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# Pott's disease and scrofuloderma as presentation of clinically extrapulmonary tuberculosis in 15 years old girl: a case report



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## ABSTRACT

**Backgrounds:** Extrapulmonary tuberculosis (TB) accounts for 15–20% of all tuberculosis cases, with the lymph node being the most commonly affected organ. Spinal tuberculosis and scrofuloderma are rare manifestations of extrapulmonary TB and constitute less than 5% of the cases. Establishing the diagnosis is a challenge. Eradication and prevention of sequelae complicate further management of the disease. This study aims to describe the clinical manifestation and the management of spondylitis tuberculosis and scrofuloderma in a child.

**Case presentation:** A 15-year-old girl came with a chief complaint of lower extremities weakness for a week following monthly chronic low back pain. She also reported a painless lesion on her back with no sign of acute inflammation. She also complained of intermittent fever with night sweats and lost 3 kilos of body weight in the last two months. From physical examination, we found a kyphotic posture and severe

malnutrition. Skin examination revealed an open wound on her right lower back, size 4x3 cm, with no sign of inflammation. The tuberculin skin test was positive with 15 mm induration. The spinal magnetic resonance imaging revealed destruction of the vertebral corpus thoracal segment, hyperintense sacrum, and right ileum bone with a cold abscess, suggesting spondylitis TB. A wound biopsy with hematoxylin-eosin stain concluded chronic granulomatous inflammation and showed multinucleated giant cell Langhans-type, suggestive of cutaneous tuberculosis. Microbiology examination (PCR GeneXpert) from wound biopsy and ziehl neelsen stain failed to detect *Mycobacterium tuberculosis*. The patient was diagnosed with Pott's disease and scrofuloderma. Anti-tuberculous therapy commenced, and the patient responded well after several months of treatment.

**Conclusions:** The diagnosis of extrapulmonary TB manifests as spinal tuberculosis, and scrofuloderma could be made from clinical manifestation and imaging.

**Keywords:** children, Pott's disease, scrofuloderma.

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## INTRODUCTION

Tuberculosis can infect any place in the body besides the lungs, such as bones, brain, skin, and other inner organs.<sup>1</sup> This condition is called extrapulmonary tuberculosis. Extrapulmonary tuberculosis constitutes 15–20% of all cases of tuberculosis, with the commonest site being the lymph node.<sup>1</sup> Spinal tuberculosis affected approximately only 1 to 5% of all cases of extrapulmonary TB.<sup>2</sup> It causes bone destruction, spinal deformity, and neural complications. Scrofuloderma is a rare condition which accounts for between 0.5 - 2 % of extrapulmonary tuberculosis.

It occurs due to direct involvement and breakdown of skin from contiguous tubercular foci-like infected lymph nodes or bone. The diagnosis is based on clinical, microbiological confirmation, and radiological evidence, particularly in the endemic areas of the world. The treatment targets are to confirm the diagnosis, eradicate the infection, achieve decompression of the spinal canal material, and correct or prevent spinal deformity and a possible sequel. This study aims to describe the clinical manifestation and the management of spondylitis tuberculosis and scrofuloderma in a child.

## CASE PRESENTATION

A 15-year-old girl came to our emergency room with the chief complaint of weakness in both her legs. Her leg became paraesthesia at first in the following 7 days. She also presents with low back pain since 2 months ago. At that time, she still could walk normally, but in the last 3 days, her legs became weak, and she could not walk anymore. This condition affected her routine because she had limited walking ability. The patient still had good movement and strength in both her arms.

The patient was also complaining about a painless lesion on her back for

about 2 months. In the beginning, the patient reported that she had a small mass on the back before the installation of the ulceration. There was no acute inflammation on the wound lesion. She also reported an intermittent fever with night sweats and a weight loss of 3 kg over 2 months. Further anamnesis revealed that her grandmother is a confirmed case of a pulmonary tuberculosis patient.

