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Case Report

Diagnosis behind the mask: A rare case of infected Charcot's spine ☆

Giedre Kučinskaite, MD[a], *Theodor Lutz, MD*[b], *Sönke Frey, PhD, MD*[c], *Mark Wetterkamp, MD*[d], *Tobias L. Schulte, PhD, MD*[d,1,*], *Carsten Lukas, PhD, MD*[e,f,1,*]

[a] *Department of Diagnostic and Interventional Radiology, St. Josef Hospital, Ruhr University of Bochum, Gudrunstrasse 56, Bochum, 44791, Germany*
[b] *Department of Diagnostic and Interventional Radiology, St. Marien Hospital Hamm, Germany*
[c] *Department of Orthopedics, Emergency Surgery and Hand Surgery, Florence Nightingale Hospital, Dusseldorf, Germany*
[d] *Department of Orthopedics and Trauma Surgery, St. Josef Hospital, Ruhr University of Bochum, Bochum, Germany*
[e] *Institute of Neuroradiology, St. Josef Hospital, Ruhr University of Bochum, Bochum, Germany*
[f] *Department of Neurology, St. Josef Hospital, Ruhr University of Bochum, Bochum, Germany*

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ABSTRACT

Charcot's spine is a very uncommon long-term complication of spinal cord injury. Infection of the spine is a common pathology, but infection of a Charcot's spine is rare and is challenging to diagnose, especially in differentiating between the Charcot defect and the osteomyelitis defect. Surgical reconstruction has to be extremely individualized. A 65-year-old man with a history of thoracic spinal cord injury with paraplegia 49 years ago was admitted to our hospital with high fever and aphasia. After a thorough diagnostic process, destructive Charcot's spine and secondary infection were diagnosed. This report additionally reviews the surgical management of secondary infected destructive lumbar Charcot's spine and follows the patient's recovery and postoperative quality of life.

Abbreviations: SPB, Streptococcus pyogenes bacteremia; CRP, C-reactive protein; T, thoracic vertebra; L, lumbar vertebra; CT, cranial computed tomography; MRI, magnetic resonance imaging; S, Sacrum.

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* Corresponding author.
E-mail address: giedre.kucinskaite@klinikum-bochum.de (G. Kučinskaite).

[1] Contributed equally.

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Introduction

Spinal neuroarthropathy, also known as Charcot's spine, is a very uncommon progressive osseous and ligamentous disease of the spine secondary to loss of deep sensation and proprioception, frequently after a traumatic event, occurring a mean of 17.3 years after the onset of neurological impairment [1,2,6]. The usual clinical presentation is progressive thoracic or thoracolumbar kyphosis, usually causing a loss in the patient's height. In addition, patients may develop an increase in spasticity in the lower limbs as well as back or leg pain. Due to impaired sensation, the degree of destruction and instability is often much more severe than expected in relation to the minor clinical presentation [6]. To the best of our knowledge, there have only been a few reported cases of secondary infected Charcot spine [2–5].

Case presentation

A 65-year-old man was admitted to our hospital with high fever and aphasia. Laboratory investigations showed elevated C-reactive protein (CRP) (119.8 mg/L). During the physical examination, the patient did not show any signs of pain. Some erubescence in the right groin area was noticed, potentially indicating intra-abdominal pathology. The patient was known to have suffered from complete paraplegia after traumatic spinal cord injury 49 years earlier, with fracture of the vertebral body at T11 and previous surgical instrumentation and fused levels T8-L2. With the exception of aphasia and the known paraplegia, the further neurological examination was unremarkable. Intracranial pathologies were excluded by cranial computed tomography (CT).

However, X-ray, abdominal CT scanning and magnetic resonance imaging (MRI) revealed extensive destruction of L3 and L4, with extreme instability. In addition, very large abscess formations at L3-L4 were diagnosed on the right side, including complete vertebrae and paravertebral tissues, descending down to the groin (Figs. 1, 2, and 3).

Earlier radiological images indicated that after instrumented fusion following the trauma 49 years previously, the patient had first developed a degenerative lumbar scoliosis that ultimately led to destruction of the L3 and L4 vertebrae, corresponding to a Charcot's spine. Radiographs taken 2 years before the current presentation already revealed this destruction, without any signs or symptoms of infection (Fig. 4). In conclusion, a diagnosis of a severely infected Charcot's spine was made.

