



A Rare Charcot Neuro-Osteoarthropathy of Hip with an Uncommon Cause

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Abstract

Keywords

- Charcot neuroarthropathy
- diabetes
- osteomyelitis

Neuro-osteoarthropathy often called as Charcot joint results from decreased sensory innervations of the involved joint resulting in severely damaged and disrupted joints and involvement of adjacent soft tissues. Charcot joint is characterized by the “6Ds,” which are i) distended joints, ii) density increase, iii) debris production, iv) dislocation, v) disorganization, and vi) destruction. Hip joint involvement is very rare probably because of rich nerve supply compared with other peripheral joints. To minimize the joint deformity and loss of function, early diagnosis is of great importance in which radiological imaging plays a major role.

Introduction

Charcot neuro-osteoarthropathy is a progressive destructive disorder of bones and joints. The disease is often associated with disorders causing loss of normal pain sensation and abnormal proprioception leading to recurrent joint injuries followed by severe damage and disruption. Joints of foot and ankle are most commonly affected and most of such cases are seen in patients of diabetic neuropathy. Herewith we describe a rare case of Charcot neuro-osteoarthropathy of hip joint secondary to pelvic primary retroperitoneal germ cell tumor.

Case Report

A 37-year-old gentleman, known case of retroperitoneal germ cell tumor, underwent debulking surgery and colostomy followed by chemoradiation in 2017. He had recurrent left pelvic mass in 2020 and later developed limping and shortening of the left lower limb and minimal pain in the left hip joint. He was referred to our department of radiology for radiograph of both hip joints to evaluate the unexplained symptoms of lower limb and PET CT scan with the suspicion

of disease progression. Laboratory parameters were within normal limits. Clinical examination revealed wasting of gluteal and adductor group of muscles with left lower limb shortening. Tendon reflexes were normal.

Radiograph showed diffuse osteopenia in the left femur with extensive destruction of left hip joint with disorganization and superior subluxation of joint (→ Fig. 1).

On PET CT imaging, low-grade FDG avid ill-defined soft tissue thickening was seen along the left lateral pelvic wall, extending to the presacral space and encasing left external and internal iliac arteries and compressing the iliac veins.

Destruction of the roof of the acetabulum, head and neck of left femur was seen with extensive bone resorption. Thinning and atrophy of the residual neck of the femur was seen giving a “licked candy appearance.” Cortical irregularity, destruction, and osseous debris were observed in the roof and the posterior column of acetabulum extending to the anterior column. The left hip joint was deformed with antero-superior subluxation. Moderate FDG activity was noted around the residual neck of femur and hip joint. The femoral tuberosities were spared. Diffuse osteopenia was seen in visualized shaft of the left femur. Fluid density was

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Fig. 1 X-ray of both hip joints showing diffuse osteopenia in the proximal left femur with destruction of the head and neck of the left femur and roof of the left acetabulum. There is disorganization and superior subluxation of the joint.

seen in the medial aspect of the joint space. Atrophy and fatty changes were seen in adductor muscles of left thigh and in leg muscles.

Rest of the body including the brain, spine, chest, and upper abdomen were normal on PET CT imaging.

PET CT imaging was suggestive of residual disease with post treatment changes in the left side of the pelvis, with features of neuropathic joint of the left hip. Moderate FDG uptake in the left hip joint was of concern for disease extension. So, further MR imaging was performed (→ Figs. 2 and 3).

MR imaging confirmed chronic atrophic neuropathic arthropathy of the left hip joint with extensive joint deformity, complete resorption of the head of the femur and antero-

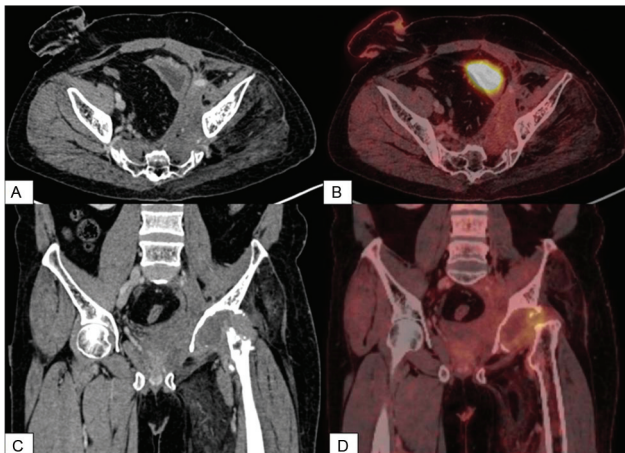


Fig. 2 PET CT images (A) CT Axial soft tissue window, (B) PET CT Axial Fusion image, (C) CT coronal soft tissue window, (D) PET CT coronal fusion image: Low-grade FDG avid ill-defined soft tissue thickening along the left lateral pelvic wall, extending to the pre-sacral space and encasing the exiting left sacral nerve roots, external and internal iliac arteries, and compressing the iliac veins.

