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Case Report

When rare conditions collide: plantar vein thrombosis in hereditary spherocytosis

Budak Akman[1], **Behic Celik**[1], **Betul Uzunoglu**[2]

[1] Yeditepe University Faculty of Medicine, Orthopedics and Traumatology Department, Atasehir, Istanbul.
[2] Yeditepe University Faculty of Medicine, Radiology Department, Atasehir, Istanbul.

Abstract

Plantar heel pain is a common orthopedic complaint, but less common etiologies such as plantar vein thrombosis (PVT) are often overlooked. We present a 27-year-old female patient with a history of hereditary spherocytosis who developed left plantar foot pain shortly after starting combined oral contraceptives. Initially suspected to be plantar fasciitis, conservative treatment provided no relief. Imaging studies, including Doppler ultrasound and magnetic resonance imaging, revealed a thrombus in the medial plantar vein. The patient was successfully treated with a 30-day course of enoxaparin anticoagulation therapy, followed by acetylsalicylic acid. Her symptoms resolved completely, and she remained asymptomatic at a one-year follow-up. This unique case highlights the importance of considering PVT in the differential diagnosis of atypical foot pain, particularly in patients with a history of thrombotic risk factors, to ensure early and effective treatment.

Level evidence: Level IV.

Keywords: Venous thrombosis; Spherocytosis, Hereditary; Contraceptives, Oral, Combined.

Introduction

Plantar heel and arch pain is one of the most common complaints that we encounter in our orthopedic practice. There are various differential diagnoses for plantar heel pain, including traumatic, inflammatory, arthrosis, or systemic causes[1]. The most common cause of plantar heel pain continues to be plantar fasciitis, but other causes are often overlooked for this reason[2]. A lesser-known diagnosis of plantar heel pain is plantar vein thrombosis (PVT), and in our literature review, only approximately 30 cases have been reported in the last 20 years. In this article, we present a case of PVT occurring immediately after oral contraceptive use in a young female patient with a history of hereditary spherocytosis. Written informed consent was obtained from the patient for publication and accompanying images.

Case description

A 27-year-old, 167 cm tall, and 52 kg (BMI: 18.6 kg/m^2) female patient presented to our outpatient clinic with pain in the plantar aspect of her left foot that increased with movement and was intermittent. The patient stated that her pain had been present for the last week, which also radiated to her heel, and that she had no history of any traumatic event that caused her pain. She stated that she had rowing training the day before her complaint and that she might have strained her foot. The patient works in the radiology department as a doctor and has a known history of hereditary spherocytosis and has undergone a splenectomy operation in 2006 in her medical history. She applied to the gynecology outpatient clinic due to menstrual irregularity one month before the diagnosis of PVT and started using combined oral contraceptives (Ginera ® - ethinylestradiol and dienogest).

Upon physical examination, there was pain on palpation, especially in the heel and medial arch of her left foot. In addition, there was minimal swelling and stiffness on the medial side of the foot compared to the other foot. Plantar fasciitis and/or tendinopathy were considered in the differential diagnosis. Conservative treatment was planned, and rest, ice application, and anti-inflammatory drug treatment were recommended. In

Study performed at the Yeditepe University Hospital, Atasehir, Istanbul.

Correspondence: Behic Celik. İcerenkoy, 34752, Atasehir, Istanbul. **Email:** behic13@gmail.com. **Conflicts of interest**: none. **Source of funding:** none. **Date received:** August 02, 2025. **Date accepted:** 2025-09-26.

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the first few days of follow-up, the pain decreased but did not go away, and the patient continued to complain of difficulty in walking. She was scheduled for magnetic resonance imaging (MRI) to rule out other possible diagnoses. Since the patient is a radiologist herself, she also underwent a Doppler ultrasound (USG) examination. In the MRI report, the muscular and tendinous structures adjacent to the joint were evaluated, and mild tenosynovitis at the level of the posterior medial tendons was noted. The Achilles tendon was intact. The plantar fascia was normal. On the plantar side of the foot, an increase in calibration and intraluminal hypointense signal changes were noted in the localization corresponding to the medial plantar vein, and the findings were evaluated as compatible with PVT (Figure 1). In the gray scale (B-mode) and the Doppler USG, there was an appearance of thrombus within the lumen in this area (Figures 2 and 3). The posterior tibial vein was intact. The patient was consulted with the hematology and cardiovascular surgery departments regarding her current diagnosis, treatment, and management. The parameters in the complete blood count were within normal limits. In addition, the patient was requested to undergo laboratory tests to assess genetic risk factors. The patient's Lupus parameters, Homocysteine, Anticardiolipin, and Anti-phospholipid antibody values were found to be in the normal range. D-dimer (quantitative) level was found to be positive above normal at 1.26 μg/mL. As treatment, the patient was prescribed Clexane® (enoxaparin sodium) 0.4ml SC 1*1 (30 days) and then Coraspin® (acetylsalicylic acid) 100 mg tb 1*1 (30 days) along with monitoring of D-dimer levels. The patient continued Clexane® treatment as a single daily dose for 30 days, and then completed her treatment up to 60 days with Coraspin®. D-dimer levels were monitored every ten days and discontinued at the end of the first month; they returned to normal limits. Treatment with Coraspin® was continued for another 30 days and then stopped. Weekly USG imaging showed that the thrombus started to resolve in the second week (Figure 4). After the completion of the 6-week blood-thinning drug protocol, a follow-up MRI without contrast was requested to re-evaluate her condition. This was less pronounced than the previous examination and showed that the previous thrombosis had at least partially resolved, and that the mild swelling/edema of the surrounding soft tissues had decreased in severity. The patient returned to an active lifestyle and sports at the end of the third month. Although one year has passed since the follow-up, the event has not recurred, and no clinical complaints have appeared.

