

## Case Report

# Bilateral Calcaneal Insufficiency Fractures due to Chronic Carbamazepine use for Trigeminal Neuralgia: A Case Report

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### ABSTRACT

Stress fractures of calcaneus are uncommon cause of heel pain. Stress fractures could be seen in risk groups such as metabolic diseases/medications causing poor bone quality and exposing repetitive microtrauma. Anti-epileptic drug (AED) use is related with poor bone quality and increased fracture risk. *Although carbamazepine-induced stress fracture is a well-known entity and there are case reports in other bones such as the femoral neck, bilateral calcaneal insufficiency fractures is an extraordinary location. To the best of our knowledge, this is the first case reporting an insufficiency fracture involving calcaneus in the relevant literature. Due to the rarity of both conditions, we decided to present and discuss this patient.* When patients receiving AED treatment present with heel pain without previous plantar fasciitis history or traumatic event, insufficiency fractures should be kept in mind. This case highlights the importance of screening adverse effect of CBZ on bone metabolism in patients with long CBZ use. We report here a 41-year-old lady suffering from bilateral heel pain without trauma history. Her complaining did not respond to analgesics and stretching exercises of plantar fascia. In her past medical history she reported ongoing carbamazepine (CBZ) use over 8 years for trigeminal neuralgia. She had had low bone mineral density; defined as osteopenia. Both calcaneus MRI revealed bilateral stress fractures of calcaneum. She had been advised immobilization for 6 weeks, vitamin D and calcium supplements. CBZ has been stopped by neurology specialist and she had undergone microvascular decompression surgery for intractable pain of trigeminal neuralgia. She is doing well with full recovery from heel pain and trigeminal neuralgia at the end of one year. CBZ use causes poor bone quality through vitamin D metabolism. Heel pain without traumatic event, objective findings of plantar fasciitis and calcaneal spur syndrome in an CBZ using patient insufficiency fracture of calcaneus should be remembered and evaluated rigorously.

**KEYWORDS:** Calcaneal insufficiency fractures, carbamazepine, heel pain, trigeminal neuralgia

## INTRODUCTION

Heel pain is a common condition in adults that may cause significant disability in daily activities. A variety of soft tissue, osseous, and systemic disorders can cause heel pain. Mechanical etiologies are the most commonly detected etiologies causing heel pain in adults.<sup>[1]</sup> As stress fractures are also becoming a common cause of heel pain especially in risky groups; patients who receive repetitive microtrauma with normal

bone structure and patients with impaired bone quality receiving normal physiologic loads are candidates for unusual presentation of heel pain.<sup>[2]</sup> Here, our patient experienced fracture with lower bone quality under


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physiological loads. Calcaneus insufficiency fractures with underlying osteopenia etiology have been reported previously. To our best knowledge, there is no report yet of bilateral calcaneal insufficiency fracture due to CBM use. Herein, we report a case of bilateral calcaneal insufficiency fracture due to long lasting carbamazepine (CBZ) use for trigeminal neuralgia (TN).

Causative relationship between antiepileptic drug (AED) use and osteopenia has been widely studied. AEDs increase velocity of vitamin D hydroxylation to metabolites which results in increased rates of bone loss.<sup>[3]</sup>

## CASE REPORT

We report this case after the informed consent form was signed by patient.

A 41-year-old non athlete lady was referred to outpatient clinic with a complaint of ongoing bilateral moderate heel pain and difficulty in walking for 6 weeks. On examination, there was tenderness on the lateral-posterior-superior of both calcanei. When compression was applied on both calcaneus painful regions; this also aggravated pain. There were no nerve compression findings, acute traumatic skin lesions.

In her medical history, it was learned that she had been using CBZ regularly for 7 years due to TN. Moreover, she did not have smoking history, and she did not use a medication except CBZ as well. Routine laboratory blood tests were within normal values except decreased vitamin D levels.

She underwent radiologic work up including weight bearing lateral x-rays and magnetic resonance imaging (MRI). Plain radiographs showed a perpendicular irregular radiopaque line to calcaneal superior edge MRI confirmed these findings with broad calcaneal edema surrounding fracture line in T2 weighted images [Figure 1].

The patient was diagnosed as calcaneal insufficiency fracture due to chronic CBZ use so she underwent

bone mineral density measurement of the femoral neck and lumbar spine, both had T scores under -2,5. She reported low score of AOFAS Ankle Hindfoot score, which was 62.<sup>[4]</sup> She was consulted to neurology specialist and CBZ treatment had been discontinued. She was advised immobilization for 6 weeks, vitamin D supplementation (daily 600 IU combined with 1, 2 gr elementary calcium) and then she underwent microvascular decompression surgery for TN. At the one year visit, she was pain free. She reported full recovery of symptoms.