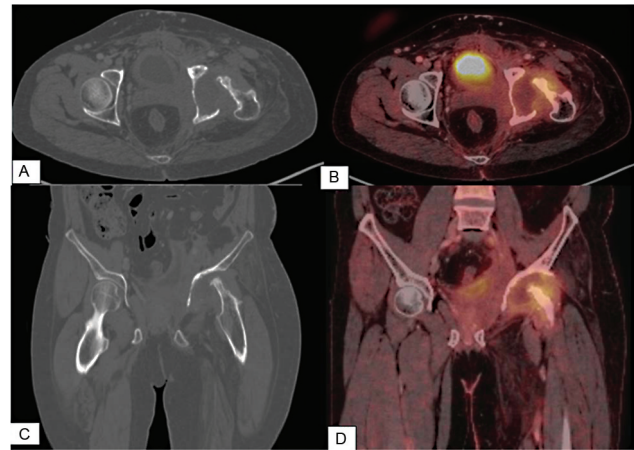


Fig. 3 PET CT images (A) CT Axial bone window, (B) PET CT Axial Fusion image, (C) CT coronal bone window, (D) PET CT coronal fusion image: destruction of the roof of the acetabulum, head, and the neck of the left femur with extensive bone resorption. Thinning and atrophy of the residual neck of femur giving a "licked candy appearance." There is antero-superior subluxation of deformed left hip joint. Moderate FDG activity noted around the residual neck of the femur and hip joint.

superior subluxation. No malignant lesion was seen around the left hip. Diffuse thickening of the synovium was seen corresponding to the moderate FDG uptake in the left hip joint, which appeared hypointense on T1, T2 and heterointense on STIR imaging and showed enhancement. Fluid collection was seen in the joint space with tiny loose bodies, which are hypointense on all pulse sequences. Left gluteal and adductor compartment muscles of the proximal left thigh were atrophic and show fatty replacement, which appear hyperintense on T1 and T2WI. Post-treatment changes with residual disease were seen on the left side of the pelvis extending to the presacral space and S1-S2 left neural foraminae encasing left external and internal iliac vessels and exiting left S1, S2 and S3 nerve roots (→ Figs. 4–6).

Discussion

Charcot osteoarthropathy is a destructive disorder of bones and joints secondary to neurosensory deficit that produces progressive degeneration of the joint, marked by bony destruction and resorption eventually leading to deformity.¹

Charcot joint can be caused by various underlying etiologies and common etiologies vary by the involved joint. Diabetes mellitus is the overall most common cause² and commonly affects the foot and ankle joints, of which tarso-metatarsal joint (medial > lateral) is the commonest joint involved. Syringomyelia is the most common predisposing etiology in the upper extremity and shoulder joint is mostly involved. Neurosyphilis/tabs dorsalis was much more common in the past, commonly affecting the hip and knee joints.³ Traumatic spinal cord injury is the most common cause in the spine Charcot. Other common etiologies include poliomyelitis, leprosy, tumors compressing, or involving the spinal cord or peripheral nerves, alcoholism, amyloidosis, multiple

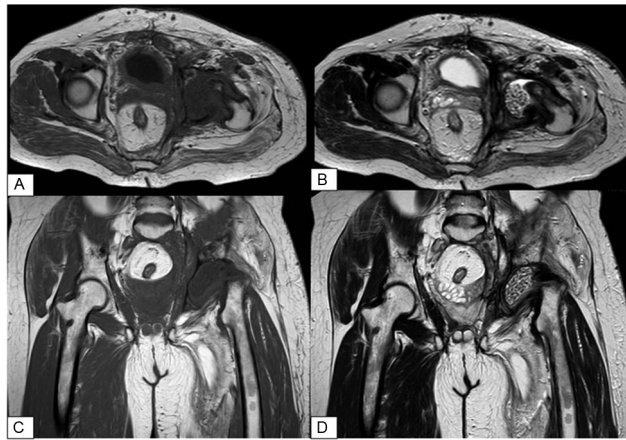


Fig. 4 MRI images (A) Axial T1, (B) Axial T2, (C) coronal T1, and (D) coronal T2: extensive joint deformity, complete resorption of the head of the left femur, and antero-superior subluxation was seen. Diffuse thickening of the synovium in the left hip joint, which appears hypointense on T1 and T2WI. Joint effusion is seen with multiple loose bodies/debris (B and D). Volume loss and fatty infiltration of gluteal and adductor muscles of the left thigh is seen.

sclerosis, steroid use and congenital insensitivity to pain (Table 1).

