Radiographic Presentation of Delayed Diagnosis of Charcot Foot in a 56-Year-Old Patient With No Reported History of Diabetes: A Case Report

Yuri Korvatko, DS, MS, DACBR

ABSTRACT

Objective: The purpose of this case report is to describe the radiographic presentation of a patient with neuropathic osteoarthropathy.

Clinical Features: A 56-year-old white man presented to the diagnostic imaging department of a chiropractic teaching institution for the radiographic examination of the left foot. He had a 6-month history of left foot pain unresponsive to chiropractic care. The patient had reported no history of diabetes and denied any trauma, surgery, or malignancy.

Intervention and Outcome: Based upon the history, a 3-view radiographic examination of the left foot using computed radiography was performed. Radiographic abnormalities included osseous resorption, architectural foot disorganization, and dissociation and collapse of the anatomical alignment between the intertarsal and tarsal-metatarsal articulations. Intra-articular osseous debris formation with periarticular edema of the midfoot was noted. After the radiological diagnosis, the patient was subsequently investigated and diagnosed with type 2 diabetes mellitus. Thus, there were radiographic findings of poorly controlled or untreated diabetes mellitus with the resultant complication of neuropathic osteoarthropathy.

Conclusion: Some patients with type 2 diabetes may present with covert manifestations and other symptoms, such as a history of unexplained foot pain. The patient in this case with diabetes-induced neuropathic osteoarthropathy demonstrates that these findings should raise suspicion and result in an appropriate follow-up investigation. (J Chiropr Med 2019;18:61-66)

Key Indexing Terms: *Diabetes Mellitus*

INTRODUCTION

Neuropathic osteoarthropathy or Charcot joint may develop in patients with multiple neurological or systemic conditions. This pathology will typically manifest as a spectrum of destructive and disabling osseous and articular changes that currently in most cases is associated with poorly controlled or advanced diabetes mellitus (DM). Diabetes with its focal or systemic complications may lead to socioeconomic impacts associated with major disabilities

and costly health care services. It is estimated that about 30.3 million people or 9.4% of the population in the United States experience DM.¹ If current trends continue, approximately 1 in 3 adults in the United States will be affected by DM, with 5% diagnosed as type 1 and the remaining 90% to 95% as type 2 DM.¹ In addition, estimates show that there are about 7.2 million people or 23.8% of people with diabetes in the United States who remain undiagnosed and completely unaware of their condition. The latter scenario may result in delayed diagnosis and advanced clinical complications.¹

In a relatively large proportion of patients with diabetes-induced Charcot foot (CF), the initial diagnosis may be delayed.^{2,3} Thus, clinical awareness of early radiological and clinical detection of CF is important because early diagnosis and prompt elimination of weight bearing will reduce progressive articular destruction and major disability.^{2,3} With the current epidemic in type 2 DM, there are likely cohorts of patients with undetected or poorly controlled diabetes who may present with such complication as CF. Therefore, the purpose of this case report is to

Clinical Sciences at National University of Health Sciences, Lombard, IL.

Corresponding author: Yuri Korvatko, DC, MS, DACBR, SSE (Hans) Osteopathy, Diplomate, Clinical Sciences at National University of Health Sciences (NUHS), 200 E Roosevelt Road, Lombard, IL 60148. Tel.: +1 630 396 0262. (e-mail: ykorvatko@nuhs.edu).

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describe a patient with a radiographic diagnosis of CF without a previous diagnosis of DM.

CASE REPORT

A 56-year-old white man presented to the diagnostic imaging department of a chiropractic teaching institution for radiographic examination of the left foot. He had a 6-month history of left foot pain unresponsive to chiropractic care. The patient has reported no history of diabetes and denied any trauma, surgery, or malignancy.

After this patient's referral, conventional 3-view radiographic examination of the left foot using computed radiography was performed. The most notable radiographic abnormalities revealed osseous resorption, architectural foot disorganization,

dissociation, and collapse of the anatomical alignment between the inter-tarsal and tarsal-metatarsal articulations. In addition, an appreciable intra-articular osseous debris formation with periarticular edema of the midfoot was noted (Figs 1A and 1B).

Based on the radiographic evaluation, the diagnostic imaging report stated the differential diagnoses of neuroarthropathy or a more remote possibility of bone or joint infection. After the establishment of this patient's radiological diagnosis, the patient's referring physician confirmed that the patient was subsequently investigated and diagnosed with type 2 DM. Unfortunately, no laboratory test results such as complete metabolic profile with glucose values or glycosylated hemoglobin A1C were available.