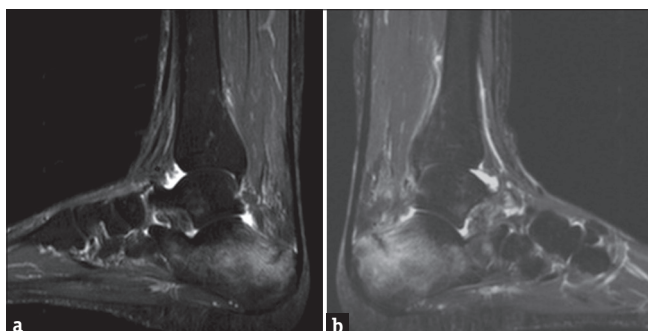
## DISCUSSION

Heel pain that does not respond to regular medical and rehabilitative procedures should be evaluated with suspicion of atypical causes of heel pain. Nerve entrapment, tumor, infection, inflammatory arthropathies, radiculopathies and stress fractures can cause heel pain and most of them could be eliminated with detailed patient history, physical examination and imaging studies.<sup>[1]</sup> Insufficiency type of stress fractures should be kept in mind especially in risky groups who exposed to repetitive micro-trauma and in patients with systemic disorders that affect the bone metabolism.

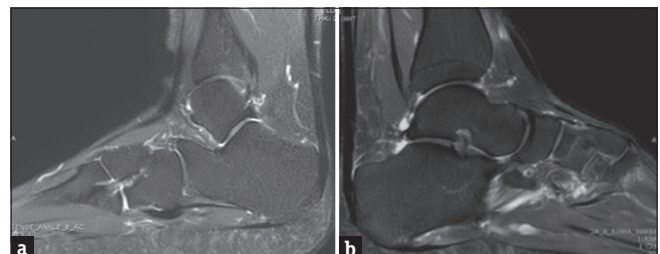
MRI detects stress fractures with high sensitivity and specificity at early stages. In addition to this, some authors used radionuclide bone scanning (Tc-99m) as first line imaging modality but low specificity and radiation exposure, limits its use for detection of stress fractures.<sup>[5]</sup> Timing of MRI has not been well defined. In the literature in case of no response to medical treatment 2 to 6 weeks and unusual presentation of heel pain has been used to an indication for MRI.<sup>[5,6]</sup>

Previously reported insufficiency type stress fractures in lower extremities indicated low bone quality caused by metabolic disorders,<sup>[5]</sup> medications which caused osteopenia.<sup>[6]</sup>

Here, our patient had a history of CBZ use more than seven years for TN. TN is a clinical condition characterized by a sudden usually unilateral, brief, stabbing recurrent pain with one or more divisions of the trigeminal nerve. According to the American



**Figure 1:** (a) Right and (b) Left ankle MRI showing broad calcaneal body edema surrounding fracture line in T2 weighted images



**Figure 2:** (a) Right and (b) Left Bilateral ankle MRI showed normal findings in T2 sequence images at one year follow up

Academy of Neurology (AAN)- European Federation of Neurological Society (EFNS) recent guidelines, CBZ is the first step medical treatment for pain control in TN.<sup>[7]</sup>

CBZ is an enzyme inducing AED and enzyme inducing AEDs have been shown to be responsible for osteopenia through vitamin D metabolism. It is also reported that direct inhibitor effect on osteoblast proliferation, decreased calcium absorption, and endocrinopathy are the other suggested mechanisms of AED use on bone metabolism. AED use has been shown related with hypocalcemia, decreased vitamin D levels in hospitalized patients and children in previous reports.<sup>[8]</sup>

The AAN-EFNS guidelines recommended that patients unresponsive or that cannot reach the therapeutic dosage of the drug because of adverse events should be informed about the surgical treatment of TN. In this guideline, adverse events defined as central nervous system related symptoms (somnolence, dizziness and postural unbalance), but bone metabolism complications of CBZ usage has not been well defined because of lack of literature knowledge.<sup>[8]</sup> *Bone metabolism complications may be another contraindications for AED use and surgical intervention would be necessary if so.* Our patient received microvascular decompression surgery. After one year follow-up, she had full recovery from both TN and bilateral calcaneus insufficiency fractures [Figure 2].

Bilateral calcaneal fractures caused by fall from height on heels is a well-known issue.<sup>[9]</sup> Charles *et al.* reported interesting case of a bilateral calcaneus fracture in a patient with Diamond Blackfan anemia.<sup>[10]</sup> Suggested mechanism was steroid usage and red cell aplasia. AED use and insufficiency fractures firstly reported by Sariyilmaz *et al.* in a long term CBZ use with epileptic young lady.<sup>[6]</sup>

## CONCLUSION

In conclusion, heel pain with unusual presentation should remind the physicians other causes. When patients receiving AED treatment present with heel pain without previous plantar fasciitis history or traumatic event, insufficiency fractures should be kept in mind. This case highlights the importance of screening adverse

effect of CBZ on bone metabolism in patients with long CBZ use.

## Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Nil.

## Conflicts of interest

There are no conflicts of interest.

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