Two main theories explaining pathophysiology.

- **Neurotraumatic theory:** Loss of joint sensation and normal innervation leading to repeated trauma resulting in progressive damage.
- **Neurovascular theory:** Loss of sympathetic vascular tone causes regional vasodilatation and results in increased bone resorption and destruction.

There are two radiographic patterns.

- **Atrophic pattern**—predominant findings are bone resorption, osteopenia and osteolysis described as “licked candy stick” appearance.
- **Hypertrophic pattern**—conventionally described as “6Ds” of hypertrophy according to Yochum and Rowe,⁴ which are distended joints, density increase,

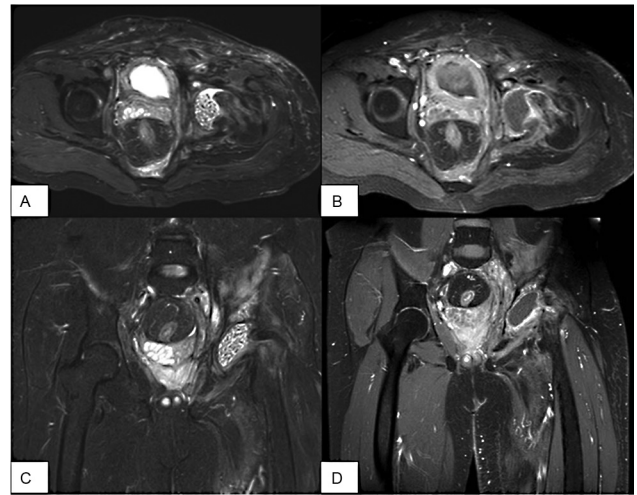


Fig. 5 MRI images (A) axial STIR, (B) axial post contrast, (C) coronal STIR, and (D) coronal post contrast: diffuse thickening of the synovium is seen in the left hip joint and appears heterointense on STIR imaging. There is smooth peripheral enhancement. No malignant lesion around the left hip.

debris production, dislocation, disorganisation and destruction.

The modified Eichenholtz classification⁵ is most commonly used for staging.

Stage 0: Acute Inflammatory/Prefragmentation Stage

Clinical signs such as swelling, warmth, and erythema are seen with normal radiograph.

Stage 1: Acute Charcot/Developmental/Fragmentation stage

Periarticular fracture, subluxation, and dislocation of joint leading to unstable and deformed foot.

Stage 2: Subacute Charcot/Coalescence Stage

Resorption of bone debris, sclerosis, and consolidation of larger bone fragments.

