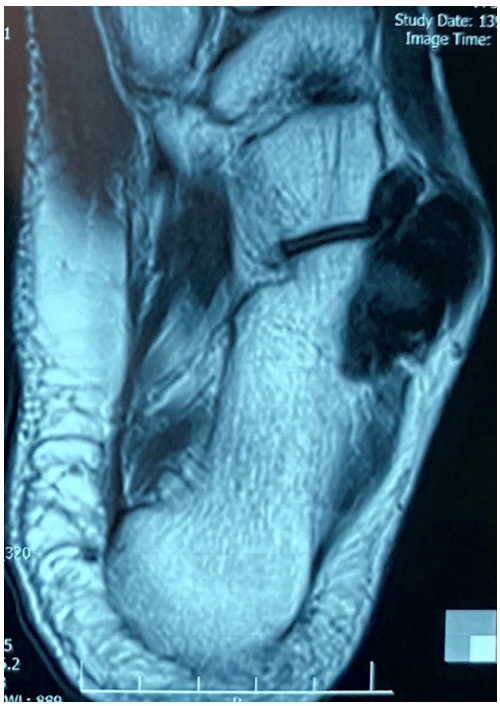


**Figure 3. Magnetic Resonance Imaging, axial T1-weighted image, a low signal soft tissue mass in the lateral side of calcaneocuboid joint.**

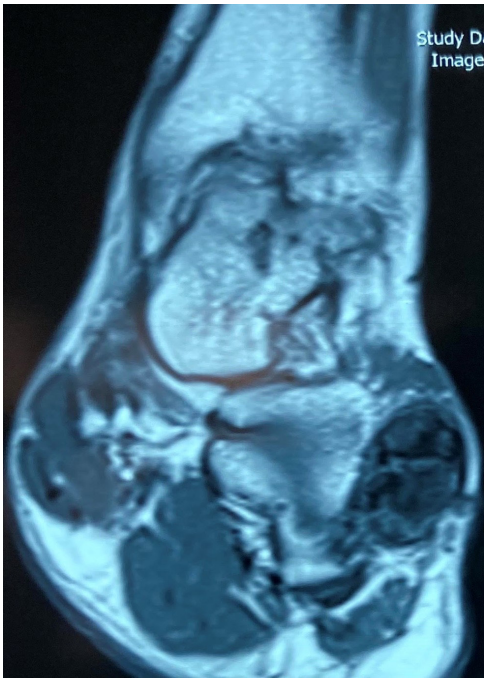

**Figure 4. Magnetic Resonance Imaging, coronal T1-weighted image, a low signal soft tissue mass in the lateral side of the calcaneocuboid joint**

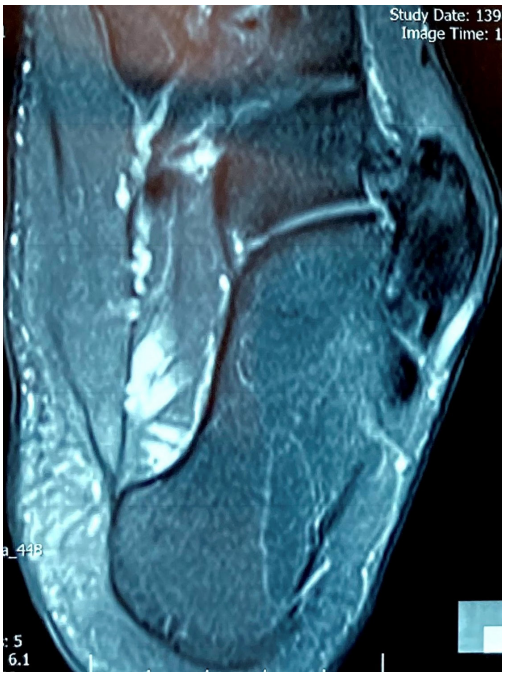


**Figure 5. Magnetic Resonance Imaging, axial T2-weighted image, a low signal soft tissue mass in the lateral side of the calcaneocuboid joint**

deposition and multinucleated giant cells which were compatible with pigmented villonodular synovitis [Figures 9-11]. The postoperative period was uneventful; moreover, the patient was completely asymptomatic and there was no evidence of recurrence at the follow-up visit which happened more than one year later.