Due to the patient's severe clinical condition and vast abscess formation, the first goal was to immediately start treatment with surgery in an emergency setting, during which the abscess was surgically drained via an open posterior lum-

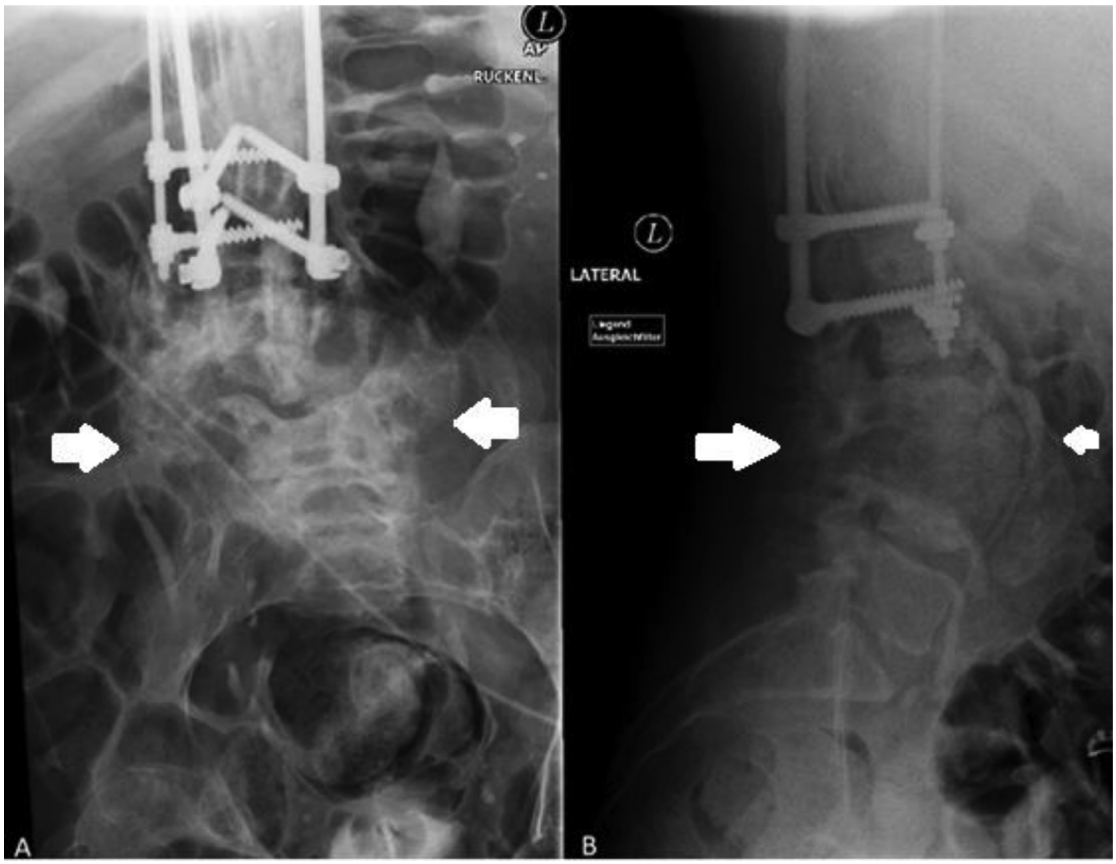

Fig. 1 – Preoperative lateral (A) and frontal (B) radiographs showing a fused spine at the T8-L2 level with destruction of the L3 and L4 vertebra (white arrow).

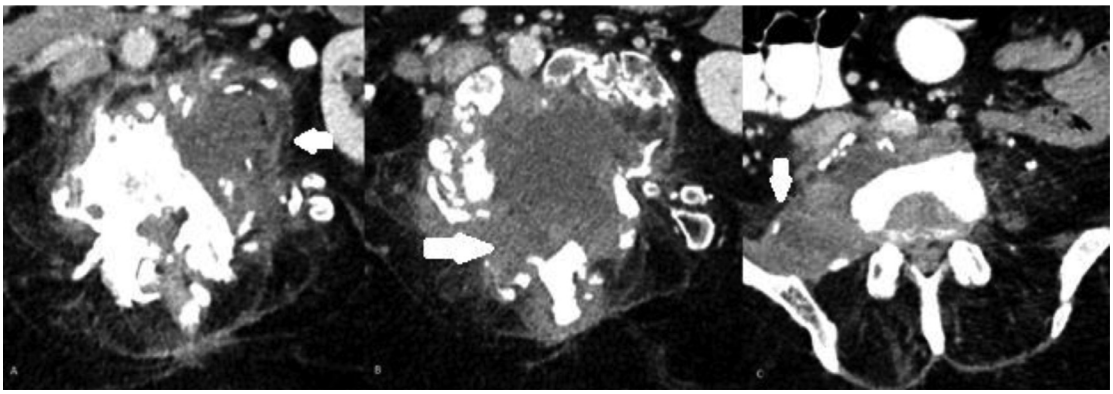

Fig. 2 – The preoperative CT scan (A) showed inflammatory bone damage below the spinal fusion and extend of infection in the paravertebral tissue (B) with abscess formation in the right psoas muscle (C) (white arrows).