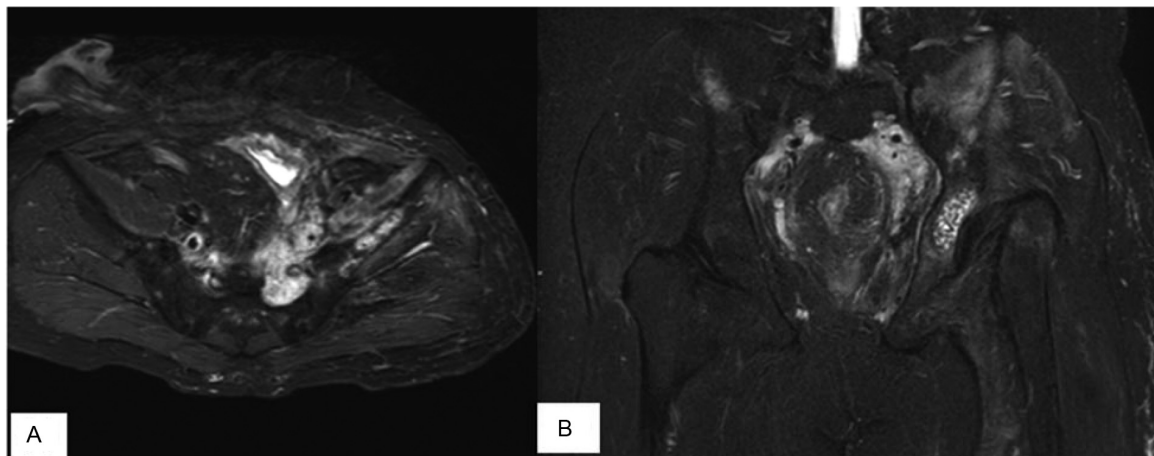


Fig. 6 MRI images (A) axial STIR, (B) coronal STIR: post-treatment changes with residual disease are seen on the left side of the pelvis extending to the presacral space and S1-S2 left neural foraminae encasing the left external and internal iliac vessels and the exiting left S1, S2, and S3 nerve roots.

Table 1 Causes of Charcot joint

Diabetes mellitus
Syringomyelia
Syphilis
Congenital insensitivity
Cerebral palsy associated sensory loss
Alcoholic neuropathy
Leprosy
Post-traumatic sensory neuropathy
Tumors compressing sensory nerves

Stage 3: Chronic Charcot/Consolidation/Reparative Stage

With fusion of involved fragments, arthrosis and fibrous or bony ankylosis leading to restabilization of the foot. Stable joint with evident deformity in radiographs.

In the fragmentation stage, earliest radiological finding is subchondral osteopenia.⁶ This is followed by bony fragmentation (with debris formation that is seen as intra articular loose bodies) and malalignment of joint due to ligamentous laxity (subluxation or dislocation). Bony consolidation begins in the coalescence stage, which is accompanied by subchondral sclerosis and periosteal bone formation. In this stage, absorption of the smaller fragments and fusion of the larger fragments is a common radiological finding. In consolidation stage, bony remodeling and ankylosis leads to permanent deformity. MRI plays an important role in the early detection of disease, assessment of the disease extent, and diagnosis of complications.⁷ On T1WI, the involved joints show decreased signal intensity and on T2WI, joint effusion shows increased signal intensity. STIR imaging helps in the early detection of marrow edema, which shows increased signal intensity. On post-contrast imaging, enhancement of inflammatory areas seen with central non-enhancing necrotic areas.

Subluxation or dislocation in early stages, which progress to bony remodeling and ankylosis, finally resulting in joint destruction and permanent deformity are the common complications of Charcot.⁸ Super added infection with gas-producing organisms can also be seen as a complication. It can cause major morbidity including limb amputation, especially in diabetes mellitus patients. Charcot affecting foot joints can lead to Rocker bottom feet deformity. In shoulder joint Charcot, upper extremity deep vein thrombosis and acromial stress fracture are rare complications following surgical interventions. Spine Charcot can lead to neurological deficits and paraplegia.

Charcot neuro-osteoarthropathy of the hip joint is very rare. Nicholas et al have described a case of neuropathic arthropathy of the hip as a sequela of undiagnosed tertiary syphilis,² where serological evidence of active infection with imaging features of neuro-osteoarthropathy were found including the destruction of the left hip with fluid and soft tissue densities in the joint space, along with extensive bone

resorption from the left acetabulum and femoral head, and significant osseous debris. Similarly, hip joint involvement was seen in our case with the destruction of the roof of the acetabulum, head and neck of left femur with extensive bone resorption.

Gunay et al have described an unusual presentation of Charcot arthropathy caused by syringomyelia mimicking a soft tissue tumor.⁹ It was a case of neuropathic arthropathy secondary to syringomyelia, which was misdiagnosed and treated as a soft tissue tumor. The patient had clinical and radiological signs of syringomyelia and imaging of the left shoulder revealed destruction of the humeral head, synovial hypertrophy, a large amount of joint effusion, and an irregular mass formation. Similarly in our patient, the post-surgical changes were mimicking a left pelvic soft tissue mass, but with later imaging found to have neuropathic left hip joint.

