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Vertebral destruction in an 11-month-old child with spinal tuberculosis: a case report and review of literature

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Abstract

Background: The incidence of tuberculosis is increasing especially in endemic countries. Spinal tuberculosis represents nearly the 50% of reported cases of skeletal tuberculosis. This is the youngest case of spinal tubercular disease that has been reported. The objective of this report is to describe a spinal tuberculosis case in an infant in thoracic spine, in order to show the importance of early diagnosis in this population, to limit the progression of this highly destructive disease and reduce the severe sequelae that this disease is associated.

Case presentation: An 11-month-old infant previously healthy born in the northeast Mexico. Physical examination revealed a mass lesion in the dorsal region, fixed to deep planes, indurated. Neurological examination found Frankel C paraparesis showing muscle strength 2/5 on the Lovett scale in both lower extremities, anal reflex present, and preserved sensitivity.

In the magnetic resonance of the spine, hyperintensities in the vertebral bodies of D6-D9 were observed in the T2 with destruction of the D7 and D8 bodies. A thoracotomy was performed with total mass resection with corpectomy of vertebrae D7 and D8, medullary decompression, and placement of fibula allograft between vertebrae D6 and D9. In the histopathological sample, a chronic granulomatous inflammatory process associated with acid-fast bacilli was observed, in addition to presenting a positive result in quantitative real-time PCR GeneXpert MTB/RIF sensitive to rifampicin. Twelve months later, he presented 5/5 muscular strength, without alterations in sensitivity, in addition to presenting ambulation onset at 18 months of age.

Conclusion: The spinal tuberculosis is a disease that occurs in endemic countries. A prompt diagnosis is necessary to limit the progression of a highly destructive disease. In addition, the fact of presenting at an early age produces hard making decisions for the adequate treatment of the disease and reduces the adverse effects of these procedures.

Keywords: Spinal tuberculosis, Pott's disease, Infant, Spine surgery

Background

The incidence of tuberculosis is increasing especially in endemic countries; skeletal tuberculosis is found in nearly 10% of patients; therefore, spinal tuberculosis represents nearly the 50% of reported cases of skeletal tuberculosis [1]. In a previous study in a pediatric reference center of infectious diseases in Mexico, 85% of the patients presented extrapulmonary tuberculosis, and 29% presented involvement at osteoarticular level [2].

Spinal tuberculosis is a secondary infection that usually occurs through hematogenous spread. In contrast to the other spinal infections, 95% of spinal tuberculosis starts in the anterior vertebral body. The infection spreads from

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the arteries in the paravertebral area or the valveless venous plexus (Batson paravertebral plexus) to the central vertebral body. The infection tends to spread under the ligaments below the anterior longitudinal ligament and into the posterior vertebral body. Due to the lack of proteolytic enzymes inherent in bacillus, the intervertebral disc is usually the last to be affected. The clinical manifestations of spinal tuberculosis may be related to osseous or non-osseous involvement of the spine or both, with osseous (vertebral body) disease (also known as Pott's spine) being the most common manifestation [3–6].

Usually, spinal tuberculosis is characterized by the destruction of the intervertebral disc space and adjacent vertebral bodies. The type of spinal deformity depends on the location of tuberculous vertebral disease. Kyphosis is the most common spinal deformity that occurs in thoracic lesions [7].

Spinal tuberculosis occurs mostly in children and young adults [8, 9]. The TB spine lesion in children causes more destruction as most of the vertebral bodies are cartilaginous.

The treatment targets are to confirm the diagnosis, eradicate the infection, achieve a decompression of the spinal canal material, and correct or prevent spinal deformity and possible sequelae [3, 5].

This is the youngest case of spinal tubercular disease that has been reported. The objective of this report is to describe a spinal tuberculosis case in an infant in thoracic spine, in order to show the importance of early diagnosis in this population, to limit the progression of this highly destructive disease and reduce the severe sequelae that this disease is associated.

Case presentation

The patient is an 11-month-old infant, without significant family history, previously healthy born in the northeast region in Mexico with complete vaccination schedule for his age, receiving 1 dose of Bacillus-Calmette-Guérin (BCG) vaccine at birth.