From physical examination, we found a kyphosis posture and severe malnutrition patient with a BMI of 10 kg/m<sup>2</sup>. *Bacillus Calmette-Guerin* (BCG) scar on the arm (+). A dermatological examination found an open wound on her right lower back that was as high as the lumbosacral level, size 4x3 cm, and there was no sign of inflammation (Figure 1). There were no palpable lymph nodes. Her lower extremity strength was 4/5 for the right leg and 3/5 for the left leg. Laboratory examination showed leucocyte 10.61 x10<sup>3</sup> u/L; neutrophil 89.49%; lymphocyte 9.7%; haemoglobin 13.68 g/dL; haematocrit 46.99%; platelet 336 x 10<sup>3</sup> u/L; sodium 140 mmol/L; potassium 4.44 mmol/L; chloride 101.1 mmol/L; and calcium 9.7 mg/dL. Serologic testing for human immunodeficiency virus (HIV) was negative. The tuberculin skin test was positive (15 mm induration). Chest X-ray AP/ Lateral showed a fibrotic line on the basal zone of the left lung, which was suspected of old inflammation (Figure 2). The spinal magnetic resonance imaging (MRI) revealed destruction of the vertebral corpus thoracic segment with cold abscess, hyperintense on T2W1 at right ileum bone and S1 with abscess para lumbar and para sacral region suggested spondylitis TB (Figure 3). Sputum gene Xpert was not available. Meanwhile, wound gene Xpert MTB did not detect *Mycobacterium tuberculosis*, but wound biopsy with hematoxylin-eosin stain concluded chronic granulomatous inflammation and showed multinucleated giant cell Langhans (Figure 4). The histopathological features were suggestive of cutaneous tuberculosis. Unfortunately, the ziehl neelsen stain failed to show acid-fast bacilli.

From these findings, we established a diagnosis of Pott's disease and scrofuloderma. Anti-tuberculosis therapy



Figure 1. Painless Wound lesion.



Figure 2. Chest X-ray showed fibrotic area on basal zone left lung.

was started, consisting of isoniazid (H), rifampicin (R), ethambutol (E), and pyrazinamide (Z). Even though the confirmed bacteriologic test was negative, the patient showed good responses after several months of anti-tuberculosis treatment.

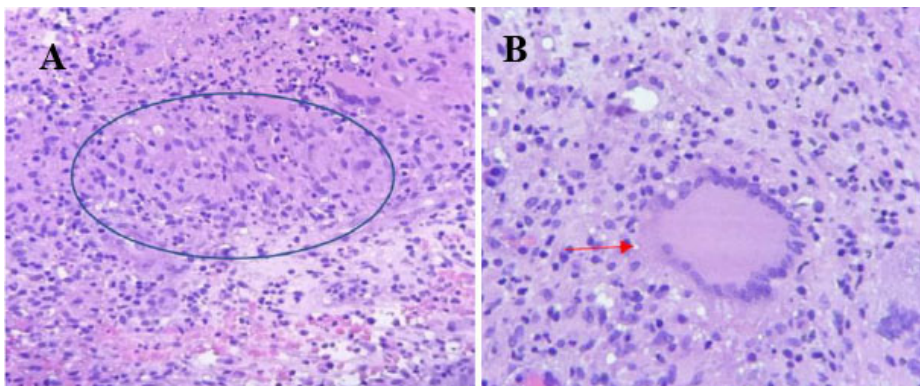
## DISCUSSION

This case report is a 15-year-old girl with Pott's disease and scrofuloderma. The accuracy of diagnosis and treatment are the most important prognostic factor of the patient. Tuberculosis is still one of the

main health problems and one of the top ten common causes of death in the world. Based on the World Health Organization (WHO) global TB report in 2020, the TB morbidity rate in the world is around 10.0 million people, and 1.2 million of them died. Moreover, WHO also stated that 70% of patients were new cases, and more than 85% of deaths were reported in Southeast Asia and Africa. The Sustainable Development Goals (SDGs) issued in 2015 include the End TB program with the goal of ending the TB epidemic in the world. This program has three main success indicators. There is a reduction in



**Figure 3.** (A) Magnetic Resonance Imaging (MRI) showed destruction of the adjacent thoracic vertebrae (white arrow) and (B) hyperintense of right ileum bone to sacral (white arrow).



**Figure 4.** (A) Chronic Granulomatous Infection and (B) Multinucleated Giant Cell Langhans (red arrow).

the incidence of TB in the world by 80% in 2030 compared to 2015, a reduction in mortality rate by 90% compared to 2015, and 0 (zero) costs that need to be incurred by TB patients in treating their disease. In Indonesia, TB is one of the national priorities for disease control programs because it has a wide impact on the quality of life and the economic burden. World Health Organization noted that Indonesia ranks second for the most TB incidences in the world after India.<sup>3,4</sup>

Tuberculosis in children cannot be separated from the TB control program because the number of children less than 15 years old is 40-50% of the population, and there are approximately 500,000

children in the world who suffer from TB every year. According to WHO 2020, tuberculosis in children less than 15 years accounted for 12% of all cases of TB. Moreover, some studies stated the highest prevalence of TB in children at ages 1- 4 years.<sup>3,5,6</sup> In our case, the patient was 15 years old girl. Although the highest prevalence of tuberculosis ranges from 1 to 4 years old, older children who live in endemic areas such as in our country are susceptible to infection.