Differential Diagnosis

Osteomyelitis is another potentially limb-threatening complication of diabetic neuropathy similar to Charcot neuro-osteoarthropathy.¹⁰ Imaging plays a major role in diagnosis because the approach to treatment is quite different. Active Charcot foot shows a peri-articular pattern with subchondral bone edema and usually involves several bones and joints.¹¹ While osteomyelitis has a tendency to involve a single bone with diffuse marrow involvement mostly associated with skin ulcerations and sinus tracts. Fluid collections can be present in both, while it can be larger in osteomyelitis and diffusion-weighted imaging might help to differentiate an abscess from a non-infected fluid collection. Subcutaneous fat is preserved in Charcot while overlying cellulitis is often seen in osteomyelitis. The presence of subchondral cysts and intra-articular loose bodies are typical features of Charcot which indicates the absence of infection. While these are not seen in osteomyelitis because these gets disappeared in the presence of infection due to resolution or obscuration by surrounding inflammation. "Ghost sign"¹² that indicates disappearance of bone on T1WI, which become distinct on T2 or post contrast imaging – is positive in osteomyelitis. Neuropathic arthropathy and osteomyelitis may coexist in some patients,¹³ where there will be overlapping clinical signs and imaging findings such as cellulitis, joint effusion, and soft-tissue and bone marrow edema. Imaging plays a major role in differentiating the two because the approach to treatment is quite different (► **Table 2**).

Septic arthritis resulting from an intra-articular infection leading to destructive arthropathy¹⁴ is another differential diagnosis for Charcot. In septic arthritis, severe symptoms such as pain and decreased range of motion are seen usually associated with fever. Here the underlying pathology is acute rapidly progressive, unlike Charcot. Large joints are commonly involved because of the abundant blood supply to the metaphyses making them prone to bacterial infection. Radiograph shows narrowing of the joint space and joint effusion may be seen. Juxta articular osteoporosis and destruction of cartilage may be seen; however, bony

Table 2 Differences between Charcot joint vs. osteomyelitis

Feature	Charcot		Osteomyelitis
Location	Most common site - tarsometatarsal and metatarsophalangeal joints		Most common site - metatarsophalangeal and interphalangeal joints, calcaneus and cuboid bone
Bone marrow pattern	Periarticular pattern with subchondral bone edema		Bone destructive disease with diffuse bone edema
Adjacent skin ulcerations and sinus tracts	Usually not seen		Often seen
Fluid collections	Present Usually smaller with no restricted diffusion		Present Usually Larger (abscess) with restricted diffusion
Subcutaneous fat	Often preserved		Often present with cellulitis
Subchondral cysts and intra articular loose bodies	Typically present		Typically not seen due to resolution or obscureness by surrounding inflammation
"Ghost sign"	Negative		Positive

involvement and destruction are much less than those seen in Charcot. Hallmark features of Charcot such as subchondral cysts and intra-articular loose bodies are absent in septic arthritis. Prompt treatment is required in this condition to avoid permanent joint damage.

Inflammatory arthritis caused by an influx of inflammatory cells include a group of joint disorders seen associated with synovial inflammation and hyperplasia.¹⁵ Radiographic features include synovial hyperplasia, joint effusion, and bone erosions. Synovial involvement is not seen in Charcot while subchondral cysts and intra articular loose bodies are absent in inflammatory arthritis. The presence of extra-articular inflammation may also be seen such as tenosynovitis and enthesitis and often inflammatory arthritis are seen associated with inflammatory conditions of other organs, which may also help to differentiate from Charcot.

Chondrosarcoma of the hip is another rare condition, which may mimic Charcot, where is chondroid matrix is seen instead of bony debris.¹⁶ Long tubular bones are commonly affected, because of the abundance of cartilage and femur is the most commonly involved bone. Plain radiograph shows lytic lesion with popcorn calcification and endosteal scalloping. Higher grade tumors show permeative appearance with significant periosteal reaction. Chondrosarcoma of the proximal femur with extension to the hip joint may mimic Charcot hip joint; however, the presence of a well-defined lytic lesion with soft tissue component and absence of typical radiological features of Charcot will help in differentiation between the two.

Conclusion

Though Charcot neuro-osteoarthropathy was not considered as a primary diagnosis in our patient, imaging features were confirmatory for atrophic pattern of neuro-osteoarthropathy of the left hip joint consequent to chronic compression and encasement of sacral nerve roots with no other predisposing

Table 3 Features of typical Charcot joint vs. our case

Feature	Typical Charcot	Our case
Common joints involved	Ankle and foot joints	Hip joint
Bone resorption and osteolysis	Present	Present
Joint destruction	Present	Present
Subluxation/dislocation	Present	Present
Synovial thickening	Present	Present
Joint effusion with debris	Present	Present
Joint deformity	Present	Present
Atrophy of adjacent muscles	Present	Present

factors such as diabetes, syphilis, syringomyelia, or other spinal cord disorders were seen.^{17,18} Knowledge about the etiologies and radiological appearances of Charcot joint will help in early diagnosis and prevention of further progression, leading to permanent joint deformity (► **Table 3**).

Conflict of Interest

None declared.

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