Based on the radiological diagnosis and current literature review, it was prudent to consider that a subclinical diagnosis of poorly controlled or untreated DM with resultant complication

Fig 1. In a 56-year-old white man with chronic nontraumatic left foot pain, non-weight-bearing dorsal to plantar (A) and lateral (B) radiographs of the left foot reveal an abnormal break and collapse of tarsal-first metatarsal line owing to spontaneous fracture and dislocation of tarsal-metatarsal articulations of the foot. Subtle dissociation between the medial cuneiform and the base of the second metatarsal bone known as Lisfranc fracture-dislocation is suspected. An appreciable periarticular osseous fragmentation and debris formation are additionally noted about midfoot articulations. Moderate soft tissue edema is present. The above constellation of radiographic findings is characteristic of neuropathic osteoarthropathy with the differential diagnosis of bone or joint infection.

of neuropathic osteoarthropathy was plausible in this patient. The patient gave consent for the publication of this study.

DISCUSSION

As far as the author is aware, there are currently 3 published cases of CF in chiropractic literature. All reported cases were presented with a well-established diagnosis and long history of DM. To date, no published cases of CF without previous diagnosis of DM were reported in the chiropractic literature. For example, Haran and coworkers reported a case of a 71-year-old woman with diabetic peripheral neuropathy and long-standing history of type 2 DM who presented with left foot deformity as a result of advanced neuropathic osteoarthropathy.⁴ Similarly, a different case presented a 55-year-old woman with well-established type 2 DM and increasing left foot pain after “stepping into a hole” that resulted in rapidly progressing CF complicated by advanced foot ulcers.⁵ Likewise, most cases in general medical literature tend to indicate that the diagnosis of CF is predominately established in patients with a longstanding history of DM. There are, however, some rare reports that discuss the diagnosis of CF in patients without a previous history of DM when further clinical investigation revealed abnormality of blood glucose and glycosylated hemoglobin.⁶

Neuropathic osteoarthropathy is most commonly identified in the lower extremity and currently in most cases, it develops in direct association with type 2 DM.⁷ Diagnostic features of CF may vary from acute onset of warmth, erythema, and swelling of the affected extremity⁷ to potentially chronic progressive destructive arthropathy resulting in rocker-bottom foot deformity, foot ulcerations, osteomyelitis, cellulitis, and gangrene.⁸ Regarding clinical diagnosis of CF, it always should begin with a detailed history and physical examination. Although lacking in specificity, some diagnostic tests such as complete blood count, C-reactive protein, and erythrocyte sedimentation rate may help to differentiate CF from potential cases of osteomyelitis, septic arthritis, or inflammatory/crystal-induced arthritis.⁹ In addition, such laboratory evaluation as complete metabolic profile and hemoglobin A1C may help to reveal an underlying abnormality of blood glucose and poor glycemic control that seem to be elevated in most patients with active diabetes-induced CF.⁹ The initial imaging evaluation of CF should begin with plain-film radiography and if radiography appears unrewarding, it can be followed by magnetic resonance imaging, especially to aid radiographically occult cases with some of the earliest features of CF or its potential delayed complications.¹⁰

The pathophysiology of neuropathic osteoarthropathy was thought to develop because of progressive neurosensory deficit of the affected joint.¹¹ Currently, however, there is ample evidence that implicates diabetes-induced sympathetic neurovascular injury as a major step in the cascade of

pathological changes associated with Charcot joint.⁸ Other causes, such as syringomyelia, tertiary syphilis, multiple sclerosis, alcoholism, amyloidosis, congenital insensitivity to pain, or potentially other systemic, metabolic, or neurological conditions may result in neuropathic osteoarthropathy.¹¹ The precise etiology of neuropathic osteoarthropathy remains uncertain. After its original description by Jean Mary Charcot in 1868, most reports have emphasized the meaning of disturbed proprioception and diminished protective pain reflexes known as “neurotraumatic theory” in its main etiology.¹² However, as stressed earlier, the role of diabetic neurovascular injury is currently being viewed as one of the major factors in the development of CF. The latter theory states that owing to direct damage to the sympathetic nerve fibers controlling regional blood vessels, there may be vasodilation and resultant blood-to-bone overflow. In turn, the latter will be followed by bone hyperemia and release of proinflammatory cytokines with subsequent activation of osteoclasts and resultant osseous dissolution and breakdown in osteoligamentous architecture, especially during repeated stresses of daily weight bearing.^{2,8,13,14}