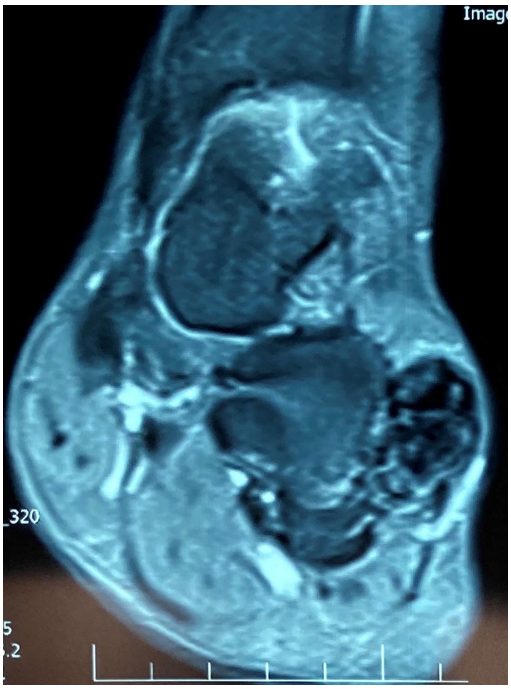

**Figure 6. Magnetic Resonance Imaging, coronal T2-weighted image, a low signal soft tissue mass in the lateral side of calcaneocuboid joint.**