His condition began 3 months prior to admission when he presented progressive volume increase in the dorsal region. Subsequently, 2 months prior to admission, he began with a setback in developmental milestones (he achieved standing at 9 months, without the beginning of ambulation), as well as a decrease in muscle strength in the lower extremities, for which he gone to medical consultation, who requested a simple magnetic resonance imaging of the spine where a swelling was observed at the D7–D8 level, for which he was referred to our hospital for its diagnostic approach. The patient did not present signs of tuberculosis such as anorexia, night sweats, fever, and weight loss.

Physical examination revealed a mass lesion in the dorsal region, fixed to deep planes, indurated, painful, without erythema. Neurological examination found Frankel C paraparesis showing muscle strength 2/5 on the Lovett scale in both lower extremities with increased tendon reflexes and bilateral Babinsky present, anal reflex present, and preserved sensitivity.

Within his admission laboratory studies, the hematologic analysis found hypochromic microcytic anemia and reactive thrombocytosis: hemoglobin 9.87 g/dL, mean corpuscular volume 75.4 fL, mean corpuscular hemoglobin concentration 31.9 g/dL and platelets 781 K/uL, C-reactive protein (CRP) 13 mg/L (normal value less than 10 mg/L), and erythrocyte sedimentation rate (ESR) of 28 mm/h (normal value less than 15 mm/h in men under 50 years of age).

In the imaging studies, a loss of size of the vertebral body of D7 and D8 with anterior wedging of both was observed in anteroposterior radiography of the dorsal column (Fig. 1). In the simple CT of the dorsal spine, destruction of the vertebral bodies D7 and D8 was observed (Fig. 2), and finally, in the contrast magnetic resonance of the spine, hyperintensities in the vertebral bodies of D6–D9 were observed in the T2 with enhancement after the administration of gadolinium and destruction of the D7 and D8 bodies, as well as involvement of the spinal canal at these levels; there was also no lesion in the posterior arches (Fig. 3).

At 2 weeks, he presented a decrease in muscle strength (1/5) in both lower limbs, and 3 weeks later, he presented neurogenic bladder data, in addition to multiple episodes of urinary tract infection treated with antibiotic therapy.

A diagnostic thoracoscopy was performed for a minimally invasive approach due the age of the patient; however, we could not obtain a sufficient sample for the diagnosis by anatomopathological study. Because of that, a thoracotomy was performed with total tumor resection with corpectomy of vertebrae D7 and D8, medullary decompression, and placement of fibula allograft between vertebrae D6 to D9 (Figs. 4 and 5).

In the histopathological sample, a chronic granulomatous inflammatory process associated with acid-fast bacilli was observed, in addition to presenting a positive result in quantitative real-time PCR GeneXpert MTB/RIF sensitive to rifampicin.

Intensive phase treatment was started with isoniazid at 10 mg/kg, rifampicin 20 mg/kg, pyrazinamide 25 mg/kg, and ethambutol at 25 mg/kg for 2 months, followed by a maintenance phase with 2 drugs (isoniazid at 10 mg/kg and rifampicin at 20 mg/kg) during 10 months, as it is stipulated in the World Health Organization's guidelines [10]. Neuromuscular rehabilitation was started. A hepatic