Considering potential pitfalls of delayed clinical diagnosis, most health care providers still may accept the notion that neuropathic osteoarthropathy is potentially a painless condition. On the contrary, it appears that pain and local inflammation may manifest as some of the earliest features of CF. For example, studies show that during early inflammatory osteolysis, capsular synovium may become infiltrated with reactive neutrophils and mononuclear cells that may manifest as nonseptic synovitis, resulting in focal inflammation and pain.¹⁵ However, the ultimate tissue damage associated with most regional or systemic complications of DM is the result of poorly controlled hyperglycemia and progressive tissue glycosylation by the advanced glycosylation end products.¹⁶ The latter typically will result in direct collagen damage and poor removal of damaged collagen, eventually causing structural and functional impairment of the ligaments, joint capsule, muscle tendons, blood vessels, and other important collagen-containing structures. Thus, regarding specific mechanisms contributing to CF, daily ambulation primarily depends on structural integrity of osseous anatomy and ligamentous restraints that typically are coupled with physiologically intact elastic tissue recoil of the foot. In patients with poorly controlled DM, most likely owing to a combination of neurovascular and proprioceptive tissue damage combined with repeated stresses of weight bearing, an accelerated rate of bone and joint destruction with subsequent structural collapse may occur.

Diagnosis of CF

Early diagnosis of CF may pose some clinical challenges. For example, it is thought that most patients who develop CF have experienced DM for decades. However, as demonstrated by this report, there may be a

substantial cohort of patients with the diagnosis of CF who may precede clinical diagnosis of DM or on occasions manifest as its initial clinical feature.¹¹ As stressed earlier, another diagnostic difficulty may arise because in its traditional form neuropathic osteoarthropathy is considered potentially “painless arthropathy” and thus not included in the differential diagnosis of painful foot, especially during its early or acute presentation or radiographic stage 0. As demonstrated by this case, pain in association with CF is reported in over 50% of patients.¹⁷ Therefore, health care providers need to bear in mind that any history of unexplained foot pain with suspect or well-established DM should prompt a thorough clinical and radiological investigation of CF. Regarding its imaging characteristics, neuropathic osteoarthropathy demonstrates a very broad differential diagnosis. For example, neurosyphilis or tabes dorsalis, congenital insensitivity to pain, spinal cord paralysis, multiple sclerosis, alcohol-induced neuropathy, leprosy, or any other major neurological or systemic conditions may lead to neuropathic osteoarthropathy. However, as stressed earlier, owing to ongoing epidemic in type 2 DM, diabetes remains the number one cause of CF. In addition, there are about 250 000 undiagnosed cases of CF annually with the initial diagnosis delayed for as long as 29 weeks or longer.^{2,3} Another limiting factor in prompt and accurate diagnosis of CF is that it often may present as a hot, tender, and swollen joint, potentially mimicking such common arthropathies as osteoarthritis, gout, rheumatoid arthritis, or any other common arthritic disorder.³ As stressed earlier, the initial imaging investigation of CF should begin with plain-film radiographs.⁸ Radiographic abnormalities associated with this pathology can present as atrophic or hypertrophic bone changes and can be divided into 3 distinct stages. For example, the radiographic stage 1 may present as well-observed architectural bone resorption, osseous dissolution, and subsequent osseous fragmentation. Stage 2, on the other hand, may present with osseous debris coalescence that will typically progress to stage 3, signifying advanced findings of bone reorganization and remodeling. However, some of the earliest findings of bone hyperemia and edema described as radiographic stage 0 in most cases of CF remain radiographically occult.¹⁸ Thus, MR imaging is considered the most sensitive imaging modality, allowing early identification of bone marrow edema and surrounding soft tissue changes.¹⁸ Magnetic resonance imaging also may aid identification of delayed complications of CF, such as osteomyelitis, septic arthritis, or cellulitis. When considering imaging diagnosis, in over 60% of cases of CF, the most radiological changes occur in and around tarsal-metatarsal and intertarsal articulations of the midfoot.¹⁷ Additional radiological features characteristic of rapidly progressing CF may include bone demineralization, osseous fragmentation, articular dislocations, increased osseous debris formation, and visible distention of surrounding soft tissues. In addition, regional medial arterial sclerosis of small- to medium-size arteries known as Mönckeberg’s arterial