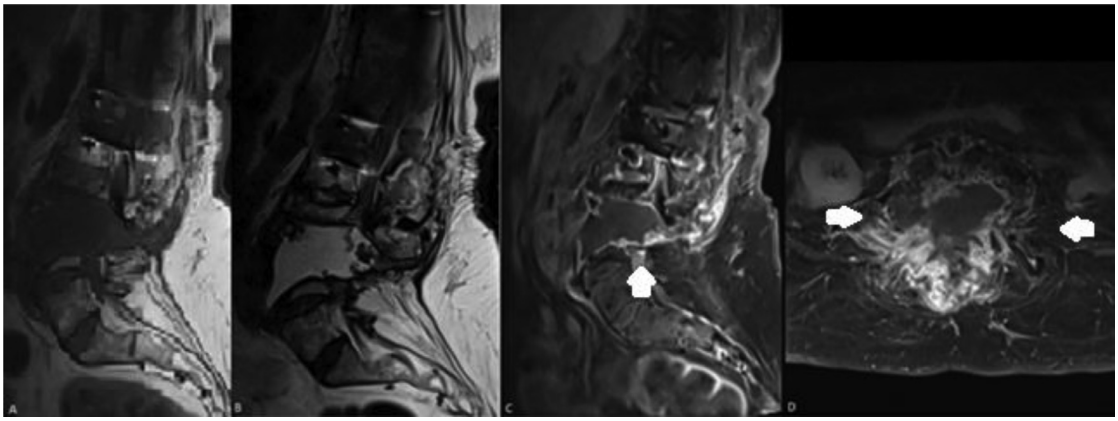

Fig. 3 – The MRI T1- and T2-weighted images (A ,B) and images after contrast administration (C, D) show the extent of infection in the paravertebral tissue, with abscess formation in both psoas muscles as well as spinal empyema at the L2-L4 level (white arrows).

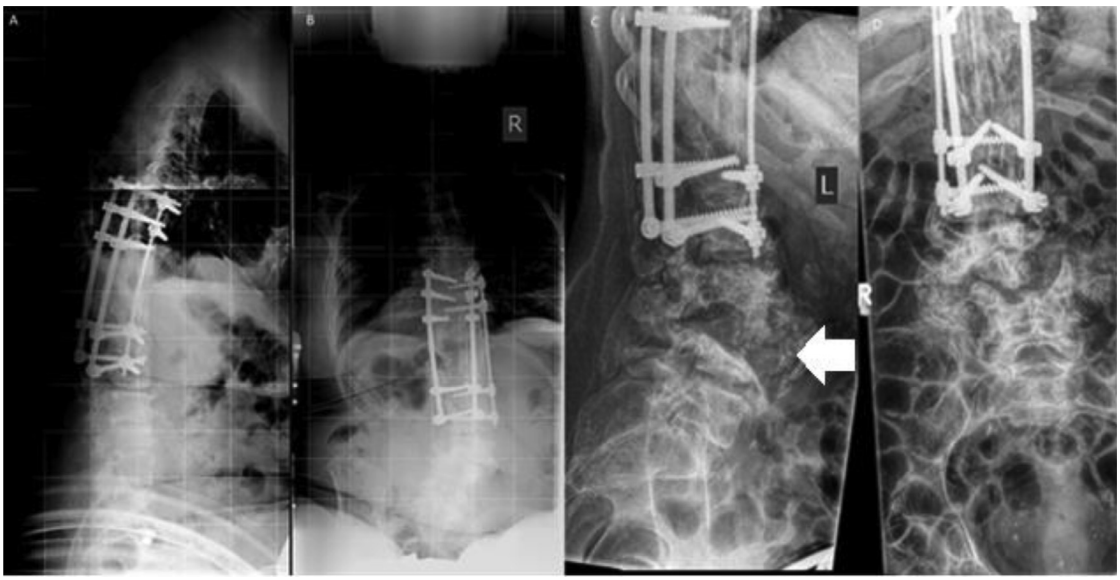

Fig. 4 – Lateral (A) and frontal (B) radiographs 42 years after the initial trauma in 2011, showing a fused spine at the T8-L2 level with degenerative lumbar scoliosis. The lateral (C) and frontal (D) radiographs 5 years later show new destruction of the L3 and L4 vertebra (white arrow).