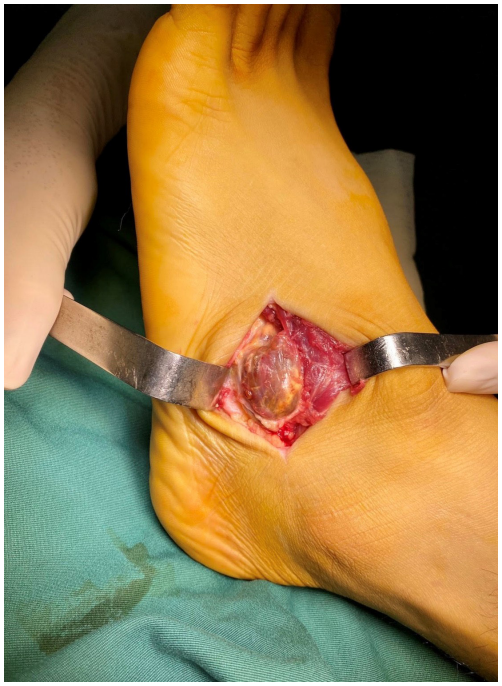

**Figure 7. Photograph of surgical incision shows a dark brown mass with areas of yellow discoloration.**

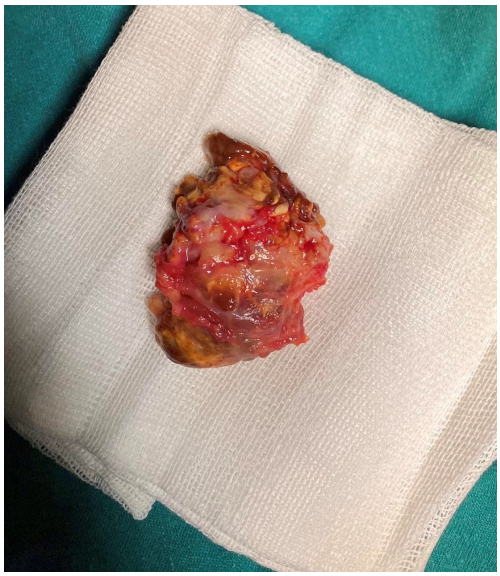

**Figure 8. Photograph of excised lesion, dark brown mass with areas of yellow discoloration.**

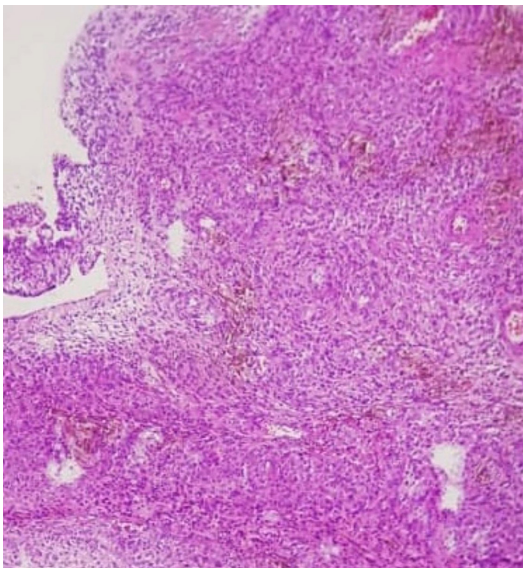

**Figure 9. Histological study of the specimen, large amounts of mononuclear cells with hemosiderin deposition.**

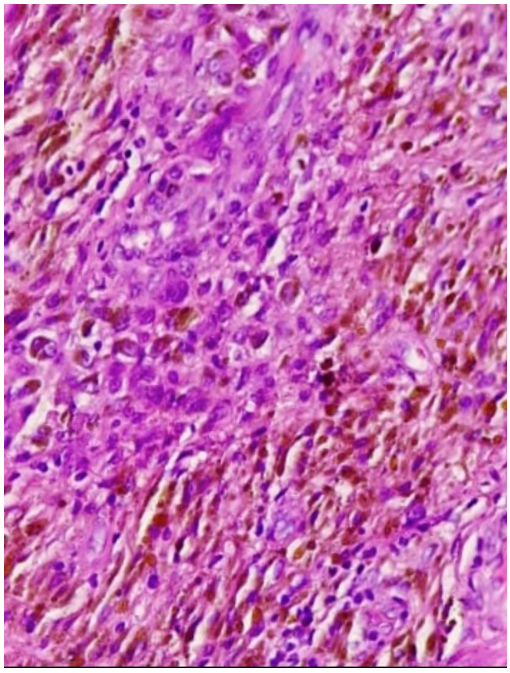

**Figure 10. Histological study of the specimen, large amounts of mononuclear cells with hemosiderin deposition.**

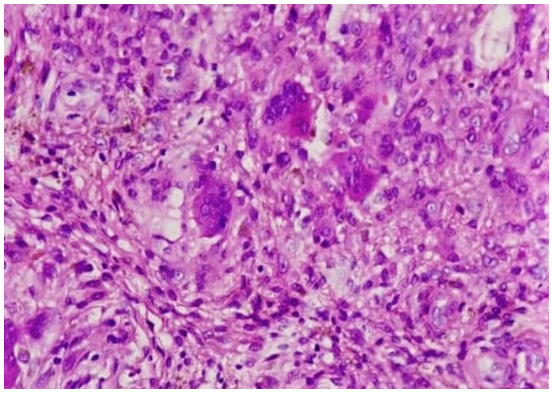

**Figure 11. Histological study of the specimen, multinucleated giant cells.**

## Discussion

The PVNS is a rare neoplastic lesion with an incidence rate of 1.8 per million (5,9). Foot and ankle area is not a common site for PVNS involvement (1,3,7–9). Most of the articles about PVNS of the foot are in form of case reports or case series (2,3,7,9,11). The PVNS usually involves patients in age ranges of 30-40 and is rare in adolescents (3,7,12). However, our case was a 20-year-old male with a history of foot mass from three years before presenting to us; therefore, he had been involved since he was 17.

A ganglion cyst is the most common soft tissue mass around the foot and ankle joints and may imitate PVNS clinically and on MRI (3,13). In children and adolescents, the other causes of soft tissue mass in the foot include the ganglion cyst, desmoid tumor, giant cell tumor of the tendon sheath, lipoma, hemangioma, schwannoma, fibroma, and some malignant lesions, like synovial sarcoma, liposarcoma, and fibrosarcoma. Most of these lesions are more common than PVNS in the foot (14); therefore, differentiating this lesion from the others is important and necessary.

Clinical manifestation of PVNS is not characteristic and is similar to other causes of benign foot lesions presenting with painless soft tissue mass since it is usually firm and

immobile (9). Our patient was asymptomatic for three years, and it was only after joining the military, wearing army boots, and longtime standing that the problems arose, including local discomfort and enlargement of the lesion.

Nonspecific clinical manifestations reveal the role of imaging in the diagnosis of PVNS. Radiography in chronic cases shows sclerotic cortical erosion in the juxta-articular site of foot bones and the shadow of soft tissue mass which are not specific (3). Computed tomography scanning cannot determine anything other than bony involvement. The MRI is known as the imaging modality of choice in the diagnosis of PVNS (7). The soft tissue involvement and extent of the lesion can be determined based on the MRI results. As seen in the present study and literature, the pathognomic pattern of PVNS on MRI is a soft tissue lesion with low to intermediate intensity signal on both T1 and T2 images (3).

In the localized type of PVNS, the accepted treatment in most of the studies includes the complete excision of the lesion with synovectomy of the involved joint and curettage of the bone cyst or bony erosion site (2,7–9,12,15). We also selected this method of surgical treatment in our case. Treatment of the diffused type of PVNS in the foot and ankle is more challenging and controversial. Diffused form of PVNS usually involves more than one joint in the foot; therefore, complete excision of the lesion is more difficult and sometimes requires more than one surgical approach. Some other adjuvant and neoadjuvant therapies, such as irradiation and cryotherapy, are recommended in excessive diffuse forms, incomplete excision, and localized recurrence (8).

Finally, in more aggressive cases with excessive joint destruction, arthrodesis is an optional treatment (2,11). According to the reviewed literature, the recurrence rate is within the range of 0-56%, and cases with incomplete excision of synovial tissue and diffused type of PVNS are more susceptible to recurrence. We had no clinical evidence of recurrence of the lesion after the one-year follow-up visit; nevertheless, the clinical and imaging exams will be continued every six months.

In our opinion, differentiation of soft tissue lesions in the foot and ankle region is an important issue that should be considered. Ganglion cysts of the foot are more common than PVNS and can imitate it clinically and on imaging tests. The PVNS is more locally aggressive and can lead to joint destruction. Clinical misdiagnosis of these two lesions is common, while the exact diagnosis of these two lesions which are different in prognosis and treatment is crucial. Imaging, especially MRI, is a useful modality to determine the foot lesion more accurately and select a proper treatment.

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