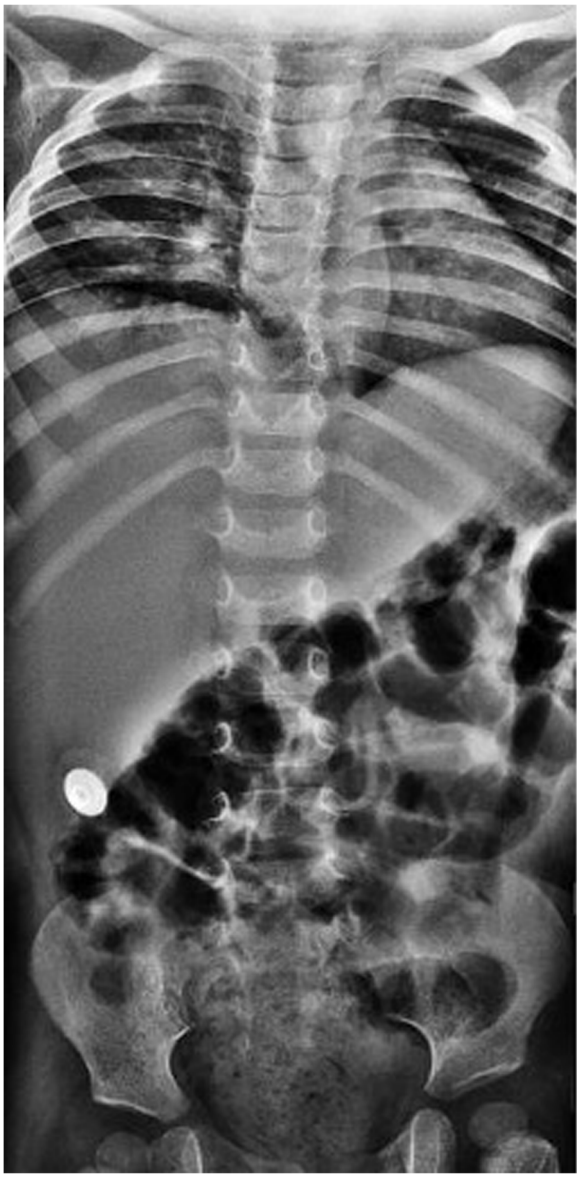

Fig. 1 Ap view of a complete spine radiograph with paravertebral swelling and destruction of D6

assessment was systematically carried out every 3 months until the end of the treatment protocol.

One week after the start of treatment, he presented 4/5 motor improvement in both lower extremities, as well as improvement in urinary symptoms. Primary immunodeficiencies were ruled out: quantitative determination of immunoglobulins: IgA 122.3 mg% (13–102 mg%), IgG 959.1 mg% (349–1139 mg%), IgM 216.1 mg% (20–229 mg%), IgE 32.1 UI/mL (0–60 UI/mL), HIV negative. He was discharged 2 weeks after surgery with a double assembly corset.

Twelve months later, he presented 5/5 muscular strength, without alterations in sensitivity, without neurogenic bladder data, in addition to presenting ambulation onset at 18 months of age (Fig. 6).

Discussion

Tuberculosis is regarded as a disease of vulnerable groups, including low socioeconomic status and fragile health conditions. Immune weakened patients, patients at extreme ages, diabetic patients, smokers, oncologic patients, and alcoholics are at increased risk (WHO, 2017). This is the younger case described of spine tuberculosis destruction syndrome, based in all the cases reported and indexed to PubMed and science direct.

Although elevated leukocyte, ESR, and CRP values have been observed in patients with both pulmonary and extrapulmonary tuberculosis, our patient presented a slightly elevation of the ESR of 28 mm/h. There have been described in the literature that up to 26% of patients with tuberculosis have a normal value of these parameters. Although they have a very high sensitivity, they are not highly specific parameters, which is why these are primarily used in monitoring disease activity and evaluating response to treatment [11].

Patients with spine pain should be kept under observation in an endemic region for tuberculosis and sequentially assessed with X-rays, every 4 to 6 weeks; in case of reduction of disc height they should be performed an MRI. Spinal tuberculosis lesions have been described as predominantly localized lesions in the thoracic region, mainly at the D6 level that can be confidently labeled as TB spine on MRI if the vertebrae show low signal in T1WI-weighted and hyper-intense signal in T2WI, suggestive of inflammation with a septate pre and paravertebral abscess and a contiguous vertebral body involvement with preserved intervertebral disc; these findings were similar to our patient helping us to evaluate the extension of the disease and start the treatment for the limitation of the spinal destruction [12].

The TB spine lesion in children causes more destruction as most of the vertebral bodies are cartilaginous [4, 5]. Our case was the youngest case of spinal tubercular disease that has been reported. The lack of information of the general population and the scarce symptoms that presents at the onset of the disease, usually delays diagnosis; if we add the aggressiveness of the disease in this age group, due to the innate characteristics of the disease, it causes to be diagnosed in advanced stages with high bone destruction. In a study carried out in a Mexican population, all the patients with Pott's disease presented a deformity of the spine cord at the time of diagnosis, with an average time of diagnosis of 8 months with a range of 1 to 48 months [2]. Similarly, an UK cohort reported