Discussion

Unilateral plantar pain can be caused by a variety of diagnoses. The most common cause is plantar fasciitis, a condition characterized by microtears in the medial tubercle that lead to degeneration of the plantar fascia's root. There are many possible causes of foot pain besides plantar fasciitis. Stress fractures, bone cysts, and tendonitis are among the most common. However, in current practice, PVT is infrequently reported; only 30 cases have been reported in the last 20 years, and therefore, it is not included in standard DVT investigation protocols. Therefore, awareness of this entity and the anatomy of the plantar veins is crucial for making a diagnosis that is often difficult to detect.

Barros and Labropoulos[3] described the characteristics and clinical outcomes of patients diagnosed with PVT. Of the 11 patients diagnosed, seven (63%) were female. All patients had pain, and eight (72%) had swelling. The most common risk factors were surgery in four patients (36%) and three patients using oral contraceptive pills (27%), followed by malignancy and air travel. The patient in this article had oral

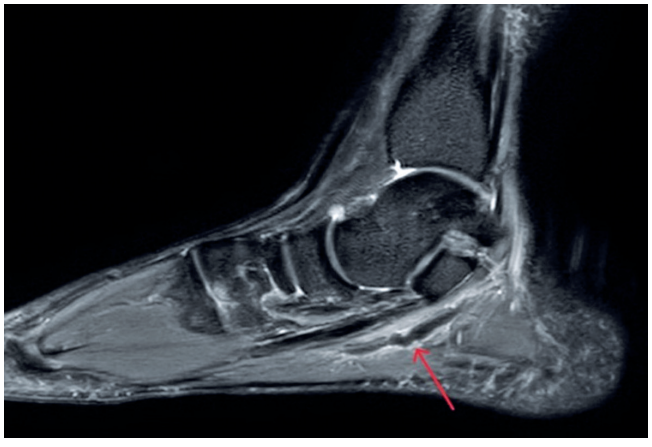

Figure 1. Sagittal STIR MRI of the foot and ankle showing findings consistent with plantar vein thrombosis. On the plantar aspect, a hypointense intraluminal signal is noted within the medial plantar vein (red arrow), accompanied by increased venous caliber. These imaging findings are suggestive of plantar vein thrombosis.

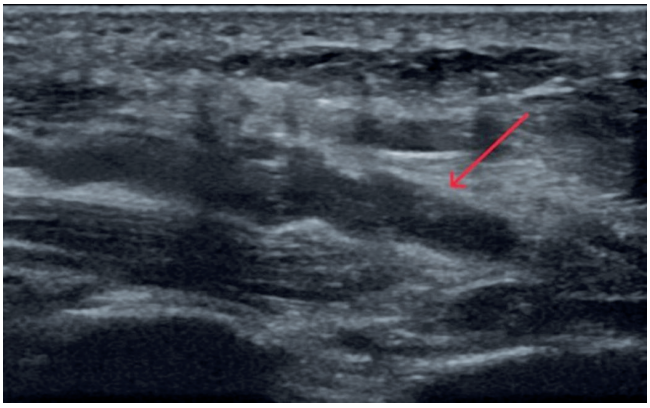

Figure 2. Longitudinal gray-scale (B-mode) ultrasonography of the medial plantar vein demonstrating plantar vein thrombosis. A hyperechoic intraluminal structure representing thrombus (red arrow) is seen along the course of the vein, which shows increased caliber. Surrounding soft tissue echogenicity is also noted, consistent with perivascular edema.