Extrapulmonary tuberculosis is tuberculosis outside of the lungs that can be presented as tuberculosis meningitis, abdominal tuberculosis, skeletal tuberculosis, including Pott's

disease (spine), scrofuloderma (skin), and genitourinary tuberculosis. It is estimated to account for 15–25% of all cases of TB. In general, extrapulmonary tuberculosis is more difficult to diagnose than pulmonary TB and often requires invasive procedures to obtain tissue or fluid samples. Besides possible fever and weight loss, the symptoms and signs generally relate specifically to the affected organ system. Approximately 10% to 20% of extrapulmonary TB involves the bone structures. The common locations are the spine (spondylitis TB), followed by the hip joint (coxitis) and the knee joint (gonitis).<sup>7-9</sup>

*Mycobacterium tuberculosis* enters the human body through the respiratory or gastrointestinal system. Pott's disease is usually secondary to hematogenous spread from a primary site of infection or lymphatic spread. It usually occurs at thoracic or lumbar vertebrae. Spinal TB predilection might be due to good vascularization at the vertebral region. The spread of infection to the vertebral through arteries, venous, or percontinuum. An arterial arcade in the subchondral region of each vertebra is derived from anterior and posterior spinal arteries; this arcade forms a rich vascular plexus. This vascular plexus facilitates the hematogenous spread of the infection in the paradiscal regions.<sup>9-11</sup> In our cases, spinal TB was suspected from the hematogenous and lymphatic spread of primary site infection that might be from the lung, which showed a fibrotic line from the chest x-ray examination.

Leg weakness, limping, and difficulty standing are the most common complications of spinal tuberculosis. This is caused by the formation of debris, pus, and granulation tissue due to the destruction of bone and intervertebral disk. In the most severe form of Pott's disease, spinal deformation and collapse can compress the spinal cord, thus causing neurological disorders such as paresis and paralysis. The incidence of neurological deficit in spinal tuberculosis varies from 23 to 76%. The other clinical feature that can be found is back pain. Back pain is the most frequent symptom of spinal tuberculosis. The intensity of pain varies from constant mild dull aching to severe disabling. The pain may be worsened by spinal motion,



coughing, and weight-bearing because of advanced disk disruption and spinal instability, nerve root compression, or pathological fracture. In longstanding cases, there may be multiple levels of vertebral body collapse, resulting in a gibbous deformity. The other symptoms of spinal TB are classical constitutional features of tuberculosis, such as malaise, loss of weight and appetite, night sweats, evening rise in temperature, generalized body aches, and fatigue. Spondylitis TB in pediatrics usually presents with an insidious onset, slow course, and mild initial symptoms.<sup>8,9,12,13</sup>

In our case, the patient has weakness of the limb and low back pain as her complaint. She also caught intermittent fever, night sweats, and weight loss for several months, which indicates the classical features of tuberculosis infection.

The investigation of patients with spinal tuberculosis includes hematological examination, tuberculin test, microbiological examination, and radiological examination, including computed tomography (CT) scan and magnetic resonance imaging. Tuberculin test is positive in 63% to 90% of the patients with TB. It does not differentiate the active infections from the latent ones or induced reactions due to the vaccine BCG (*Bacillus Calmette-Guérin*). A positive Mantoux test will require detailed investigation, and a negative test cannot rule out the diagnosis of TB.<sup>14,15</sup>

The cytological and microbiology confirmation analysis of a vertebral lesion via neuroimaging guided-needle biopsy from the affected site is the gold standard technique for the early histopathological diagnosis of spinal tuberculosis. An open biopsy of the spine is usually performed when either closed techniques have proved insufficient or other procedures, such as decompression and possibly arthrodesis, are planned. Histologic studies confirm the diagnosis of spinal tuberculosis in approximately 60% of patients. The most common cytological findings observed are epithelioid cell granulomas (90%), granular necrotic background (83%), and lymphocytic infiltration (76%). Scattered multinucleated and Langhans giant cells may be seen in up to 56% of cases. False-negative results of biopsy

are common, and therefore, diagnosis of spinal tuberculosis must be made based on clinical manifestations and radiology when bacteriology proves negative.<sup>9,14</sup>