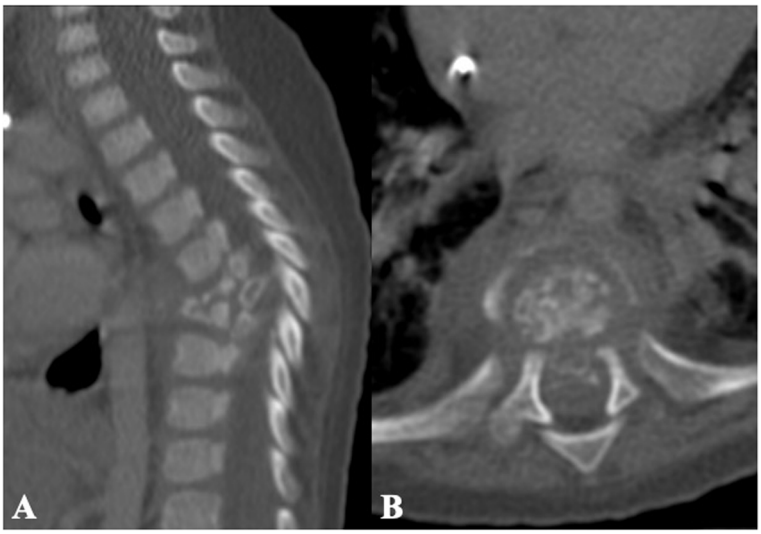

Fig. 2 Dorsal CT scan in bone window, the vertebral destruction of D7 and D8 is observed in **A** sagittal view and **B** axial view

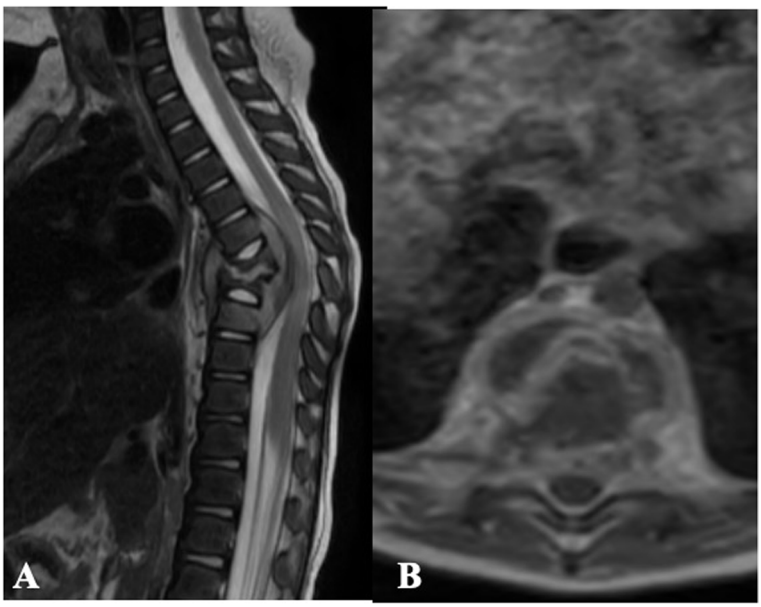

Fig. 3 **A** Sagittal T2-WI and **B** axial post contrast T1-WI. A prevertebral and epidural collection with enhancing wall with destruction of the D7 and D8 bodies. Also, the related spinal cord signal is compressed displaying abnormal bright signal signifying myelopathic changes

a median duration of symptoms before diagnosis of 7 months; whereas, the Taiwan cohort reported a median time of 2 months [13, 14].

Our patient did not presented constitutional tuberculosis symptoms, according to literature, only 20 to 30% of the patients with spinal TB have constitutional symptoms [1] making the initial approach to this type of patient even more difficult. Our patient presented neurological deficit at the time of the diagnosis; prevalence of these symptoms has been observed more than twofold times in developing countries; this is probably because of the lack of education of these syndromes around the population, as well as low economic resources that restrict the access to specialized studies for their diagnosis [1, 5]. In this type of patients, the neurological deficit depends proportionally to the size of the mass and the location, being the