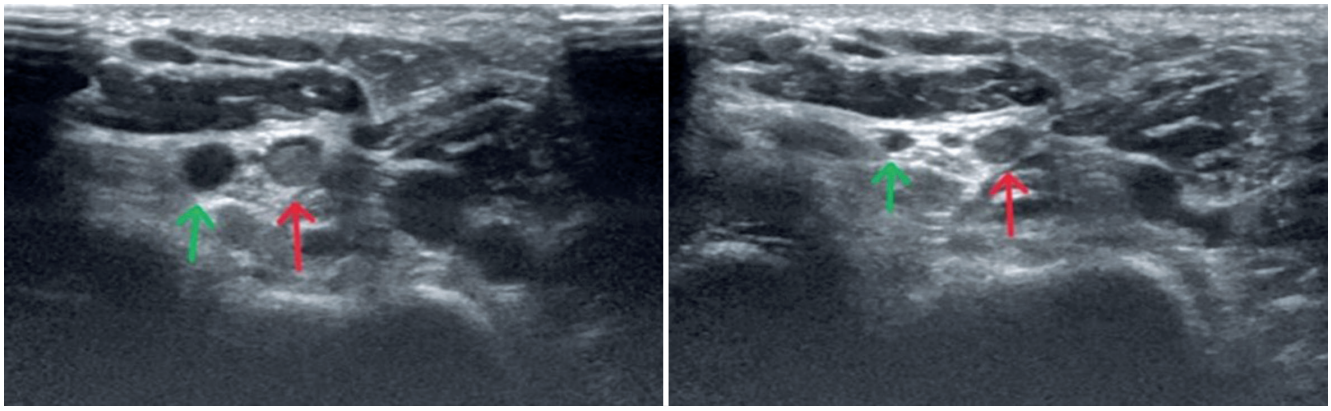

Figure 3. Axial gray-scale (B-mode) ultrasonographic images of the medial plantar vein pair and the centrally located artery between them. On the left, the pre-compression image shows an open lumen in the left vein and intraluminal echogenicity in the right vein, consistent with thrombus. On the right, the compression image demonstrates that the medial plantar vein on the left collapses with compression (green arrow), whereas the vein on the right remains non-compressible (red arrow), supporting the diagnosis of thrombosis.

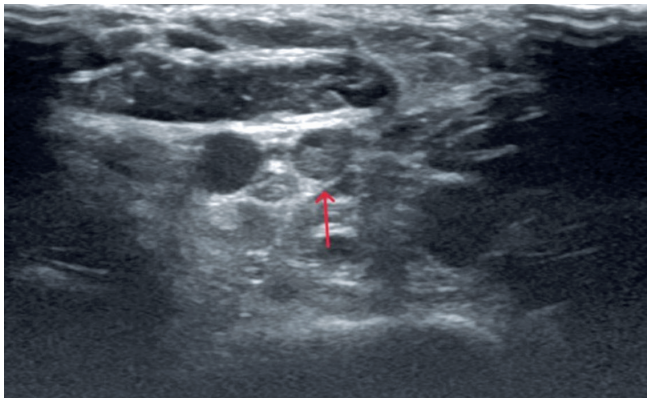

Figure 4. Follow-up axial gray-scale (B-mode) ultrasonographic image obtained during the second week. The medial plantar vein pair and the centrally located artery are visible. The left vein (red arrow) contains hyperechoic thrombus material with internal hypoechoic areas, suggestive of early recanalization and partial thrombus resolution.

contraceptive drug use as an etiological factor. Czihal et al. [4] presented the largest case series to date, a retrospective series of 22 patients diagnosed with PVT with sole pain. They observed proximal spread of thrombus in one-third of patients (27%). They found no etiological cause in 11 patients (50%), and occult malignancy was diagnosed later in two of these patients. It occurred in seven patients (32%) in association with mechanical strain on the foot. All patients were treated with anticoagulants. No cases of pulmonary embolism were reported, and no post-thrombotic syndrome was observed during a mean follow-up of 21 months.

There is no consensus for the treatment of this condition. It is usually left to the physician's preferences. Some use anticoagulation agents while others use only non-steroidal anti-inflammatory drugs (NSAIDs). The three cases reported by Barros and Lapropoulos[3] that presented signs of thrombosis progression were all treated with NSAIDs.

In our opinion, the plantar veins are part of the distal deep venous system and should be treated with anticoagulants, as in DVT. After anticoagulant therapy, during the patient's one-year follow-up, thrombosis did not recur, and no clinical complaints occurred.

Conclusion

As in this case, the development of plantar vein thrombosis in a patient diagnosed with hereditary spherocytosis who is using combined oral contraceptives has not been described in the literature to our knowledge. With this case report, we aimed to emphasize and remind of the rare diagnosis of PVT in patients with foot pain.

The clinical importance of PVT extends beyond local symptoms to include the potential risk of pulmonary embolism. PVT should be suspected in patients presenting with foot pain, especially if they have significant risk factors or lack typical plantar fasciitis findings. Early and accurate diagnosis, coupled with appropriate treatment, can improve patients' quality of life and help prevent serious complications.

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