Chest X-rays should be done in patients with spinal tuberculosis to search for the primary infection that most commonly occurs in the lungs. Conventional radio imaging gives enough information to diagnose spinal TB, especially in areas with fewer sources, but it should be noticed that radiolucent lesions only appear on the X-ray, where 30% of the mineral density of the bone has been lost. The initial phase of the disease can cause the normal finding of plain imaging and does not exclude TB infection. Spinal Tuberculosis usually presents with osteopenia of the vertebrae, narrowing of the articulation segment, and loss of definition of the paradiscal margins of the vertebral bodies. The central type lesions generally present with destruction, ballooning, and concentric collapsing of the vertebral bodies. In the infection of the posterior elements, the destruction of the pedicles and lamina, the erosion of adjacent ribs, and the posterior cortical of the vertebral body can occur. The cold paravertebral abscesses are observed on simple X-rays as shades on the soft tissues adjacent to the column. In the case of longer-duration abscesses, the imaging shows concave erosions in the anterior margin of the vertebral bodies.<sup>14</sup>

The computed tomography (CT) scan model could be showing vertebral alterations earlier in time than the X-ray, giving the characteristics in a more detailed way regarding the osseous lesions, from the involvement of the posterior elements, the involvement of the craniovertebral junction and the cervicothoracic junction and the sacroiliac articulations. Four patterns of destruction can be observed in a CT: fragmentary, osteolytic, sclerotic, and subperiosteally. CT also allows the evaluation of the involvement of the soft tissues and paravertebral abscesses, being an excellent method to detect abscess calcifications and is also useful in the evaluation of the medullar compression by the inflammatory tissue or sequestrum.<sup>14</sup>

Magnetic resonance imaging (MRI) is a better sensitivity tool than plain radio imaging and is more specific than CT. The sensitivity and specificity described

for this method of image are 96% and 93%. MRI also allows for the rapid determination of the mechanism for neurologic involvement. It is useful for an earlier diagnosis than any other method of image. The use of contrast increases the precision of the MRI, particularly at early stages. Characteristic findings of spinal tuberculosis include destruction of two adjacent vertebral bodies in opposing end-plates with the vertebral disc relatively well preserved or the reaching of multiple vertebral bodies, edema of the vertebral body seen as hyperintense signal on T2 and STIR (short tau inversion recovery) images, extension of the infection beneath the longitudinal anterior ligament and the presence of pre-vertebral, para -vertebral, intraosseous or epidural abscesses that are generally smooth with thin walls. The subligamentous spread of a paraspinal mass and the involvement of multiple contiguous bones and intramedullary spinal changes can be very well demonstrated by MRI. MRI also presents high precision to differentiate granular tissues from cold abscesses and allows assessing in a detailed way the involved tissues, the anatomical localization of the abscesses, the involvement of the neurological structures, and the vertebral non-continuous disease.<sup>9,14,16</sup>

According to the 2013 revised World Health Organization (WHO) case definitions, spinal TB was classified as a bacteriologically confirmed case and or clinically diagnosed case. Confirmed cases were those patients for whom the presence of *M. tuberculosis* in a spine biopsy was demonstrated by a positive Xpert MTB/RIF test, positive culture for *M. tuberculosis*, or positive smear microscopy for acid-fast bacilli. Clinically diagnosed cases were those patients for whom spine imaging, clinical presentation, and, in some instances, confirmed TB from other sites indicated spinal TB.<sup>17</sup>

In our case, we did not do the spinal biopsy for the spinal lesion. The MRI showed destruction of the adjacent thoracic vertebrae, narrowing the intervertebral space, sacrum and ileum bone marrow edema, and cold abscess. Based on these findings, we diagnosed the patient as a clinical case of spinal tuberculosis.