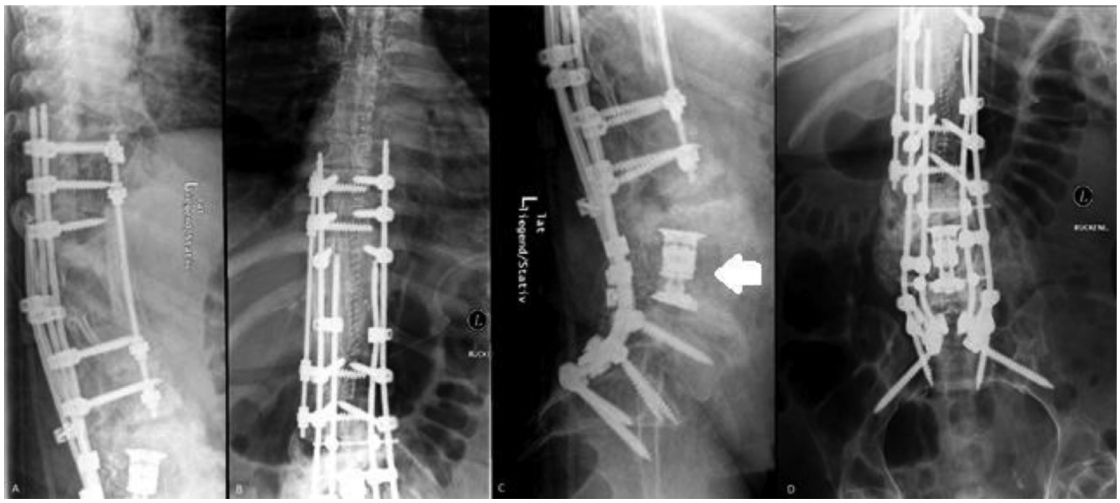

Fig. 5 – Lateral (A, C) and frontal (B, D) postoperative radiographs after instrumentation at T8-S2/ilium and intermittent reconstruction of the anterior spine using a titanium cage inserted from a posterior approach (white arrow).

bar approach, a biopsy was taken, and broad-spectrum intravenous antibiotic treatment was started. Two days later, a second operation was performed via a posterior approach: the old pedicle screw and rod construct at T8-L2 was removed, posterior instrumentation at T8–S2/ilium was performed, and approximately 70% of the L3 and L4 vertebrae was removed. A very unusual finding was the fact that a thecal sac was not present inside the former spinal canal. It had apparently atrophied due to the spinal cord lesion many years previously. The titanium cage for intermittent reconstruction of the anterior column could therefore be inserted from posterior "through" the spinal canal to achieve primary stability (Fig. 5).

After surgery, the patient recovered without any postoperative complications and was transferred to a rehabilitation center 1 week after the initial admission.

A third operation was performed 3 weeks later, after the patient had recovered from the systemic infection, in order to reconstruct the anterior column properly and biologically. Via an anterior transperitoneal approach, the vertebral body replacement cage that had been intermittently inserted from posterior to achieve short-term anterior stability was removed, radical debridement was completed, and a new cage was inserted from an anterior approach. Additionally, a fibula graft from the patient was placed anteriorly to the cage at L2-L5 to promote fusion biologically (Fig. 6). The postoperative recovery was again uneventful, without any complications. At a 1-year follow-up examination, the patient showed no signs of infection and no pain. Radiologically, there were no pathological findings (Fig. 6). The instrumented spine was stable.

Discussion

The diagnosis is usually made on the basis of similar patterns, although in the present case the differential diagnosis was more complicated due to severe sepsis causing neurological impairment, that is, aphasia. True Aphasia is an impairment of language caused by damage to the language area of the brain, primarily Broca and Wernicke areas. Main reasons include ischemic events or intracerebral hemorrhage which could be ruled out in the present case by cranial CT [7]. As such, the causes of neurological symptoms can be seen as secondary related to a systemic infection with consequent septic encephalopathy being the most frequent complication in the acute stage of the disease [8].

Inguinal erubescence suggested an abdominal pathology, and this led to abdominal CT being carried out. Spondylodiscitis or pyogenic osteomyelitis were the primary suspected diagnoses until the X-rays from 2011 and 2016 were received, clearly indicating Charcot's spine in the past. The main clues that led to the diagnosis were the history of trauma and the patient's imaging history and many years of paraplegia [5].