sclerosis is detected in over 94% of patients with DM and may potentially aid radiographic and clinical diagnosis of diabetes-induced CF.¹⁹ Similarly, as was suspected in this case, identification of a particular pattern of fracture dislocation between the medial cuneiform and the corresponding second metatarsal base may indicate a possibility of Lisfranc fracture-dislocation that will typically portend irreversible changes observed in advanced cases of CF.¹⁸ In summary, to arrive to a successful and early clinical diagnosis of CF, health care providers should be encouraged to exercise high level of clinical suspicion, especially in those patients who present with early or acute presentation of Charcot joint. The latter is important because early diagnosis and prompt immobilization of the affected extremity may reduce osseous and articular destruction and promote bone healing with potential reduction in clinical complications and disability. Alternatively, continued weight bearing may cause progressive osseous destruction and articular collapse, leading to previously mentioned rocker-bottom foot deformity, foot ulcerations, osteomyelitis, cellulitis, and gangrene.

Management of CF

Based on its conflicting etiologies and limited randomized placebo-controlled trials, most guidelines about the management of CF are largely a result of different professional opinions rather than well-documented clinical evidence.⁸ Nevertheless, there is a general agreement that timely diagnosis and cessation of weight-bearing activity are crucial for the prevention of rapid osseous destruction and future complications of CF. For example, Perrin and associates reported a unique case of a middle-age man with diabetes with bilateral CF when the diagnosis in the right foot was delayed by approximately 3 months. As the result of such delay, osseous destruction in the patient’s right foot was more extensive than in the left, causing major structural deformity and increased risk of skin ulcerations and gangrene.³ Therefore, once the diagnosis of CF is established, it requires prompt immobilization and complete cessation of weight bearing, often referred to as extremity “offloading.” The latter is typically achieved through an introduction of a short-duration total-contact cast. In addition, the patient may require weekly monitoring with repeated radiographs and being occasionally supplied with crutches or a wheelchair until the edema, foot temperature, and radiologic changes have improved.⁸ Similarly, after prompt immobilization of the affected extremity, the patient may require prescriptive boots or weight-bearing braces and regular re-evaluation to ensure successful bone healing. However, health care providers should be aware that prolonged immobilization of patients with CF may lead to disuse osteoporosis, muscular atrophy, and general fitness loss, including other complications.

Because of the potentially rapid nature of diabetes-induced inflammatory osteolysis, the extent of osseous destruction may be quite substantial. Therefore, in addition

to extremity offloading, some providers recommend the use of antiresorptive medications such as bisphosphonates that specifically target the activity of osteoclasts. However, despite their good antiresorptive properties, there is insufficient data showing that bisphosphonates may contribute to long-term improvement in the ambulatory status of patients with CF.²⁰ Moreover, prolonged use of bisphosphonates may potentially lead to bisphosphonates-induced insufficiency fractures and other regional or systemic complications. Unfortunately, surgical care may be required as a last resort, especially in cases of advanced articular deformities, osteomyelitis, and gangrene. However, if surgery becomes necessary, it should be considered as a potential contraindication during active CF or inflammatory osteolysis to minimize the potential risk of poor wound healing, infection, or surgical appliance failure within the demineralized bone.

Limitations

One major limitation of this study is its single-subject design. It represents anecdotal evidence that may lack important comparative analysis and statistical evaluation. Considering the current epidemic in type 2 DM, more research and clinical education are required regarding early diagnosis and evidence-based therapeutic regimens of this potentially treatable pathology.

CONCLUSION

This case provides an important clinical lesson that despite that CF traditionally is considered a delayed complication of DM, some patients present early or remain undiagnosed. Such delay in clinical intervention may result in major disability and advanced complications of CF.

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No funding sources or conflicts of interest were reported for this study.

CONTRIBUTORSHIP INFORMATION

Concept development (provided idea for the research): Y.K.
Design (planned the methods to generate the results): Y.K.
Supervision (provided oversight, responsible for organization and implementation, writing of the manuscript): Y.K.

Data collection processing (responsible for experiments, patient management, organization, or reporting data): Y.K.
Analysis (responsible for statistical analysis, evaluation, and presentation of the results): Y.K.
Literature search (performed the literature search): Y.K.
Writing (responsible for writing a substantive part of the manuscript): Y.K.
Critical review (revised manuscript for intellectual content): Y.K.

Practical Application

- Early clinical and radiological recognition of neuropathic osteoarthropathy of the foot will prevent advanced complications and major disability of patients by initiating appropriate patient management.

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