The other extrapulmonary TB form

is Scrofuloderma. It is the most common form of cutaneous TB among children. Scrofuloderma arises due to the contiguous spread of an underlying tuberculous focus to the overlying skin. Lymph nodes are the most common underlying foci, and cervical lymph nodes are the most common group involved. Systemic TB foci are seen in up to 66% of cutaneous TB cases. Bones, joints, testes, breast, and lacrimal glands are the other underlying foci of infections leading to scrofuloderma. Clinically, scrofuloderma is characterized by asymptomatic subcutaneous swellings that persist for several months before softening and ulcerating to form discharging sinuses and ulcers. Typically, the ulcers are shallow with undermined and bluish edges.<sup>18</sup> In our case, the patient had a painless lesion at the skin overlays on the right ileum bones as the underlying focus of infection. The lesion began with a small subcutaneous mass before becoming a shallow ulcer.

Scrofuloderma histopathology includes chronic granulomatous inflammatory infiltrate associated with caseous necrosis, Langhans giant cells, and the detection of acid-fast bacilli. Tubercular epithelioid cell granulomas with lymphocytes and Langhans-type giant cells are the hallmarks of cutaneous TB. In response to the tuberculosis infection, the activated macrophages, cytokine interferon (IFN), and T cell activity produce a type IV reaction. This reaction, combined with ischemia, results in central caseation necrosis in the tuberculous granuloma. Demonstration of the *Mycobacterium tuberculosis* can be done with Ziehl-Neelsen staining but has low sensitivity (14%). This is due to tuberculosis infection in children, which is often in the form of paucibacillary, so examination smears of acid-fast bacilli often give negative results. Moreover, skin tissue is slightly favorable to the reproduction of the bacilli, unlike pulmonary tissue growth. Another modality for microbiological confirmation is culture and real-time polymerase chain reaction (RT PCR).<sup>18-21</sup>

In our case, the gene Xpert MTB/RIF of soft tissue specimens showed negative results. Histopathological examination from a wound biopsy using a hematoxylin-eosin stain concluded

chronic granulomatous inflammation and showed multinucleated giant cell Langhans, but the ziehl neelsen stain failed to find acid-fast bacilli.

Based on the WHO guidelines, tuberculosis should be treated with a multidrug regimen. The dosages of anti-TB medicines that should be used daily for the treatment of TB in children are Isoniazid (H) 10 mg/kg (range 7–15 mg/kg); maximum dose 300 mg/day, Rifampicin (R) 15 mg/kg (range 10–20 mg/kg); maximum dose 600 mg/day, Pyrazinamide (Z) 35 mg/kg (range 30–40 mg/kg), and Ethambutol (E) 20 mg/kg (range 15–25 mg/kg).<sup>22</sup>

Children with suspected or confirmed extrapulmonary TB, except meningitis TB and osteoarticular TB, should be treated with a four-drug regimen (HRZE) for 2 months, followed by a two-drug regimen (HR) for 4 months. Meanwhile, osteoarticular TB should be treated with a four-drug regimen (HRZE) for 2 months, followed by a two-drug regimen (HR) for 10 months, the total duration of treatment being 12 months. The wound treatment of scrofuloderma focuses on hygiene and topical ointment to prevent secondary infection. Other topical treatments include compressing with normal saline solution on the wound surface area.<sup>17</sup>

In our case, the patient was taking 12 months duration of treatment divided into a four-drug regimen (HRZE) for 2 months, followed by a two-drug regimen (HR) for 10 months. No major adverse event developed during the observation. The patient showed good responses after several months of anti-tuberculosis treatment. The skin lesion was treated by a plastic and reconstructive surgeon, who applied antimicrobial dressing and hygiene around the wound area. The limitation of this study is microbiology confirmation analysis from vertebral lesions as gold standard could not performed due to technical issues.

## CONCLUSION

We reported one case of spondylitis concomitant with scrofuloderma. Establishing the diagnosis is challenging due to the diversity of the symptoms, clinician experience, and difficulty in obtaining an adequate sample

for confirmation. It could be made from clinical manifestations such as paraesthesia, limping, painless chronic wounds, investigation of radiology such as destruction vertebral body, and appearance of multinucleated giant cells Langhans of wound biopsy when the proof of bacteriologic confirmation was not found.

## CONFLICT OF INTEREST

There is no competing interest regarding the manuscript.

## FUNDING

None.

## ETHICS CONSIDERATION

Ethics approval has been obtained from the Ethics Committee, Faculty of Medicine, Universitas Udayana, Prof. dr. I.G.N.G Ngoerah Hospital, Bali, Indonesia, before the study was conducted.

## AUTHOR CONTRIBUTIONS

All authors equally contribute to the study from the conceptual framework, data acquisition, and data analysis until the study results are reported through publication.

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