

**CASE REPORT**


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## ISONIAZID-RESISTANT TUBERCULOUS SPONDYLODISCITIS ASSOCIATED WITH SCROFULODERMA

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**ABSTRACT**

The extrapulmonary forms of tuberculosis are responsible for about 20% of cases. Scrofuloderma is the cutaneous manifestation secondary to infection in some subcutaneous foci. A 33-year-old patient was admitted to the Clinical Hospital with exudative skin lesions on the back and thorax, initiated 10 months previously, associated with daily fever, and constipation. Spine resonance showed a paravertebral pseudotumoral lesion with T4 and T9 invasion, including vertebral canal and sub-ligament extension. The lesions presented fistulas for paravertebral muscles, lung and skin. Polymerase chain reaction (PCR) proved positive for *Mycobacterium tuberculosis* in the thorax wound secretion, characterizing tuberculous spondylodiscitis with scrofuloderma. Treatment was initiated with rifampicin, isoniazid, pyrazinamide and ethambutol with important clinical improvement after the first week. The febrile peaks came to an end and there was improvement in the pattern of the cutaneous lesions. The susceptibility test showed resistance to isoniazid.

**KEY WORDS:** Extrapulmonary tuberculosis; tuberculous spondilodiscitis; scrofuloderma.

**INTRODUCTION**

Tuberculosis is one of the top ten causes of death in the world, with around 10 million new cases in 2017 and 1.3 million deaths in the same period (Anderson et al., 2018). The extrapulmonary forms of the infection account for about 20% of cases. Vertebral osteomyelitis caused by *Mycobacterium tuberculosis* is one of the most common osteoarticular manifestations of this infection (Berbari et al., 2015). Cutaneous forms are rare and may occur due to exogenous inoculation, with contiguous dissemination of some subcutaneous foci or hematogenic dissemination (Hill & Sanders, 2016). They account for

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about 1% to 1.5% of cases (Punia et al., 2015). The endogenous form, with contiguous spreading, formerly called scrofuloderma, may occur secondary to infection in lymph nodes, bones, joints or epididymis (Hill & Sanders, 2016). The lesions begin as subcutaneous mobile nodules, with posterior fistulization and drainage of secretion (Punia et al., 2015). The following report demonstrates a rare case in the literature of scrofuloderma secondary to bone tuberculosis. The patient gave informed consent.

## CASE REPORT

Patient F.N.F., 33 years old, coming from Planaltina-Goiás, was admitted with a history of cutaneous lesions detected 10 months previously, associated with fever and approximately 8kg weight loss in the period. The first lesion appeared in the left lumbar region, being characterized as a mass with phlogistic signs and drainage of purulent secretion after puncture performed under medical care. During a six-month period, the lesion maintained purulent secretion without improvement. Approximately one month after admission, three new lesions appeared on the back and left hemithorax, all with the same characteristics (Figures 1, 2 and 3).



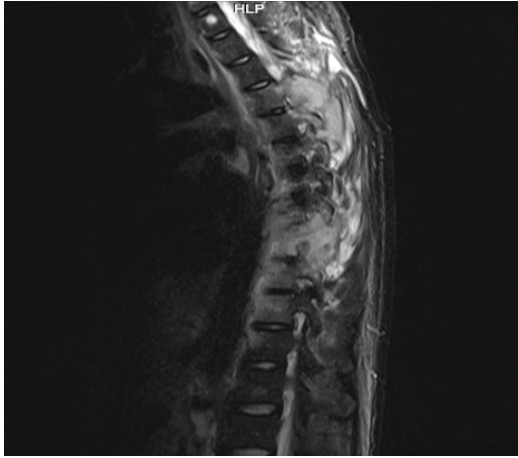
*Figure 1.* Phlogistic nodular lesions with central orifice and continuous drainage of purulent secretion.



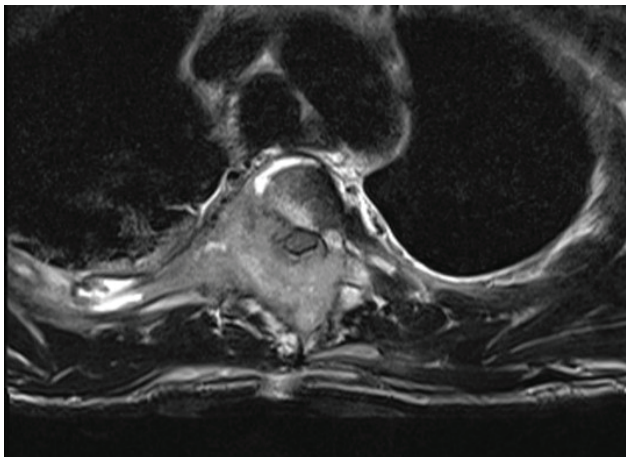
*Figures 2 and 3. Phlogistic nodular lesions with central orifice and continuous drainage of purulent secretion.*

Patient denied coughing during the period and reported contact with person with pulmonary tuberculosis about five years before. Physical examination: emaciated, eupneic; presence of 4 nodular lesions of approximately 2 cm, with central ulceration and spontaneous drainage of thick yellowish secretion. Serology for HIV, hepatitis B and hepatitis C were negative. Chest Computed Tomography (CT) revealed bilateral pleural effusion with atelectasis in the left base, as well as a small consolidation. Magnetic Resonance Imaging (MRI) of the spine showed an exuberant inflammatory process, with pseudotumoral lesions predominantly affecting the vertebral bodies and posterior elements of