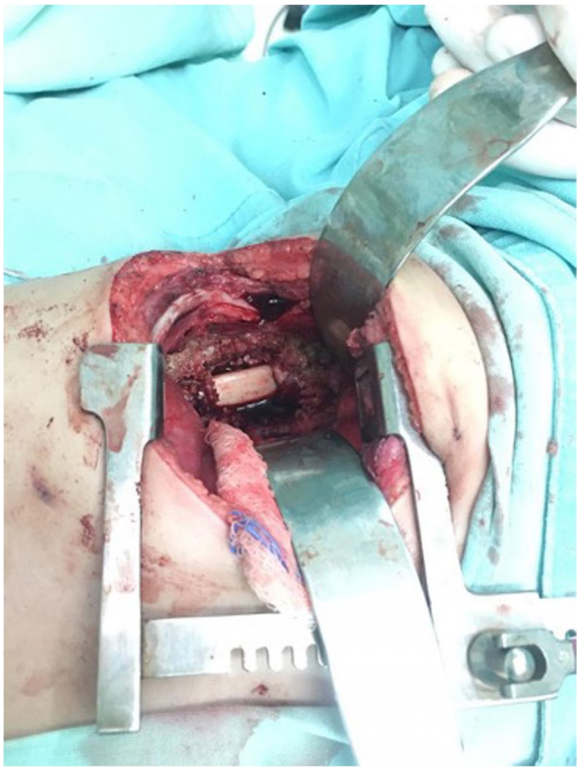

Fig. 4 Thoracotomy with corpectomy of vertebrae D7 and D8 and placement of fibula allograft between vertebrae D6 and D9

cervical and dorsal levels where it was presented in most of the patients [7, 15]. In order to get better outcomes in the developing countries, it is necessary to promote the dissemination of information in the population, in addition to carrying out an adequate approach in this type of patients.

It should be clarified that in Mexico since 1951, the BCG vaccine is part of the vaccination scheme, which is applied at birth, not recalled dose [16]. BCG vaccination is a highly cost-effective intervention against severe childhood tuberculosis [16]. Similar to our patient, multiple cases of bone tuberculosis have been described in children despite having the vaccine [17, 18]. This lack of protection must be assessed to reduce this catastrophic disease in this vulnerable group.

The base of the treatment is the antitubercular medication suggested for 12 months, starting during the first 2 months an intensive phase with isoniazid at 10–15 mg/kg, rifampicin 10–20 mg/kg, pyrazinamide 20–25 mg/kg, and ethambutol at 15–25 mg/kg. Subsequently, a support phase for the remaining 10 months based on isoniazid and rifampicin at the same previous doses in most of the patients, without surgery indications, responds to conservative treatment in 90–95% of the cases [10, 19–21].

The destruction of the vertebral body and endplate combined with the continuation of growth in the spine can cause deformity to progress. In 40%, the kyphosis

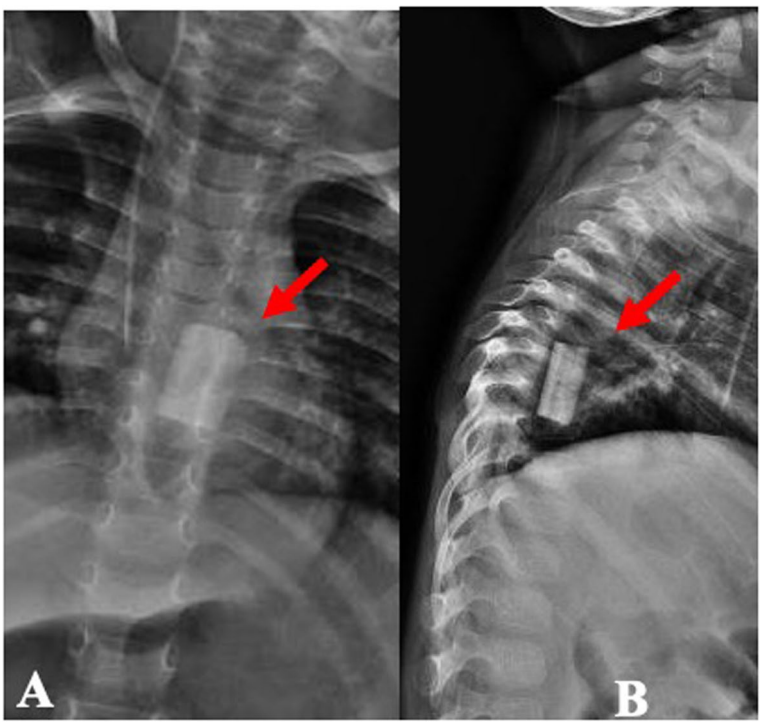

Fig. 5 **A** Ap view and **B** lateral view of a dorsal spine radiograph post-surgery with fibula allograft (red arrow) between vertebrae D6 and D9