Following a confirmed diagnosis of Charcot's spine, the aim of treatment is usually to eradicate the infection, restore and preserve the structure and function of the spine, and alleviate pain [10,12].

In cases of uncomplicated spondylodiscitis, conservative therapy is the first-line treatment. Severe vertebral osteomyelitis with relevant instability usually requires radical debridement, instrumented stabilization, and decompression of neural structures [9,11].

In the present patient, who had a combination of Charcot's spine and vertebral osteomyelitis, the situation was complex. The initial goal was to treat the urgent sepsis by relieving and draining the abscess formations via a posterior approach in the emergency situation. After that, the goal was to stabilize and reconstruct the spine via posterior and anterior approaches.

During the further diagnostic workup Streptococcus pyogenes bacteremia (SPB) could be confirmed by positive blood culture tests. SPB is a severe condition with high mortality most commonly occurring in the setting of skin and wound infections, followed by respiratory tract infections [13]. Extensive physical examination revealed a gluteal decubitus with surrounding erythema. Although the gluteal erysipelas is very

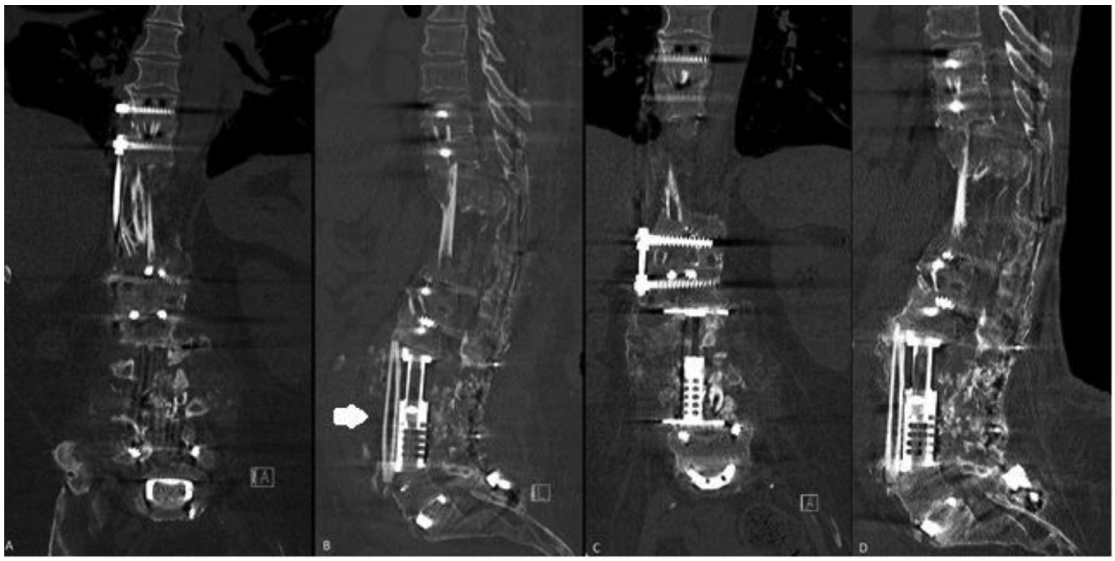

Fig. 6 – Postoperative CT (A, B) scans after the third operation, with a new cage and a fibula graft anterior to the L2-L5 (white arrow). One year after the last operation, the CT scans (C, D) do not show any visible changes.

rare it has been associated with pressure ulcers in elderly often bedridden or immobilized patients and in our case a likely origin of hematogenous seeding into the spine. Hence, adjustments of antibiotic therapy according to the results of final blood cultures led to quicker recovery [14].

Conclusion

In summary, spinal neuroarthropathy, that is, Charcot's spine, is a rare disease. It must always be considered in paraplegic or tetraplegic patients with a history of spinal fusion, even in the absence of infection. Infection of a Charcot's spine is even more rare, but needs to be taken into account in Charcot's spine patients with a septic clinical presentation. Special forms of reconstruction and stabilization may be necessary due to the massive destruction present.

Author contributions

Giedre Kučinskaite: conceptualized and drafted the case report, made design of the work and analyzed it. Theodor Lutz: conceptualize and analyzed the case report, approved the submitted version. Sönke Frey: conceptualized and substantively revised the case report, approved the submitted version.

Mark Wetterkamp: analyzed and revised the case report, approved the submitted version. Tobias L. Schulte: analyzed and substantively revised the case report, also approved the submitted version. Carsten Lukas: conceptualized, drafted and substantively revised the case report, approved the submitted version.

Patient consent

The patient gave full consent. We can provide signed copy of our patient consent permission form if needed.

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