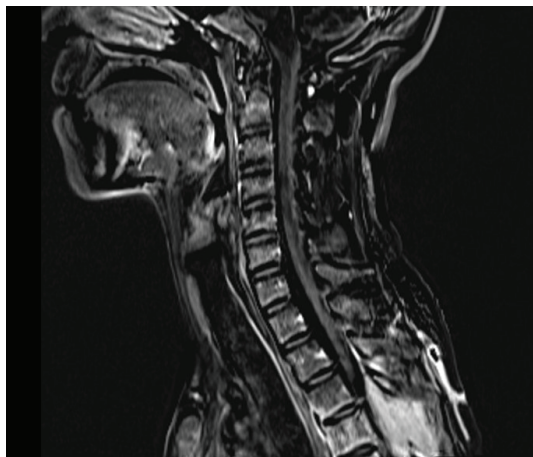
T4 and T9, with subligamentar and epidural extension, compromising supra and infra adjacent vertebral segments. This inflammatory process invaded paravertebral soft parts, including the regional and subcutaneous musculature, with fistulization at T2 level (Figures 4, 5 and 6).



*Figure 4.* Magnetic Resonance Imaging of the spine, sagittal cut, demonstrating paravertebral pseudotumoral lesions (arrows) with vertebral body invasion of T4 and T9 and sub-ligament extension. Contiguous spread to soft parts and skin.



*Figure 5.* Magnetic Resonance Imaging, axial cut, pseudotumoral lesion, with invasion of T4 vertebral body and medullary canal



*Figure 6.* Spine Magnetic Resonance Imaging, sagittal cut, demonstrating fistulization of skin pseudotumoral lesion, resulting in lesion of Image 1.

The alcohol-acid resistant bacillus was positive in the skin secretions, associated with positive PCR (GeneXpert®) for tuberculosis in the same material, confirming the diagnosis of tuberculosis. Direct examination and culture for bacteria and fungi were negative. Before diagnosis by PCR, a cutaneous lesion biopsy and a lymph node biopsy were performed. The histological examination showed chronic granulomatous inflammation with bacilloscopy and culture positive for *Mycobacterium tuberculosis*. After starting treatment with rifampicin, isoniazid, pyrazinamide and ethambutol (RIPE scheme), there was some clinical improvement, with remission of fever and reduction of drainage from the lesions. In the third month of treatment, using the drugs of the second phase of the tuberculosis scheme (rifampicin + isoniazid), there was worsening of the lesions and the exudation returned. The susceptibility test, performed by the proportion method in Löwenstein-Jensen medium, showed resistance to isoniazid, justifying the worsening of the condition, since the patient had only one active drug. In outpatient care, the patient reported crural paraparesis. He was hospitalized, but escaped, causing follow-up loss.

## DISCUSSION

Scrofuloderma most commonly occurs due to lymph node fistulization, especially in the cervical and axillary chain (Pinto et al., 2016; Saral et al., 2005) and children are more affected than adults (Punia et al., 2015; Hill & Sanders, 2016). In the case reported, the primary lesion was in the bone, with subsequent dissemination by contiguity to the lung, soft tissues and skin. About 50% of cases of bone tuberculosis occur in the spine, especially in the thoracic spine. The vast majority of cases are associated with paravertebral abscesses (Pinto et al., 2016). There are few cases of scrofuloderma resulting from fistulization of bone infection. None of the reported cases showed germ resistance (Ermertcan et al., 2011; Pinto et al., 2016; Punia et al., 2015; Saral et al., 2005). Polymerase Chain Reaction (PCR) for *Mycobacterium tuberculosis* allowed rapid diagnosis, however, the patient was only examined after arriving at a tertiary care unit. In this case report, we observed a primary isoniazid resistance which enhances the importance of cultures for susceptibility testing, since molecular testing does not always provide the standard of resistance to all tuberculostatics. Resistance is more frequently observed in patients previously treated with tuberculostatics (Anderson et al., 2018). However, in this case, resistance was detected even without any previous treatment for tuberculosis.

In this report we emphasize the importance of clinical suspicion regarding uncommon etiologies in cutaneous abscesses. The patient presented ten months of lesion evolution, an unusual period in cutaneous infections by nonspecific bacteria. The main differential diagnoses for lesions of this nature are: sporotrichosis, coccidioidomycosis, actinomycosis, chronic bacterial osteomyelitis, suppurative hidradenitis and conglobate acne (Frankel et al., 2009). The patient probably suffered vertebral pain for a long period before skin lesions appeared. If there had been an early diagnosis of the spinal injury, the dissemination of the lesion to the lung and skin might have been avoided; however, the patient had no complaints at this site, and the spinal lesions were identified after visualization of alterations during chest tomography, performed to evaluate the presence of pulmonary disease. It is important that cutaneous tuberculosis be included in the differential diagnosis of chronic skin conditions, recalling the importance of cultures to identify any resistance to drugs recommended for treatment, since WHO estimates that 3.5% of new tuberculosis cases are multi-resistant (Anderson et al., 2018).

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