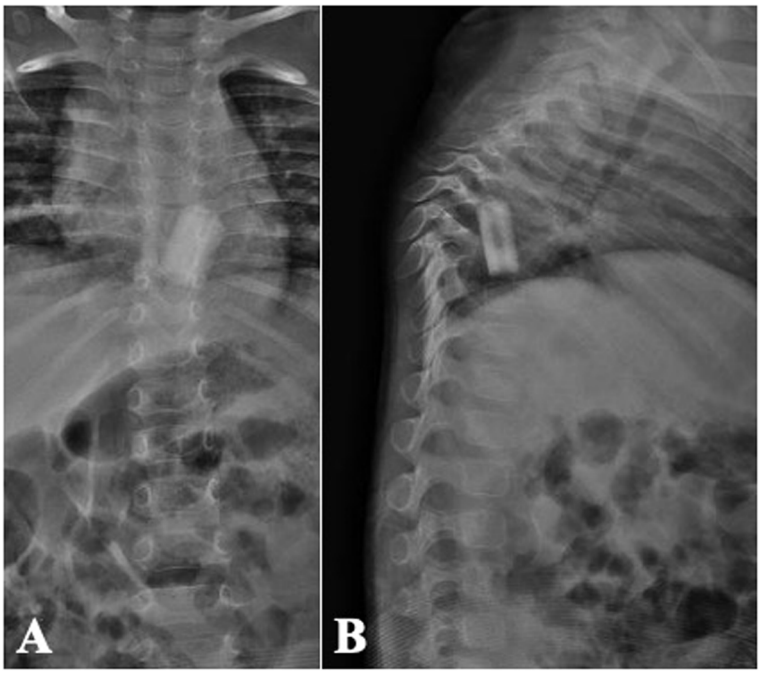

Fig. 6 **A** Ap view and **B** lateral view of a dorsal spine radiograph 12 months after the surgery with fibula allograft between vertebrae D6 and D9

worsens, 40% improves, and in 20% stays constant [3]. Consequently, surgery is indicated for complications such as deformity, neurological deficit, instability, huge abscess, diagnostic dilemma, and in suspected drug resistance to *Mycobacterium tuberculosis*. In our patient, the surgery was indicated for multiple complications as deformity, neurological deficit, and instability [5]. In children less than 7 years, that have 3 or more vertebral body involvement in a dorsal or dorso-lumbar spine, the deformity is severe and tends to progress further [5]. Our patient presented 2 of 3 clinical risk factors for developing progressive kyphosis, such as age less than 7 years, more than three vertebral body affection, only the lesion at distal dorsal, and dorso-lumbar junctional area was not present.

It should be noted that the growth potential is also disturbed when the disease focus is surgically approached. At the decompression of the spinal cord, the apophyseal ring is partially or completely damaged; consequently, the growth potential is partially or totally affected [1]. This suggests that posterior elements do contribute to growth and as far as possible anterior radical resection should be avoided in children [22, 23]

Therefore, an anterior decompression with total mass resection with corpectomy of vertebrae D7 and D8 was realized, with complete neurologic improvement. The "DiMeglio formula" states that for each vertebral segment, the person loss 0.7 mm/year of longitudinal growth after posterior arthrodesis [5]. For this reason, the posterior fusion of the thoracic spine to limit posterior vertebral growth and control the kyphosis deformity is pending.

In conclusion, spinal tuberculosis represents a huge challenge for its diagnosis because of its insidious clinical onset without specific clinical manifestations. However, this disease has been associated with catastrophic complications in pediatric age, due to the anatomical characteristics of the spine at this age, such as a severe deformity, an increased risk of vertebral collapse and neurological alterations. For this reason, a prompt diagnosis is necessary to limit the progression of this highly destructive disease. In addition, the fact of presenting at an early age produces hard making decisions for the adequate treatment of the disease and reduces the adverse effects of the procedures.

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Authors' contributions
AKLL managed the patient and made a thorough search about the disease. JRPM analyzed the patient data regarding the spinal disease and was a major contributor in writing the manuscript. PRMF analyzed and interpreted the patient data regarding the spinal disease and realized the spinal surgery. VMPM and FFMT reviewed and made important corrections of the manuscript. JICB analyzed and interpreted the patient data of the infectious disease and made a thorough search about the disease. All authors read and approved the final manuscript.

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Availability of data and materials
All data is available for future research.

Declarations

Ethics approval and consent to participate
Ethics committee School of Medicine, Universidad Autonoma de Nuevo Leon, does not require ethical approval for reporting individual cases or case series.

Consent for publication
The parents agree with the publication of the case

Competing interests
The authors declare that they have no competing interests

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